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EDITED BY

LARAINE MASTERS GLIDDEN

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Laraine Masters Glidden

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Preface

Volume 32 follows two thematic volumes, number 30 on Neurotoxicity and number 31 on Personality and Motivation. Although Volume 32 is eclectic, several of the chapters share common themes such as etiology of intellectual disability and cognitive characteristics of persons with intellectual disability. These themes are prominent in the field of mental retardation and developmental disabilities more generally. In addition, two chapters, the first and second, have special significance. The lead chapter by Leonard Abbeduto, Yolanda Keller-Bell, Erica Richmond, and Melissa Murphy on language development and mental retardation is the second of two being featured as an outgrowth of the 2003 theme of the Gatlinburg Conference on Research and Theory in Intellectual and Developmental Disabilities. In that year, keynote speakers were asked to assume a historical approach to understanding the development and trajectory of different research domains by addressing the past, present, and future of research in their respective areas of specialization within the developmental disabilities field. The first of these two chapters was published in Volume 29, on behavioral phenotypes by Bob Hodapp.

Len Abbeduto and his coauthors do a masterful job in tracing approximately 40 years of research on language development in persons with mental retardation, using a general approach as well as an etiologys-pecific emphasis. They review theoretical positions that were dominant at different times during these 40 years, and draw conclusions about the viability of different theories given the evidence that has been amassed. They communicate the vibrancy of this research area, the substantial progress that has been made, and they outline important directions for its future.

The chapter on residential services research by Steve Holburn and John Jacobson also has a special significance. It includes almost two decades of research that was largely unpublished because it was conducted as part of a

statewide evaluation program and was described and analyzed primarily in technical reports. It was a major corpus of work by the authors who were writing this chapter when John Jacobson was diagnosed with lung cancer. He died just a few months later. John and Steve Holburn worked frenetically and conscientiously to finish this chapter before John's death, and Steve made all the subsequent revisions. All of us in the field who knew John and respected his work see this chapter as part of the invaluable legacy that he left. Thank you, John.

The next two chapters, one by Eric Emerson, Hilary Graham, and Chris Hatton, the other by Laura Glynn and Curt Sandman consider correlates and possible etiology of intellectual disability. In their focus on poverty and socioeconomic status, Emerson et al. point to both neglect and confusion about the treatment of variables relating to these critical life circumstances. They are critical because they are both correlates of mental retardation and factors that influence the lives of persons with mental retardation. This neglect is a somewhat recent phenomenon, driven by both the intense interest in genetic causes of mental retardation as well as the political desire to dissociate disability and class. Nonetheless, it does not help either science or persons with mental retardation to ignore the lives of poverty that many live, and to ignore, as well, the adverse consequences of that poverty.

Laura Glynn and Curt Sandman review the most recent research on low birth weight and developmental disability and conclude that at least some of the adverse consequences are permanent and not simply reflections of a delayed developmental trajectory. They then link the low birth weight to prenatal stress, a relatively understudied connection especially given the influence that prenatal stress might have on the developing structure and function of the brain. Although Glynn and Sandman do not directly address poverty in their chapter, certainly environmental stressors associated with very low socioeconomic status might be part of a conceptual model that would entail the study of prenatal stress.

Clancy Blair and Megan Patrick in one chapter, and Natalie Sinn and Carlene Wilson in another, both focus on cognitive functioning but from quite different perspectives. Blair et al. provide a convincing argument that fluid cognition must become more central to understanding the deficits of persons with mental retardation. Fluid cognition includes those aspects of intelligence that involve executive function and working memory and that are largely responsible for performance in tasks such as abstraction, relational reasoning, and planning and problem solving. More importantly, they then link individual differences in fluid cognition to differential experience of stress and emotion as studied within a developmental neuroscience framework. They also propose that these foundational variables may

explain the adaptive behavior deficits that are a central defining feature of mental retardation.

Sinn and Wilson are also concerned with cognitive development and with a developmental neuroscience approach to studying it and to studying attention deficits. In particular, they argue that problems in attention and learning, characteristics of distractibility, lack of impulse control, and maladaptive social behavior may result from difficulties in neurotransmitter production and use. Although they recognize that the evidence is still quite sparse, they suggest that dietary interventions with highly unsaturated omega-3 fatty acids may correct the underlying neurotransmitter problems and therefore alleviate some of the behavioral difficulties. Clearly, more well-done science is needed in this area.

The last several decades have seen an explosion of research and writing about autism. Between 1975 and 2005, the number of references to autism in the PSYCINFO database has increased by approximately 1600. I recognized this upsurge by editing a thematic volume on autism, published in 2001 as Volume 23 of the *International Review of Research in Mental Retardation.* In that volume, we addressed the issue of diagnosis. However, Kylie Gray, Bruce Tonge and Avril Brereton bring us up-to-date on screening for autism, reviewing many of the more recent studies including their own work in this area. They provide very useful summaries of different screening instruments and their efficacies, and I have no doubt that their chapter will be frequently consulted by both novice and experienced diagnosticians and practitioners.

Chapters "People with mental retardation and psychopathology: Stress, affect regulation and attachment: A review" and "Diagnosis of depression in people with developmental disabilities: Progress and problems" both focus on behavioral and psychological problems of persons with mental retardation. Carlo Schuengel and Cees Janssen, in their chapter, propose that many of the challenging behaviors that are exhibited by children and adults with mild mental retardation are the result of failures in the coping and adaptation to life stresses. They hypothesize that these failed adaptations may result from cognitive deficits, affect dysregulation, and difficulties stemming from the absence or lack of support from attachment figures. They review empirical evidence on attachment, especially with children with Down syndrome, and conclude that there is some support for their hypothesis.

Ann Poindexter, in her chapter, first convinces us that the incidence and prevalence of depression is higher in persons with mental retardation than in intellectually typical individuals, and then describes and summarizes the issues with regard to assessment and to treatment. She concludes that much more research on treatment outcomes is necessary in order for psychiatric practice to be evidence based.

Finally, as always, this volume would not have been possible without an international array of individuals who were willing to lend their expertise and review and comment on manuscripts. The following persons, listed alphabetically, all contributed extensive advice and guidance that helped make the chapters reviewed for this volume as good as they are: Arnold Birenbaum, John Borkowski, Margaret Burchinal, Doug Detterman, Anton Dosen, Elisabeth Dykens, Glenn Fujiura, Mike Guralnick, Linda Gottfredson, Maureen Hack, Linda Hickson, Ann Kaiser, Connie Kasari, Craig Mason, James McCracken, Bill MacLean, Ted Nettelbeck, Johannes Rojahn, MaryAnn Romski, Lynn Ward, and Marilyn Yeargin-Allsopp. Thank you, all.

LARAINE MASTERS GLIDDEN

Research on Language Development and Mental Retardation: History, Theories, Findings, and Future Directions*

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In this chapter, we briefly review some of the theoretical shifts and most important findings to emerge in the study of language development in individuals with mental retardation since the inception of the Gatlinburg Conference on Mental Retardation Research and Theory in 1968. This period corresponds to an especially vibrant era of research on language and mental retardation, and one that has been shaped in profound ways by the nativist theory espoused first by Noam Chomsky (1965) and later by Fodor (1983), Pinker (1996), and others. Research on language development among those

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with mental retardation has not only been shaped by nativist theory but this research has also yielded data that have proven important for evaluating the merits of the theory, as in the case of research on Williams syndrome (Bellugi, Lai, & Wang, 1997). In our review, we have by necessity been selective, focusing largely on those concerns and findings that have been motivated by, or are relevant to, the claims of nativist theory or the alternative theories that have risen in prominence in recent years. The alternatives we consider are the social-interactionist (Bruner, 1983) and emergentist (MacWhinney, 1999) approaches, which grew out of work in typical child development and cognitive science, respectively, and the genetic syndromes approach (Hodapp & Dykens, 1994), which emerged from work within the field of mental retardation. Our goal is to provide a brief history of research on language and mental retardation, demonstrate the bidirectional relationships between that research and the dominant theoretical approaches of our time, and outline future directions for research.

I. THEORETICAL APPROACHES TO LANGUAGE DEVELOPMENT

A. Nativist Approach

The nativist approach was first introduced in the 1960s (Chomsky, 1965). According to the nativists, there is something "special" about language that reflects the unique character of the processes by which it is learned. Moreover, this uniqueness is thought to be biologically based, arising from the innately given organization and structure of the human mind (Chomsky, 1988). Although there are different variants of the nativist approach (Chomsky, 1975, 1988; Fodor, 1983; Pinker, 1996), all include the same four key claims:

1. The human brain is well designed to learn language and every child is born with the capacity to learn a language (i.e., there is species universality). This capacity consists of a tacit, or implicit, knowledge of the properties common to all languages and of the constraints on the ways in which languages can differ (i.e., the so-called universal grammar). This "advance" knowledge leads the child to generate only a limited number of tightly constrained hypotheses about the input language rather than relying on, for example, trial and error.
2. The capacity to learn language operates in modular fashion in that this capacity involves mechanisms that are especially tuned to processing linguistic representations and require little if any input from more general cognitive processes or other mental functions. It is even proposed that there is modularity of one component of language from the next (e.g., syntactic rules are

thought to be unconstrained by the meanings represented in the semantic component of the language).

3. The child needs to encounter only a limited number of key examples in the input language to arrive at the necessary language-specific categories and rules. These key examples represent very basic "facts" about language and are likely to be available in virtually all environments. Thus, normal variations in children's environments are unlikely to have much of an impact on language development.

4. The "goal" of language learning is mastery of an internalized set of abstract, deterministic, and context-free rules that define the elements of language and govern their combination into higher-order units. Although the knowledge represented in these rules is the basis of speaking and listening, it is represented only imperfectly in such performances because of the "distorting" influences of various peripheral constraints such as those on memory and attention (i.e., the rules represent *competence*, which is only one of several determinants of *performance*). Nativists are interested largely in competence and see performance factors as trivial.

More detailed analyses of the assumptions of the nativist approach can be found in Abbeduto, Evans, and Dolan (2001); Abbeduto and McDuffie (in press); Chapman et al. (1992); Karmiloff and Karmiloff-Smith (2001); Rondal (2003); and Rosenberg and Abbeduto (1993).

Many of the nativist claims are controversial and have prompted the formulation of alternative theories. In addition, although the nativist approach outlines in broad strokes the mechanisms that shape language development, it is surprisingly devoid of details about how language development occurs in real time (Karmiloff & Karmiloff-Smith, 2001). In other words, the steps by which the child moves from being a nonspeaking being with the potential to acquire language to possessor of the end-state grammar is not specified by the nativist account in part because of "simplifying" assumptions that make the problem of language learning logically possible (e.g., the assumption of simultaneous access to key exemplars in the input language and the instantaneous acquisition of the end-state grammar). This lack of detail has also prompted the articulation of alternative theoretical accounts.

B. Alternatives to the Nativist Approach

1. SOCIAL-INTERACTIONIST APPROACH

In the mid- to late-1970s, researchers began to question the nativist claims about the environment's contributions to language learning by documenting differences in the quality of talk that adults direct to children (Snow, 1977). From these beginnings, the social-interactionist approach emerged.

This approach emphasizes that the social uses and contexts of language are important for development and are a source of difference among children as regards their trajectories and outcomes. Although not necessarily inconsistent with the nativist claim that children are biologically prepared to learn language, the approach is distinguished in all its variants by the claim that environmental variations are important in language development.

Bruner (1983) was among the earliest and most influential architects of the social-interactionist approach. Bruner suggested that a crucial component of the language acquisition process is the *format*—a patterned, routinized, and familiar interaction that enables the child and his or her adult care provider to communicate with each other successfully. The linguistic exchanges that take place in the format constitute the input from which the child learns grammar, how to refer and mean, and how to realize his or her intentions in language. The format also provides a system that frames, or structures, the input of language and interaction, making both more comprehensible to the child and thereby helping him or her to learn language. Especially important is the notion, that within the format, the care provider scaffolds the child's participation, helping the child to participate at a level appropriate to his or her current skills, gradually decreasing the amount of scaffolding as the child is capable of more independent participation. Bruner referred to the system of formats that parents create for their children as a Language Acquisition Support System.

Bruner illustrates these claims in a longitudinal study focused on a mother–infant pair in the process of acquiring standard lexical labels from the 8th to the 18th month of the infant's life (Ninio & Bruner, 1978). The most striking characteristic of this labeling activity was that it took place in structured interactional sequences that had the texture of dialogue such as book reading. During these activities, the mother and child would frequently engage in a standard three-step routine (the attentional vocative, the query, and the label). For the majority of the routines, the child would play an active role in the routine, often prompting the mother to initiate a new turn and to engage in more labeling activity. This research demonstrated that a crucial element in the achievement of reference by the child is his or her mastery of the reciprocal dialogue rules and conventions that govern the format. The child acquires these rules from the mother, who scaffolds the dialogic exchange so that the child moves from prelinguistic referential activity to mastery of verbal reference (Franklin & Barten, 1988).

Another pioneer of the social-interactionist approach was Snow (1977), who suggested that adult caregivers tailored their language to the changing capabilities of the children with whom they interacted, with this special register coming to be known as *child-directed language* (CDL). In a now classic study of maternal talk to infants, Snow (1977) demonstrated that

maternal talk was typically designed to elicit responses, suggesting that mothers believed that their infants were capable of reciprocal communication. Moreover, mothers would fill in responses for the babies in the conversation, taking the baby's turn. By 7 months, when the babies had become more active partners in these exchanges, initiating turns and participating in language games, mothers seemed to "require" more sophisticated, and clearly intentional, responses than they had when their infants were younger.

More generally, research on CDL has documented that maternal speech to infants and young children, relative to speech to other adults, is characterized by simpler syntactic constructions, more concrete and less diverse vocabulary, less propositional complexity, and greater fluency (Snow, 1999). Moreover, the attention patterns of the infant as well as his or her language ability appear to guide the complexity of the adult's language and interaction (Snow, 1977). Many of these characteristics are also displayed by older children speaking to younger ones and by nonparent adults (Shatz & Gelman, 1973). In short, CDL appears to be "designed" to enable the child to control the flow of the interaction and simplifies the processing demands posed by the language in accordance with the child's level of competence.

Some social-interactionists have argued that if participation in social interaction is important for language learning, then the child's emerging social-cognitive system must constrain what is learnable at any particular point in development. For example, many researchers have argued that the infant's emerging understanding that other people are intentional agents is a precursor to lexical development (Baldwin & Tomasello, 1998; Tomasello, 1995, 1999; Tomasello, Kruger, & Ratner, 1993). These researchers argue that this understanding allows the infant to achieve joint attention with persons in his or her environment by pointing and using other gestures that emerge before the first word (Bates, Camaioni, & Volterra, 1975) and to be motivated to participate in context-embedded language games or routines (Bruner, 1983; Ninio & Bruner, 1978; Ninio & Snow, 1996).

The Child Talk model proposed by Chapman et al. (1992) further distances the social-interactionist approach from the nativist perspective. Chapman et al. argue that the nativist approach incorrectly characterizes the fundamental nature of the language-learning process by assuming that its purpose is the mastery of linguistic rules. Instead, Chapman et al. propose that children are not acquiring a set of abstract, context-free rules but rather are learning how to participate in real-world social contexts. Moreover, the linguistic knowledge that children acquire is thought to be procedural in nature, inextricably tied to the social contexts of its use, and is no different from any of the other cultural practices that must be learned in order to participate in the social world. Finally, children are thought to draw on the

full range of their linguistic and nonlinguistic capabilities to acquire these practices.

In summary, four claims emerge from the various frameworks falling within the social-interactionist approach and distinguish it from nativism:

1. Language development is motivated by the child's desire for, and attempts to acquire, a means of participation in social interaction with familiar people. Language is thus embedded within the broader context of the acquisition of cultural practices and is just one among many such practices to be learned.
2. The interactions and relationships that the child has with other, competent, language users, especially parents or other linguistically skilled care providers, are critical for determining the rate and course of language development. In the ideal case, care providers scaffold the child's learning, providing opportunities to acquire and practice more advanced behaviors for speaking and listening with appropriate contextual support.
3. There is considerable variation among children as regards their environments. Not all care providers will be as inclined to, or as adept at, scaffolding the child's language learning. This suggests (given the first two claims) that wide variations in language learning are possible and result, in part, from environmental causes.
4. Because the process does not depend on language-specific mechanisms, it can be influenced by numerous dimensions of the child's psychological and behavioral profile, including auditory memory skills, attentional capabilities, knowledge of the social and physical worlds, and the presence of various forms of psychopathology (e.g., anxiety). Impairments or psychopathologies in the social realm should be especially deleterious in light of the need to process social-contextual information.

2. EMERGENTIST APPROACH

Emergentism owes its origins, in part, to the connectionist, or neural network, models of Elman (Elman, 1995, 1999; Elman, Bates, Johnson & Karmiloff-Smith, 1996), Seidenberg (Seidenberg, 1997; Seidenberg & MacDonald, 1999), and others. Emergentism falls somewhere between the nativist and social-interactionist approaches on the theoretical spectrum, incorporating aspects of both approaches in its account of language development. Thus, emergentists, like nativists, assume that children are well prepared by virtue of the design of their brains to learn language. Like the social-interactionists, however, emergentists ascribe an important role to the environment with the pattern and sequence of environmental inputs

to the child determining the nature of what is learned. In general, the emergentist approach is distinguished by three additional claims:

1. This preparedness to learn language is a property of the child's domain-general cognitive system. Thus, language learning depends on the same mechanisms as does the learning of other types of skills; there is no language-specific learning module.
2. Learning is a process of determining which properties of language regularly co-occur in the input language.
3. The ways in which language is represented are intimately connected to, and shaped by, the human input and output systems.

More technically, the mind is conceptualized by emergentists as a network of nodes and associations among them (much like the physical brain is a system of neurons and connections among them). Thus, language learning is a process of forming associations of varying strengths among nodes in the network, with each node representing a feature in the input language and with a link to output (comprehension or production) nodes. Associations among nodes and their strengths are determined by the frequency of co-occurrence of the language features they encode in the language that the child hears. Moreover, the system is dynamic, encoding its history of encounters with the environment to that point in time. In the emergentist approach, therefore, cognitive variations, environmental variations, and the constraints inherent in the output (or performance) systems all shape the nature and course of language development.

The emergentist approach and the types of data on which it relies are exemplified in investigations demonstrating that young children track statistical co-occurrences in the input language and use multiples sources of correlated information to identify language-relevant units. For example, Saffran, Aslin, and Newport (1996) demonstrated that 8-month olds can develop expectations for words based on the statistical relationships contained in sequences of previously heard sounds. Saffran et al. found that infants were able to track the transitional probabilities from one sound to the next, which are typically high within words and lower between words, to identify word boundaries. In a similar vein, studies of language comprehension have demonstrated that toddlers and young children are sensitive to different sources of information in the linguistic input (e.g., prosody, syntactic position, and context) and have the ability to abstract higher-order units of language structure, such as words and phrases, and discover the relations embedded within the grammar (Bates, Bretherton, & Snyder, 1988; Hirsh-Pasek & Golinkoff, 1996). In fact, toddlers as young as 16 months have been

found to conduct semantic-distributional analyses and to use word order cues to comprehend speech before they have the ability to produce this speech (Hirsh-Pasek & Golinkoff, 1996). And finally, research on children with specific language impairment (SLI) has shown that problems in using grammatical morphology are related to the frequency with which the words to be marked occur in the input (Marchman, Wulfeck, & Ellis Weismer, 1999) and to the processing demands associated with the discourse context of those words (Bishop, 1994; Evans, 1996; Evans & Craig, 1992), and are not due to an underlying grammatical deficit (Evans, 2001).

3. GENETIC SYNDROMES APPROACH

Language has long assumed a central role in behavioral research on mental retardation, at least in part because verbal skills are represented in most definitions and measures of intelligence and adaptive behavior (Abbeduto, 1991). However, research on language in mental retardation has historically not been especially concerned with exploring possible differences in language development or responsiveness to language intervention across different etiologies. There are many reasons for this state of affairs, not the least of which being that in the past, etiology could not be determined for most instances of mental retardation. The popularity of nativist theories, with their claim of species universality, however, also discouraged investigations of etiological differences in language development among individuals with mental retardation (Abbeduto et al., 2001).

Advances in molecular genetics and related disciplines have now made it possible to identify the causes of many more cases of mental retardation (Brodsky & Lombroso, 1998). In fact, more than 1000 genetic causes of mental retardation have now been identified (Dykens, Hodapp, & Finucane, 2000), with the number growing annually. In the field of behavioral research on mental retardation, this has led to an increasing interest in understanding behavioral (including linguistic) variability among persons with mental retardation in terms of etiology (Hodapp & Dykens, 1994, 2001). Such research often has as its goal the specification of the *behavioral phenotype*, or profile of strengths and weaknesses, which characterizes individuals with the syndrome and distinguishes them from people whose mental retardation has a different etiology. Elisabeth Dykens and Robert Hodapp have been among the most influential and articulate proponents of a syndrome-based approach for research on developmental disabilities (Hodapp & Dykens, 1994, 2001). They have offered the useful caution that it is unlikely that any given aspect of a behavioral profile will be unique to the syndrome or shared by all individuals with the syndrome (Dykens, 1995, 1999). Instead, these

theorists argues for conceptualizing a behavioral phenotype as a probabilistic profile; that is, a profile that is more likely to be evidenced by those with the syndrome than without it, but not necessarily universal among, those with the syndrome.

Although the syndrome-based approach emphasizes the genetic bases of language (and other dimensions of behavior), it also ascribes an important role to the environment, with the assumption that the behavioral phenotype of an individual reflects the contributions of genes, environment, and their interaction on human development (Hodapp & Dykens, 1994, 2001). Despite sharing the biological emphasis of the nativist approach, the genetic syndromes approach, at least as espoused by Dykens and Hodapp, is distinguished by the following three claims:

1. Genetic differences can lead to different developmental trajectories of language and different degrees of impairment in language relative to other psychological and behavioral domains (Abbeduto et al., 2001).
2. Language development is shaped by impairments in cognition and other domains of functioning. The development of language (or any other dimension of behavior) can be understood only in relation to the other dimensions of functioning that define the syndrome's behavioral phenotype (Murphy & Abbeduto, 2003).
3. The characteristics and behaviors of any individual with mental retardation affect, and are affected by, his or her environments (Hodapp, 1997; Murphy & Abbeduto, 2005).

In syndrome-based research on language, therefore, genetic variations, cognitive and behavioral variations, and environmental variations all matter for the development of language and communication. Moreover, linguistic performance is thought to mediate some environmental effects on the individual's subsequent language development (Murphy & Abbeduto, 2005).

II. NATIVIST ASSUMPTIONS AND RESEARCH ON MENTAL RETARDATION

The debates engendered by the various claims of the nativist approach have shaped research on language development and mental retardation in profound ways in the past and continue to do so today. At the same time, however, data on language development in individuals with mental retardation have often taken center stage in those debates because these data

provide tests of many of the nativist claims. In this section, we outline four research questions that have emerged from or are relevant to the nativist debates and illustrate the types of research and findings each question has provoked.

The claim that there is a species-universal capacity to learn language that is manifested in a tightly controlled and highly constrained acquisition process leads to the prediction that language learning should occur in much the same way for all children (e.g., in the same sequence of steps or achievements). This does not mean that all children will learn language at the same rate. Variations in systems that are "external" to the language-learning module but that underlie linguistic performance (e.g., the perceptual system involved in hearing) can lead children to traverse the same steps at different rates. In the case of children with mental retardation, language development may occur more slowly and may end prematurely (i.e., without acquisition of the typical end-state grammar), however, it should not be different from that of typically developing children in any fundamental way. In other words, quantitative but not qualitative differences among populations of children are possible. These predictions have led to numerous studies addressing the question:

1. Is language development delayed or different (deviant) in individuals with mental retardation?

The claim that the innate capacity for language operates in modular fashion, independently of other cognitive processes, leads to the prediction that dissociations between language and cognition are possible, perhaps even commonplace. Such dissociations would be manifested as asynchronous development across language and nonlanguage domains, with more rapid development in one domain than in the other. This does not mean that there will be no correlation between measures of linguistic and cognitive growth because any measure of language is subject to the influence of various performance factors, which are likely to reflect the contribution of cognitive abilities. Nevertheless, the nativist position predicts less synchrony between linguistic and nonlinguistic abilities than any other theoretical approach to language learning. In fact, the most interesting possibility as regards the asynchrony of these two domains—and the one that has proven key to the tenability of the nativist position—is the possibility of intact language abilities in the face of substantially impaired cognitive functioning. This is why the early accounts of spared language in individuals with Williams syndrome (Bellugi, Bihrle, Jernigan, Trauner, & Doherty, 1990) were so exciting (and controversial) when they were first reported. In short, researchers in the field of mental retardation have been keenly interested in the question:

2. What is the relation between language and cognitive development in individuals with mental retardation?

The claim that the child needs very little support for language learning from his or her environment has, not surprisingly, been arguably the most controversial of the nativist claims. This claim leads to the prediction that variations in language learners' environment will have little consequence for the development of language. In the field of mental retardation, questions about the role of the environment have focused largely on whether (i) the "natural" environments of (e.g., parent behaviors toward) individuals with mental retardation are less than optimal and thereby contribute in some way to their language-learning difficulties and (ii) significant alterations of their environments (i.e., intervention or therapy) can ameliorate some of their language-learning difficulties. In other words, the question that has been asked is:

3. Do differences in the environments of individuals with mental retardation contribute to differences in their language outcomes?

The nativist claim that the child is learning a set of linguistic rules that are abstracted away from their context of use and whose character is not shaped by the performance systems of real-time speaking and listening determines how language development is studied. In particular, it leads one to develop an abstract description of what individuals with mental retardation "know" about language rather than a description of how that knowledge is shaped by or interacts with performance constraints and the contexts in which speaking and listening occur. It, like the modularity claim, also leads to the study of language in isolation from other dimensions of the phenotype because the influence of factors, such as social impairment or psychopathology, is seen as rather trivial. In contrast, both the social-interactionist and emergentist approach argue that the context and the various psychological and behavioral systems that define the child at any point in time are critical determinants of what and how he or she learns. In the field of mental retardation research, investigators have thus asked:

4. Do performance factors affect the language learning of individuals with mental retardation?

A. Is Language Development Delayed or Different?

In general, studies focused on the delay or difference question in mental retardation have been of two types. First, some studies have been focused on what rules and categories are acquired or constructed by the individual with mental retardation at various points in development. Early studies of this type involved writing grammars (or parts of grammars) to describe the rules and categories that seemed to be expressed in the utterances produced by individuals with mental retardation and then asking whether those grammars corresponded to those of younger typically developing children.

If there was such correspondence, then the conclusion was that language development was delayed. In contrast, if no comparable grammar could be identified among typically developing children of any age, the conclusion would be that language development in individuals with mental retardation was different. In essence, studies of this first type focus on the products rather the process of language learning.

An example of a products-oriented study is that of Lackner (1968), who constructed grammars to describe the rules and categories that appeared to underlie the linguistic competence of children with mental retardation. The data on which the grammars were based consisted of expressive language samples from the children, along with some refinements in the grammars based on additional probing with tests of comprehension and imitation. Lackner then derived tests designed to elicit the knowledge represented in these grammars and administered the tests to five younger typically developing children. Lackner used the responses of the typically developing children to determine whether the order of difficulty, or complexity, of the grammars accurately recapitulated the typical developmental sequence. Lackner found that the tests for the grammar for the child with mental retardation who had the lowest developmental level were passed by all five typically developing children, whereas the test for the grammar from the most capable participant with mental retardation was passed by only the oldest typically developing participant. A similar ordering across the participant groups was obtained for the intermediate grammars as well. The fact that the order of complexity of the grammars was the same for the children with mental retardation and the typically developing children led Lackner to conclude that development was delayed and not different in the domain of syntax for individuals with mental retardation.

The second type of delay-difference study is focused less on the products of development, or on what knowledge is acquired, and more on the processes by which such knowledge is acquired by individuals with mental retardation. These studies have typically been concerned with identifying the learning strategies used or the sequence in which particular milestones are reached by individuals with mental retardation in comparison to younger typically developing children. Again, the use of strategies or the identification of sequences not found in the intellectually typical population, regardless of age, is taken as evidence of difference, whereas correspondence with typical children is taken as evidence of delay.

An example of such a process-oriented study is provided by the work of Mervis et al. (Mervis, Robinson, Rowe, Becerra, & Klein-Tasman, 2003). They investigated the developmental sequencing of referential pointing and labeling. It has been consistently documented in the literature on typical development that children's first referential acts (i.e., attempts at focusing

another's attention on an object in the world) are made through gestures, with words being used to achieve reference only some months later. Presumably, this reflects the relative simplicity of the task of mapping the adult gesture of pointing onto the referential act compared to the adult's referential use of words. The latter is more transitory in terms of duration and more variable in form (i.e., a point is topographically consistent across different acts of reference), whereas the words spoken vary across referential acts in accordance with the object to which the speaker intends to refer.

In contrast, Mervis et al. found that 9 of the 10 children with Williams syndrome that they studied longitudinally used words referentially 6 months, on average, before they produced or understood referential pointing. Mervis et al. speculated that (1) there may be multiple developmental routes to particular linguistic achievements and (2) the route chosen may be determined by one's profile of strengths and weaknesses. In the case of Williams syndrome, the child's relative facility with words (or perhaps auditory information more generally) leads the task of sound-to-meaning mapping to be simpler than that of gesture-to-meaning mapping. In the Williams syndrome case, therefore, this facet of language development is not delayed but rather different relative to typical development (and even other etiologies of mental retardation).

In general, there is evidence of both delay and difference in the language development of individuals with mental retardation (Rosenberg & Abbeduto, 1993). On balance, however, the weight of the evidence to date supports a delay rather than a difference characterization of the language development of those with mental retardation (Abbeduto & Boudreau, 2004). It is important to note, however, that this may simply be a reflection of the fact that most studies have been designed to characterize the *products* of language development, with those products usually being defined in a rather gross manner such as Lackner's (1968) study in which grammars were judged to be passed or failed by children in all-or-none fashion. Such studies might obscure differences in the *processes* by which individuals with mental retardation learn those products (Karmiloff-Smith, Scerif, & Thomas, 2003). In general, however, studies of the processes of language learning have been rare in the field of mental retardation (Abbeduto & Boudreau, 2004; Abbeduto et al., 2001). This is unfortunate because different types of language interventions may be needed to impart the same products depending on the processes by which an individual acquires language.

B. Is Language Development Independent of Cognition?

In general, investigations of the dissociability of language and cognitive development in mental retardation have taken one of two approaches. First, some studies have attempted to look at the relative levels or rates of

achievement in the two domains asking whether those in one domain outpace those in the other. This may be done through clinically identified cases of apparently dramatic dissociations of language and cognition or through group comparison studies that search for evidence of differences in language development for groups of participants (e.g., mental retardation versus typically developing) matched on one or more measures of nonlinguistic cognitive ability (e.g., mental age [MA]). Second, some studies have been designed to determine whether variation in nonlinguistic cognitive ability explains variation in rate of development of, or level of achievement in, language among individuals with mental retardation. Such studies may entail examination of the relative timing of achievements in language and cognition so as to determine whether one is a prerequisite for the other or whether they have a common underlying cause. Alternatively, studies of this second type may take a regression approach to determine whether variance in language outcomes is explained by concurrently or previously measured nonlinguistic cognitive ability or level.

There have been a half-dozen or so well-documented case studies of what appears to be relative sparing of language compared to cognition among individuals with mental retardation. Several of these have been compiled and summarized by Curtiss (1988). One case for which there are particularly extensive data is that of Antony, who was 6 years of age when he was studied. His nonverbal IQ was determined to be 51 and a battery of Piagetian tasks generally yielded a level of performance consistent with that of a typically developing 2-year old. Antony also displayed substantial limitations in many facets of language, including limited vocabulary (e.g., a PPVT age-equivalent score of 2.8 years and frequent lexical substitutions in conversation) and pragmatic skills (e.g., frequently and inappropriately switched topics in conversation and his speaking turn was often a simple repetition or echolalia response). Nevertheless, Antony appeared to have command of fairly sophisticated syntactic rules and forms in his expressive language. For example, he consistently and correctly marked words for past tense, number, and the like; produced phrasal and clausal patterns, such as passives and relative clauses, which are among the latest acquired by typically developing children (Rosenberg & Abbeduto, 1987); used a range of auxiliary verbs; and consistently followed subject-verb agreement rules. These syntactic features are illustrated among the following examples of Antony's utterances, which were drawn from the utterances recorded by Curtiss (1988):

- You're gonna get pushed.
- Jeni, what did you touch?
- That girl doesn't have shoes.

- He wants to chase the cat.
- Why don't you fly?
- I don't want Bonnie coming in here.
- A stick that we hit peoples with.
- I don't know who he gots.

In general, these utterances convey the sense that, from a syntactic perspective, Antony was reasonably sophisticated, which Curtiss took as supporting the nativist claim of modularity.

There are two problems, however, that complicate interpretation of Antony's case. First, although Antony's utterances do appear formally complex and sophisticated, so do at least some of the utterances of many typically developing 2- to 3-year olds. Much of the development that we see after the age of 3 or 4 years represents a consolidation, extension, and integration of previously acquired syntactic forms. These changes tend to be manifested as an increase in the frequency of use of more complex forms rather than as the emergence of entirely new forms (Chapman et al., 1992). In other words, a few (perhaps, even many) examples of sophisticated language can be culled from a large corpus of just about any language user who is functioning near a 3-year-old level. Thus, the examples from Antony are perhaps less dramatic, less indicative of "spared" skills, than it would first appear. This is a problem that characterizes many of the case studies reported to date (Abbeduto, 1997).

The second interpretive problem plaguing Antony's case is that although his cognitive skills were quite delayed in general, he displayed rather minimal delay in one area of cognition, namely, auditory short-term memory. In fact, Antony functioned at a 7-year level on a test of auditory memory span. In light of claims about the importance of auditory short-term memory for language development (Baddeley, Gathercole, & Papagno, 1998), Antony's accomplishments may well reflect the role of cognition rather than the modularity of language. It is interesting that in many of the other cases of so-called spared syntax, auditory memory remains, if not age appropriate, an area of relative strength, including, for example, Jean Rondal's (1995) well-documented case of Francoise, a young woman with Down syndrome whose native language is French.

In a group study of the relative rates of achievement in the language and cognitive domains, Abbeduto, Furman, and Davies (1989) compared the receptive language skills of individuals with mental retardation who, as a group, were heterogeneous with respect to etiology, and typically developing children matched to them on nonverbal MA. Receptive language was measured by administering the Test for Reception of Grammar (TROG; Bishop, 1982). Within each diagnostic group, the participants were organized

into the nonverbal MA levels of 5, 7, and 9 years. Abbeduto et al. found that although the typically developing children earned higher TROG scores than the participants with mental retardation, on average, there was a Group X MA interaction. The interaction reflected the fact that the difference between the diagnostic groups increased with increasing nonverbal MA. This finding suggests that language, at least when it is measured receptively, is an area of relative weakness for individuals with mental retardation, particularly when one focuses on those linguistic achievements that occur relatively late in development. This is a fairly robust finding in the literature, although the extent to which language lags behind cognition depends on etiology (as discussed in a subsequent section). Most importantly for present purposes, group comparison studies yield very different conclusions than do case studies such as those of Antony. In particular, for many individuals with mental retardation, language is an area of relative weakness and the degree of dissociation of language and cognition is not always so dramatic (i.e., with one domain being "spared").

An example of a study designed to examine the role of cognitive variation in explaining language variation is provided by Kahn (1975). This study, like many in the 1960s and 1970s, was motivated by the theory of the noted developmentalist, Jean Piaget, who claimed that certain achievements in the sensorimotor realm were prerequisites for major transitions in language in the infant and toddler years. Kahn administered a battery of Piagetian tasks (e.g., object permanence, comprehension of means-ends relationships) to children with mental retardation ranging in age from approximately 4 to 8 years. Each child was classified, based on his or her performance on the Piagetian task, as being at or below Stage 6 of sensorimotor development. Stage 6 is the crowning achievement of the first major period of development according to Piaget and is thought to reflect the emergence of symbolic understanding. Kahn also classified each child as having or not having expressive language (i.e., more than 10 words and used language to request objects). Consistent with Piagetian theory, there was some evidence that Stage 6 functioning was necessary for the emergence of expressive language: Children at Stage 6 were more likely to have expressive language than were those at lower stages. The relationship was far from perfect, however, as some children who had expressive language functioned below Stage 6. More generally, it is often found that children, both typically and atypically developing, perform at different sensorimotor stages on different sensorimotor tasks (Rosenberg & Abbeduto, 1993), a fact that is obscured in the analyses conducted by Kahn, who apparently derived an average stage for each participant. Such across-task variability has led many researchers to abandon the stage notion and to search for more narrow types of relations between language and cognition such as the finding that achievements in

object permanence underlie the learning of *gone* and other words denoting disappearance (Rosenberg & Abbeduto, 1993).

Another example of a study focused on the role of cognitive variation in explaining language variation is provided by Chapman, Miller, Sindberg, and Seung's (1996) investigation of *fast mapping* in adolescents and young adults with Down syndrome. Fast mapping refers to the process by which children create a partial representation of the meaning of a new word after hearing the word in context only a few times (Mervis & Bertrand, 1994). Fast mapping would be demonstrated by a child who selects an odd-shaped unfamiliar object rather than a cup or other familiar object when asked by an adult to *get the koob* and who later spontaneously labels the former object *koob*. Chapman et al. found that fully 41% of the variance in the fast-mapping tests scores of their participants with Down syndrome was accounted for by differences in their performance on a concurrently administered digit span task. The digit span task is thought to be a measure of important components of auditory short-term memory (Baddeley, 1986, 2001). This finding is consistent with those of many other investigations with a number of mental retardation syndromes (Robinson, Mervis, & Robinson, 2003) in suggesting that some cognitive functions play a critical role in various aspects of language learning. Here too, however, the correspondence is most often seen when fairly narrow facets of language learning and cognition are examined rather than broad all-encompassing measures of each domain. Early research, therefore, may have cast the language-cognition question too broadly.

C. Do Environmental Differences Matter?

1. PARENTAL CONTRIBUTIONS

For the most part, studies focused on environmental contributions to language development in mental retardation have been designed to determine whether parents (usually mothers) or other adult care providers of children with mental retardation are as adept at making the child-centered adjustments that define CDL as are parents and care providers of typically developing children. The motivation for such investigations has been the hypothesis that the unique needs of the child with mental retardation might make it more difficult for even the most skilled and motivated adults to engage in optimal levels of CDL over the course of development (Snow, 1995). If care providers are not adept at making such adjustments, they might be contributing to their children's challenges in the domain of language. Fewer studies, however, have sought to document the causal role of caregiver behavior on the language learning of those with mental retardation (Abbeduto & Hesketh, 1997).

The most common type of study in this area is that in which a comparison is made between the language or interactional styles of mothers of children with mental retardation and those of mothers of younger typically

developing children who are matched to those with mental retardation on some measure of developmental level (e.g., MA). The maternal behaviors of interest are usually assessed in dyadic interactions with the child, generally in free play, but sometimes in a structured task. Analyses then focus on testing for diagnostic group differences in maternal behaviors on variables thought to reflect the CDL style. The argument is that any differences observed between the two groups of mothers reflect a "failure" to appropriately adapt to their children's developmental needs. It is important to note, however, that such differences could also reflect an appropriate adaptation on the part of mother to some unique characteristics of the child with mental retardation that has not been measured in the study, but this latter interpretation is seldom considered (Rosenberg & Abbeduto, 1993). More generally, group differences in parental behavior should be seen as only the first step in understanding parental contributions to children's development; one still needs to determine why those differences have emerged and what, if any (positive or negative), consequences they actually have (Seltzer, Abbeduto, Shattuck, Greenberg, & Swe, 2004).

An example of this type of study is that of Rondal (1978), which is one of the most comprehensive analyses of parental talk to children with mental retardation to date. Rondal compared mothers of children with Down syndrome to mothers of language level–matched typically developing children and found no differences on literally dozens of variables designed to reflect the linguistic complexity of maternal talk. At the same time, however, he did find that virtually all of the maternal talk variables were correlated with child language level: More complex maternal talk occurred for more linguistically advanced children. This same pattern of within-group relationships was found for the mothers of the typically developing children. Together, these findings suggest that, like mothers of typically developing children, mothers of children with Down syndrome adjust their linguistic interactions to suit the capabilities of their children. Importantly, however, other studies, which have focused less on the linguistic character of mother–child interaction and more on the social uses of maternal talk within those interactions, have uncovered differences between parents of those with and without mental retardation.

Perhaps the most active area of research on the social uses of talk by parents has been that focused on parental directiveness (Abbeduto & Hesketh, 1997). Directiveness refers to the extent to which a care provider controls the interaction to achieve his or her own agenda rather than following and being responsive to the child's lead. More specifically, a highly directive parental style of interaction includes:

1. a failure to talk about the objects or events that are the focus of the child's attention;

2. a lack of responsivity to the child's utterances (e.g., by introducing a new topic);
3. frequent attempts to redirect the child's attention;
4. a high rate of imperatives (e.g., *Get the block*) and other utterances that function to request actions or other well-defined and highly constrained responses from the child rather than comments on the child's actions, talk, and so on.

Parents of children with mental retardation, especially those with Down syndrome, have consistently been found to be more directive than parents of typically developing children of comparable developmental levels (Abbeduto & Hesketh, 1997). This directiveness appears to be motivated by parents' desire to solicit more age-appropriate behavior from their children (Mahoney, Finger, & Powell, 1985; Mahoney, Fors, & Wood, 1990; Maurer & Sherrod, 1987). Of course, the extent to which parental directiveness inhibits language learning by children with mental retardation is not addressed by such comparative studies.

Determining the consequences of parental directiveness, and causal relationships among parent and child behavior more generally, requires a longitudinal design so that the relationship between care provider behavior at one point in time and subsequent child progress in language learning can be statistically analyzed. Such a longitudinal approach is illustrated by a path-breaking study conducted by Harris, Kasari, and Sigman (1996). These investigators were interested in the role of joint attention, which is the state in which both parties in an interaction are focused on the same object or topic and at least one of them says something about that object or topic (Murphy & Abbeduto, 2004). There is a fairly substantial literature documenting that engagement in a higher proportion of joint attention episodes is facilitative of language development, especially the acquisition of vocabulary, of older typically developing toddler and preschoolers (Akhtar, Dunham, & Akhtar, 1991; Tomasello & Farrar, 1986). Harris et al., however, looked not only at the occurrence of joint attention but also at who initiated the episode—mother or child. The participants were 28 children with Down syndrome and 17 mental age-matched typically developing children and their parents (virtually always the mother), with the language development measured twice with a 1-year interval between measurements.

Harris et al. found that the dyads including the children with Down syndrome actually engaged in more episodes of joint attention than did the dyads with typically developing children, however, this reflected the fact that mothers of children with Down syndrome were more likely to maintain episodes they themselves initiated than were the comparison mothers. Most importantly, gains in receptive language among children with Down

syndrome were greater for those children who participated in a greater rate of joint attention episodes initiated by the child but maintained by the mother and a lower rate of episodes in which mother redirected the child away from a child-selected object or topic. The typically developing children benefited more, in terms of progress in language learning, if they participated in a greater rate of mother-initiated episodes of joint attention.

In summary, children with Down syndrome appear to be especially adversely affected by parental directiveness, which is common among their care providers. It would be useful to extend the longitudinal investigation of directiveness to other groups of children with mental retardation (e.g., fragile X syndrome) to determine how child characteristics moderate and mediate this relationship (Murphy & Abbeduto, 2004).

2. LANGUAGE INTERVENTION

Research on language intervention for individuals with mental retardation has a long history (for reviews see Brady & Warren, 2003; Miller, Leddy, & Leavitt, 1999; Rondal & Buckley, 2003; Rondal & Edwards, 1997). Although this research is of obvious importance from a clinical perspective, it is also relevant to many of the nativist claims, as well as to the claims of various alternatives to the nativist approach (Abbeduto & Boudreau, 2004). In particular, language intervention research provides a basis for examining the nativist claim that the environment's role is limited to the provision of key exemplars of the language and thus, most variations in children's environments are of little consequence for language learning. Here we briefly consider the implications of language intervention research for the theoretical debates sparked by the nativist approach.

In the 1950s and 1960s, research on language intervention was dominated by behaviorism (Abbeduto & Boudreau, 2004), which posited that language was learned, like any other behavior, through principles such as operant conditioning, which were largely domain- and species-general (Guess, Sailor, Rutherford, & Baer, 1968). Although behaviorism contrasted rather dramatically with nativism in many of its fundamental assumptions, the two did share a focus on learning the forms of language isolated from the social contexts in which language is typically learned and used. The behaviorist approach entailed teaching linguistic targets within a therapeutic context that highlighted the target without the "distractions" and competing stimuli typical of natural contexts and with the nature and course of the interaction determined almost exclusively by the clinician (Camarata & Nelson, 1992). In many respects, this approach is consistent with the nativist claim that the environment's role is limited to the provision of key exemplars and that social context in which the talk occurs is largely irrelevant. It is interesting, therefore, that the behaviorist approach—at least in its *direct*

instruction varieties—met with limited success: Many children failed to master the targets of intervention and even those who succeeded often failed to generalize the newly learned behaviors beyond the training context (Warren, 1993).

Direct instruction has now been replaced as the dominant approach in the language intervention field by approaches that emphasize the need to embed language teaching in meaningful social interaction. *Interactive* approaches are child centered in that they encourage interventionists to "follow the child's lead" by allowing the child to initiate a communicative exchange and responding in a way that enables to child to control the subsequent flow of the exchange (Fey, 1986). There are a variety of interactive approaches distinguished by the extent to which they also incorporate elements of direct instruction and adult control. All interactive approaches, however, are based, at least in part, on social-interactionist theories of language learning (Paul, 2001; Yoder, Kaiser, & Alpert, 1991).

In its purest form, clinicians, parents, or other facilitators in an interactive intervention are taught how to modify their behavior to facilitate communication development (Hubbell, 1977; Mahoney et al., 1999). In the most child-centered instantiation of an interactive intervention, the facilitators do not have explicit communication goals nor do they try to teach specific language skills (Fey, 1986). Instead, their goal is to establish interaction patterns that facilitate and maximize opportunities for the child to use his or her language skills within natural communication settings (Girolametto & Tannock, 1994; Hubbell, 1977; MacDonald, 1989; Tannock & Girolametto, 1992; Tannock, Girolametto, & Siegel, 1992). Achieving this goal is thought to require that the facilitator match his or her style and type of interaction to the child's current level of communication skill, or zone of proximal development (Paul, 2001; Snow, 1995; Vygotsky, 1978).

Focused stimulation is a type of interactive intervention approach that includes some adult control (Fey, Cleave, Long, & Hughes, 1993; Girolametto, Weitzman, & Clements-Baartman, 1998; Leonard, 1981; Pearce, Girolametto, & Weitzman, 1996). Although the interventionist continues to follow the child's lead, he or she also provides multiple models of the targeted language behavior that are selected according to the child's current level of functioning. Girolametto et al. (1998) examined the effectiveness of a focused intervention targeting expressive vocabulary with 12 toddlers with Down syndrome and their mothers. In the experimental group, parents learned how to use focused stimulation procedures and basic principles of interactive intervention. Girolametto et al. (1998) reported that the toddlers in the experimental group produced more of the targeted vocabulary than those in the control group.

Milieu approaches also embed the intervention in social interactions in which the child exerts a considerable degree of control, however, there are more

adult-imposed constraints on the child's environment and the adult's responses to the child's attempts at communication (Hart, 1985; Rogers-Warren & Warren, 1980). Halle, Marshall, and Spradlin (1979), for example, used time delay and modeling to increase spontaneous requests in three adolescents with mental retardation and to facilitate generalization to other settings. Similarly, Miller, Collins, and Hemmeter (2002) used a time-delay procedure to teach three adolescents with developmental disabilities to initiate use of manual sign language. All three participants showed an increase in the spontaneous use of the target signs within 3 days of participating in the intervention program. In a study involving three preschoolers with developmental disabilities, Yoder, Kaiser, Alpert, and Fischer (1993) used model and mand-model procedures to increase the comprehension and production of nouns.

In keeping with social-interactionist theories, interactive approaches to intervention have also employed parents, siblings, and other people familiar to the child as interventionists. This practice reflects the view that language is typically learned from others with whom the child has meaningful, personal relationships that are motivated and shaped by goals other than the teaching of language. In a study of 32 mothers and their preschool children, Tannock et al. (1992) trained parents to modify their interaction style so as to become more responsive and less directive. A similar approach characterizes the Ecological Communication Organization Program (MacDonald, 1989; MacDonald & Carroll, 1992), which has been used with children who have various developmental disabilities, including Down syndrome and autism. Facilitators are encouraged to follow the child's lead to match his or her communication abilities and to wait responsively to increase turn taking.

Likewise, Hanen Centre programs, such as *It Takes Two to Talk* (Manolson, 1992), train parents, teachers, and other child care providers to use interactive approaches to facilitate language development in children with language delays and other developmental disabilities (Girolametto, 1995, 1998; Girolametto, Verbey, & Tannock, 1994; Girolametto et al., 1998). Girolametto (1988) trained the mothers of preschoolers with developmental delays in an 11-week interactive intervention program. The mothers who received the training were less directive and more responsive than were the mothers in the control group. Although the preschoolers did not differ in terms of overall language development, the children in the experimental group demonstrated greater turn-taking skills, were more responsive to their mothers' attempts at interaction, and exhibited greater lexical diversity at the end of the intervention.

Kaiser and colleagues have trained siblings, parents, and teaching professionals (Alpert & Kaiser, 1992; Hemmeter & Kaiser, 1994; Kaiser, 1993; Kaiser, Hemmeter, Ostrosky, & Fischer, 1996; Kaiser, Ostrosky, & Alpert, 1993). In an investigation by Hancock and Kaiser (1996), the older siblings

of three children with cognitive disabilities were trained to use modeling and mand-modeling in a play context. Each sibling was able to learn and implement the milieu teaching techniques in a play setting, and two of the three siblings generalized their behaviors to snack times. The children with cognitive disabilities, ranging in age from 56 to 72 months, demonstrated gains in the number of utterances produced and the spontaneous use of language targets at the end of the intervention and during the follow-up period.

Although interactive interventions have demonstrated the importance of the environment, particularly a child-centered and socially rich environment, for language learning, they also have demonstrated that the environment does not act on an infinitely malleable child. Yoder et al. (1991), for example, compared the effectiveness of a direct-instruction approach, the Communication Training Program (Waryas & Stemel-Campbell, 1983), with a milieu intervention approach, the Milieu Language Teaching Program (Kaiser, Hendrickson, & Alpert, 1991). Yoder et al. (1991) found that children who scored higher on pretreatment measures, such as percentage of self-initiated utterances, benefited more from the direct-instruction approach. The participants who scored lower on pretreatment measures exhibited greater gains with the milieu approach. In another study, Yoder and Warren (2002) implemented the Hanen Centre (Manolson, 1992) parent education program in conjunction with a milieu approach to increase the communication skills of prelinguistic toddlers. Using the program, parents learned how to respond to the child's communicative acts as intentional and to facilitate language development. Yoder and Warren found that the toddlers who produced fewer canonical vocalizations and comments before treatment demonstrated the greatest gains in the number of child-initiated comments and lexical density. In contrast, participants who frequently produced canonical vocalizations and comments at the start of the intervention exhibited a decrease in comments and lexical density after the intervention. These studies demonstrate that the child's level of development and personal characteristics and behavioral profile moderate the effects of language intervention.

Intervention research has also illustrated the complexity of the environmental systems in which children live and on which their development depends. In a study by Yoder and Warren (1998), for example, interest was in the effectiveness of an intervention approach they called Prelinguistic Milieu Teaching (PMT). The approach entails creating opportunities for the prelinguistic child with mental retardation to engage in communication, with the clinician attempting to encourage the child to engage in triadic eye gaze (i.e., to coordinate looking at an object of interest and the adult partner so as to create joint attention). These investigators compared PMT to a Responsive Small Group (RSG) approach, which entailed largely the clinician responding appropriately to communicative overtures from the child, in a randomized

experimental design. Pre- and posttreatment measures of communication in a variety of contexts and with a variety of partners were taken. The finding of greatest interest was that the children who benefited from PMT the most were those who had mothers who were found to be most responsive to their children prior to the intervention. In contrast, the children with mothers who seemed least responsive prior to treatment tended to benefit more from RSG. Such findings underscore the connectedness of the contexts of children's lives and support models, such as that of Bronfenbrenner (Bronfenbrenner & Morris, 1998), that conceptualize the environment in terms of systems rather than as concatenations of stimuli or experiences.

In summary, studies of language intervention for individuals with mental retardation have clearly indicated that the environment can play an important role in language development. Moreover, many children with cognitive limitations learn best when the targets of intervention are embedded in meaningful social interaction and the targets themselves have social utility rather than being abstract, decontextualized language forms. Less capable children, however, may need new behaviors to be taught, at least initially, in a less socially rich context; however, generalization of those behaviors will eventually require opportunities to practice them in meaningful social activities. At the same time, these studies demonstrate that any theoretical account that supposes that the environment functions to mold a passive child, as was the case in early behaviorist accounts, will fail to capture the essence of language development. And finally, research on language intervention has dramatically illustrated the need for a developmental perspective, which as noted previously, is lacking in most nativist accounts. Simply put, the characteristics and behaviors of the child are an important determinant of the extent to which the environmental arrangement represented in an intervention will be effective in fostering language development.

D. Do Performance Factors Matter?

Studies of performance are not concerned with the particular elements or rules of language that have been acquired, but rather with whether and how individuals with mental retardation use their knowledge of language when participating in important social contexts. In general, studies of performance have focused on two questions:

1. Can individuals with mental retardation use whatever language they have learned to accomplish important communicative tasks in social interaction—tasks like turn taking, comprehension monitoring, making polite requests, and producing sufficiently informative messages?

2. Is the use of particular aspects of language by individuals with mental retardation constrained by, and thus variable across, communicative tasks or contexts?

Studies addressing the first question often involve an analysis of naturally occurring interactions (Abbeduto & Rosenberg, 1980), usually interactions between individuals with mental retardation and their parents (Tannock, 1988). Although these studies have high external validity, it is generally difficult to draw conclusions about the performance capabilities of the participants with mental retardation. The difficulty arises because one seldom can discern whether successful performance is due to the capabilities of the participant with mental retardation or to the provision of "extraordinary" scaffolding by the parent or other care provider (Rosenberg & Abbeduto, 1993).

An alternative to naturalistic studies, which we have employed in our own research (Abbeduto, 1984; Abbeduto, Davies, & Furman, 1988; Abbeduto, Davies, Solesby, & Furman, 1991; Abbeduto, Short-Meyerson, Benson, & Dolish, 1997; Abbeduto, Short-Meyerson, Benson, Dolish, & Weissman, 1998), is to create laboratory-based tasks that strip away the scaffolding to determine what the participants can accomplish independently (or with a minimum of support). In Abbeduto et al. (1991), for example, we created a task in which the participant responded to simple directions from the examiner. By design, however, some of the directions were inadequate. We then measured the occurrence of explicit signals of noncomprehension from the participant (e.g., *Which balloon?*), with such signals indexing the extent to which the participants were able to monitor their comprehension and devise plans for resolving problems. The results indicated that children and adolescents with mental retardation were less skilled in this domain than were younger typically developing children matched to them on nonverbal mental age. The participants all had the linguistic skills needed to produce at least rudimentary signals of noncomprehension.

In addressing the question of task or context variations in performance, we have created structured tasks that contrast in terms of the extent to which they challenge particular language capacities. For example, we have found that dyadic conversation imposes relatively minimal demands on the speaker's syntactic capabilities compared to the task of storytelling or narration (Abbeduto, Benson, Short, & Dolish, 1995). In contrast, conversation often elicits more diverse vocabulary from the speaker than does narration (Abbeduto et al., 1995). As a result, narration often highlights differences in the syntactic capabilities of different speakers, which are not apparent in conversation, whereas the reverse is true for lexical capabilities. In our research on etiological differences in language, for example, we found that adolescents with Down syndrome had less syntactically mature performance

than cognitively matched adolescents with fragile X syndrome but only in narration (Abbeduto & Murphy, 2004). Unfortunately, most previous research on etiological differences has relied on conversational language exclusively, with the result being that important etiology-related differences in syntactic capabilities were probably missed (Murphy & Abbeduto, 2003).

In summary, research on linguistic performance has clearly documented that performance factors are important for understanding language learning in individuals with mental retardation:

1. Individuals with mental retardation often fail to use the language they have learned to accomplish many of the linguistic tasks that comprise everyday social interaction.
2. The use of particular language forms and rules varies according to the processing demands of the task or context in which language must be used.
3. The extent to which linguistic performance is impaired relative to language mastery or other domains of functioning varies across etiologies.

Such findings are at odds with the trivial role attributed to performance factors in the nativist approach. Performance factors, however, play a central role in the social-interactionist and emergentist approaches.

III. FUTURE DIRECTIONS IN RESEARCH ON LANGUAGE LEARNING AND MENTAL RETARDATION

We believe that future research on language development in mental retardation will be strongly influenced by the genetic syndromes approach described in a previous section. This approach has already led to many interesting insights, not the least of which is that language development is different in important respects across many syndromes. At the same time, however, it is important to acknowledge that establishing that there are different profiles and trajectories of language development does not by itself provide insight into the mechanisms producing those differences. The pathways by which the genes involved in these syndromes lead to phenotypic outcomes are complex and indirect (i.e., mediated, in part, through person–environment transactions). We believe that understanding these pathways will require understanding the environments of affected individuals and thus will be informed by the social-interactionist and emergentist perspectives, which have already proven invaluable in understanding typical language development. In the following sections, we briefly and selectively discuss research on three syndromes—Down syndrome, Williams syndrome, and

fragile X syndrome—pointing out some of the questions and challenges for future research as we do so.

A. Down Syndrome

Down syndrome, which is most often caused by a trisomy of chromosome 21, is the leading genetic cause of mental retardation, with a prevalence of 1 in 900 (Rozien, 1997). It has been well documented that language is especially challenging for individuals with Down syndrome, with delays relative to cognition emerging early in development (Chapman & Hesketh, 2000). All facets of language are delayed relative to chronological age, but the delays are greater in production than in comprehension and greater in syntax than in vocabulary (Abbeduto et al., 2003; Chapman et al., 1992, 1996).

For many years, it was thought that children with Down syndrome reached a plateau in their language development, at least in the domain of syntax, around the age of 10–12 years (Fowler, Gelman, & Gleitman, 1994). This observation was based largely on cross-sectional and short-term longitudinal studies conducted by Fowler et al. and involving small samples of participants. Nevertheless, it became well accepted by the field, fitting with Chomskyan claims about the innate bases of language, which also gave rise to notions of a sensitive period for language learning (Lenneberg, 1967). Presumably, children with Down syndrome move beyond the sensitive period before completing their language development, hence the plateauing of their skills. From a clinical perspective, this notion of a Down syndrome plateau is often the basis for decisions to reduce or eliminate speech and language services for high school–age adolescents with Down syndrome, who presumably cannot benefit from speech-language services. Research, however, suggests that language development may not plateau for most individuals with Down syndrome.

Perhaps the most compelling evidence on the issue of plateaus comes from a long-term longitudinal study of children and adolescents with Down syndrome conducted by Chapman (2003). Chapman, Hesketh, and Kistler (2002) reported language production data for a relatively large sample of participants ($n = 31$). In addition to its large sample size, the study is notable for its coverage of a wide age span (i.e., 5–20 years). Chapman et al. documented continued growth in syntax, measured by Mean Length of Utterance (MLU), from childhood at least until the adult years. This finding is especially interesting because Fowler et al. had also relied on gross measures of syntactic maturity derived from production, and it suggests that a plateau of expressive syntax may be the exception rather than the rule for this population. At the same time, however, Chapman et al. also found that

the comprehension skills of their participants with Down syndrome, as a group, appeared to actually regress in the early adult years. This regression may reflect the early phases of the precocious aging and decline that occurs in Down syndrome (Silverman & Wisniewski, 1999).

More generally, the findings for Down syndrome point to the need to conduct longitudinal studies over relatively broad age ranges. Such studies are more feasible now with the advent of Hierarchical Linear Modeling and other multilevel modeling statistical techniques that offer more flexibility (e.g., with regard to missing observations) compared to the standard-repeated measures ANOVA approach of the past. Moreover, results of studies, such as those by Chapman et al., also document the need to take a comprehensive approach to measuring language change and to considering the possibility that development may be characterized by both improvement and decline.

B. Williams Syndrome

Williams syndrome is the result of a microdeletion on chromosome 7 at 7q11.23, with at least 17 genes mapped to the deletion site thus far (Dykens et al., 2000). It is relatively frequent as a cause of mental retardation, although rare in the general population (1 per 20,000 births). Interest in Williams syndrome among language researchers began in earnest in the 1980s following some intriguing findings reported by Bellugi and her colleagues (see Bellugi et al., 1997 and Mervis et al., 2003, for reviews). In her initial studies, Bellugi studied a small sample of individuals with Williams syndrome who despite having moderate to severe mental retardation had excellent, if not age-appropriate, language, including syntactic capabilities. Such findings seemed to provide strong empirical support for the nativist approach.

It has since been found that these initial claims were incorrect on several counts. Most importantly, it does not appear that syntactic skills are "spared." True, syntax, and language in general, may be an area of relative strength in Williams syndrome, however, that strength is far less dramatic than Bellugi's initial reports suggested.

The bulk of the evidence for a more moderate view of the Williams syndrome linguistic strengths comes from the research of Mervis et al. (2003). In one study with 7- to 17-year olds with Williams syndrome, Mervis et al. administered the Differential Ability Scales, which includes relatively (statistically) distinct verbal, nonverbal, and spatial clusters of subtests. It was found that, for the sample as a whole, verbal skills were, on average, two standard deviations below the mean of the standardization sample. Moreover, the asynchrony that existed was not really one of relative strengths in the verbal domain but of relative weakness in the spatial domain. Verbal and

nonverbal standard scores were quire similar for the sample. Similar results were obtained for a sample of toddlers with Williams syndrome using the Mullen Scales of Early Learning. Importantly, these findings are not limited to the use of standardized tests. Parent report measures suggest that early vocabulary development is quite impaired in toddlers with Williams syndrome as well, for example, all of the 13 children studied by Mervis et al. were found to be below the 5th percentile for the age at which parents reported them to have an expressive vocabulary of 10 words.

How are findings such as those reported by Mervis et al. (2003) to be reconciled with the early reports from Bellugi et al. (1997)? Two reasons for the inconsistency can be offered. First, it is now clear that there is a wide range of affectedness in Williams syndrome. Many of the early studies in this area included quite small samples, with many of the participants quite likely being referred to researchers precisely because they had relatively strong language skills. As a result, it may be that relative strength in language is characteristic of only a small proportion of all individuals with Williams syndrome. Second, in many of the early studies in this area, the participants with Williams syndrome were compared to those with Down syndrome, however, individuals with Down syndrome, as already noted, have a relative weakness in language, especially in the syntactic domain. As a result, the relative strengths in language of the (nonrepresentative) samples of individuals with Williams syndrome were exaggerated by their comparison with Down syndrome.

More generally, the research on Williams syndrome illustrates two challenges for research on language development in mental retardation. First, it is important to recognize that recruitment procedures can result in the participation of individuals who are "unusual," or nonrepresentative, of the syndrome more generally, especially when the design of the study requires the use of a relatively small sample. Second, not all comparison groups are equivalent in their profiles of language and other skills and thus the choice of a comparison group affects interpretation of the findings. Future research should strive to study larger, more representative samples reflective of the full range of syndrome affectedness and to include comparison groups chosen to be maximally informative as regards the research question(s) of interest.

C. Fragile X Syndrome

Fragile X syndrome is the leading inherited cause of mental retardation (Sherman, 2002). It is caused by a mutation in a single gene (FMR1) on the X chromosome (Brown, 2002). In the full mutation, a repetitive sequence of trinucleotides, which is typically characterized by 54 or fewer repeats,

expands to 200 or more (Oostra, 1996). This expansion results in substantially lower levels of FMRP, a protein thought to play a critical role in experience-dependent maturation and functioning of neural synapses (Greenough et al., 2001). The prevalence of affected individuals is 1 in 4000 males and 1 in 8000 females (Crawford, Acuna, & Sherman, 2001).

The behavioral phenotype of fragile X syndrome has been investigated for the past three or more decades, although research on language has been relatively limited (Murphy & Abbeduto, 2003). Nevertheless, it is has been proposed that perseveration is a core feature of the linguistic phenotype of fragile X syndrome (Cornish, Sudhalter, & Turk, 2004). Perseveration is the excessive self-repetition of a sound, word, phrase, or topic (Sudhalter, Cohen, Silverman, & Wolf-Schein, 1990). Numerous studies have documented more perseveration among males with fragile X than their mental age-matched peers with Down syndrome, autism, or typical development (Ferrier, Bashir, Meryash, Johnston, & Wolf, 1991; Belser & Sudhalter, 2001; Sudhalter, Maranion, & Brooks, 1992; Sudhalter, Scarborough, & Cohen, 1991; Sudhalter et al., 1990), and there is some evidence of perseveration by females as well (Madison, George, & Moeschler, 1986). Research that we and our colleagues have conducted, however, suggests that there is much about perseveration and the mechanisms that produce this phenomenon yet to be worked out.

We designed a communication task to examine the ability of adolescents with fragile X syndrome to make clear the referents of their talk in a non-face-to-face context (Abbeduto et al., 2006). The task, based on the barrier task of Glucksberg, Krauss, and Higgins (1975), required the participant to play the role of speaker and describe one of several abstract shapes to a listener so that the listener could select that same shape from a larger set. The speaker and listener were separated by an opaque partition. Each shape recurred on multiple trials over the course of the session so that we could examine consistency in the words or phrases used to describe a given shape each time it occurred. In this task, the use of the same description for a shape each time it recurred would decrease the listener's burden compared to the situation in which a new description was created for each recurrence. In a sense, the tendency to perseverate would actually benefit the speakers with fragile X syndrome by leading them to be consistent in their descriptions.

Surprisingly, we found that adolescents and young adults with fragile X syndrome were significantly less consistent in their descriptions than either of two MA-matched comparison groups: Down syndrome youth and typically developing children. For example, relative to the comparison groups, the participants with fragile X syndrome were more likely to refer to a shape as a "house" on one trial and as an "envelope" on a subsequent trial. In this

task, then, youth with fragile X syndrome were not perseverative in a context in which perseveration would actually have been beneficial. This finding is important in that it suggests that youth with fragile X syndrome are not always perseverative. More importantly, the findings suggest that task or contextual factors are likely to shape many dimensions of linguistic performance, including the so-called core features of fragile X syndrome. Such a finding is difficult to reconcile with nativist claims of linguistic modularity and the trivialization of performance factors.

IV. CONCLUSIONS

The past four decades have seen dramatic shifts in the theories dominating research on language development, both in typical children and in individuals with mental retardation. At the same time, research in the field of mental retardation has provided empirical data that have informed the theoretical debates. It now appears that although humans are innately equipped to learn language, the mechanisms driving that learning are general ones that facilitate learning in other domains as well. Moreover, it appears that the power of those mechanisms resides in their sensitivity to patterns of environmental stimulations and thus the nature of a child's learning environment is an important determinant of the course of language learning. It also appears that the centrality of language to human interaction motivates the young language learner to marshal all of his or her skills and knowledge to master it, although acquiring the language is only a means to an end, namely, interacting with others. Individuals with mental retardation have difficulty learning language, but the reasons vary across individuals due, in part, to the etiology of their retardation. Even in the case of genetic syndromes, however, language problems do not reside in the genes; instead, those problems reflect the complex ways in which genes and environments interact in complex and often indirect ways to shape all domains of the child's competence.

This brief history of research on language and mental retardation was meant to coincide with the history of the Gatlinburg Conference on Mental Retardation Research and Theory. It is worth noting, therefore, that the Conference has been central to much of the theory and research on language learning presented in the foregoing pages. The Conference has throughout its history provided an intellectually invigorating context in which researchers interested in language development and intervention have been able to discuss their findings and theoretical frameworks. Moreover, language researchers attending the Conference have been informed and to some extent transformed by their exposure to research and theory on the diversity of

topics, from memory to families to brain–behavior relationships, that populate the Conference program each and every year. We expect that the Conference will continue to play this role for language researchers for years to come.

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Residential Services Research in the Developmental Disabilities Sector*

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I. INTRODUCTION

In contrast to clinical research, services research is ultimately concerned with the organization and financing of services at national, state, or provincial, and levels of service organization as mediating influences on service processes and outcomes (Newman, Howard, Windle, & Hohmann, 1994). Given this wide purview, services research spans an immensely broad range of issues (Jacobson, 1991; Jacobson & Schwartz, 1991). Examples of service research include organization and delivery of service patterns, characteristics and needs of service recipients, cost effectiveness, and changing philosophies of care and treatment. Collectively, these examples underscore the unremitting need in our field to reconcile diverse systemic pressures with socially desirable and responsible policy priorities (Newman, Kellett, & Beail, 2003; Todd et al., 2000).

*Author note: The views expressed in the chapter do not necessarily represent those of the sponsoring agency.

†Deceased, May 8, 2004. This manuscript was written during the last months of John Jacobson's life and stands as a tribute to his longstanding interest in improving residential services for individuals with developmental disabilities.

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A. Some Trends in Published Residential Services Research

Compared to the mental health sector, the amount of published residential services research in developmental disabilities has been modest. One area that did receive significant attention beginning in the 1970s was deinstitutionalization, the outcomes of which have continued to be studied and reported (Kim, Larson, & Lakin 2001a,b; Spreat & Conroy, 2002; Stancliffe, Hayden, Larson, & Lakin, 2002; Young, Ashman, Sigafoos, & Grevell, 2001). During the same decade, studies of program policy and demography emerged, which also have continued through the present and have remained the primary index of program policy implementation in the United States and elsewhere. Examples of contemporary key reports include Braddock and Mitchell (1992), Braddock and Hemp (1997), Gilmore, Schuster, Timmons, and Butterworth (2000), Lakin, Anderson, and Prouty (1998), Lakin, Hill, and Anderson (1991), Mansell, Ashman, Macdonald, and Beadle-Brown (2002), and Stancliffe (2002).

In the decades of the 1990s and 2000s, research on costs and financing of residential developmental services has become a major aspect of services research. Studies during the early 1990s reflecting national or state practices included Braddock and Fujiura (1991), Gettings and Chapin (1992), O'Neill et al. (1990), Schalock and Fredericks (1990), and Shiell, Pettipher, Raynes, and Wright (1992). Research by Braddock and colleagues has been foundational, establishing a national picture of financing dimensions, practices, and trends. In contrast, there has been increased emphasis on the relationship of costs (or expenditures) and outcomes since the mid-1990s, including utilization of billing records for services as a source of cost estimation and measures of outcomes such as enriched and varied lifestyles for people with intellectual disabilities (ID) (Braddock, 2002; Campbell & Heal, 1995; Felce, Lowe, Beecham, & Hallam, 2000; Hatton, Emerson, Robertson, Henderson, & Cooper, 1995; Knobbe, Carey, Rhodes, & Horner, 1995; Lakin, Prouty, Polister, & Coucouvanis, 2003; Stancliffe & Lakin, 1998).

A persistent administrative problem in providing residential services for people with developmental disabilities has been direct service staff turnover. This problem became an evident and virtually universally recognized threat to continuity and overall quality of care (Jacobson & Schwartz, 1991). During the early 1990s, relevant research focused on influences upon rates of staff turnover including related organizational factors, alleviation of turnover and absenteeism, and issues surrounding compensation for direct service staff (Briggs, 1990; Felce, Lowe, & Beswick, 1993; Mitchell & Braddock, 1993, 1994; Razza, 1993).

Researchers have also examined residential service as a function of the type of residential service provided, including the intermediate care facility

(ICF/MR), Medicaid waiver, supported housing, and independent living models. With respect to the ICF/MR model, investigators have examined resident demographics (Anderson, Lakin, & Prouty, 1997; Chen, Bruininks, Lakin, & Hayden, 1993); impacts on residents (Crinella, McCleary, & Swansom, 1998; Konarski, Riddle, & Walker, 1994; Stancliffe et al., 2002); services provided (Jacobson et al., 1984); management (Holburn, 1992; Mayer, Heal, & Trach, 1992); quality improvement (Dura, Aman, & Mulick, 1988; Poindexter, 1989; Reid, Parsons, Green, & Schepis, 1991; Sparr & Smith, 1990); and costs and reimbursement (Chapin, Rotegard, & Manard, 1991; Conroy, 1996). A small number of studies have reported on the implementation of Medicaid waiver services for people with ID in the United States (Gardner, 1986; Lutzky, Alecxih, Duffy, & Neill, 2000; Smith, Prouty, & Lakin, 1996; West, Revell, Kregel, & Bricout, 1999). The impact of waiver-funded services has also been an area of investigation (Campbell, Fortune, & Heinlein, 1998; Stancliffe, Abery, & Smith, 2000).

A more recent residential model to attract investigation is the supported housing model in which individual or several-person housing is provided with more tailored services and supports than may typically occur in group homes (Howe, Horner, & Newton, 1998). During the early 1990s there was little research published on supported housing, with studies by Lowe and de Paiva (1991) and Schalock and Genung (1993), exemplifying the work that was disseminated through journals. From the mid-1990s to the present, interest and opportunity to research these settings has increased (Braddock, Emerson, Felce, & Stancliffe, 2001; Felce & Perry, 1996; Howe et al., 1998; Perry & Felce, 2003; Rapley & Beyer, 1996).

There has been persisting interest in the design and impacts of independent living arrangements for people with intellectual and developmental disabilities during the early 1990s (Lozano, 1993; O'Brien, 1994; Rose, White, Conroy, & Smith, 1993; Saunders & Spradlin, 1991; Siperstein, Reed, Wolraich, & O'Keefe, 1990), and more recently (Dyer, Kneringer, & Luce, 1996; Schwartz, 1995; Webber, Jenkinson, & McGillivray, 2002). Independent living requires less financial support than does supported housing; however, there may be considerable overlap in the substance of services and housing typified as supported housing or independent living. Inconsistencies and vagaries exist in how small residential contexts are typified in studies. Frequently, these stem, at least in part, from differences in the character and terminology among state, provincial, or national service systems, and increased flexibility in separation of housing and services within financing mechanisms.

Although the number of studies cited earlier suggests a significant amount of residential services research on four types of residential models alluded to here, the body of research is more accurately typified as cursory, relative to their level of utilization over the past three decades. Similarly, relative to the

strategic potential of flexible funding allocation represented by the Medicaid waiver mechanism, published assessments of its parameters of implementation and impacts have been superficial and insufficient to contribute to informed policy formulation within state or local systems. Moreover, services research, at least the research published in journals, has often (although not entirely, e.g., Braddock, Hemp, Bachelder, & Fujira, 1995) been divorced from decision making surrounding some of the toughest issues facing administrators of developmental services. Few comprehensive and generalizable studies have been conducted that address pressing concerns such as administrative and pragmatic outcomes of comprehensive strategies for addressing waiting lists for services, or for accommodating or enhancing services for people with co-occurring intellectual and mental disorders (or others requiring integration of resources and services across formally defined governmental service sectors).

The three state-commissioned research projects that are described in this chapter exemplify research conducted for administrative purposes that yields valuable information that is not likely to be found in research journals or other peer-reviewed sources. Following a cursory review of selected lines of residential research that can be obtained in the published literature, we discuss residential services research conducted through public-agency interests and contrast it with the traditional published studies. We present methods and excerpted findings from several state-level services research projects to illustrate the types of issues that are of pragmatic interest within the larger body of commissioned services evaluation studies.

B. Administratively Commissioned Residential Services Research

Although a large number of peer-reviewed publications fall within the category of residential services research, it is not necessarily accurate to consider these studies as representative of the larger body of residential services research. Many services research projects are administratively commissioned and conducted in conjunction with services reform or diversification initiatives, or as monitoring reports, sometimes in response to legislative, executive, or judicial mandates. Many relevant studies, meeting procedural criteria similar to those applied to professional publications, undoubtedly are carried out through contracts by state agencies, often in conjunction with universities. There may be few incentives, or in some instances, administrative disincentives, to their dissemination through publication or posting on governmental or quasi-governmental websites.

Public-agency–commissioned studies differ in substantial ways from studies that are published in professional journals or versions of these types of

studies that in some instances appear in journals. First, the format is not bound to traditional publication protocol. The research tends to be largely descriptive, often describing a wide range of characteristics of services and participants. Findings from measures tend to be broken down into their components or items rather than primarily summarized. Data are usually analyzed for management and administrative implications. Peer-reviewed articles that might be derived from these studies would present findings with respect to summary measures, in a less descriptive manner. Findings would more likely be summarized through predictive models (e.g., regression) and interpreted in the form of quite general policy implications.

To some extent these differences represent different priorities and information needs of consumers of the two types of documents (e.g., reports and articles), but both formats present quandaries that are often underestimated. In particular, the most critical quandary is presented by the need to translate programmatic and services research findings into administrative and managerial recommendations. The difficulty in this context is that services researchers, whether academics or public-agency technical staff, are frequently not administrators or managers and so they must often interpret the implications with incomplete understanding of the character and parameters of administrative or managerial practices that are priorities for professionals with those responsibilities. In the fortunate circumstances where researchers are also trained administrators or managers, analyses may be subject to distortion because of differing perspective on the nature of administrative and managerial functions, and a limited understanding of the parameters of possible actions that can be taken by administrators in the larger policy and program context of senior public administration within state and federal systems. In particular, services research analyses that do not include, or address, financial issues and administrative oversight processes related to program innovation or system change are unlikely to result in substantial systems impacts through administrative adoption or local management initiatives.

Whereas services researchers are engaged in depicting both positive and negative outcomes of managerial and clinical activity, administrators are concerned with system maintenance and improvement including protection of financial resources and iterative rather than revolutionary processes of system change. They, thus, may be more narrowly interested in identification of processes and activities that lead to more effective services. This differing emphasis may be one factor that disposes administrators, as suggested by Oliver, Crayton, Holland, and Hall (2000) to development and support of services research that entails more descriptive, qualitative, and vivid depiction of effectiveness (see also Anderson, 1999, indicating features of evaluative research conducive to public service consensus processes and administrative priorities) rather than more critical analyses.

II. THREE ILLUSTRATIVE RESIDENTIAL SERVICES RESEARCH STUDIES

In the next section of this chapter we present methods and selected findings from three state-level services research projects. Specifically, they address: (1) staff turnover in group homes, (2) perspectives of residents in supported housing, and (3) individualized support services (ISS). The purposes of these studies overall was to advance administrative understanding of the nature of services provided, patterns of resulting utilization, and factors that differ among service settings that impede or enhance the stability and quality of services.

A. Staff Turnover in Group Homes: The Best Practices Research Project, 1986–1988

The Best Practices Research Project (BPRP) was a comprehensive study of employee, organizational workplace, resident, and service aspects of group homes in New York State serving fewer than 15 people each and certified as either community residences or ICF/DDs (intermediate care facilities for persons with developmental disabilities). This project resulted in a series of technical reports (Jacobson & Ackerman, 1989a,b, 1990a,b) some of which have been published as a journal article or book chapters (Jacobson & Ackerman, 1992a,b,c). The BPRP was commissioned by the New York State Committee on Workforce Enhancement and Productivity (CWEP), a labor-management relations committee within New York State government composed of state agency and union representatives. The study was instituted because of labor concerns about continuity of employment for state workers in the context of then-continuing deinstitutionalization and rapid growth of not-for-profit provision of group home services that exceeded the development pace for state-operated group homes. The Office of Mental Retardation and Developmental Disabilities (OMRDD) was approached to conduct the study and agreed to do so.

The portions of the BPRP reported here focus on staff turnover. Although the specific impacts of turnover on staff of developmental services, residential programs of any type had not been measured at the time the BPRP began, researchers had concluded from the organizational literature and experience that high turnover will result in an array of adverse organizational outcomes in intellectual disability facilities and other human services settings (George & Baumeister, 1981; Price, 1977; Wandersman & Moos, 1981; Zaharia & Baumeister, 1978). However, these perspectives remained untested in group homes by objective data or formal evaluation measures at the time of this study (Burchard & Thousand, 1988; McCord, 1979).

A broad range of turnover rates had been reported in the literature for group homes, and Lakin and Bruininks (1981), who studied a random national sample of 104 group homes, observed a mean annual turnover rate of 54.2%. In small group homes (serving 16 or fewer persons), mean annual turnover rates were 61% (public) and 54% (private).

Studies of turnover among group home staff had often concentrated on the work situation and management issues. For example, in their study of turnover rates in community programs in Massachusetts, Coleman and Craig (1981) posited that turnover was a function of compensation, recruitment, supervision, training, and job structure. In exit interviews of staff, the Developmental Disabilities Program (1983) in Minnesota found that the most prevalent reasons for leaving employment were personal reasons, to get higher pay, and job dissatisfaction. In Ohio, higher pay and opportunities for advancement were most frequently cited by group home workers as reasons for leaving their jobs (Fritz, 1986).

The Lakin and Bruininks (1981) national study found that, among group home staff who left their jobs, 49% quit for a better job, higher pay, or other job-related reasons; 19% left for family-related reasons; 16% were fired, and the remaining 16% left for other reasons. Separation was more closely associated with lower satisfaction with circumstances and supervision (extrinsic satisfaction) than with satisfaction with the work itself (intrinsic satisfaction). The best predictors of turnover for facilities were the county unemployment rate and the starting salary for staff indexed to the average income in the county. Higher unemployment rates and higher relative income were associated with lower turnover (see also Butterfield, Barnett, & Bensberg, 1966). Other complementary research on turnover had been conducted by that time (Bersani & Heifetz, 1985; Burchard & Thousand, 1988; Holburn & Forrester, 1984; Hom, Katenberg, & Hulin, 1976; Mobley, Griffith, Hand, & Meglino, 1979; Porter & Steers, 1973).

Although job satisfaction was implicated as a contributor to turnover, it was also clear that its impact could be mediated or possibly vitiated by other variables. Clearly, a wide range of other variables were implicated, including social environment (e.g., resident-oriented versus staff-oriented care practices), management (e.g., influence of staff in decision making), staff elements (e.g., attitudes toward the work and residents), resident factors (e.g., psychological well-being), and service-related factors (e.g., content and quality of individual service plans, participation in community activities). These variables also reflected a wide range of concerns that were to be considered within the scope of the BPRP, and analysis of the relation of these personal and environmental states and characteristics to personnel turnover was a natural focus of analysis within this data set, and within this diverse sample.

Participants for this study included the staff, mangers, and residents of the group homes. At that time, April 1, 1986, there were 1274 group homes in operation in New York. Participation by employees and residents in publicly and privately operated programs was voluntary, and informed consent was obtained for resident interviews. The questionnaire completion methods assured anonymity of response and structured to indicate that consent was implicit. Within the initial sample, alternative sites were obtained in a total of eight instances, by random sampling within the same cell of the frame. One site provided grossly incomplete data and logistics prevented visits and interviews at another site, and these were dropped from the study (one private and one public community residence). The 38 programs served a total of 298 residents with developmental disabilities ($M = 7.8$); residents had an average of 3.3 functional limitations, on a scale ranging from 0 to 7.

In each of the respondent programs data were collected from managers (1 per site), from direct-care staff (from 2 to 15 per site), and from residents (2 per site), using questionnaires, interviews, and information abstracted from residents' records. Both standard research instruments and purpose-constructed research instruments were used in the questionnaires and interviews.

Variables were classified as predictor, control, or outcome variables. Turnover rate was the predictor variable, defined as the full-time equivalent number of staff who left the program during the 12 months preceding or following questionnaire completion, divided by the number of full-time equivalent staff. The number of staff who left the program was determined by the manager through a personnel-record review. The general design entailed correlating the rate of prior turnover from Time 1 to Time 2 with outcome variables measured at Time 2 to predict impacts of turnover, and the rate of subsequent turnover from Time 2 to Time 3 with outcome variables measured at Time 2 to predict precursors of turnover. Outcome variables corresponded to domains of social environment, management practices, staff, and resident services and activities.

In May of 1996 agencies were contacted and agreed to participate. Completion of questionnaires preceded site visits, which were 2 days in duration and were conducted by interviewers employed solely for this purpose. The interviewers were registered professional nurses and students recruited from undergraduate and graduate programs in psychology and special education. All interviewers had previous developmental disabilities service experience, were trained to complete all protocols by the evaluators, and were briefed on the purpose and dimensions of the study by the evaluators in a 3-hour training session. Each interviewer conducted five separate, individual, interviews at each site, with two residents, two staff, and the manager. During the visit the interviewers also reviewed and abstracted clinical records and collected completed questionnaires.

To examine the precursors and impacts of turnover, Pearson product moment correlations and partial correlations controlling for type of program, operator, staff tenure, and resident functional limitations were computed. For site and program variables the site was the unit of analysis ($N = 38$ maximum). For other variables the unit of analysis was staff and managers interviewed, staff and managers who completed questionnaires, or residents. Correspondingly, statistical power was greater for the latter analyses, and greater for assessment of impacts (in relation to prior turnover) than for precursors (in relation to subsequent turnover). Specific predictions were made for a wide range of variables but are not detailed here.

1. PRIOR TURNOVER

In total, 269 staff completed questionnaires, including 38 managers, representing 76% of all staff and 100% of the managers. Overall, 45.0% of staff had held their positions for less than 2 years, and 34.9% of the largely (74%) female staff were age 29 years or younger, and 37.9% had a high school education (Jacobson & Ackerman, 1992a for further information).

The average prior 12-month turnover rate was 34.9%, with a range of 0% to 181% (SD = 38.3). In four sites no turnover (0%) occurred in the prior year, and in three sites the rate was in excess of 100%. Primary reasons for staff departures, documented in the course of manager interviews, were 55.1% to take another job in developmental disabilities services, 15.8% fired, 14.0% to take another job in other services or the for-profit sector, and 11.4% for family or economic reasons.

2. SUBSEQUENT TURNOVER

Subsequent turnover was determined for 21 programs that provided follow-up data, dispersed about equally among the sampling strata. These programs represented 21 managers and 139 staff who had completed questionnaires. Eighty-three staff had left their jobs during the year following the site visits. The average subsequent turnover rate for these homes was 45% (SD = 51), with a range from 0% to 214%. The overall correlation between prior and subsequent turnover was $r(19) = .65$ ($p < .01$), with $r(7) = .70$ ($p < .02$) in community residences and $r(7) = .75$ ($p < .01$) in private programs; these turnover rates were uncorrelated in ICF/DDs or public programs. Among the staff who departed their positions during the year, 82% were full-time employees. They left primarily to begin work in other services or the for-profit sector (38%), transferred to another location in the same agency (28%), or were fired (12%). Altogether, 72% of reasons for departure reflected loss of staff to the service system. The job performance of departing staff was often rated as above average (28%) or average (41%) compared to coworkers.

In order to consider staff stability, the ratio of staff departures to new hires was calculated for day, evening, night, and rotating shifts, and for "other schedules." These calculations indicated that, relative to night and rotating shifts, stability of day, evening, and other schedule staff was relatively low. Departing from one's position was associated with working in a private program or community residence and with working other than day shift. Lower tenure, younger age, and greater formal education were associated with leaving, as was working with residents with fewer functional limitations. Leaving was also associated with lower social support from one's manager, but also with greater extrinsic satisfaction, and uncorrelated with intrinsic satisfaction, commitment, staff attitudes toward residents, or job characteristics ratings.

3. CORRELATES TO PRIOR AND SUBSEQUENT TURNOVER

Partial correlations between variables and prior and subsequent turnover rates were conducted separately for private and public programs, and for ICF/DDs and community residences. For private and public programs, partialling controlled for program type, resident functional limitations, and staff tenure. For ICF/DDs and community residences partialling controlled for operator, resident functional limitations, and staff tenure. Turnover was uncorrelated with social environment scale scores from the Group Home Management Schedule. Findings with respect to all variables were summarized in the project technical report. Here, selected findings are highlighted.

For private programs, higher prior and subsequent turnover were both associated with lower skill variety, less feedback from completion of work tasks, greater intensity of feelings of depersonalization by staff, and decreased feelings of accomplishment by staff, reflecting job characteristics and burnout.

Although there were seven correlations between variables and both higher prior and subsequent turnover for public programs, five of these were inverse relationships (e.g., positively related to prior turnover and negatively related to subsequent turnover). These five variables were commitment to the job, positive job attitudes, task identity, number if ISP goals, and quality of ISP goals, reflecting job characteristics and service planning. The two exceptions to this pattern were service adequacy indices: lower adequacy of health and therapy services and of community support were both associated with higher prior and subsequent turnover.

The only variable consistently associated with prior and subsequent turnover in ICF/DDs was extent of unmet needs for health and therapy services. Counter to intuition, higher turnover was associated with the presence of

fewer unmet needs for therapeutic services. It is possible that this finding reflected higher turnover in settings in which fewer service needs were effectively identified, or which provided more extensive services to all residents.

Several variables were consistently associated to prior and subsequent turnover in community residences. Increased day-to-day operational problems, lower social support by the manager for staff, lower significance of work tasks, and greater unmet needs for both health and therapy services and community support were associated with higher staff turnover both before and after the site visits.

4. SUMMARY

Subsequent turnover was related to more variables than was prior turnover. This finding may be attributed to selection of measures based on previous research, which had focused primarily on the relationship of program characteristics to subsequent turnover, rather than on impacts of prior turnover. Notably, extrinsic and intrinsic satisfaction and social support from one's supervisor were more consistently associated with subsequent turnover than with prior turnover. Job characteristics were, expectedly, associated with both prior and subsequent turnover, but generally the characteristics evidencing relationships to prior turnover differed. Compared to job and work characteristics, burnout of staff appeared less closely related to turnover. Resident satisfaction measured by series of facial expression icons anchoring a five-point Likert scale showed some relationships to staff turnover, but neither relationships were found between turnover and a composite scale of resident satisfaction nor a scale of resident self-rated well-being. Resident positive descriptions of the home where they lived were related to prior turnover in ICF/DDs and public homes, and to subsequent turnover in private homes.

A variety of general conclusions were possible from this study, with the qualification that degrees of freedom were limited for some analyses: (1) depending on whether the site, manager and staff interview, manager and staff questionnaire, resident interview, or clinical record was used as the unit of analysis and (2) sites studied with respect to subsequent turnover were a subset of those for which prior turnover was measured. Nonetheless, even with reduced power for analysis of subsequent turnover a greater number of variables evidenced relationships to subsequent than to prior turnover. Degrees of freedom and statistical power were further diminished by studying subsets of residences, by type of operator or certification. It is also important to note that numerous correlations were conducted and no correction factor was applied, because the purposes of the study were descriptive and exploratory, at least with respect to impacts of prior turnover. On the other hand, power was also reduced in that the obtained relationships were

identified after influences of type or operator of site, extent of resident disability, and staff tenure (variables that had been previously identified as related to turnover in various studies) had been partialled out. In particular, tenure was found to be related to numerous study variables in separate analyses (Jacobson & Ackerman, 1992a).

Turnover rates were highest for private programs and community residences, but persistent high turnover was not detected in the homes that were studied, in that prior and subsequent turnover were unrelated in the subset of settings for which both were measured. Of particular interest were relationships of prior and subsequent turnover to ISP measures and measures of service and community support adequacy, the latter reflecting the sum of listings of possible service or support needs that had not yet been addressed and were collected annually as part of the DDIS database. These findings suggested that there were compelling service quality, as well as workplace or work life issues presented by higher staff turnover, a concern of relatively minor focus in the previous research on staff turnover. Management and service delivery implications of the study findings were extensively interpreted and discussed in the original technical report. More succinct findings and guidance were provided and adapted for use by managers and clinicians in the field through follow-up briefs on management, staff impacts, and service delivery. Several internal memoranda were developed that further posed interpretations and recommendations for administrative consideration.

B. Resident Perspectives: The Supported Housing Evaluation, 1992–1995

In 1992 through 1995 the New York State OMRDD conducted a comprehensive evaluation of supported housing projects in New York State, funded as demonstrations by the Developmental Disabilities Planning Council (DDPC; Jacobson & Ackerman, 1993b; Jacobson, Schwartz, & Ackerman, 1995). The DDPC demonstrations were initiated because, as part of periodic hearings in which information was gathered toward development of the state's federal developmental disabilities plan, housing concerns were frequently raised, and supported housing and supportive (minimally supervised) residential settings were noted by some participants as a viable means of providing flexible, out-of-home care varying greatly in intensity. The dimensions of overall utility, suitability, and characteristic operations and impacts of supported housing had not been thoroughly studied at that time. As a consequence, the DDPC issued a request for proposals to operate supported housing project demonstrations from the provider agency sector and for an evaluation of these projects.

1. CONTEMPORANEOUS PERSPECTIVES ON SUPPORTED HOUSING

Although there was a long-standing literature on the use of supported housing by people with developmental disabilities, supported housing for this population had not become as widely available as more service-intensive forms of small community residential settings on either a state or national scale. Increasingly, however, states were moving toward individualized approaches to the provision of supports and services. Accordingly, supported housing with minimal supervision or a supportive advisory relationship between providers and participants was recognized as an important and distinct alternative within the broad array of individualized service approaches.

Sponsored community living with supports for people with disabilities in noncertified, (natural) settings had been identified as an important lifestyle option for people with physical disabilities (DeJong & Wenker, 1979), psychiatric disabilities (Anthony & Farkas, 1989), and people with developmental disabilities (Halpern, Close, & Nelson, 1986; Jacobson & Mulick, 1990, 1991; Schalock, Harper, & Carver, 1981). Within rehabilitation services, living alone or with a companion in regular housing with supports has been termed *independent living*. Jacobson and Mulick (1990, p. 5) described the equivalent developmental services model as "assisted independent living." Carling (1992, p. 20) referred to this lifestyle as "normal housing with supports." To distinguish the services and supports provided within the DDPC-funded demonstrations from these other alternatives, the designation of "supported housing" was adopted for these projects, underscoring that regular, or normal, housing with supports at least partially defines this lifestyle option. The model set forth for supported living stipulated that participants would not be supervised by others; rather, they would live in their own home or apartment, and lead an independent lifestyle, which might include counseling or training if needed and desired.

Although serving to support people with disabilities in their independence, or beneficial interdependence, supported housing was recognized to pose some concerns for family members and professionals. The existing literature suggested that many concerned individuals, including advocates, feared that people with developmental disabilities living in supported housing would be victimized or shabbily treated by others. These fears appear to have been largely unjustified, but there were some lifestyle features in supported living that remained. People with developmental disabilities living with their families aspired to supported living largely to gain freedom of choice, whereas their families often viewed the goal of supported living as a venue for skill development (Flynn & Saleem, 1986). One of the major familial and professional concerns that emerged was that people who enter supported housing

will become socially isolated, although it has been noted that these same individuals, whether living with family or in a community residence, are relatively isolated from social contact with people who are neither relatives nor paid companions (Catermole, Jahoda, & Markova, 1988; Dalgleish, 1985; Donegan & Potts, 1988). Thus, one rationale for not promoting more independent living for some people with developmental disabilities was the belief that in the group home residents had peers with whom to socialize.

Research by Edgerton (1988), Halpern et al. (1986), and Schalock et al. (1981) had shown that, despite some economic difficulties and "hard knocks," people with developmental disabilities who have been living more or less independently valued their independence and were relatively satisfied with their lifestyles (but see Brown et al. [1988] on valid differences of opinion on lifestyle priorities between people with disabilities and advocates or families). Such findings suggested that substantial investment in research is warranted to better understand both the risks and the benefits of supported housing.

2. APPROACH

Questionnaires to gather data for the evaluation were developed using protocols from the National Institute of Mental Health National Study of Supported Housing conducted by the Center for Community Change Through Housing and Support in Burlington, Vermont, and interview protocols from the BPRP and the Vermont Community Adjustment Study of the University of Vermont as basic resources (see Jacobson et al., 1992). The final protocols reflected a combination of the substance of those protocols and of primary concerns identified jointly by the DDPC and the NYS OMRDD Planning Bureau.

In the first phase of the evaluation, agency directors responded to questionnaires in 1991–1992, as did service co-ordinators (i.e., case managers). In addition, service co-ordinators provided information on participant characteristics at the same time, using the Developmental Disabilities Profile (DDP—Brown et al., 1986). Participants (consumers) were interviewed during early 1992, following granting of informed consent. Interviews typically took place at the main office or a satellite office of the agencies sponsoring supported housing projects, and with the exception of cases when manual communication, articulation difficulties, or primary language necessitated the assistance of a translator; interviews were generally conducted without agency staff present. Interviews were conducted by Qualified Mental Retardation Professionals (QMRPs), including two doctoral level QMRPs in psychology. Interviewers were employees of the central or district offices of the OMRDD.

In the second phase of the evaluation, agency directors and service co-ordinators again completed questionnaires. For participants who had been in supported housing during the first and second phases, the service co-ordinators completed abbreviated questionnaires (i.e., updates). For the participants who began to take part in supported housing during the second phase, the service co-ordinators completed the initial, more extensive, questionnaire used in the first phase. Consumer interviews were again conducted, with comparably trained and experienced interviewers including the primary evaluators. Agency director, service co-ordinator, and interview data were collected during the spring of 1993. Individual DDP information forms were completed for new participants during the second phase.

3. PARTICIPANTS

Sponsoring agencies reported serving a total of 253 people in the supported housing projects over the first 2 years. For the 2 years of supported housing services, sponsoring agencies reported information on the characteristics of a total of 139 participants using the DDP data system. Their average age was 33.2 years (SD = 10.5, range = 13–68 years). Altogether, 44% of the participants were female and 19% were members of racial and ethnic minority groups. The primary disability conditions reported were: intellectual disability (56.4%), learning disability (19.4%), cerebral palsy (8.3%), and other (15.7%). Other conditions included Apert syndrome, epilepsy, hearing impairment, neurofibromatosis, neurological impairment, spina bifida, and traumatic brain injury. Altogether, a history of seizure disorder, but not necessarily current seizures, was reported for 11.5%. Some participants had chronic medical conditions: 15.1% respiratory conditions and 18.0% cardiovascular conditions. Another 10.1% had some form of psychiatric disorder and 7.2% took antipsychotic or antidepressant medications. Another 4.3% took anticonvulsant medication and, in total, 20.1% took some other form of maintenance medication for medical reasons. In total, 27.3% of the participants evidenced one or more behavior problems monthly or more frequently, a low rate of occurrence in the overall population with developmental disabilities.

Just over one-half of the participants (53.3%) lived alone. Participants who did not live alone lived with an average of two persons, of whom 66.9% were also people with some form of disability. In 64% of the cases, the participant held the lease or contract for their housing. Generally, participants had incomes and benefits considered to be sufficient to pay for basic costs of living. Many were employed (62.3%). In the majority of instances (70.6%), the participants paid their rent entirely (using entitlements like SSI, SSDI, and Public Assistance). A small minority (9%) of participants used

some form of a Section 8 housing assistance certificate and some individuals (11.5%) received some financial assistance from relatives. The average total monthly income of participants, including entitlements, was US$ 678.39; an average of US$ 8140.68 annually, or 119% of the poverty level threshold (at that time) for a one-person household in New York. Twenty-one participants who had Medicaid coverage also had private health insurance.

4. PARTICIPATING AGENCIES

A total of 11 agencies were studied in this evaluation. Nine of the agencies were employing a "place-and-train" model for supported housing and two were using a "train-and-place" model. Projects were located throughout the state in urban ($N = 6$), rural ($N = 2$), suburban ($N = 1$), and metropolitan ($N = 2$) areas. Over the period of 1 month, case managers and other agency staff met face-to-face with participants an average of 4.5 times and for an average of 2.2 hours weekly. All agencies used some form of admissions or eligibility criteria for supported housing. The most highly developed admissions criteria involved 14 specific criteria, in addition to ability to use a telephone, ability to meet daily basic needs, and requirements for no more than 5 hours weekly of support services. Additional criteria employed by one or more agencies included being considered capable of functioning independently within 1 year of training, being at risk of movement from family or independent living to a supervised setting, being age 16 years or older, desiring a more independent lifestyle, or being a graduate of an agency's supervised community residence.

Interviews were conducted with 63 participants. They provided information about the extent of choice and influence by others in the selection of their residential situation, their satisfaction with where they lived and their work, their psychological well-being and social network, and services and supports that were desired, needed, and available to them. Some of the questions asked in interviews duplicated those to which case managers responded to obtain the consumer's point of view.

Participants generally indicated they felt they were doing "OK" (38.1%) or "pretty well or very well" (57.1%) in terms of day-to-day living in their apartments or homes. Although three-quarters felt that they had a lot of choice in selecting whether to live alone or with others, only about one-half felt that they had a lot of choice about where to live. Individuals who tended to feel that they had a lot of choice also felt that others exerted little influence over their decision. Nevertheless, about one-third felt that others had exerted a lot of influence over the selection of the specific place where they would live. In addition, almost half (44.3%) reported that their apartment or home was the only place they were shown when they were looking for a place to

live. The great majority (83.3%) reported that they received "just the right amount of help" in finding a place to live.

5. RESIDENTIAL AND WORK SATISFACTION

Participants reported that they never (25.4%) or only once in a while (52.4%) found themselves sitting around with nothing to do. However, 15.9% reported that they did this most of the time and 6.3% all of the time. On a measure of residential satisfaction, which was scored according to percent of scoreable responses that were rated positively, participants reported an average satisfaction level of 93.2% out of a possible 100% (SD = 7.0, $N = 60$). Similarly, the average level of satisfaction with work and day activities was high at 93.9% (SD = 10.4, $N = 60$). About 10% reported that they disliked where they lived greatly, and 16% reported that they disliked where they lived somewhat, with most citing either the size of their apartment or the character of the neighborhood as dissatisfactions. Only one participant reported the lowest level of work dissatisfaction, with 11% reporting that they disliked their work somewhat.

6. PSYCHOLOGICAL WELL-BEING AND SOCIAL NETWORKS

On a measure of psychological well-being that was scored by subtracting scoreable negative responses from scoreable positive responses and creating a percent of possible positive responses score, participants scored an average of 73.5% (SD = 17.6, $N = 62$), indicating, on balance, more positive than negative self-descriptions. However, there was a considerable range of scores with 11 persons rating themselves at 90% or above and 6 persons rating themselves below 50%.

Participants reported social networks consisting on the average of 4.03 people (SD = 2.21, $N = 64$). Members of these networks were balanced among relatives (Mean = 1.48, SD = 1.38), staff (Mean = 1.29, SD = 1.05), and friends/peers (Mean= 1.33, SD = 1.55). However, individual social networks were sometimes quite different from these means. Most social network members lived locally (83.3%), and only 3.4% lived in the same household. Participants reported lending or giving things to network members for only 33.2% of the people they mentioned. However, they reported giving emotional support to 68.3% of their network members. When asked how they felt about each network member, participants rated 86.7% of their network members positively, rather than mixed or negatively. When strong positive relationships, defined by positive responses to all three measures for the network member (lending, giving emotional support, and positive feelings), were considered, such relationships were present for only 19% of network members.

7. ASSISTANCE NEEDED AND RECEIVED

Table I displays participants' reports of: (1) various aspects of help needed, (2) whether it was provided, and (3) if it was enough. Generally, participants reported that they received enough help, with relatively high needs in dealing with upsets or crises, money management, personal hygiene, shopping, and cooking meals. In nearly all areas of help needed, more help was provided than participants identified as needing. The only area in which participants reported receiving less help than they needed was in dealing with upsets or crises, the highest reported area of need. A relevant portion of the Supported Housing Evaluation, not detailed here, compared case manager questionnaires to consumer interviews on help needed and help provided. These analyses showed that there were no significant differences, overall, between the extent that case managers and participants felt that the participants needed help and received the help that was needed.

Participants also responded to a set of supports that would potentially be helpful in assisting them in living independently in the community. As detailed in Table II, helpful supports for over 70% of participants were being able to reach staff by telephone day or night, income or subsidy improvements, transportation availability, regularly scheduled visits by staff, and money for deposits. Between 44% and 58% of all participants reported that they did receive these supports, suggesting some unevenness among agencies in terms of the extent to which subsidies and money for deposits were acquired for participants.

TABLE I
PARTICIPANTS REPORTS OF ASSISTANCE NEEDED AND PROVIDED

	Need help?	Someone helps?	Enough assistance?
Aspect of help	Yes (%)	Yes (%)	Yes (%)
Dealing with upsets and crises	64.8	25.4	92.1
Managing money	50.0	58.7	82.5
Shopping	44.3	55.6	85.7
Personal hygiene	38.7	47.6	84.1
Housework	35.5	46.0	88.9
Cooking meals	33.3	35.5	85.7
Laundry	27.4	31.7	92.1
Making friends	19.7	25.4	90.5
Managing medications	15.4	20.6	95.2

TABLE II
PARTICIPANT REPORTS OF HELPFUL SUPPORTS AND EXTENT OF RECEIPT

Potential supports	Helpful to you? Yes (%)	Received? Yes (%)
Reach staff by telephone day or night	79.4	58.7
More money/income/subsidy	79.4	49.2
Available transportation	74.6	52.4
Regular appointments by staff to my home	71.4	55.6
Have staff come to my home any time	68.3	47.6
Get money for deposits	71.4	44.4
Get a telephone	65.1	61.9
Have staff help me get benefits	61.9	58.7
Get furniture	55.6	54.0
Get household supplies	55.6	41.3
Have staff help me find roommate/housemate	31.7	15.9
Have staff live with me	11.1	1.6

8. SOCIAL AND RECREATIONAL ENGAGEMENT

More than one-half of the participants reported having visited a friend, dining out, or going for recreational walks during the preceding 2 weeks. Nearly one-third participated in a variety of other recreational activities. With the exception of dining out, and going to a movie, sports event, concert, or a club meeting (activities which are often attended in a small group), participants engaged in each of the recreational activities either alone or in the company of a companion at least half of the time. The overall pattern of recreational involvements suggested that participants had access to and made use of a range of community recreational resources, although not necessarily at a high frequency.

9. SUMMARY

The findings presented here reflect segments of a major project report (Jacobson & Ackerman, 1993a,b) in which agency directors and case managers also provided information about supported housing. Consumers' viewpoints of their supported housing experiences were provided by individuals who were receiving supported housing services in a total of 11 agencies in New York State. Just over one-half of the participants lived alone. About a quarter of the sample reported disliking where they lived, although reported levels of satisfaction were generally high across multiple life aspects, and these tended to be correlated highly with work experience.

With respect to social and recreational engagement, participants made use of a wide range of community recreational resources, although they tended to occur as a group activity and not necessarily at a high frequency. Participants reported social networks on average of about four people, within which the category of friends accounted for only 1.3 people on average. This average poignantly underscores a void in the lives of these individuals. Interestingly, 22% of participants were receiving training in making friends, whereas their case managers reported that 44% of participants needed such training. However, when considering various aspects of anticipated need, participants reported that they were receiving enough help, with the exception of the need for assistance in dealing with upsets or crises.

The findings from the supported housing evaluation project illustrate the types of information gathered that is often of general administrative interest and of specific interest to agency personnel engaged in system change activities. A series of additional reports, although not a full third-year report, was developed from this project, as well as management finding memoranda and graphic summaries of findings over the course of the project (Jacobson & Ackerman, 1993c; Jacobson et al., 1994). Furthermore, a detailed report was produced with respect to the full range of variables measured in this project, and mentioned earlier, contrasting first year participants to themselves during the first and second year of their enrollment and contrasting first year enrollees to second year enrollees (Jacobson et al., 1995). All reports that were issued contained management summaries several pages in length presenting key findings, and these summaries were distributed as freestanding documents as well as within reports.

C. An Early Review of Statewide Individual Support Services: 1994–1996

In 1992, the New York State OMRDD initiated Individual Support Services (ISS). In that year, according to Braddock et al. (1995), there were only 21 states offering supported housing services similar to ISS, serving only 14,779 people with developmental disabilities nationally, although these initiatives in some states dated back to the mid-1980s. As a support to the dissemination of ISS activities and Home and Community Based Services (HCBS) in New York State, individualized services practicums were held that year, across the state.

Individual support services were developed in New York to assist adults with developmental disabilities to establish their own households or independent living by renting or owning a home and were not intended to assist people with developmental disabilities who were living with their families. Although originally intended as supports for people who were not eligible

for the HCBS waiver, in practice, ISS could be used to fund supports for people taking part in HCBS services, such as rent subsidies or particular vocational supports that were not reimbursed through the HCBS waiver. ISS was targeted to address several converging needs: (1) to provide supports for people with developmental disabilities for whom supervised living was not indicated; (2) to accommodate selected needs for people who required and desired a small amount or narrow range of supports; (3) to enhance choice regarding residence, supports, and work place and colleagues; (4) to avoid provision of unwarranted and unneeded services; and (5) to increase use of appropriate, existing generic, nondevelopmental disabilities sector, entitlements, and financial supports. The common strand running through these uses was specific support for the development and continuation of living in regular housing with supports. At the time of the ISS evaluation (1993–1996; Jacobson & Ackerman, 1996), findings regarding independent living and related supports for people with developmental disabilities had also been presented by Halpern et al. (1986), Lozano (1993), Schalock and Genung (1993), and Schalock et al. (1981).

1. DEFINITION OF THE ISS PROGRAM MODEL

When ISS was implemented in 1992, guidance materials were issued that defined major features and administrative expectations for ISS. The underpinning principles of ISS included the aspirations that individuals should choose where and with whom they live, select their own services, and have access to integrated, safe housing. Housing and services should be separated to allow individuals to live in housing of their choice.

Each ISS participant had a case manager, with a wide range of duties including implementation of a support plan that reflected individual choice, identification of service providers, assistance in housing negotiations, resource management, technical assistance to landlords, and access to generic community supports and services. The support plan was to address needs such as rent, utilities, telephone, insurance, transportation, food, clothing, leisure and recreation, health, staffing, employment and education, and savings. Agency management provisions included a plan for 24-hour emergency coverage, as well as housing safety. The provisions also assured that generic benefits and entitlements were optimized, that services were appropriate and of high quality and that participants have input into the agency's ISS program and their own individual support plans.

2. THE REVIEW APPROACH

Provider agencies were originally solicited for participation in this evaluative review based on their ISS service provision status as of November, 1993. At that time there were 76 organizations, including local offices of the state

agency that were providing ISS. Of these, 18 agencies (24%) served 10 or more people through ISS. A letter of invitation to participate in the evaluation was sent to each of these agencies; 12 providers agreed to participate.

Participation entailed: (1) completion of a participant roster, (2) completion of DDP forms (Brown et al., 1986) for participants not previously registered in the DDP individual client database, and (3) provision of information on previous and current participants, their services and associated expenditures, and a description of features of the ISS approach by the executive or ISS director of each agency. In addition, agencies completed questionnaires and facilitated face-to-face participant interviews conducted by research assistants. Prior to the interviews, informed consent was solicited and obtained from all 102 participants.

3. PARTICIPANT CHARACTERISTICS

Case mangers reported that the average age of the 250 participants was 34.4 years (SD = 12.5, range = 4–72 years), and 52% of participants were female. The current living situations of participants were a single family house (23%), two family houses or duplex (20%), apartment building (37%), or apartment complex (19%). At the time of the evaluation, on average, participants had lived in their current type of living situation for 23 months and been involved in ISS for 10 months. Developmental Disabilities Profile data indicated that most participants had mild intellectual disability or neurological impairment, and very small proportions were reported with cerebral palsy, epilepsy, or as using wheelchairs.

4. CASE MANAGER AND AGENCY DIRECTOR REPORTS

Generic (nondevelopmental disabilities sector) human services were used at relatively high rates by participants in ISS. Case managers reported that one-third of the participants had received social services, one-fifth had received mental health services, and one-sixth had used special transportation, within the past month. With respect to health care utilization, 14% each visited a clinic or used personal care services, 12% used emergency room services, and 7% made use of public health services.

Agency directors were asked to indicate the needed generic supports that were otherwise available but that were nonetheless paid for with ISS because of barriers to access. The generic supports they listed primarily were rental subsidies, getting qualified for or services from Medicaid or SSI sources, accessibility and availability of transportation, and secondarily involved basic health, safety, adjustment, and protection concerns. In most instances the barriers clearly reflected delayed access to supports, in that the supports were not available when needed. Case managers reported that there was a wide range of support needs that were not addressed due to delays in access to

generic services, but generally these needs were infrequent and idiosyncratic to the individual. Most frequently, these entailed needs for transportation.

Case managers reported that from 90% (managing money) to 40% (making friends) of participants did not require training in 11 areas of activities of daily living or personal skills. Although more than three-quarters of participants required either training or assistance with shopping, dealing with crises or upsets, and managing money, less than one-half required such supports with respect to making friends, managing medications, or personal hygiene. For each ADL or personal skills area, about equal proportions of participants were receiving training or receiving assistance, possibly indicating the training capacity of provider agencies. Virtually all people who evidenced training needs either were involved in training or received continuing assistance with these activities.

The evaluation also investigated the nature of the living situations of ISS participants prior to receiving ISS, and for those who departed during the study period, the nature of their subsequent living situations. Prior to ISS, about 30% lived with family, and about 40% lived independently, with the remainder having lived in various supervised settings, including state facilities or ICFs-MR. Among those who discontinued participation in ISS, 58% subsequently lived independently and 23% lived with family. A small number moved into small, supervised settings, but not into congregate care (i.e., state facilities or ICFs-MR).

Case managers also reported whether a variety of training services and professional services were paid for through ISS, provided by their agency, or provided by another developmental disabilities or generic agency. Only recreational therapy, leisure activities, or lifestyle and social supports were provided to more than 40% of the participants. Eighty percent of the participants engaged in recreational or leisure activities that were provided by the ISS agency (regardless of whether these were reimbursed through ISS mechanisms), whereas 40% were provided with activities by generic agencies (indicating multiple sources of activities for some participants). Of the subgroup that did receive lifestyle and social supports, 75% received these supports from the ISS agency and seldom from another developmental disabilities or generic agency. Social skills training was provided as an individual support service for about one-quarter of the participants, and three-quarters of these people received this service from the ISS agency. The only other support services provided to more than 10% of participants and paid for via ISS were self-advocacy training and personal care.

Less than 5% of participants received adult education, rehabilitation counseling, physical or occupational therapy, psychological services, or speech therapy that was reimbursed by ISS, and overall, participants seldom used these services. To the (generally low) extent that participants used services from other

developmental disabilities agencies, these usually involved recreational or leisure activities or vocational supports (at 8–9% of participants). With the exception of recreational or leisure activities, counseling, adult education, and psychological services (range of 44–8%), less than 8% of participants made use of training and professional services provided by generic agencies.

Participants relied heavily on federal benefit programs including Medicaid (76%), SSI (63%), and Food Stamps (59%). Between 20% and 30% received SSDI or supports from lead state vocational rehabilitation agency. Between 10% and 20% received benefits such as supplemental utility payments, Medicare, or Public Assistance. Less than 5%, however, made use of the family reimbursement/voucher program, community development block grant supports, or Aid for Dependent Children (AFDC).

With regard to vocational activities and finances, case managers reported that 38% of participants did not work and that 44% were engaged in sheltered or supported work. Less than 5% were full-time students, and less than 15% were competitively employed or occupied as homemakers. Among those who worked, participants worked an average of 25 hours weekly for an average of about US$ 160 per month. A range of 80–90% of participants were reported by case managers to usually have sufficient funds to cover basic living expenses such as medical care, food costs, housing costs, and clothing costs. However, one-third usually did not have enough money to cover basic transportation costs or basic costs for social activities.

Case managers reported on contacts with the participants during a 1-month period. Typically, case managers had contact with participants nine times during the month (five in person, four by telephone), and spent 10 hours monthly with each participant. About one-third of these contacts were planned. Case managers reported that the current living situation was safe for about 90% of participants. When the situation was judged to be unsafe, this characterization was based on considerations such as inadequate or inappropriate housing, an inhospitable or risky environment, an unsafe neighborhood, or, in three instances, an abusive or potentially abusive social environment in the living situation.

5. PARTICIPANTS' REPORTS OF BENEFITS AND EXPERIENCES

More than one-half of the respondents indicated that someone helped them in each of six aspects of activities of daily living, whereas about one-quarter or less reported that someone helped them with getting around to places. More than half reported that they required either *some* or *a lot of* assistance with managing medications, cooking meals, dealing with upsets, or shopping. More than half of the respondents reported that they did not need assistance with housework. Participants indicated overwhelmingly that

they got enough help with managing medications, housework, laundry, cooking, and getting around to places. Fewer than 50%, however, reported that they got enough help dealing with crises, shopping, or making friends. Respondents indicated generally there were no basic living expenses for which they were unable to pay. But meaningful proportions, between 10% and 20%, reported that they sometimes had problems paying for social activities and needed food, telephone use, and transportation.

More than one-half of the participants indicated that transportation was available, they could reach staff by phone or request that they could come to the home as needed, or they could reach friends when their assistance was needed. Fewer than half reported that they had friends who they could depend on for social support. Fewer than one-fifth reported that they were required to come to staff offices for appointments or that they usually had problems getting to the places they wanted to go.

Almost all participants had landlords, and 90% of them got along with their landlords well. Similarly, virtually all of the participants had case managers, and 95% got along with them well. About two-thirds of respondents wanted to continue living where they were living at the time of the interview. About two-thirds of the respondents reported having a job, although slightly over one-third of these people would have preferred to work at a different location and about 40% would have preferred to be engaged in a different kind of work.

About 9 of 10 respondents reported that they were doing OK or Well in terms of day-to-day living. Ninety-five percent indicated that they felt safe where they lived, either all or most of the time. However, one-quarter reported that they had little choice in selecting the places that they moved into, almost one-third reported little choice over selecting the neighborhoods that they moved into, and one-sixth indicated that they did not get the amount of help they needed in finding a place to live. A substantial proportion (27%) reported that they found themselves sitting around with nothing to do either all or most of the time.

Among respondents who said they would prefer to live elsewhere, about one-third of the reasons had to do with relocation to a different town, village, or city that was close to work or resources. About 10% of the reasons each had to do with moving to a different neighborhood, a bigger or smaller apartment, a certified but nonrestrictive setting, or to live with companions or different companions.

6. AGENCY DIRECTOR REPORTS OF ISS OPERATIONS

Agency directors reported that staff training to support ISS activities was provided in the areas of innovative housing practices, person-centered planning, and legal aid, social security, and financial issues. Other topics

addressed in preparation of staff for ISS were lifeline services, community action, alcoholism, mental health, and sexuality issues.

The strategies that agency directors employed to enhance the community integration and participation of people engaged in ISS were skill-building activities, encouragement, support groups, peer social activities, and activities involving actual community presence. Generally, providers tried to capitalize on participants' interests in developing community presence; they sought to engage participants in use of neighborhood stores, generic transportation, community events, and linkages to individuals and existing community social and recreational groups. Examples of opportunities involved volunteering in Head Start, with Literacy programs, or at Nutrition sites, providing private home care for others, and enrolling in CPR training.

Agency directors also indicated which supports were likely to be received under ISS. Staffing, rent subsidies, and telephone and utilities subsidies were the supports that were most commonly received. In addition, transportation assistance, help paying for food, and household management were provided to 10% or more of current participants. Although costs for insurance (e.g., renters' or homeowners' insurance), health-related care, employment or education, and personal savings had been allowed in the guidance materials for ISS, less than 4% of current or previous participants received any of these types of supports, at least through ISS.

Providers were required to complete a quality assurance checklist twice annually under the requirements issued by the state developmental services agency. Agency directors indicated that their quality assurance efforts went far beyond these requirements, entailing use of applicable incident review procedures, case manager involvement or review on a quarterly, monthly, or as needed basis, and involvement of multiple personnel in activities with direct implications for quality improvement and assurance.

7. SUMMARY AND CONCLUSIONS OF THE ISS REVIEW STUDY

The primary purpose of ISS was to provide specific support for the development and continuation of living in regular housing with supports. The range of annual costs per person was from US$ 4101 to US$ 5316. Most agencies used ISS to support people with mild, but real, functional limitations, whereas a few used ISS to serve people with more severe functional limitations. Participants ranged from preschoolers to people in their 70s. Training was necessary for activities of daily or independent living, sometimes by very few participants and with some activities, by a majority of participants. Virtually all of the people who evidenced training needs either were involved in training in progress or received continuing assistance with these activities.

Generally, participation in ISS tended to encourage greater independence, with increases in the proportion of people engaged in individual residential arrangements, family care living, and, especially, engagement in independent living. ISS participation, however, did not systematically lead to individual movement from more restrictive and expensive services to less restrictive and less expensive services. This finding reflects the manifold uses of ISS, ranging from selected to comprehensive supports in the individual case.

Generic human services were used at relatively high rates by participants in ISS. More than one-half of the participants were enrolled in Medicaid, or in SSI, or received Food Stamps. With respect to professional services, only recreational therapy or leisure activities or lifestyle and social supports were provided to more than 40% of the participants. Participants reported that they generally had sufficient funds to cover basic living expenses. They indicated that there were no basic living expenses for which they were frequently unable to pay, but from 10% to 20% reported that they sometimes had problems paying for social activities, and for food, telephone use, and transportation. Most frequently, respondents reported being engaged in some type of work at community human services, vocational, or developmental disabilities agencies. The low rate of competitive employment in the sample was considered to merit further exploration by provider agencies because the profile of functional skills of many ISS participants suggested that they had well-developed skills compared to their peers with developmental disabilities. Case managers reported contact with participants nine times during a month (five in person, four by telephone), and spent 10 hours monthly with each participant. This finding appeared to indicate that a ratio in the range of 1 case manager to 16 participants in ISS would permit an appropriate level of involvement by case managers in supporting independent living through ISS.

ISS was used to compensate for delays in getting other supports. The generic supports that were acquired using ISS because of delays were primarily rental subsidies, getting qualified for or services from Medicaid or SSI, accessibility and availability of transportation, and secondarily involved basic health, safety, adjustment, and protection. Although findings indicated that ISS was a viable means to compensate for delays in delivery of supports, they also pointed to the need to address the system problems that perpetuate or exacerbate these delays.

More than one-half of the participants reported that they required either some or a lot of assistance with managing medications, cooking meals, dealing with upsets, or shopping. Less than one-half, however, reported that they got enough help dealing with crises, shopping, or making friends. Generally, however, participants tended to report that they received insufficient amounts of the services that they most commonly needed. This pattern

suggested that idiosyncratic, personal needs were relatively well addressed, but that training shared by many participants needed to be provided more intensively or broadly (e.g., increased development of social networks) than was currently the case.

More than one-half of the respondents indicated that transportation was available, they could reach staff by home or have staff come to the home as needed. However, less than half reported that they had friends who they could depend on for social support. This finding underscored that although agencies did specifically address socialization concerns in their ISS-related activities, for a substantial proportion of participants these efforts had not yet been successful in garnering meaningful social support from other community members.

A large minority of participants would have preferred to be engaged in different types of work or jobs than they were at the time of the interviews. One-quarter reported that they had little choice over selecting the places that they moved into, almost one-third reported that they had little choice over selecting the neighborhoods that they moved into, and one-sixth indicated that they did not get the amount of help they needed in finding a place to live. Over one-quarter reported that they found themselves sitting around with nothing to do either all of the time or most of the time. Although these findings suggest that most participants had choice in important decisions they also suggested that agencies needed to work further to enhance substantive individualization and choice making for ISS participants.

III. DISCUSSION AND FINAL REMARKS

This chapter presents excerpts from three state-commissioned research projects in the area of residential services in the developmental disabilities sector. This type of research, conducted for administrative purposes, is contrasted with the more widely circulated published services research typically found in journals. In recent years, a number of researchers, within the latter format, have attempted to link more closely issues that reflect policy aspirations in developmental services (e.g., choice and individualization) to processes of care, treatment, and socialization at the individual level and at the point of service, as well as to organizational factors and costs of services (Becker, Dumas, Houser, & Seay, 2000; Emerson et al., 2000; Felce et al., 2000; Hallam et al., 2002; Hatton et al., 1999, 2001; Rhoades & Altman, 2001). These types of studies have likely increased administrative interest in applying findings from services research within state and provincial systems, but it is still the case that very few studies address service and development processes in the same terms, and in the context of the same constructs, that

are components of everyday administrative and managerial decision making in ID services (Green et al., 1993; Parsons et al., 1989; Reid et al., 1991, 2001).

It is difficult to estimate the impact of statewide services research relative to the more general published services research. All services, of course, take place at the local level, the focal point of the administrative investigations, yet such investigations are at least partially influenced by findings from more general studies that have implications for service design, implementation, and even the mission of the state and local agencies. No doubt, the local research projects inform and influence the policies and practices in the targeted region within which they are undertaken, but much of this research might have implications that extend beyond the scope of the local project. Such studies, for example, likely hold administrative utility for other states seeking to answer questions about efficient and effective allocation of resources, despite the other states' differing configurations of funding, services, and organization. Unfortunately, such research may be difficult to identify and locate. One promising outlet is the Internet. For example, the Administration on Developmental Disabilities website provides links that connect to state developmental disabilities planning councils and to funded projects of national significance. A sampling of state council websites suggested that these sites contain a preponderance of policy and advocacy publications that are readily accessible and that in some instances services research reports are also available.

Another consideration that might explain why commissioned research findings may not be well disseminated pertains to the gap between the progressive aspirations generated by the changing philosophies of care in the field of developmental disabilities and the existing status of service provision. Researchers have consistently stressed that there are marked discrepancies between publicly stated aspirations for delivery of services and the substance of service delivery as it occurs. In addressing the nature of this discrepancy, they have reached some stark conclusions that are not consistent with the broadly positive portrayal of progress and status of services common to representations by public agencies to legislators and constituents in public-agency publications. Parmenter (1999) argued that social policy in disability services is far ahead of our current capacity to achieve the outcomes promised by the policies (p. 322). This perspective was echoed by Bigby and Ozanne (2001) who stated "The field is characterized by major gaps between policy and practice and high levels of unmet need" (p. 179). In their discussion of services in Victoria, Australia they noted that "People with intellectual disability are more often located in communities, but physical presence seldom equates with social connectedness and community inclusion" (p. 179). Such assertions do not constitute condemnation

of community services or vitiate our efforts as a whole to advance the quality of life of individuals with developmental disabilities. However, despite the immense progress in developmental services and the responsiveness of publicly funded services both in the United States and internationally, such assertions do indicate that much more remains to be done to improve services. They also indicate the differences in the tenor of conclusions that might be drawn from publicly disseminated and administratively based analyses. It is not unusual for researchers in developmental disabilities to call attention to the gap between the aspirations and the realities, but note that such assertions are typically found in professional journals, not the more locally oriented services research. Perhaps if the latter were as strident in illuminating the gap between policy and services in residential research, the contrast would be less discrepant.

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The Measurement of Poverty and Socioeconomic Position in Research Involving People with Intellectual Disability

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In this chapter, we argue that our understanding of the health, well-being, and life experiences of people with intellectual disabilities would be enhanced significantly if we paid more serious attention to issues relating to their socioeconomic position (SEP), including their exposure to poverty. We then go on to discuss ways in which the measurement of SEP/poverty can be incorporated in research undertaken with people with intellectual disabilities. First, however, we briefly review current knowledge about the association between SEP and health, well-being, and life experiences among the general population.

I. PERVASIVE NEGATIVE IMPACT OF LOW SEP

We know from research in high-income societies that the chances of living a long and healthy life are related to socioeconomic position (SEP), to an individual's place in the social hierarchies built around education, occupation, and income. In these societies, educational qualifications mediate access to paid employment which, in turn, is the major source of income for individuals and families.

Figure 1 uses occupation (manual/nonmanual) to map absolute inequalities in men's health in five countries in Europe. It shows the probability of dying between ages 45 and 65, a probability higher in all countries in manual when compared to nonmanual occupational groups. Figure 1 also makes it

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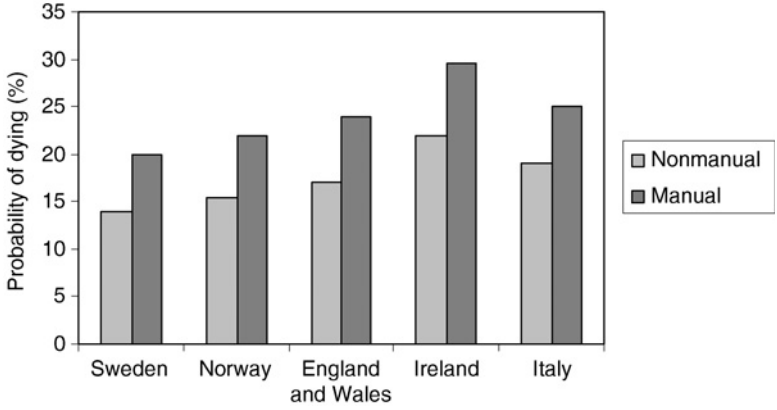

FIG. 1. Probability of dying between ages 45 and 65 among in manual and nonmanual groups in five European countries (source: Kunst et al., 1998).

clear that while health inequalities are ubiquitous, both poorer and richer groups endure poorer health in some countries than others. For example, although Sweden and Britain match each other in terms of national wealth (as measured by gross domestic product per person), the probability of dying for manual and nonmanual occupational groups is higher in England and Wales than in Sweden (Kunst, Groenhof, Mackenbach, & EU Working Group on Socioeconomic Inequalities in Health, 1998).

While most health research is focused on high-income societies, 80% of the world's population lives in middle- and low-income countries. In the poorest countries like Zambia and Malawi, life expectancy is below 40 years. It is appreciably higher in middle-income countries like India (62) and China (70) but still below the levels reached in the United States and United Kingdom (77), Sweden (80), and Japan (81) (World Bank, 2003). Shorter lives are often those spent in poor health and with more disability. In sub-Saharan Africa, the region of the world with the lowest life expectancy, it is estimated that 18% of total life expectancy is spent living with disability, whereas in countries where life expectancy is high, around 8% of the average life span are years spent living with disability (Mathers, Sadana, Salomon, Murray, & Lopez, 2000).

With a large proportion of the population engaged in subsistence agriculture and the informal economy, standard measures of SEP, such as education and income, are less useful in lower income countries. However, where data are available, marked socioeconomic inequalities in health have been found. Mortality rates among young children in low- and middle-income

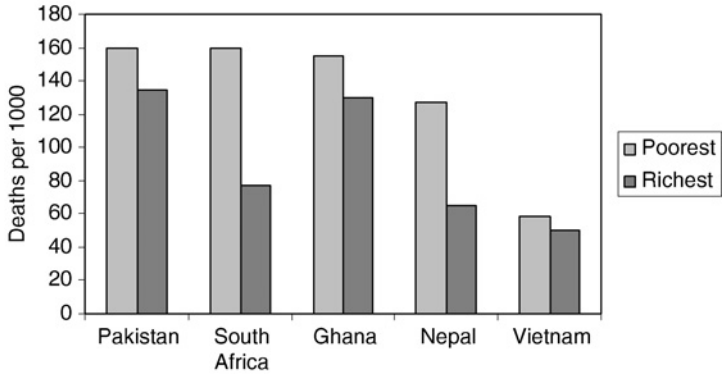

FIG. 2. Under-five mortality (deaths per 1000) in poorest and richest quintile in low- and middle-income countries (source: Wagstaff, 2000).

countries provide an example (Fig. 2). As in Fig. 1, both the absolute levels of health and the magnitude of health inequalities vary among countries. Inequalities in health are matched by inequalities in access to the determinants of health, with publicly-funded health care failing "to reach the poor in almost all developing countries" (Wagstaff, 2002, p. 102). A study using the international poverty line (living on <US$ 1.0 a day/US$ 1–2 a day/US$ >2 a day) revealed clear income gradients in health risks, with poverty associated with child malnutrition and limited access to water and sanitation (Blakely, Hales, Kieft, Wilson, & Woodward, 2005).

Health inequalities persist across time as well as across place. Historically, the process of industrialization has been associated with changes in patterns of disease. In nineteenth century Britain, for example, infectious and nutrition-related diseases, spread by lack of access to clean water, sanitary housing, and an adequate diet, took the heaviest toll on poorer urban communities. Improvements in sanitation, living standards, and diet reduced the burden of mortality from these diseases, with the result that life expectancy rose. However, mortality from noninfectious diseases like cardiovascular disease and lung cancer increased, with mortality initially higher among affluent groups. But, in a pattern repeated across high-income societies, the socioeconomic profile of these chronic diseases has changed, as risk factors like smoking and obesity increasingly become markers of poverty.

In poorer countries, infectious and nutrition-related diseases are still major causes of death, including malaria, measles, tuberculosis, and HIV/AIDS. But the drive for economic growth has disrupted traditional food systems and led to increased consumption of processed foods and manufactured cigarettes.

The result is "a double burden of disease," with high rates of acute, infectious diseases persisting alongside an increase in chronic noninfectious diseases (Monteiro, Moura, Conde, & Popkin, 2004; Yusuf, Ounpuu, & Hawken, 2004). As in richer societies, chronic diseases initially appear as diseases of affluence. But there is evidence that as poorer countries get richer, the socioeconomic profile of risk factors, such as smoking and obesity changes, with higher rates becoming apparent in lower socioeconomic groups (Blakely et al., 2005; Efroymson et al., 2001; Monteiro et al., 2004).

As this historical perspective suggests, the link between SEP and health is maintained even when new risk factors emerge and new diseases take hold (Link & Phelan, 1995). How can such persisting health inequalities be explained?

One line of inquiry has examined whether health-related social mobility is the cause of health inequalities. Thus, for example, there is considerable evidence that people with chronic illnesses and disabilities are at risk of downward social mobility, moving out of the labor market and into poverty. But, while important for individuals struggling with illness, such health-related mobility has been found to play only a modest role in the SEP-health association across the population as a whole (Power, Matthews, & Manor, 1996). Attention is therefore focusing on the social and physical environment as a determinant of health, from before birth and across the course of people's lives.

In this "lifecourse" perspective, people's social and physical environments leave imprints on body systems (Kuh & Ben-Schlomo, 2003). Thus, poverty before and during pregnancy can deprive the mother of key nutrients, compromising fetal development, and increasing the risk of chronic disease in later life (Barker, 1998). After birth, poor children continue to be at greater risk of exposure to health-damaging environments and behaviors both in childhood and in adulthood (Fig. 3). As a result, death rates in Britain among middle-aged men and women born into poorer families are double those of men and women growing up in better-off circumstances, with the increased risk of death remaining after adjustment for adult SEP (Kuh, Hardy, Langenberg, Richards, & Wadsworth, 2002). Circumstances in adulthood have an additional health effect, with disadvantage further increasing the risk of poor health and premature death (Blane, 1999). As this research suggests, children who escape from poverty have better health in adulthood than those who endure persisting disadvantage across their lives.

Although circumstances at all life stages influence health, childhood is especially important. This is in part because, as a period of rapid development, there is heightened sensitivity to environmental influences. It is also because childhood and adolescence are the formative periods for adult SEP,

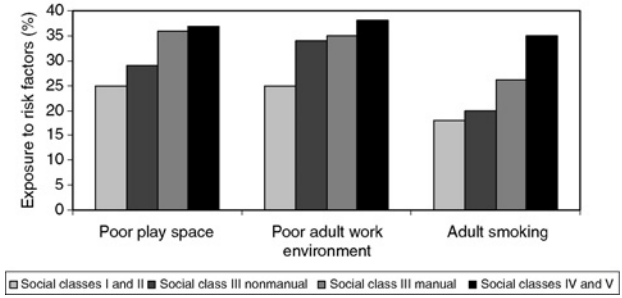

FIG. 3. Poorer beginnings: exposure to risk factors in childhood and adulthood by childhood social class (father's occupation at birth) in the 1958 British birth cohort study (source: Power & Matthews, 1997).

with educational trajectories influencing career opportunities and lifetime earnings. These periods are also ones in which adult health behaviors, such as tobacco use, are established (Graham & Power, 2004).

Lifecourse research has relied primarily on data on individuals and the households in which they live. But attention is also being paid to the influence of the wider environment—of the school, workplace, and neighborhood—on health. The relative contribution of individual and contextual factors is difficult to disentangle. But there is some evidence that neighborhoods exert an influence on health over and above the characteristics of those who live there. For example, neighborhood effects have been identified for aspects of child development, like cognitive development, readiness for school and behavioral problems, which remain after adjustment for family-level social position (Leventhal & Brooks-Gunn, 2000).

II. SEP IN THE WORLD'S RICHER HIGHLY STRATIFIED SOCIETIES

Inequalities in life chances and living standards are found in all societies, but their magnitude varies over time and between societies. Between 1950 and 1970, income inequalities narrowed in the United States and United Kingdom, reducing the proportion of adults and children living in relative poverty. From the 1970s to the mid-1980s, poverty and inequality increased sharply—across decades in which other rich nations experienced either no change or a fall in inequality. From the mid-1980s to the late 1990s, income inequality rose in almost all high-income countries, including the United States and United Kingdom, which continue to have the highest rates among rich European and English-speaking countries (Smeeding, 2002).

Underlying these patterns are changes linked to the globalizing economy, including shifts in the labor market from low-skilled to high-skilled labor. But domestic policies and welfare institutions mediate the effects of economic change and explain variations in levels and trends of income inequality among countries. They do so because investment in welfare services, including public education, housing, and health and welfare benefits, is redistributive: it has a greater proportionate impact on the living standards of poorer than richer households (Graham, 2002, 2004). Such welfare services are particularly important for the socioeconomic circumstances of people with intellectual disabilities, leveling up income, and equalizing access to education and employment.

Poverty-alleviating policies in high-income countries provide an example of the redistributive effects of domestic policies. Figure 4 maps childhood poverty rates before and after payment of tax and receipt of welfare benefits (transfers), based on a poverty line of 50% of median income. While taxes and benefits reduce poverty rates in all countries, the effectiveness of their welfare systems varies. The contrast is particularly marked between Nordic systems (Sweden, Finland, Norway) based on inclusive provision of benefits pegged to average incomes and systems that rely on low income and means-tested benefits (the United States and United Kingdom). In Sweden childhood poverty rates fall by 78% (from 18% to 4%) after tax and transfers; in contrast, in the United States, rates fall by only 19% (from 27% to 22%). Lifecourse studies suggest that policies that expose large numbers of children to poverty are likely to exact a heavy burden on future health as these children move into adulthood.

There is also increasing evidence that the degree of inequality within a society may itself be an important cause of poor health and mortality, over and above

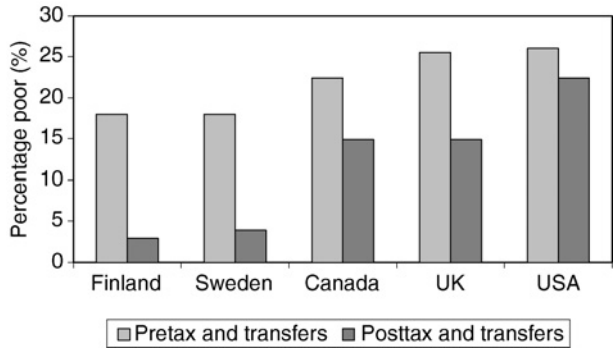

FIG. 4. Percentage of children in poverty (in households below 50% of median national income) before and after income transfers (tax and welfare benefits) (source: UNICEF, 2005).

the impact of absolute levels of poverty/SEP within that society (Wilkinson, 2005). The argument proposed by researchers, such as Wilkinson, is that more socioeconomically unequal societies result in greater numbers of people in relative poverty and greater social status differences. Greater inequalities have an impact at the level of societies (e.g., more authoritarian relations and hostility between social groups), communities (e.g., less involvement in community life and trust in others), and individuals (e.g., feelings of stress, depression, insecurity, shame, and social anxiety), resulting in more violent and antisocial societies and progressively poorer health for people at progressively lower levels in societal hierarchies (Wilkinson, 2005). On this argument, limited redistribution of resources to reduce poverty may have a small or even negligible impact on health in the absence of broader measures to tackle resource inequalities in society as a whole.

III. LOW SEP DISPROPORTIONATELY AFFECTS PEOPLE WITH INTELLECTUAL DISABILITIES

Epidemiological research undertaken in the world's richer countries has consistently reported an association between SEP and the prevalence of less severe intellectual disability or intellectual disability of unknown cause (Leonard & Wen, 2002; McLaren & Bryson, 1987; Roeleveld, Zielhuis, & Gabreels, 1997). For example, Leonard et al. (2005) reported that within a cohort of 2871 children with intellectual disability of unknown cause born between 1983 and 1992 in Western Australia, children of mothers in the most socioeconomically disadvantaged 10% had more than five times the risk of mild and moderate ID when compared with those in the least disadvantaged 10%. Similarly, Emerson, Graham, and Hatton (2006) have reported that within a nationally representative cross-sectional sample of 10,438 British children aged 5–15 years in 1999, children in the most socioeconomically disadvantaged 20% of families had more than four times the risk of intellectual disability when compared with those in the least disadvantaged 20%.

This association between SEP and intellectual disability is also evident in studies undertaken in the United States and United Kingdom that describe the social and economic contexts of families supporting a child with intellectual disabilities (Emerson, 2003a; Emerson & Hatton, 2005; Fujiura, 1998; Fujuira & Yamaki, 2000). For example, we have reported that within a nationally representative cross-sectional sample of 7070 British families supporting children aged 0–16 years in 2002, families supporting a child with intellectual disabilities were 42% more likely to be living in poverty, 70% more likely to have no savings and to worry about money "all the time," and over twice as likely to be in debt and suffer material hardship (Emerson & Hatton, 2005).

Less information is available on the SEP of adults with intellectual disabilities. The information that is available, however, indicates high levels of unemployment and poverty, at least among those living independently or with their relatives (Edgerton, 1975; Edgerton, Bollinger, & Herr, 1984; Flynn, 1989; Institute of Applied Health & Social Policy, 2002). Emerson, Malam, Spencer, and Davies (2005), for example, have reported that adults with intellectual disabilities living independently or with their relatives were significantly more likely than their nondisabled peers to be living in deprived neighborhoods, to be unemployed, and to experience material hardship.

There are no reliable data on the global distribution of intellectual disabilities (Durkin, 2002; Institute of Medicine, 2001). There are, however, very plausible reasons to suggest that exposure to potential *environmental* causes of developmental disabilities is likely to show substantial regional and international variation, thereby leading to variations in incidence and prevalence (Emerson, Fujiura, & Hatton, in press). Epidemiological studies undertaken in poorer nations have also reported a clear link between socioeconomic deprivation and an increased prevalence of intellectual disability in Pakistan (Durkin, Hasan, & Hasan, 1998; Yaqoob et al., 1995), Bangladesh (Durkin et al., 2000), and India (Ganguli, 2000). In addition, Fujiura, Park, and Rutkowski-Kmitta (2005) have reported increased rates of exposure to poverty (when compared with the general population) for people with intellectual disabilities in Bulgaria and South Africa (but not Nicaragua). They also reported significantly higher levels of unemployment among adults with intellectual disabilities in Nicaragua, Albania, Bulgaria, and South Africa.

In short, the available evidence suggests that people with intellectual disabilities in the world's richer countries (and probably elsewhere) are at significantly greater risk of living in poverty than are their nondisabled peers. This relationship is likely to reflect the operation of a number of distinct processes. First, it is clear that low SEP is associated with an increased incidence of intellectual disabilities (Leonard & Wen, 2002; McLaren & Bryson, 1987; Roeleveld et al., 1997). Second, supporting a child with intellectual disabilities may entail significant direct and indirect or "opportunity" costs for families (Dobson & Middleton, 1998; Dobson, Middleton, & Beardsworth, 2001; Leonard, Brust, & Sapienza, 1992; Meyers, Lukemeyer, & Smeeding, 1998; Parish, Seltzer, Greenberg, & Floyd, 2004). Direct costs include, for example, the additional costs of transport, child care arrangements and equipment, costs that may not be adequately compensated by welfare benefits. Indirect or opportunity costs include the costs arising from family accommodation to raising a child with intellectual disabilities including, for example, the reduced rate of employment among mothers of children with intellectual disabilities. These costs may have an impact on poverty dynamics

by increasing the chances of families descending into poverty and decreasing the chances of them escaping from poverty. Third, having an intellectual disability is, in most countries, likely to significantly increase the risk of exclusion from the workforce, the possibility of long-term unemployment, and consequential poverty (Emerson et al., 2005; Fujiura et al., 2005).

IV. SEP AND THE HEALTH, WELL-BEING, AND LIFE EXPERIENCES OF PEOPLE WITH INTELLECTUAL DISABILITIES

In the preceding sections, we have presented evidence to suggest that: (1) the experience of poverty or low SEP (especially during childhood) has a widespread negative impact on people's health, well-being, and life experiences; (2) a substantial proportion of the world's population experience poverty/low SEP; and (3) people with intellectual disabilities are significantly more likely to experience poverty/low SEP than their nondisabled peers. There is also now abundant evidence to suggest that when compared to their nondisabled peers, people with intellectual disabilities experience poorer physical health (Ouellette-Kuntz, 2005; US Department of Health and Human Services, 2002), poorer mental and emotional health (Dekker, Koot, van der Ende & Verhulst, 2002; Emerson, 2003b; Linna, Piha, Kumpulainen, Tamminen, & Almqvist, 1999), and experience more negative life experiences (Emerson et al., 2005; Hatton & Emerson, 2004).

Taken together, these observations suggest that the experience of poverty/low SEP may be an important factor in understanding the negative life experiences and high rates of emotional, behavioral, and health problems of people with intellectual disabilities (Emerson, 2004; Graham, 2005; Ouellette-Kuntz et al., 2005). It is also worth noting that within societies with high levels of inequality, people with intellectual disabilities are likely to be perceived (and to perceive themselves) as at the lower end of status hierarchies, with all the concomitant negative psychological and health consequences (Wilkinson, 2005). Later we will consider three specific ways in which investigation of the experience of poverty/low SEP may be relevant to research undertaken in the field of intellectual disabilities. First, the increased risk of exposure to poverty/low SEP of people with intellectual disabilities may account, at least in part, for their increased rates of negative life experiences, including emotional, behavioral, and health problems. Second, as in the general population, the experience of poverty/low SEP may be a risk factor for a range of negative outcomes *within* the population of people with intellectual disabilities. Third, the experience of poverty/low SEP may act to moderate the

impact of relationships that have commonly been reported within research undertaken in the field of intellectual disabilities. Later we will use examples from our research to illustrate these three possibilities.

A. SEP and Increased Rates of Morbidity and Negative Life Experiences

Previous research has reported an increased prevalence of antisocial behavior among children and adolescents with intellectual disability (Frison, Wallander, & Browne, 1998) and an increased prevalence of predominantly minor offending behavior among adults with intellectual disability (Hodgins, 1992; Lindsay, 2002; Simpson & Hogg, 2001). Research in the general population has indicated that antisocial behavior and minor offending are linked to a number of social and environmental risk factors such as familial and neighborhood poverty and social deprivation (Coleman & Hendry, 1999). Dickson, Emerson, and Hatton (2005) examined the rates and predictors of self-reported antisocial behavior in a nationally representative sample of 4174 adolescents in Britain. They reported that while rates of self-reported antisocial behavior were significantly higher among adolescents with intellectual disability (43% versus 32%; OR = 1.58; $p < 0.05$), there were no statistically significant differences between the two groups when relevant between-sample child characteristics (gender, mental health, age), social (family functioning), and socioeconomic factors (poverty, maternal educational attainment) were statistically controlled.

More recently, we have attempted to estimate the extent to which the elevated risk of poor physical and mental health among children and adolescents with intellectual disabilities may be mediated by their more disadvantaged SEP (Emerson & Hatton, in press). We examined the contribution of SEP to the health and mental status of children and adolescents with intellectual disabilities in a sample of 10,438 British children. As expected, intellectual disability was a risk factor for poorer general physical health, emotional disorders, and conduct disorders. However, between-group differences in SEP and household composition accounted for 19% of the increased risk of poor physical health, 21% of the increased risk for conduct disorder, and 35% of the increased risk for emotional disorder.

Similarly, research with families of children with intellectual disabilities has consistently reported poor health outcomes for parents (Blacher & Baker, 2002). As we have seen earlier, however, such parents are also at increased risk of exposure to poverty/low SEP (Emerson, 2003a; Emerson & Hatton, 2005; Fujiura, 1998). Emerson, Hatton, Blacher, Llewellyn & Graham (in press) investigated levels of happiness, self-esteem, and self-efficacy among a

nationally representative sample of 6954 British mothers. They reported that although mothers of children with intellectual disabilities were significantly less happy and had significantly lower rates of self-esteem and self-efficacy when compared to other mothers, there were no statistically significant differences between the two groups on measures of happiness and once relevant between-sample differences in maternal characteristics (maternal disability, lone parent status) and socioeconomic factors (poverty, maternal educational attainment) were controlled for.

We are not, of course, suggesting that all differences between the health status and life experiences of people with intellectual disabilities and their nondisabled peers can simply be accounted for by their adverse socioeconomic situation. The above evidence does, however, suggest that such factors may in part account for some of the inequalities faced by people with intellectual disabilities.

B. SEP and Variation in Morbidity and Negative Life Experiences

Research undertaken in the general population has documented a consistent relationship between poverty/low SEP and an increased prevalence of a range of psychiatric, emotional, and behavioral problems among children and young people (Meltzer, Gatward, Goodman, & Ford, 2000; Melzer, Fryers, & Jenkins, 2004; Muntaner, Eaton, Miech, & O'Campo, 2004; Power, Stansfeld, Matthews, Manor, & Hope, 2002). However, remarkably little work has been undertaken on the association between poverty/low SEP and the mental health of young people with intellectual disabilities. The majority of studies have either failed to report analyses of the association between socioeconomic circumstances and the prevalence of mental health disorders among children with intellectual disabilities (Dekker et al., 2002; Einfeld & Tonge, 1996; Linna et al., 1999; Stromme & Diseth, 2000) or have tended to employ crude indicators of SEP (e.g., raw income, simple two or three point scales of SEP; cf., Dekker & Koot, 2003; Molteno, Molteno, Finchilescu, & Dawes, 2001).

In a series of studies we have begun to explore more systematically the extent to which poverty/low SEP may constitute a risk factor for psychiatric, emotional, and behavioral problems among children with intellectual disabilities. First, we reported an association between markers of low household SEP (equivalized household income, occupational status, lone parent status) and conduct disorder, emotional disorder, and Attention-deficit/hyperactivity disorder among a nationally representative sample of 264 children and adolescents with intellectual disabilities (Emerson, 2003b). In two separate studies undertaken in different cities, we reported an association

between neighborhood indicators of deprivation (see later section): (1) conduct disorders among sample of 615 children and adolescents with intellectual disabilities (Emerson, Robertson, & Wood, 2005) and (2) a range of emotional and behavioral disorders among children from Black and South Asian minority ethnic communities (Emerson, Robertson, & Wood, in press). The results of the latter study also suggested that ethnicity may moderate the association between socioeconomic deprivation and child well-being in that, for South Asian children, higher rates of emotional and behavioral disorders were found in *less deprived* areas. This "inverse" relationship between deprivation and morbidity is consistent with what has previously been reported in the public health literature as ethnic or group density effects (Karlsen, Nazroo, & Stephenson, 2002; Smaje, 1995). These may reflect the weakening of the protective functions of within-group social networks as individuals from minority ethnic communities start to become more assimilated into the wider society and move away from poorer neighborhoods.

At a more general level, we have reported robust associations within a nationally representative sample of 2898 adults with intellectual disabilities in England between household-level hardship and/or neighborhood-level deprivation and housing quality, employment, use of day services, being bullied or a victim of crime, contact with relatives and friends, participation in community activities, self-rated health, prevalence of long-standing illness or disability, smoking, and a range of psychological outcomes including happiness, anxiety, helplessness, and confidence (Emerson et al., 2005).

C. SEP as a Moderating Variable

The previous two examples of the possible impact of poverty/low SEP have been examples of "main effects" (i.e., that exposure to poverty/low SEP may increase the risk of particular negative outcomes). An alternative mechanism is that poverty/low SEP may act to moderate the association between two other variables.

As noted previously, research with families of children with intellectual disabilities has consistently reported poor health outcomes for parents (Blacher & Baker, 2002). A consistent finding in this research literature has been an association between child behavior problems and parental distress, the former being assumed to have a direct causal influence on the latter (Hastings, 2002). However, our analysis of a representative sample of 245 British mothers supporting a child with intellectual disabilities suggested that socioeconomic circumstances may moderate this relationship, with associations between child behavior problems and maternal distress only evident among families with higher equivalized household incomes or families living in more affluent neighborhoods (Emerson, 2003a). More recently, we have attempted to replicate this result in a sample of 136 South Asian parents

of a child with severe intellectual disabilities (Hatton & Emerson, submitted for publication). Our results suggest that indicators of SEP based on employment, income, and housing quality moderate the relationship between child behavior problems and maternal anxiety, depression, and self-rated health but not between child behavior problems and overall maternal distress. The mechanisms underlying these moderating effects are far from clear. It is possible, however, that having a child with intellectual disabilities who also has behavioral problems may be a greater threat to social status and family dynamics among families with higher SEP.

V. CURRENT PRACTICE IN RESEARCH IN INTELLECTUAL DISABILITIES

While recognizing a number of notable exceptions (Birenbaum, 2002; Fujiura, 1998; Fujuira & Yamaki, 2000; Landesman Ramey & Ramey, 1999; Park, Turnbull, & Turnbull, 2002; Richardson & Koller, 1996), it is our contention that research in the field of intellectual disabilities has failed to pay sufficient attention to the possible role that poverty/low SEP may have in contributing to the range of negative life experiences and outcomes faced by people with intellectual disabilities.

To examine this issue further, a content analysis was conducted on the 2004 issues of four intellectual disability journals: the American Journal on Mental Retardation (AJMR), the Journal of Applied Research in Intellectual Disabilities (JARID), the Journal of Intellectual Disability Research (JIDR), and Mental Retardation (MR). Empirical articles including information on challenging behavior, physical or mental health problems, or well-being experienced by children or adults with intellectual disabilities or carers (either family or paid) were examined to determine whether they reported any information relevant to SEP, what information was reported, and whether this information was simply used descriptively or used in further analyses. We used a generous definition of information relevant to SEP, including information such as type of education or the living situation of people with intellectual disabilities. Such information would not ordinarily be considered as indicators of SEP in the mainstream health inequalities literature (see in a later section) but were included here to provide a comprehensive picture of contemporary intellectual disability research practice.

Table I reports the results of this content analysis. Because findings were very similar across the four journals, consistent conclusions can be drawn from data collated across the four journals. Only 29 of 75 relevant articles (39%) reported any information relevant to the SEP of people with intellectual disabilities, and only 18 articles (24%) used any of this information in further analyses. However, by far the most common information reported was current living

TABLE I
SOCIO-ECONOMIC INDICATORS INCLUDED IN EMPIRICAL JOURNAL ARTICLES[a]

	AJMR	JARID	JIDR	MR	
	n	*n*	*n*	*n*	Total *n* (%)
Participants with intellectual disabilities:					
N. relevant empirical papers ...	23	12	34	6	75 (100%)
that report SEP	6	5	14	4	29 (39%)
Income	1	0	0	2	3 (4%)
Employment status	2	2	0	2	6 (8%)
Education status/situation	5	2	2	0	9 (12%)
Living situation	5	1	11	3	20 (27%)
Area-level indicators	0	1	2	0	3 (4%)
that use SEP in analyses	3	3	8	4	18 (24%)
Participants family or paid carers:					
N. relevant empirical papers ...	7	10	7	11	35 (100%)
that report SEP	6	7	6	11	30 (86%)
Household income	4	2	2	5	13 (37%)
Subjective appraisal	1	1	1	0	3 (9%)
Occupation/employment	4	1	4	2	11 (31%)
Education-level	5	5	3	7	20 (57%)
Area-level indicators	1	1	0	0	2 (6%)
Home ownership	0	0	0	1	1 (3%)
that use SEP in analyses	3	4	4	7	18 (51%)

[a]Inclusion criteria: All papers published in 2004 in American Journal on Mental Retardation (AJMR), Journal of Applied Research in Intellectual Disabilities (JARID), Journal of Intellectual Disability Research (JIDR), and Mental Retardation (MR) assessing challenging behaviors, physical health, or mental health status in children or adults with intellectual disabilities or their carers.

situation (20 articles; 27%) and educational status (e.g., special versus mainstream school; 9 articles; 12%), which are not typically considered as socioeconomic indicators in the health inequalities literature. Excluding these indicators, only 9 articles (12%) reported any information concerning the income, employment status, or local area of people with intellectual disabilities, with 6 articles (8%) using any of these variables in further analyses.

Table I shows that the picture in research concerning carers of people with intellectual disabilities is quite different, with 30 of 35 relevant articles (86%) reporting some information relevant to the SEP of carers and 18 articles (51%) using this information in further analyses. The range of socioeconomic indicators reported was also broader than that reported for people with intellectual disabilities. Carer education (educational history or qualifications) was the most common indicator reported (20 articles; 57%),

followed by income (13 articles; 37%) and occupational or employment status (11 articles; 31%). Very few articles reported subjective appraisals of finances (e.g., difficulty in paying bills; 3 articles; 9%), area-level indicators of deprivation (2 articles; 6%), or home ownership status (1 article; 3%).

In summary, this content analysis reveals that the SEP of people with intellectual disabilities is rarely described in current empirical research and even more rarely considered analytically as a potentially important influence on outcomes. While aspects of the SEP of carers are more frequently reported in contemporary research, the indicators used typically lack sophistication and are often used as control variables rather than as key variables for analytic purposes.

The relative neglect of poverty/SEP is not unique to the field of intellectual disabilities. For example, Ensminger and Fothergill (2003) examined the extent to which the general literature on children and adolescents reported and examined the potential impact of SEP in three leading journals between 1991 and 2000. They reported that although more than 80% of papers in Child Development and the Journal of Health & Social Behavior reported SEP data, only 55% of papers in the American Journal of Public Health did so. The most commonly reported measures of SEP related to parental education (45% of studies), economic resources (28%), and parental occupation (14%). Furthermore, Ensminger and Fothergill found that fewer than 50% of the studies that reported SEP included SEP as either an independent or control variable in their analyses.

The systematic neglect of SEP in these fields will reflect a number of factors including existing custom and practice, editorial policy, and the socialization of academics and researchers within particular disciplinary world views. Particularly relevant to research in the field of intellectual disabilities is the dominance within the research community of psychologists and educationalists, neither of whom have tended to view SEP (or other social contextual factors) as key explanatory variables.

VI. IMPROVING PRACTICE: THE MEASUREMENT OF SEP IN INTELLECTUAL DISABILITY RESEARCH

A. Definitions and Approaches to Measurement: SEP, Poverty, Social Exclusion, and Inequality

As this chapter has demonstrated, an individual's health is shaped by his or her social and material circumstances, with poor circumstances increasing the risk of poor health and premature death. Labor market position is a major determinant of an individual's circumstances and, in high-income societies particularly, educational qualifications govern labor market position.

"Socioeconomic position" is increasingly used to signal these multiple dimensions—occupational and educational, social, and material. It is beginning to replace both "social class" and "socioeconomic status" as the preferred term to describe an individual's location in the socioeconomic structure (Kreiger, Williams, & Moss, 1997; Lynch & Kaplan, 2000). Social *class*, the term traditionally favored in United Kingdom studies, is seen as too specifically linked to labor market position; socioeconomic *status* is seen to obscure this aspect by giving emphasis to social standing and prestige. SEP serves as an aggregate category that, at least conceptually, can encompass both economic and social dimensions.

As this suggests, SEP is inherently multidimensional. It encompasses material circumstances (living and working conditions) and command over resources (wealth and assets). The mechanisms through which such dimensions operate are likely both to vary across health outcomes and to be mutually reinforcing. Further, the health effect of SEP will be mediated by other structures of inequality, like "race" and gender, making causal pathways hard to disentangle.

Such complexity raises problems for the measurement of SEP. Developed for the white, male, and nondisabled population, the standard measures have proved to be less reliable guides to the circumstances of less advantaged groups. For example, the income associated with a given level of education and occupation is greater for Whites than African Americans in the United States and for African Caribbeans in the United Kingdom. As a result, education and occupation provide inflated estimates of the living standards of minority ethnic groups compared with the white population (Kaufman, Cooper, & McGee, 1997; Nazroo, 2003).

In the following sections, we will review the main approaches to measuring poverty/SEP. First, we will discuss approaches based at the level of individuals or households in relation to economic resources, occupation, education, and composite measures. Following this we will briefly discuss approaches based on neighborhoods, communities, and nations.

B. Individual and Household-Level Approaches

Whereas SEP and poverty are most commonly considered to be attributes of individuals, most approaches to measurement are based on households or family units.

1. ECONOMIC RESOURCES AND MATERIAL CIRCUMSTANCES

Most contemporary approaches to measuring poverty are based on household-level measures of economic resources (Flaherty, Viet-Wilson, & Dornan, 2004; Gordon, 2000; Lister, 2004; Saunders & Tsumori, 2002).

Economic resources are also considered a key, if not the most central, component of SEP (Duncan & Magnuson, 2003; Hoffman, 2003; Kreiger, Williams, & Moss, 1997). Thus, for example, the World Bank employs a consumption-based definition of poverty, "the expenditure necessary to buy a minimum standard of nutrition" but operationalizes this definition in terms of income that allows an expenditure of $1 per day. In the world's richer countries, most official "poverty lines" are also based on measures of net household income that are typically adjusted (equivalized) to take account of household size and, sometimes, housing costs.

In the European Union (EU), for example, poverty is defined as living in a household with an equivalized net income falling below 60% of the national median. Other approaches include definitions based on income falling below a level necessary to buy a "standard" basket of goods or maintain an "acceptable" standard of living (Orshansky's US "poverty line" and the Australian "Henderson line"; Glennerster, 2000; Saunders & Tsumori, 2002).

The adjustment (equivalization) of net household income to take account of household size is common practice. It is based on the assumptions that household income is, in some sense, a shared resource, that the expenditure "needs" of children and adults will differ and that there are economies of scale related to household size. The measurement of child poverty in the EU, for example, uses the modified Organization for Economic Co-operation and Development (OECD) equivalization scale to adjust net household income. This gives a weight of 1 to the first adult in the household, 0.5 to each subsequent adult and each child aged 14 years or over, and 0.3 to each child aged under 14 years. So, for example, a household income of $50,000 would translate into an equivalized household income of $50,000 for a single person living alone (weight = 1), of $38,462 for a lone mother with one child under 14 (weight = 1.3), and of $16,667 for a couple with three children over 14 (weight = 3).

Equivalization scales have been criticized for their failure to take account of the additional costs directly associated with disability (Burchardt, 2004; Zaidi & Burchardt, 2003). As such, their use is likely to underestimate poverty rates among families including a person with disabilities, especially those families supporting a person with more severe and complex disabilities. Nevertheless, they do go some way to addressing the impact of household composition on the translation of income to attaining an "acceptable" standard of living.

Household income is also sometimes adjusted to take account of housing costs (i.e., net income is determined prior to and following the subtraction of basic housing costs). The rationale for such an approach is that housing costs often represent a substantial and relatively fixed outgoing that demonstrates marked regional variation (e.g., the average costs of a semidetached house (duplex) in England and Wales in the first quarter of 2005 varied

by a factor of 3.6 from $155,000 in Merthyr Tydfil to $560,000 in Greater London).

Income (especially equivalized income) is, however, a relatively volatile measure of SEP as it reflects changes in both economic and/or family circumstances. As a result, increased attention is being paid to understanding the dynamics of poverty over time (Apospori & Millar, 2003; Bradbury, Jenkins, & Micklewright, 2001). For example, in an analysis of the persistence of poverty in the United Kingdom between 1991 and 1999, Jenkins and Rigg (2001) reported that for an average 4-year period 19% of children were poor in 3 or 4 years compared with 58% of children who were never poor (the remaining 23% being poor in 1 or 2 of the 4 years). Over the full 9-year period they reported that 10% of children were poor in at least 7 of the 9 years (Jenkins & Rigg, 2001). There are also inequities in the distribution of persistent poverty. The United States, for example, has been reported to have one of the lowest exit rates for child poverty (the percentage of poor children who escape from poverty within a given time period; Bradbury et al., 2001) and higher rates of persistent and intergenerational poverty among children from minority ethnic communities. For example, Magnuson and Duncan (2002) reported that while childhood poverty was associated with significantly increased risk of experiencing poverty in early adulthood, 1 in 10 European Americans compared with 1 in 4 African Americans who grow up in poverty also experience poverty as young adults. These factors indicate the potential value of measuring income over more extended periods of time.

There are a number of limitations associated with income-based measures (cf., Flaherty et al., 2004; Gordon, 2000; Lister, 2004). First, they may be difficult to collect in some circumstances, especially in the level of detail that would allow for appropriate equivalization. Second, they have been criticized for failing to take account of key resources (e.g., savings, support from family and friends) that may moderate the association between income and the ability to access goods or activities that are deemed essential or appropriate, or to maintain an "acceptable" standard of living. Third, due to the very low rates of employment among people with intellectual disabilities, the income distribution of adults with intellectual disabilities may show little variation. This may prove problematic for studies that attempt to identify associations between SEP and outcomes *within* the population of adults with intellectual disabilities.

Nevertheless, income and the purchasing power associated with it does represent a key indicator of social standing in many societies and a key factor in determining a person's capacity to attain an "acceptable" or greater standard of living. As noted previously, we have successfully used equivalized household income in some of our studies on the situation of families

supporting children with intellectual disabilities (Emerson, 2003a,b). Income-based measures, especially those based on relative equivalized income, are also highly suitable for use in international comparative studies.

As a result of some of the difficulties associated with income-based measures of economic resources, alternative approaches have been developed based on subjective appraisals of the sufficiency of economic resources (Barrera, Caples, & Tein, 2001; Gordon et al., 2000), an individual's or family's actual ability to access activities or goods that consensus deems necessary or essential (Gordon, 2000; Gordon & Pantazis, 1997; Gordon et al., 2000; Mack & Lansley, 1985; Nolan, Maitre, & Watson, 2001), and analyses of household assets (Filmer & Pritchett, 1999). The use of *deprivation indices* provides a more direct measure of the adequacy of a family's resources to maintain an "acceptable" standard of living. Gordon et al. (2000), for example, replicated and updated the methods used in the "Breadline Britain" surveys (Gordon & Pantazis, 1997; Mack & Lansley, 1985) to: (1) identify items and activities that a representative sample of people in Britain considered to be "necessities" for an acceptable standard of living (e.g., to be able to have two meals a day, to have friends or family round for a meal) and (2) determine what proportion of the population wanted but could not afford these items or activities. Such an approach has been incorporated into the official approach to measuring child poverty in the United Kingdom (Department for Work and Pensions, 2003) and forms half of the items of a composite measure of SEP developed in New Zealand (Salmond, Crampton, King, & Waldegrave, in press).

We have successfully used similar deprivation indices in two studies involving people with intellectual disabilities. In a study of maternal well-being (Emerson et al., in press), we found that deprivation indices were stronger predictors of maternal well-being than equivalized household income (either before or after controlling for housing costs). More recently, we used an abbreviated nine item deprivation scale to measure deprivation among a nationally representative sample of English adults with intellectual disabilities. This involved asking informants whether they (1) had, (2) did not have because they could not afford, or (3) did not have because they did not want, nine items (new clothes, new shoes, food, heating, telephoning friends and family, going out, visits to the pub or club, a hobby or sport, a holiday). Adults who reported that they would like but could not afford two or more of these items were significantly more likely in multivariate analyses to live in unsuitable accommodation, report less access to and satisfaction with supports and services, engage in fewer social and community activities, report being bullied or being a victim of crime, and report poorer health and well-being (Emerson et al., 2005).

There is, however, relatively little overlap between groups identified on the basis of relatively low income, deprivation, and subjective appraisal (Bradshaw & Finch, 2003). This suggests that, at present, it may be most appropriate to use multiple measures of economic resources in order to identify which indicators in isolation or combination may have the strongest association with the life experiences and health status of people with intellectual disabilities and their families.

Deprivation and *asset indices* have also been developed to operationalize the measurement of absolute and relative poverty in low-income countries. For example, *deprivation indices* have been developed in the domains of food, safe drinking water, sanitation, health, shelter, education, information, and basic social services to measure rates of "absolute" poverty in low-income countries (Gordon, Nandy, Pantazis, Pemberton, & Townsend, 2003). Alternatively, the use of *asset indices* represents the most common approach to the measurement of SEP in low- and middle-income countries (Filmer & Pritchett, 1999; Gwatkin, Rustein, Johnson, Pande, & Wagstaff, 2000). These represent the weighted sum of household assets (e.g., ownership of a radio, television, refrigerator, motorcycle, car) and housing characteristics (e.g., access to electricity, source of drinking water, type of toilet) where weights are determined by principal components analysis (Filmer & Pritchett, 1999).

2. OCCUPATION

Historically, occupational status has been the main measure of SEP in the United Kingdom literature. Occupation is an indicator of income, status, prestige, and power that is very easy to identify and code and relatively stable over time. However, there are obvious problems with using occupation as an indicator of SEP for people outside the labor market, including women and some people with disabilities. While it may be possible to use parental occupation data as a proxy indicator of SEP for people with intellectual disabilities, accurate parental occupation data may be difficult to obtain from people with intellectual disabilities and may also have little impact on the SEP of adults with intellectual disabilities who are living outside the family home. Occupational status is measured by classifying an occupation into one of a number of mutually exclusive hierarchical categories, reflecting the relative income, status, prestige, and power associated with that occupation in a particular society. The most commonly used occupational classification systems include: (1) *Socioeconomic Index of Occupations* (cf., Bornstein, Hahn, Suwalsky, & Haynes, 2003), developed by Duncan (1961) with further updates and revisions (Nakao, Hodge, & Treas, 1990) for use in the United States. This places occupations into a 0–100 ordinal scale according to occupational prestige, with occupational prestige being calculated from data

combining education and income levels associated with each occupation and (2) *The National Statistics Socioeconomic Classification* (NS-SEC: Rose & Prevalin, 2001, 2003; http://www.statistics.gov.uk/methods_quality/ns_sec/default.asp) for use in the United Kingdom. This has been developed for use in official United Kingdom statistics assessing SEP based on occupation, and replaced the United Kingdom Registrar General's classification of social class in 2001. The version of the classification that is likely to be most commonly used classifies occupations into one of eight categories, ranging from "higher managerial and professional occupations" to "never worked and long-term unemployed"; and (3) *International Standard Classification of Occupations* (Elias & Birch, 1994; Ganzeboom & Treiman, 1996), developed for international use. This has been developed on a similar basis to the NS-SEC and has 10 categories ranging from "legislators, senior official, and managers" to "elementary occupations."

3. EDUCATION

Parental educational experience or qualifications, classified hierarchically, have been the most common indicator of SEP used in the child development literature (Ensminger & Fothergill, 2003). Education data of this type are very easy to collect, relatively stable over time, and robustly associated with many relevant factors, including parental lifestyle, parenting beliefs, and parental behaviors. However, there are problems with developing stable SEP classification systems based on education. First, education systems (e.g., school-leaving age, access to university) and qualification frameworks within countries change over time, a particular problem with mixed age samples due to changing educational policy and practice. Second, there are huge variations in educational policy and practice between countries. Although education has been widely used as an SEP measure with parents of children with intellectual disabilities (Table I), the utility of education data for this purpose with adults with intellectual disabilities is extremely limited, as there is likely to be very little meaningful variation in this population for the purposes of assessing SEP.

4. COMPOSITE MEASURES

Some researchers have attempted to develop composite measures of SEP, combining multiple indicators of SEP across multiple individuals in a household. The most commonly used of these composite measures of SEP is the *Hollingshead Four Factor Index of Social Status* (cf., Bornstein et al., 2003), which combines data on the educational attainment and occupation of all employed adults in a household into a single score. Although widely used in US child development research (Ensminger & Fothergill, 2003), the Hollingshead Four Factor Index has a poor theoretical or empirical rationale.

There is an emerging consensus within the child development literature that it is necessary to measure multiple components of SEP and that these should be used in analyses separately rather than in composite scales (Duncan & Magnuson, 2003; Ensminger & Fothergill, 2003).

C. Area-Based Measures

In addition to household-level measures of SEP/poverty there exist an increasing number of well-constructed measures of area or neighborhood-level deprivation. These are of interest for two reasons. First, in many societies deprivation and affluence are inequitably geographically distributed. As a result, it is possible to use area-based measures of deprivation as proxy measures for household-level deprivation. This is particularly relevant as such measures can often be easily derived from census-defined areas, or postal or zip code data. It should be kept in mind, however, that the association between household poverty and neighborhood poverty is often complex. For example, in the United States only 15% of poor children live in high poverty urban neighborhoods, with twice as many living in low poverty urban neighborhoods (Magnuson & Duncan, 2002). The conjunction between family and neighborhood poverty does, however, vary considerably with ethnicity, with 27% of poor African American children and 20% of poor Hispanic children compared with just 3% of poor European American children living in poor neighborhoods (Magnuson & Duncan, 2002).

Second, the impact of area-level or "neighborhood effects" is of interest in its own right. Thus, for example, there is now clear evidence to suggest that, once any effects due to household-level SEP/poverty are taken into account, growing up in poorer neighborhoods is itself associated with poorer educational attainment and increased risk of adverse behavioral and emotional outcomes for children (Leventhal & Brooks-Gunn, 2000; Sampson, Morenoff, & Gannon-Rowley, 2002; Seccombe, 2000).

For example, *English Indices of Deprivation 2004* (Noble et al., 2004) are available for local areas or neighborhoods with a population of approximately 1500 people. These indices include an overall index of multiple deprivation score as well as scores for the seven constituent domains: income; employment; health and disability; education, skills, and training; barriers to housing and services; living environment; and crime. In the United Kingdom similar indices are available for Scotland, Wales, and Northern Ireland (Wood & Emerson, 2005). Parallel developments in other countries include the Socioeconomic Indexes for Areas (SEIFA) in Australia (Trewin, 2003).

For the United States, several area-based SEP indicators based on the 2000 census are available at the level of census block-group (average approximately 1000 people), census tract (average approximately 4000 people) and

above through the interactive American Fact Finder service.[1] SEP indicators (available in table SF3 for both 1990 and 2000 census data) include indicators of occupation, income, poverty, wealth, and education (Kreiger et al., 1997). Updated poverty and income data are available at county and school district-level through the *Small Area Income and Poverty Estimates* (SAIPE) programme.[2] Finally, Eibner and Sturm (in press) describe the development of census tract-level indices of deprivation similar to those developed in the United Kingdom.

There also exist a number of indicators of the extent of *inequality* (rather than deprivation) as measured at regional or national levels (Kawachi & Kennedy, 1997). These include the Gini coefficient (range 0 [perfect equality] to 1 [perfect inequality]) and the Robin Hood index (the proportion of a society's income that would have to be redistributed to attain equality). Such measures are of use in disentangling the effects of exposure to deprivation (e.g., living in poverty) and the impact of living in cities, regions, or nations that vary with degree to their overall level of stratification or inequality (Wilkinson, 2005).

We have successfully used area-level indicators of deprivation in two studies of the emotional and behavioral needs of young people with intellectual disabilities (Emerson, Robertson, & Wood, 2005, in press) and in a study of the life experiences of adults with intellectual disabilities (Emerson et al., 2005). The results of these analyses suggested that while controlling for household-level deprivation (see previous section), people who lived in poorer neighborhoods were more likely to live in unsuitable accommodation, be less satisfied with their education, have been bullied at school, have less access to services, be an unpaid carer for another adult, engage in a more restricted range of community-based activities, not feel safe, be a victim of crime, smoke, and be less happy.

VII. CONCLUSIONS AND RECOMMENDATIONS

In this chapter, we have demonstrated the relevance of poverty and SEP for research involving people with intellectual disabilities and provided some information on measures of poverty/SEP that investigators can apply to their own work. Despite the obvious health inequalities and early mortality experienced by people with intellectual disabilities, research examining these constructs has for too long neglected the central importance of poverty and

[1]http://factfinder.census.gov/home/saff/main.html?_lang=en.
[2]http://www.census.gov/hhes/www/saipe/overview.html.

inequalities in SEP experienced by many people with intellectual disabilities across the lifespan. Researchers wanting to build these issues into their empirical work need to consider the implications for theory, research design and sampling, measurement, and analysis.

A. Theory

General health researchers are developing increasingly sophisticated theories linking poverty and SEP to health inequalities (Bornstein & Bradley, 2003; Graham, 2002; Marmot & Wilkinson, 2005; Wilkinson, 2005). It may be possible to place many of the current family and psychosocial theories used by intellectual disability researchers within these broader health inequalities frameworks to provide a richer understanding of the socioeconomic contexts within which people with intellectual disabilities live. This may involve revising current theories that either ignore the role of poverty/SEP or assume simple additive effects of SEP on personal or family health and well-being. It is also possible that research with intellectual disabilities will shape broader health inequalities theory, for example, by examining the role of stigma and social exclusion alongside poverty/SEP.

B. Research Design and Sampling

To examine levels of poverty/SEP systematically amongst people with intellectual disabilities and their families, nationally representative samples are required, ideally with sufficient numbers of people in gender, age, and ethnic groups to allow meaningful comparisons. This clearly requires sampling strategies and numbers beyond the reach of most intellectual disability researchers, although both primary research (Emerson et al., 2005) and secondary analyses of general population datasets (Fujiura & Yamaki, 2000) have been conducted concerning people with intellectual disabilities.

To investigate the potential role of poverty/SEP systematically, researchers using both quantitative and qualitative methods need to ensure that their samples are varied in terms of SEP and include sufficient numbers of people in poverty or experiencing low SEP to allow meaningful analysis. This may require more proactive or persistent sampling strategies or action research designs, as people and families experiencing low SEP may be less likely to be in contact with formal services and also less likely to initially volunteer for research projects. Longitudinal studies may also be required to examine both the dynamics of poverty/SEP among people with intellectual disabilities and their families over time and the impact of early experiences on health across the lifecourse.

C. Measurement

SEP researchers have acknowledged that there is no single best measure of SEP (Krieger et al, 1997; Liberatos, Link, & Kelsey, 1988). Key issues involved in selecting an appropriate SEP measure or measures (multiple SEP measures are preferred) include the theoretical or conceptual relevance of the measure, its applicability to the populations and time period being investigated, the reliability and validity of measures, the feasibility and ease of data collection, and comparability to other studies or national data.

Researchers working with people with intellectual disabilities and their families also need to consider the relevance of the measure to the population of people with intellectual disabilities and the utility of the measure for analysis. For children with intellectual disabilities living with their families, standard SEP measures based on parental, household, and area-level data are all likely to be valid, reliable, and practical, with numerous comparisons available to other research and national data. For adults with intellectual disabilities the picture concerning the measurement of SEP is considerably more complex. Many standard measures of SEP will show little or no within-population variability with regard to personal income, occupation, or education due to the pervasive social exclusion experienced by people with intellectual disabilities. There is some evidence that for adults living independently or with relatives, household or area-based measures show reasonable variability, although this variability is limited for adults living in supported accommodation (Emerson et al., 2005). The pervasive impact of benefit systems and support services on the living circumstances of people with intellectual disabilities will also be reflected in SEP measures (e.g., employment, income, occupation, deprivation, neighborhood). Researchers working with people with intellectual disabilities may need to consider the development of relevant SEP indicators to allow the examination of within-population variation; such development work might also be relevant to other socially excluded populations.

D. Analysis

As our content analysis (Table I) demonstrated, both quantitative and qualitative researchers are extremely unlikely to ask questions about the poverty/SEP of people with intellectual disabilities or to include such constructs in their analyses of the behavior, health, or well-being of their samples. Family researchers are more likely to include some simple measures of parental/household SEP, although these measures are typically used as control variables or covariates in within-sample or between-sample analyses, rather than highlighted as important constructs for the purposes of analysis.

Where relevant, researchers need to use conceptually appropriate SEP measures that are integral to the theory being tested or developed and ensure that these SEP measures are equally integral to the analytic strategy. Given that SEP measures may have moderating as well as mediating effects (Emerson, 2003a,b), analyses will need to be conducted to allow such moderating effects to emerge.

It is our contention that understanding poverty and SEP is crucial for understanding the lives (and deaths) of people with intellectual disabilities. It is our hope that this chapter will encourage other researchers working with people with intellectual disabilities and their families to systematically investigate poverty and SEP.

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The Influence of Prenatal Stress and Adverse Birth Outcome on Human Cognitive and Neurological Development*

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As described by Barker (1998), in Britain at the turn of the nineteenth century there was a noticeable decline in the population. Birth rates were low and infant mortality was high. Moreover, approximately two-thirds of the young men who volunteered to fight in the South African war were reported to have been rejected because of their physical frailties. The enlightened government response was to recruit and train an ''army'' of midwives to interview and assist all women during pregnancy and through infancy until 1 year of age. One extraordinary consequence of this effort was meticulous record keeping of pregnancy history and birth outcomes in this population. These very early life-history records, combined with national health records from later in life, are perhaps the first data allowing the systematic examination of programing during the fetal period and provide convincing demonstrations of the profound impact of prenatal and early life experiences on later morbidity and mortality.

Intrauterine experience and birth outcome have been linked to a wide range of disease outcomes including hypertension, coronary heart disease, diabetes, and polycystic ovary disease (Cresswell et al., 1997; Godfrey & Barker, 2000; McCance et al., 1994; Roseboom et al., 2000; Savona-Ventura & Chircop, 2003), as well as psychiatric illnesses including schizophrenia, depression, anxiety disorders, and suicide (Gale & Martyn, 2004; Gunnell, Rasmussen, Fouskakis, Tynelius, & Harrison, 2003; Indredavik et al., 2003; Mittendorfer-Rutz, Rasmussen, & Wasserman, 2004; Van Os & Selten, 1998). The prenatal experience also appears to exert persisting and widespread influences on cognitive and neurological function later in life. The focus of this chapter is on the influence of preterm birth (children who are born prior to

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37 completed weeks of gestation) and low birth weight (those that are born at a weight of less than 2500 g) and prenatal maternal stress on cognitive, motor, and neurological development in the human.

I. BIRTH OUTCOME AND DEVELOPMENTAL DISABILITY AND DELAY

There is no question that those born early or small are at risk for developmental disability ranging from quite mild to severe (Anderson, Doyle, & The Victorian Infant Collaborative Study Group, 2003; Cooke & Foulder-Hughes, 2003; Hack & Fanaroff, 1999; Hack et al., 1994; O'Brien et al., 2004; Saigal, Hoult, Streiner, Stoskopf, & Rosenbaum, 2000; Wood et al., 2000). Absolute numbers of those with disability related to these birth outcomes are not decreasing but in fact increasing due to larger number of survivors at lower gestational ages and lower birth weights. Advances in neonatal intensive care in the 1990s, especially the introduction of surfactant therapies, have pushed the lower limit of viability to 23 weeks' gestation and increased rates of survival at gestational ages prior to 30 weeks' (Hack & Fanaroff, 1999; Tin, Wariyar, & Hey, 1997).

In a quantitative review of studies published from 1970 to 1997 of survivors of very low birth weight (<800 g) or very preterm delivery (<26 weeks) the average rate of disability for the most prevalent causes were: mental retardation, 14.3%; cerebral palsy, 7.5%; blindness, 7.8%; and deafness, 2.9% (Lorenz, Wooliever, Jetton, & Paneth, 1998). Because of the comorbidity of many disabilities, the total number of disabilities exceeds the number of individuals with those disabilities, making it useful to consider the total number of individuals with any disability. In this same sample, the probability that a survivor of low birth weight had at least one clinically significant disability was 24.2% and the equivalent estimate for those born early was 22.1%. In a sample of almost half a million births at less than 28 weeks' gestation in northern England, it was estimated that "one in ten of all survivors has a disability so profound that he or she is never likely to become independently mobile or to communicate effectively with others" (Tin et al., 1997). Although disability rates are high in this population, Doyle and The Victorian Infant Collaborative Study Group (2001) report that the survival rate free of disability does rise significantly with each gestational week from 23 to 27 weeks.

Not surprisingly, the rates of less severe developmental delay also are increased in those born early or small. One might conclude that there is a continuum of severity associated with these birth outcomes. Even those

who escape obvious disability are likely to suffer from some more subtle impairment. In studies examining only those children who are not developmentally disabled and are apparently neurologically healthy, those born preterm or at a low birth weight have repeatedly been found to exhibit lower intelligence quotient (IQ) scores (both performance and verbal; Levy-Schiff, Einat, Mogilner, Lerman, & Krikler, 1994; Olsén et al., 1998; Rickards, Kelly, Doyle, & Callanan, 2001; Schenkin et al., 2001; Sørensen et al., 1997). Compromised motor skills are common among those with these birth outcomes (de Kleine et al., 2003; Levy-Schiff et al., 1994; Sommerfelt, Ellertson, & Markestad, 1993). Those who do not display disabilities also show specific deficits in various cognitive processes, including visual processing and visual memory (Rickards et al., 2001), episodic memory (Isaacs et al., 2000), and spatial working memory (Luciana, Lindeke, Georgieff, Mills, & Nelson, 1999). These more subtle impairments appear to have implications for achievement outcomes such as arithmetic (Rickards et al., 2001) and verbal fluency performance (Rushe et al., 2001) as well as scores on a General Certificate of Secondary Education (Pharoah, Stevenson, & West, 2003).

On average, those born preterm or of low birth weight escape major disability. However, they may have lower mean IQs and perhaps other more subtle delays, and it is often only with more precise empirical measurement that one is able to detect these differences. If one employs more general assessments of successful functioning, the differences may no longer be apparent. For example, using broader measures of outcome, such as the probability of completion of further education following compulsory schooling, low birth weight (after exclusion for disability) was not associated with nonenrollment or nongraduation (Olsen, Myhrman, & Rantakallio, 1994).

II. BIRTH OUTCOME AND THE BRAIN

There is good evidence to suggest that preterm birth and low birth weight are associated with structural brain injury (due to brain hemorrhage or ischemia), resulting in later developmental delays and deficits in motor and cognitive functioning (Fawer, Besnier, Forcada, Buclin, & Calame, 1995; Hack et al., 1994; Keeney, Adcock, & McArdle, 1991a,b; Ment et al., 2003; Perlman, 1998; Roth et al., 1993; Stewart & Kirkbride, 1996; Stewart et al., 1999; Vollmer et al., 2003). What may be of equal or greater interest are more subtle effects of these birth outcomes on the brain in the absence of such overt injury. The effects of being born early or small on the brain have been assessed in several studies both indirectly and directly. The indirect method involves inferring reduced brain volumes from measurement of

head circumference. Children born preterm or of low birth weight have on average smaller head circumferences (Cooke & Foulder-Hughes, 2003; Powls, Bottting, Cooke, Pilling, & Marlow, 1996) and smaller head size has been linked to less optimal cognitive and motor outcomes in childhood. For example, among a cohort of children born with low birth weights and who also had subnormal head size at 8 months of age, IQ scores were lower as were language and mathematics skills at 8 years of age, compared to those with normal head sizes (Hack et al., 1991). Similarly, in a study of children born preterm, those with smaller head circumferences at age 7 performed more poorly on assessments of IQ and motor skills at the same age when compared to term controls (Cooke & Foulder-Hughes, 2003). These studies suggest that preterm birth and low birth weight each are associated with reduced brain growth and that the reduced growth has implications for cognitive and motor abilities that last at least until childhood.

The direct method of assessment involves employing technologies, such as MRI, to measure differences in brain structure and function. Among survivors of preterm birth or low birth weight who do not suffer from any clinically significant neuromotor or sensory handicaps, neurological development appears to be altered. There is evidence that overall brain and regional cortical volumes are reduced in those born with these adverse birth outcomes (Abernethy, Palaniappan, & Cooke, 2002; Cooke & Abernethy, 1999; Isaacs et al., 2004; Peterson et al., 2000, 2003). Moreover, preterm infants exhibit both reduced myelination and gray-white matter differentiation at term compared to term controls (Hüppi et al., 1996). In fact, among those born early or small, many individual brain regions are reduced in size even after accounting for total brain volume. Investigators have found evidence of this size reduction for the hippocampus (Isaacs et al., 2000; Peterson et al., 2000), corpus callosum (Cooke & Abernethy, 1999), basal ganglia (Peterson et al., 2000), amygdala, and caudate nuclei (Abernethy et al., 2002).

The differences detected by structural MRI have been linked to behavioral measures of cognitive function. For example, reduced caudate and hippocampal volumes were associated with lower IQ among those born preterm (Abernethy, Cooke, & Foulder-Hughes, 2004; Abernethy et al., 2002). Reductions in cortical volumes (sensorimotor, temporal, and occipital) were associated with diminished mental and motor development in toddlers and with decreased intelligence in 8- and 15-year olds (Isaacs et al., 2004; Peterson et al., 2000, 2003). It has also been demonstrated that hippocampal volumes in preterm children were related to measures of episodic memory (Isaacs et al., 2000).

The studies describing structural differences and their behavioral consequences are complemented by the results of a functional MRI study and event-related potential (ERP) studies (Dupin, Laurent, Stauder, & Saliba, 2000;

Fellman et al., 2004; Jansson-Verkasalo et al., 2003). Peterson and colleagues (2002) examined correlates of language abilities with functional MRI in 8-year-old children. They found differences in phonetic and semantic processing between the preterm and term children. Overall brain activity was higher for the term children when processing meaningful speech than when listening to meaningless phonetic sounds. However, on average for the preterm children, there was no difference in brain activity during processing of meaningful and meaningless sounds. The authors concluded that preterm children were processing phonetic stimuli in the manner that the term children were processing semantically meaningful stimuli. Put differently, preterm children processed gibberish in the same way that term children were processing meaningful sounds. Among the preterm children, the greater the tendency to exhibit this deviant sort of processing, the lower the reading comprehension and verbal IQ scores.

Fellman et al. (2004) examined auditory ERP in infants born preterm and at term. Differences in ERP responses were detected between the preterm and term groups at 6 and 12 months of age. At 12 months, the mismatch negativity, an index of auditory preattention, was absent in the preterm group but detectable in the term children. Further, the atypical auditory ERP responses were predictive of scores on the Bayley Developmental Index at 2 years of age. A second study (Jansson-Verkasalo et al., 2003) examined auditory ERP responses in very low birth weight preterm children at the age of 4 years. Again, as with the Fellman et al. (2004) study, they found that the premature children displayed attenuated mismatch negativity compared to controls and that the mismatch negativity was associated with verbal naming ability. Data from these studies suggest that one contributing factor underlying language deficits in premature children may be atypical auditory processing.

III. NOT SO VERY PRETERM OR LOW BIRTH WEIGHT

A few studies are beginning to speak to the gestational ages or birth weights that are driving the effects of these birth outcomes on development. In other words, is it just that those born very early, or born extremely small, who drive the deficits in development observed among those born early and small? It is of course the case, as discussed earlier, that the lower extremes in birth weight and gestational age are associated with poorer outcomes. However, both for length of gestation and for birth weight, the effects are not confined to these extremes. There have been at least three studies examining the link between birth weight and later cognitive performance that utilized analyses designed to specifically address this question. Each study analyzed birth weight as a continuous variable and also repeated the

analyses after excluding all births at less than 2500 g. Schenkin et al. (2001) found a positive relation between birth weight and IQ in the normal range of birth weights in 445 Scottish children. Cognitive function in 4300 Danish men (mean age of 18) was also correlated positively with birth weight (Sørensen et al., 1997). Again, the association remained after restricting birth weights to the normal range. Similarly, Richards, Hardy, Kuh, and Wadsworth (2001) found that birth weight within the normal range predicted cognitive ability at the ages of 8, 11, 15, and 25 among British males and females. In the Sørensen et al. (1997) study, very high birth weight (>4200 g) was associated with a decrease in cognitive abilities, a finding that is consistent with the broader literature examining the links between birth weight and later health showing that very high, like very low, birth weight is a risk factor for diseases such as schizophrenia, diabetes, and polycystic ovary syndrome (Cresswell et al., 1997; Gunnell et al., 2003; McCance et al., 1994; Savona-Ventura & Chircop, 2003).

To date, we do not know of a study that contains analyses asking similar questions with regard to length of gestation. There are few studies, however, that do suggest that, like the effects of birth weight, the gestational length effects are not entirely driven by those born extremely early. In a study of 5-year-old children born preterm, a clear linear relation was seen between gestational age at birth and general intelligence, with each week between 26 and 34 weeks' gestation contributing an average increase of 1.4 IQ points (Fawer et al., 1995). Similarly, McCarton, Wallace, Divon, and Vaughan (1996) show quite convincing data that the relation between length of gestation and cognitive ability and neurological function is present among higher gestational ages (they show data ranging from 26 to 37 weeks' gestation). One study did compare cognitive performance of early (mean gestational age of 30.6) and late preterm (mean gestational age of 36.3 weeks) groups to term controls (mean gestational length of 40.1 weeks; De Haan, Bauer, Georgieff, & Nelson, 2000). At 19 months of age, those in the early preterm group showed reduced performance on an explicit memory task (with both immediate and delayed recall) compared to the term controls. The performances of those in the late preterm group fell between the early preterm and controls, and were not statistically different from either. This study further reinforces the notion that the relation between preterm birth and later development is not confined solely to the very early preterm births.

IV. SMALL FOR GESTATIONAL AGE

A discussion of the influence of birth weight on later cognitive and neurological development would not be complete without a word about those born small for gestational age (SGA). These are children who are born

at a significantly lower weight than expected for their gestational ages. The data do suggest that being born smaller than expected confers its own negative developmental consequences. McCarton et al. (1996) have suggested that "premature SGA infants are felt to be at double biological jeopardy because of a shortened gestational period compounded by intrauterine growth retardation." Data from their study and the work of others support this assertion. Cognitive impairment, neurological dysfunction, decreased IQ, diminished motor ability, and reductions in intracranial and cerebral gray matter volumes have each been linked to SGA status within successive gestational age categories in premature infants (Hutton, Pharoah, Cooke, & Stevenson, 1997; McCarton et al., 1996; Tolsa et al., 2004). Studies also have addressed this issue by examining those born SGA at term and found that SGA status at term is associated with decreased IQs (Goldenberg et al., 1996; Sommerfelt et al., 2000, 2002). Furthermore, in the Goldenberg study term, SGA children were at an increased risk of mental retardation compared to the term appropriate weight for gestational age controls. Data from this study also suggest that the reduction in IQ associated with being born SGA at term was equivalent to the reduction associated with being born prior to 34 weeks' gestation at an appropriate weight.

V. A SLOW DEVELOPMENTAL TRAJECTORY OR PERSISTENT DIFFERENCES?

The persistence of the effects on cognitive and neurological function related to preterm birth or low birth weight has yet to be fully established, as the results from longitudinal studies are not entirely consistent. Some studies find that the rates of disability or impairment increase from childhood into adolescence or early adulthood (Isaacs et al., 2004; O'Brien et al., 2004; Saigal et al., 2000), some find they stay the same (Richards et al., 2001), and some find that they decrease (Tideman, 2000). It is difficult to interpret these inconsistencies because of sample differences, differences in the outcomes that were assessed, and variations in the age ranges under study. The optimal method to address this question is a longitudinal study, and as just described, these have yielded conflicting results. However, some additional insight is also provided by studies examining the effects of birth outcome later in life and these studies do show differences in adolescence and beyond.

Among the studies that have examined the effects of preterm birth and low birth weight in adolescence (average age of 13–15 years), it has been shown that those born with these birth outcomes still exhibited lower IQs, less verbal fluency, impaired visual–motor coordination, poorer visual memory, and were four to six times as likely to have repeated a grade (Levy-Schiff

et al., 1994; Rickards et al., 2001; Rushe et al., 2001; Saigal et al., 2000). Importantly, all of these findings are present among those without clinically significant disabilities or impairments, in other words among those teens who were "apparently normal." All but one of the studies excluded children with disabilities, and Rushe et al. (2001) who did include them, conducted separate analyses only on the apparently normal children.

These behavioral findings are mirrored by three studies examining differences in brain structure among individuals in adolescence. Stewart et al. (1999) found that preterm birth was associated with an 11-fold increase in the probability of showing structural abnormalities with MRI at 14–15 years of age, most commonly in the ventricles, corpus callosum, and white matter. Cooke and Abernethy (1999) found that 15–17 year olds born with low birth weights had smaller total brain volumes and smaller corpus callosum volumes (taking into account total brain volume) using MRI. Finally, 15–17 years old who survived very low birth weight (<1500 g) without major disability were found with MRI to have smaller brains, smaller caudate nuclei, and reduced hippocampal ratios compared to term controls (Abernethy et al., 2002).

The fact that functioning differs at these later developmental stages, combined with the documented differences in brain morphology, suggests that some of the differences are permanent and not simply reflections of a delayed developmental trajectory. Consistent with this conclusion are findings from an MRI study in adolescents classified as neurologically normal and born preterm but who showed declines in IQ from age 7 to 15. The declines in verbal IQ were associated with structural changes in the frontal and temporal lobe regions (including the hippocampus) and declines in performance IQ were associated with differences in occipital and temporal regions (Isaacs et al., 2004).

Although the differences do appear to persist into adolescence and early adulthood, suggesting some persistent biological differences, it is also the case that several studies have demonstrated that the lasting negative effects of low birth weight and preterm birth can be attenuated through intellectual stimulation and positive home environment factors (Fawer et al., 1995; Goldenberg et al., 1996; Gross, Mettelman, Dye, & Slagle, 2001; McGauhey, Starfield, Alexander, & Ensminger, 1991; Vohr, Garcia Coll, Flanagan, & Oh, 1992). For example, it appears that children born early or small who come from low socioeconomic backgrounds are especially at risk for later cognitive deficits. However, the adverse effects of a low socioeconomic status environment in this population can be positively offset by the use of special early intervention services (occupational therapy, speech and language therapy, and physical therapy; Ment et al., 2003).

VI. PRENATAL STRESS AND BIRTH OUTCOMES

Before discussing the postnatal effects of prenatal stress, it must be noted that the most well-documented outcome of prenatal stress in humans is preterm birth and low birth weight. Prenatal stress measured in a variety of ways has been linked both to the outcomes of shortened gestation and reduced birth weight (Copper et al., 1996; Glynn, Wadhwa, Dunkel-Schetter, Chicz-DeMet, & Sandman, 2001; Hedegaard, Henriksen, Sabroe, & Secher, 1993; Paarlberg, Vingerhoets, Passchier, Dekker, & Van Geijn, 1995; Rini, Dunkel-Schetter, Wadhwa, & Sandman, 1999). Not yet known is the extent to which the poorer developmental outcomes of children born with these adverse birth outcomes as a result of prenatal stress are due to the associated complications of being born early or small or to having been exposed to a high-stress environment *in utero*—presumably both contribute to some extent.

VII. PRENATAL STRESS AND DEVELOPMENTAL OUTCOMES

The number of studies in humans examining the effects of prenatal stress on cognitive and neurological outcomes is so few that some attention can be given to each. (There are a few more studies examining the relation between prenatal stress and later temperament and behavior providing convincing evidence for the influence of prenatal stress or negative effect on behavior regulation: Buitelaar, Huizink, Mulder, Robles de Medina, & Visser, 2003; Davis et al., 2004a; Martin, Noyes, & Wisenbaker, 1999; O'Connor, Heron, Golding, Beveridge, & Glover, 2002; O'Connor, Heron, Golding, Glover, & ALSPAC Study Team, 2003; Van den Bergh & Marcoen, 2004). Brouwers, van Baar, and Pop (2001) examined the association between maternal anxiety during the third trimester (32 weeks' gestation) and infant development at 3 weeks and 1 and 2 years of age. They found that high levels of state anxiety were associated with lower scores on the orientation cluster of the Neonatal Behavioral Assessment Scale, a measure of attention to visual and auditory stimuli and overall alertness. At 12 months, lower scores on both the psychomotor and mental developmental indexes of the Bayley Scales of Infant Development were associated with high prenatal anxiety. At 2 years, high anxiety also predicted lower scores on the mental developmental index. Interestingly, the strength of association between mental development at the age of two and prenatal anxiety ($\beta = -.33$) was similar to that for maternal educational level ($\beta = .26$), a consistently potent predictor of childhood IQ.

Laplante et al. (2004) took a different approach and examined an objective stressor rather than perceptions of stress. They examined the effects of exposure during the prenatal period to the natural disaster of an ice storm in Québec, Canada in January 1998 on the language and intellectual development in 2-year-old children. Higher levels of objective stress exposure during pregnancy were associated with lower performances on the Bayley Mental Developmental Index. In addition, higher prenatal stress exposure was associated with the toddlers' productive and receptive language skills.

Huizink, Robles de Medina, Mulder, Visser, and Buitelaar (2003) studied the effects of prenatal stress, with both psychological and physiological markers, on infant developmental outcomes. Prenatal stress was assessed with measures of maternal daily hassles, pregnancy anxiety, and cortisol at 15–17 weeks, 27–28 weeks, and 37–38 weeks of gestation. Measures of motor and mental development were gathered with the Bayley Scales of Infant Development at 3 and 8 months of age. Results indicated that greater levels of daily hassles during early pregnancy were associated with lower mental development at 8 months. Further, pregnancy anxiety during mid-pregnancy negatively predicted mental and motor development also at 8 months. Importantly, these results were found after taking into account, statistically, gestational age at birth, birth weight, and postnatal maternal perceived stress and depression. Although the psychological factors appeared to exert influence in the early stages of pregnancy, it was only in late pregnancy that an association between cortisol and development was found. Higher early morning cortisol levels were associated with decreased mental and motor development at 3 months and mental development at 8 months.

Despite the paucity of research in this area, it does appear that research with humans is beginning to reinforce the strong findings with animal models linking prenatal stress with adverse cognitive and neurological developmental outcomes (Braastad, 1998; Schneider, Moore, Kraemer, Roberts, & DeJesus, 2002; Weinstock, 1996). The few existing human studies show that whether prenatal stress is measured as a psychological construct, or a physiological marker, it appears to have an impact on postnatal cognitive development.

VIII. PRENATAL STRESS AND THE BRAIN

There are at least two direct ways through which prenatal stress might affect the structure and function of the brain. First, there is evidence in humans that prenatal stress is associated with decreased growth including head size (Lou et al., 1994; Mulder et al., 2002). Second, areas of the brain that are particularly sensitive to stress hormones, such as glucocorticoids

(GCs), may be altered through stress exposure. There appears to be some evidence to support both views, which are not mutually exclusive.

To date, although no study exists that examines the relation between prenatal stress and brain development in humans, there is one study that links prenatal stress with head circumference. Lou et al. (1994) identified 70 women from a large population study who had experienced, during pregnancy, moderate to severe stressors according to the *Diagnostic and Statistical Manual of Mental Disorders, 3rd Edition Revised* (DSM-III-R) definitions. They also reported low social support. These women were compared to matched controls with low levels of stress and adequate social networks. Stress that was assessed during mid-gestation was associated with reduced head circumference even after accounting for the smaller total birth weights resulting from higher stress pregnancies. This suggests that prenatal stress affects brain development, a conclusion that was further supported by the finding that the prenatally stressed children in this sample also scored lower on a measure of neonatal neurological functioning.

IX. PRENATAL GC THERAPY AS A MODEL OF PRENATAL STRESS

There is some evidence to suggest that endogenous, or naturally occurring, GCs—cortisol in humans—may mediate the relation between prenatal stress and developmental outcomes (Barbazanges, Piazza, Le Moal, & Maccari, 1996; Welberg & Seckl, 2001). Production of cortisol increases in response to stress exposure, it readily passes through the placenta, and many areas of the brain are sensitive to GCs (Matthews, 2000; Seckl, 1997). A pharmacological model that can provide insight into the effects of endogenous GCs in humans is the widespread therapeutic use of exogenous or synthetic GCs for threatened preterm delivery. Administration of synthetic GCs (dexamethasone and betamethasone) to those in preterm labor is an established treatment that decreases morbidity and mortality in those children born early, largely through accelerating lung development in the fetus (Crowley, 1995).

Despite the clear therapeutic benefits of prenatal administration of GC's, questions have begun to arise addressing the costs that also may exist. It has been suggested that this therapy may have lasting, negative effects on other systems, especially the hypothalamic-pituitary-adrenal (HPA) system and growth (Davis et al., 2004b). The results of studies examining the link between prenatal GC therapy and cognitive development are mixed with some studies showing detrimental effects, whereas others do not (Kay, Bird, Coe, & Dudley, 2000). However, there are studies showing that prenatal

treatment with synthetic GCs is also associated with smaller head circumference (French, Hagan, Evans, Godfrey, & Newnham, 1999; Thorpe, Jones, Knox, & Clark, 2002) and decreased cortical volumes (Modi et al., 2001).

Although definitive answers to the existence of long-term adverse effects of prenatal GC treatment in humans await the completion of currently ongoing randomized controlled trials, primate studies do strongly support the notion that prenatal stress and GCs influence both brain function and morphology. Administration of GCs to pregnant baboons in levels similar to those given in clinical use in humans resulted in reduced levels of microtubule-associated proteins and synaptophysin in the neocortex, both essential for brain development (Antonow-Schlorke, Schwab, Li, & Nathanielsz, 2003). There also is some evidence suggesting that certain areas of the brain, especially the hippocampus, may be particular targets of prenatal GC exposure. For example, exposure of pregnant primates to dexamethasone has been shown to reduce hippocampal formation and volumes (Uno et al., 1990).

A note of caution should be given in interpreting the studies of synthetic or exogenous GCs in the context of a model of the effects of endogenous or naturally occurring GCs. Naturally occurring cortisol and synthetic betamethasone and dexamethasone all readily pass through the placental barrier. However, there is one critical difference: a placental enzyme 11 β-hydroxysteroid dehydrogenase type 2 (11 β-hsd2) converts the cortisol to cortisone, but dexamethasone and betamethasone are not adequate precursors and so pass through to the fetal adrenals. Thus, most of the naturally occurring cortisol is inactivated and unavailable to influence the fetal adrenals, whereas the exogenous GCs retain their ability to influence. With that being said, it is still the case that synthetic GC models provide insight into the potential effects of maternal GCs because not all endogenous cortisol is converted to inactive cortisone.

X. TIMING OF STRESS AND DEVELOPMENT

In considering the impact of stress during pregnancy it is necessary also to consider the timing of the stressor or insult. The effects of timing of a stressor during gestation are determined by at least three factors, two related to the fetus and one to the mother. It has long been acknowledged that timing of insults to the fetus will have different effects depending on developmental trajectories (Joseph, 2000; Selevan, Kimmel, & Mendola, 2000). That is, the timetable of development of the organ systems as well as the development of the central nervous system will determine in part the effects of stress exposure. A second sort of fetal vulnerability relates to placental 11 β-hsd2, the placental enzyme that renders cortisol to inert cortisone. As a result of its

presence in the placenta, the great majority of maternal cortisol is inactivated when crossing the placental barrier. 11 β-hsd2 activity in the placenta increases during the gestational period starting in the first trimester (Shams et al., 1998; Shcoof et al., 2001) and may begin to decrease at term >38 weeks (Murphy & Clifton, 2003). It seems plausible to suggest that when 11 β-hsd2 is most abundant will be the time period in which the fetus may be most protected from maternal GCs and also corresponds to the period in which the pregnancy-related increase in maternal cortisol levels are highest.

The timing of stress also appears to be important in the context of the maternal stress system. The placenta, a transient organ of fetal origin, plays a primary role in the changes in the maternal–fetal endocrine milieu (Petraglia, Florio, Nappi, & Genazzani, 1996). It is not widely appreciated, however, that these hormonal alterations have implications that extend beyond the reproductive process, also influencing the maternal stress system. As pregnancy progresses to term, the functioning of the maternal stress system is altered as levels of the products of the HPA axis rise: cortisol, adrenocorticotropin, and beta-endorphin increase two- to threefold and plasma levels of placental CRH increase 25-fold (Goland, Conwell, Warren, & Wardlaw, 1992; McClean, Davies, Woods, Lowry, & Smith, 1995; Sandman et al., 2003; Schulte, Weisner & Allolio, 1990). As pregnancy progresses women are physiologically and psychologically less reactive to stress (Glynn, Schetter, Wadhwa, & Sandman, 2004; Glynn et al., 2001; Matthews & Rodin, 1992; Schulte et al., 1990). The implication of these findings is that the impact of stress during pregnancy is not uniform. Stressors experienced early in pregnancy have greater impact, on both physiological and psychological responses, than stress experienced later. There is some evidence that, in humans, early stress does have a greater impact on birth outcome and on later development. Two studies have shown that insults experienced early in pregnancy are more likely to result in shortened gestation (Glynn et al., 2001; Lederman et al., 2004). In addition, a study highlights the importance of timing for developmental outcomes, demonstrating that the occurrence of a stressor during the first or second trimester, but not the third, was associated with lower levels of intellectual functioning in 2-year-old children (Laplante et al., 2004).

XI. PRENATAL STRESS, GROWTH, AND THE FETAL PARADIGM

Investigations of the influence of stress on human development can be extended into the prenatal period. There is a growing body of literature aimed at assessment and characterization of fetal development *in utero*. It does seem possible to detect cognitive or neurological disability prenatally.

For example, Johansson, Wedenberg, and Westin (1992) were able to detect hearing impaired fetuses *in utero* with an acoustic stimulation paradigm. It also has been demonstrated that these paradigms can detect more subtle influences on development. In two studies, the influence of maternal-placental endocrine milieu on fetal behavior has been shown. Both have demonstrated that indices of fetal learning are affected by maternal stress hormones. Higher levels of corticotropin-releasing hormone (CRH) were associated with depressed responses to novelty in 32-week-old fetuses (Sandman, Wadhwa, Chicz-DeMet, Porto, & Garite, 1999). Similarly, a measure of endocrine disregulation (adrenocorticotropic hormone/beta-endorphin (ACTH/BE)) predicted rate of habituation in 32-week-old fetuses (Sandman et al., 2003). Relatedly, 32- to 36-week-old fetuses of depressed women had elevated baseline heart rates and a delayed heart rate recovery following vibroacoustic stimulation (Allister, Lester, Carr, & Liu, 2001). Other work demonstrates continuity between fetal and infant motor activity (Almli, Ball, & Wheeler, 2001; Groome et al., 1999; Ratcliffe, Hellar, & Leader, 2002), temperament (DiPietro, Hodgson, Costigan, & Johnson, 1996), and autonomic function (DiPietro, Costigan, Pressman, & Doussard-Roosevelt, 2000; Fifer, Hurtado, Garcia, & Myers, 1998). Expansion of these sorts of studies during the fetal period will provide additional insight into the effects of timing of stress as well as the systems that are targeted by exposure to stress. They may even have the potential to provide tools useful for identifying those at later risk for major, and also more subtle, developmental delay or impairment.

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Fluid Cognitive Abilities Neglected Aspects of Cognition in Research on Mental Retardation

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> I do not believe that anyone would argue with the staement that a 7-year-old retarded child is less intelligent than a 7-year-old child who is not retarded. Unfortunately, when we venture even a short distance from this statement, we immediately find ourselves adrift on a sea of definitional uncertainty. For if we ask the rather basic question of what we mean by intelligence, we encounter considerable disagreement that has only intensified in recent years (Zigler, 1999, p. 1).

By definition, individuals with mental retardation (MR) are characterized by a deficit in general intelligence. Determining just what the nature of this deficit is, however, and how it is related in specific ways to the functioning of persons with MR has proven to be quite challenging. Individuals with MR exhibit developmental delay in a wide variety of cognitive processes and functions that result in IQ scores of 70 or below. Relative to typically developing individuals, those with MR exhibit delays in memory, speed of processing, inspection time, perceptual acuity, and a number of other domains. As a result, attempts to isolate a central cognitive deficit that could perhaps explain the overall deficit in general intelligence in individuals with MR have not been successful. Research on cognition in MR stands as a prominent indicator of the fact noted in the quote from Edward Zigler above, that general intelligence is poorly understood.

No doubt, there has been and continues to be some expectation that research on cognition in MR will yield valuable information on intelligence that can advance understanding of human cognitive abilities and inform the design of programs to remediate or prevent developing mental retardation.

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The purpose of this chapter is to suggest that one possible way in which this can happen is through the continued examination of aspects of cognitive ability that we will refer to under the general heading of fluid cognitive abilities. Fluid cognitive abilities are well known in the cognitive psychological literature under the terms executive function (EF) and working memory. These terms encompass processes that include the holding of information in mind, the inhibiting of prepotent or extraneous information or responding when engaged in task completion, and the appropriate shifting of attention between various goal-relevant aspects of a given task. Fluid cognitive abilities also feature largely in performance on measures of what is known as fluid intelligence. Fluid, as opposed to crystallized, intelligence refers to the ability to comprehend novel or unfamiliar stimuli and includes abstraction, relational reasoning, and planning and problem-solving abilities. The effortful, domain general nature of fluid intelligence is best seen in juxtaposition to crystallized intelligence, that associated with highly learned, acculturated, and relatively automatic aspects of cognitive function such as language ability, vocabulary, and information available in long-term memory.

The distinction between fluid and crystallized aspects of intelligence has been comprehensively described in what is known as the gF–gC theory of intelligence. This theory, first proposed by Horn and Cattell (1966) but also present in Spearman's early theorizing on intelligence, states that while fluid (gF) and crystallized (gC) aspects of general intelligence are related and can be subsumed under a single factor, they represent distinct aspects of mental functioning (Carroll, 1993; McGrew, 1997) with unique developmental trajectories (McArdle et al., 2002). Measures of fluid intelligence, such as the Raven matrices test, the performance subscales of the Wechsler batteries, or the fluid subtests of the Woodcock–Johnson tests of cognitive ability, require the application of the processes of fluid cognition. For example, the Raven matrices test is thought to be a highly effective measure of fluid intelligence because it makes demands of multiple aspects of fluid cognition simultaneously. In the matrices test, the individual is presented with patterns of increasing complexity, each of which is missing a single uniformly shaped piece. At the bottom of each pattern, six versions of the missing piece are presented, only one of which correctly fits the pattern. Correct solving of the task requires the individual to choose from among similar pieces, holding diverse aspects of the pattern in mind, shifting attention among these aspects while considering the fit of each piece to the pattern as a whole, and inhibiting any tendency to focus on or respond to only one aspect of the pattern completion problem. Consideration of the cognitive operations required for the performance of this task can lead one to the conclusion that fluid intelligence is little more than the sum of its fluid cognitive components (Carpenter, Just, & Shell, 1990; Kyllonen & Christal, 1990).

Although distinctions among gF, fluid cognitive abilities, and the general factor of intelligence remain controversial, a close look at these constructs is important for understanding the nature of intelligence and therefore mental retardation. Of particular interest for MR research is a variety of evidence indicating that while gF has in many instances been shown to be highly similar if not identical to the general factor of intelligence, in many other instances it has been shown to be clearly distinct from general intelligence. This paradoxical nature of similarity and difference between gF and general intelligence may be important for understanding the experience and development of persons with mental retardation. At the very least, the somewhat unusual empirical base on fluid cognition and general intelligence (reviewed in a later section) suggests that fluid cognition is an excellent topic of study for research on mental retardation. Surprisingly little research, however, has specifically addressed fluid cognitive functioning as an aspect of mental retardation. In this chapter, we will examine possible reasons why this has been the case and suggest that the relative neglect of this specific aspect of cognition in MR is unfortunate. Primarily, we will review evidence that suggests that a focus on fluid cognitive functioning in persons with MR is particularly interesting for what it has to say about the second criterion by which MR is diagnosed, namely adaptive behavior as manifest in a number of personality and social and behavioral characteristics. Working from a person-centered developmental perspective, we will outline how fluid cognitive abilities can be seen to play a role in the interface of social and cognitive functioning. Consequently, we will suggest that the study of fluid cognitive abilities in MR can help to chart a course out of the "sea of definitional uncertainty" noted by Zigler in the opening quote. Specifically, as an intermediary between social–emotional and cognitive functioning, fluid cognition may play a central role in developing social-emotional competence in individuals with MR through its relation with aspects of developing personality and motivation. As such, we propose that fluid cognition can help to expand the conceptual and empirical basis for definitions of adaptive behavior and constructs, such as outerdirectedness and positive and negative reaction tendencies, that have featured prominently in the holistic approach to the study of cognitive and social development in persons with mild MR.

As other reviews have focused on aspects of fluid cognition, primarily referred to as EF, in specific syndromes, such as autism (Pennington & Bennetto, 1998), this chapter will focus primarily on idiopathic, nonorganic or cultural–familial, mild mental retardation. In the first section, we will describe the study of fluid cognition in typically developing children and adults. In this description, we will outline how fluid cognition has been shown to be central to general intelligence in many instances but highly distinct in others. Here we will also pay careful attention to the study of fluid

cognition in a number of developmental disorders and examine evidence indicating dissociation of fluid cognitive abilities from general intelligence.

In the next section, we will describe the role of fluid cognition in developing cognitive and social-emotional competence from a neuroscientific perspective. In particular, we will present evidence that suggests that developmental neuroscience work on stress and emotion early in the life course helps to account for the dissociation between fluid and crystallized aspects of cognitive functioning. Of particular importance here is the way in which work in the developmental neuroscience of fluid cognition suggests that emotion and stress-related processes, in part as structured by the early caregiving environment, may play an important role in the development and utilization of fluid cognitive skills.

In the third section we will examine the idea that work linking stress, emotion, and fluid cognition provides an important integrative link between the development of cognition and the development of adaptive behavior in persons with MR. Here we will review the very limited literature on fluid cognition in the study of MR and consider the relation of this work to the study of motivational and personality characteristics in individuals with MR. In addition, we will consider implications of the study of fluid cognitive abilities for developmental versus deficit approaches to the study of mental retardation. We will also relate work on fluid cognition to an enduring aspect of research on MR, namely Zigler's motivational approach to MR.

In the fourth section we will consider some additional implications of the developmental neuroscience approach to fluid cognition for the study of mental retardation. Central to this discussion is consideration of MR as a disorder of self-regulation and the way in which a focus on the interaction between cognition and emotion in the process of development can help to expand the conceptual basis for the characterization of the self-regulatory deficit in MR. Within this discussion of self-regulation, we will conclude by examining evidence indicating the applicability of a focus on fluid cognition and self-regulation to intervention efforts directed at enhancing cognitive function and improving quality of life for persons with mental retardation.

I. FLUID COGNITION AND GENERAL INTELLIGENCE

As noted earlier, fluid cognitive abilities are those associated with relatively effortful as opposed to automatic information processing that supports the maintenance of information in working memory, the inhibition of prepotent or extraneous information and responding, and the appropriate shifting and sustaining of attention required to maintain a particular cognitive set or framework within a given task or setting. Within this tripartite framework of working memory, inhibitory control, and attention shifting, fluid cognitive

abilities have been studied developmentally under the term EF (Diamond, 2002; Zelazo, Muller, Frye, & Marcovitch, 2003) and have been examined in relation to temperament and the self-regulation of behavior, and the development of attention abilities (Posner & Rothbart, 2000). Executive dysfunction has also been examined as a feature of a number of developmental disorders and disabilities (Pennington & Ozonoff, 1996; Zelazo & Mueller, 2002).

Although there are important shades of similarity and difference in various approaches to the study of fluid cognitive abilities as embodied in the terms EF and working memory [working memory broadly defined as in Baddeley's (1992) model or Engle's (2002) working memory capacity model], a somewhat less subtle, but critical distinction for MR research concerns the wide variety of data suggesting that fluid cognitive processes are central to if not identical with general intelligence. Tests that measure fluid cognitive functions, particularly working memory, have higher g loadings than do other cognitive measures, that is, exhibit larger factor scores on the higher order g factor extracted from hierarchical analysis of mental test batteries (Colom, Rebollo, Palacios, Juan-Espinosa, & Kyllonen, 2004; Gustafsson, 1984, 1988). And measures of working memory have been shown to correlate extremely highly (r's $> .90$), with the general factor extracted from various measures of cognitive ability (Colom et al., 2004; Gustafsson, 1988; Kyllonen, 1996; see Kane & Engle, 2003 for review). Latent variable models have also shown working memory and cognitive control processes to be central aspects of general intelligence (Embretson, 1995; Süß, Oberauer, Wittman, Wilhelm, & Schulze, 2002).

Furthermore, psychometric data are not the only source of evidence indicating the centrality of fluid cognition in general intelligence. Studies examining brain structures and neural interconnectivity that support fluid cognitive functions (Braver et al., 1997; MacDonald, Cohen, Stegner, & Carter, 2000; Smith & Jonides, 1997) indicate a high degree of overlap within the brain between fluid cognition and general intelligence (Duncan et al., 2000; Prabhakaran, Smith, Desmond, Glover, & Gabrieli, 1997; Thompson et al., 2001; but also see Haier, Jung, Yeo, Head, & Alkire, 2004). Structural magnetic resonance imaging has indicated positive correlations between IQ and gray matter in the prefrontal and anterior cingulate cortices (ACC) and has shown frontal gray matter, as with IQ, to be highly heritable (Thompson et al., 2001; Wilke, Sohn, Byars, & Holland, 2003). Functional brain imaging studies have also consistently demonstrated activations in dorsolateral areas of the prefrontal cortex (PFC) in response to working memory tasks that are highly similar to those observed in response to measures of general fluid intelligence such as the Raven matrices test (Duncan et al., 2000; Prabhakaran et al., 1997).

By demonstrating relations of gray matter volumes and PFC activation to IQ, usually as measured by the full scale IQ score of the Wechsler batteries, a number of studies present an apparent neurocognitive basis for general intelligence. Although seemingly straightforward, however, the relation of activation in the PFC to performance on working memory tasks and tests of intelligence is not one in which greater activation necessarily equals higher performance. Examinations of individual differences in working memory capacity indicate higher levels of PFC activation at moderate working memory loads in adults with limited working memory capacity relative to adults with greater working memory capacity. Change in activation in the PFC in response to increasing working memory load demonstrates an inverted U shape with increasing activation at initial load levels followed by decreasing activation once a capacity set point is exceeded (Callicott et al., 1999; Goldberg et al., 1998; Rypma et al., 1999).

Increased frontal activation in individuals with lower general intelligence and working memory capacity appears to reflect cortical processing inefficiency in the PFC and is similar to the finding of an inverse correlation between cerebral glucose metabolic rate (GMR) and IQ. Specifically, initial examination of cerebral GMR in response to the Raven progressive matrices test indicated an inverse relation between whole brain GMR and fluid intelligence (Haier et al., 1988). However, in a follow-up study in which problem difficulty was matched to participant aptitude, using hard and easy backward digit span trials (a standard measure of the working memory aspect of fluid cognitive function), cerebral GMR was found to be positively correlated with fluid intelligence (Larson, Haier, Lacasse, & Hazen, 1995). These findings indicate the need to take into account effort and difficulty level when examining relations between brain activation and measures of intelligence. In an extension of these findings to the instance of low general intelligence, cerebral GMR during the performance of a continuous performance task was found to be higher in participants with mild MR relative to a control group (Haier et al., 1995). Although the small sample size in this study (approximately nine per group) precluded examination of within group variation, it is likely that as with nondelayed participants, individual differences in GMR are associated with variation in processing efficiency and performance on measures fluid cognition and attention in persons with mild MR.

II. DISSOCIATION OF FLUID COGNITION FROM GENERAL INTELLIGENCE

Paradoxically, despite what would seem fairly definitive psychometric and brain-imaging evidence indicating the similarity of fluid cognition and

general intelligence, a variety of work, including that on cognitive impairments in established disabilities, indicates that fluid cognition is in many respects distinct from psychometric g (for review see Blair, 2006). Specifically, work ranging from that on secular change in population mean IQ over the last 100 years (Flynn, 1999), to clinical neuropsychological work on individuals sustaining damage to specific areas of the PFC (Waltz et al., 1999), to that on cognitive impairments in ADHD, autism, and learning disabilities (McLean & Hitch, 1999; Pennington & Ozonoff, 1996; Zelazo & Mueller, 2002), indicate dissociation of fluid cognitive functions from other aspects of cognitive ability.

Historical data from normative samples of several widely used measures of intelligence indicate a secular rise in IQ over the past century in which gains are particularly large on tests of fluid cognitive abilities. In fact, gains are greatest, upward of 18 points in a single generation, on the Raven matrices test, a test, as noted earlier, that is highly dependent on fluid cognitive abilities and the integrity of the PFC (Carpenter et al., 1990; Gray, Chabris, & Braver, 2003). Mean IQ gains on measures more closely tied to crystallized intelligence, however, are considerably smaller and become increasingly small the closer intelligence subtests come to measuring purely crystallized aspects of cognition. The very rapid and substantial rise in scores on measures of fluid intelligence without a concurrent rise of similar magnitude in crystallized skills suggests dissociation of fluid cognitive functions from g.

Similarly, but with the emphasis on a decrease rather than a rise in fluid intelligence, adults with damage to the PFC perform very poorly on fluid cognitive tasks but exhibit measured general intelligence within the normal range (Duncan, Burgess, & Emslie, 1995; Waltz et al., 1999). Individuals with damage to the PFC exhibit scores on measures of fluid intelligence that are 1–3 SD below their scores on measures assessing primarily crystallized intelligence. Such data can easily be taken, and have been previously by many, as support for what would seem to be the erroneous conclusion that prefrontally based fluid skills are unrelated to intelligence! However, adult patients with lesions to the PFC demonstrate intact IQ relative to matched controls as assessed by the Wechsler Intelligence Scale for Adults (WAIS) while simultaneously exhibiting substantial postmorbid fluid intelligence deficits as measured by tests such as the Raven matrices test and Cattell Culture Fair Test (Duncan et al., 1995). In essence, while the crystallized IQ of these individuals is in the normal range, fluid IQ scores are in the range of mental retardation. No such discrepancy is observed among matched controls who, in fact, exhibit fluid intelligence scores equivalent to or higher than their WAIS scores. Further examination of the deficit displayed on measures of fluid IQ in patients with frontal lesions, but intact IQ as assessed by the WAIS indicates that performance is dramatically impaired by the

requirement of holding multiple relations in mind simultaneously when attempting to solve problems adapted from the Raven matrices test. Individuals with prefrontal damage exhibit no deficits on problems whose solution requires holding in mind no relations or only one relation but exhibit a near inability to solve problems involving two or more relations (Waltz et al., 1999).

Evidence of the differentiation of fluid skills from g is also provided by the study of cognitive impairment among individuals with specific developmental disorders. Studies examining a variety of developmental disorders of childhood indicate that children with attention deficit hyperactivity disorder (ADHD), early and continuously treated phenylketonuria (PKU), and specific learning disabilities (LD) exhibit impaired performance on measures of fluid cognition, but general intelligence in the normal range (Barkeley, 1997; Berlin, 2003; Diamond, Prevor, Callender, & Druin, 1997; McLean & Hitch, 1999; Stanovich, Siegel, & Gottardo, 1997; Swanson, 1999). Furthermore, some studies have identified specific patterns of fluid deficits associated with different disorders. In an examination of four developmental disorders, ADHD, autism, conduct disorder (CD), and Tourette syndrome (TS), consistent EF deficits were identified in ADHD and autism but not CD and TS. More severe deficits relative to IQ matched controls were observed in autism compared with ADHD. In contrast, children with ADHD exhibited greater deficits in inhibitory processes relative to children with autism (Pennington & Ozonoff, 1996).

Prima facie developmental evidence for the distinction between fluid cognitive functions and measures of general intelligence is provided by work on specific LD, as defined in the United States. In specific LD, deficits in fluid cognition impair learning and academic achievement, but general intelligence is in the normal range. Examination of EF in studies of both reading and math disability have indicated fluid cognitive impairments in comparisons with age-matched and to some extent ability-matched (i.e., younger) controls. Differences in the maintenance of information in working memory and in inhibitory control have been noted in the presence of measured intelligence in the normal range (McLean & Hitch, 1999; Pennington, 1997; Sikora, Haley, Edwards, & Butler, 2002; Swanson & Sachse-Lee, 2001; Willcutt et al., 2001).

Given considerable evidence for dissociation of fluid and crystallized aspects of cognition in a number of developmental disorders, it would be surprising if the dissociation were not also observed to some extent among persons for whom overall level of general intellectual functioning is low. Although the bulk of the evidence in the study of disability makes reference to fluid cognitive deficits in the presence of general intelligence in the normal range, it is certainly likely that variation in fluid cognitive ability exists in the presence of low general intelligence. Furthermore, just as deficits in fluid

abilities appear to be quite detrimental to real world competence in individuals with intact IQ, so might higher levels of fluid cognitive competence prove beneficial in the lives of individuals with mild mental retardation. To understand how this may be the case, it is necessary to turn to work that examines the relation of fluid cognitive processes to stress and to emotion and thereby to aspects of developing competence important for the self-regulation of behavior and social–emotional competence.

III. DIFFERENTIATING FLUID COGNITION AND GENERAL INTELLIGENCE: A DEVELOPMENTAL NEUROSCIENCE PERSPECTIVE

Some insight as to how fluid cognitive functions can be so similar to yet so distinct from other aspects of cognition is gained from a developmental neuroscience approach to the study of human cognitive abilities. Given that fluid cognitive functions have been associated primarily with the prefrontal and anterior cingulate cortices, it is of considerable interest that these brain areas are extensively and reciprocally interconnected with limbic and brainstem structures associated with emotional reactivity, the stress response, and autonomic nervous system function (Allman, Hakeem, Erwin, Nimchinsky, & Hof, 2001; Bush, Luu, & Posner, 2000; Drevets & Raichle, 1998; LeDoux, 1989; Paus, 2001; see edited volume by Uylings, Van Eden, de Bruin, Feenstra, & Pennartz, 2000). In combination, prefrontal, limbic, and brainstem structures integrate cognitive, emotional, and autonomic responses to stimulation with the primary implication of such reciprocal innervation and regulation being that prefrontally mediated fluid cognitive processes directly influence and, most importantly for present purposes, *are influenced by* emotional and autonomic responses to stimulation (de Kloet, Oitzl, & Joels, 1999; Erickson, Drevets, & Schulkin, 2003; Groenewegen & Uylings, 2000; Kaufman & Charney, 2001).

For example, the ACC contains areas in its ventral and dorsal segments that are associated with the processing of emotional and cognitive information, respectively. Brain-imaging studies of the processing of attention and emotion-related information in the PFC and ACC indicate the integrated and reciprocal relation between affect and cognition in the brain. Distinct dorsal regions of the ACC are activated in response to cognitive tasks and ventral regions to stimuli eliciting emotional arousal (Bush et al., 2000; Drevets & Raichle, 1998). Similarly, examinations of intentional reappraisal of emotional arousal and of changes in emotional state associated with emotionally arousing stimuli have indicated reciprocal prefrontal cortical-limbic activation (Mayberg et al., 1999; Ochsner, Bunge, Gross, & Gabrieli, 2002). With reappraisal of negative emotion and recovery from sadness and

depression, prefrontal and dorsal ACC activity is increased and limbic and ventral ACC activity is decreased. During periods of negative affect without reappraisal, however, limbic and emotional ACC activity is increased and prefrontal and cognitive ACC activity is decreased. Such reciprocal interconnectivity of emotion and cognition in the brain is highly consistent with the goal directedness of fluid cognitive functioning. Working memory and cognitive control processes are utilized in the service of specific goals related to problem solving and learning. However, at high levels of emotional arousal, fluid cognitive functions become inhibited and impairments in the control of attention, working memory, and inhibitory control are more likely to occur.

Consistent with the relation between arousal and cognitive function, studies of the stress response indicate an inverse U relation between fluid cognitive abilities and activation of the hypothalamic-pituitary-adrenal (HPA) axis stress response system. In the HPA response, stressful stimuli trigger a cascade leading from the hypothalamus to the pituitary to the adrenals to produce the glucocorticoid hormone cortisol. Levels of circulating cortisol feedback on the amygdala, the subcortical brain structure associated with fear responding, to maintain the activation of the HPA stress response system and sustain high levels of cortisol. In response to cortisol increase, however, the PFC and the hippocampus provide negative feedback to the central nucleus of the amygdala to downregulate the response to stress (Francis, Caldji, Champagne, Plotsky, & Meaney, 1999; Kaufman & Charney, 2001). In both human and nonhuman animal models, very high or low level of cortisol has been associated with impairments in fluid cognition and learning and memory. In contrast, moderate increases in cortisol have been associated with increased cognitive performance (de Kloet, Oitzl, & Joels, 1999; Erickson et al., 2003; Lupien, Gillin, & Hauger, 1999; Roozendaal, McReynolds, & McGaugh, 2004).

The inverted U shaped relation between cortisol and fluid cognition is consistent with the role of glucocorticoids in brain function. Specifically, of the two types of corticosteroid receptors in the brain, glucocorticoid receptors (GR) and mineralocorticoid receptors (MR), GR bind cortisol with less affinity than do MR and therefore remain unoccupied at low levels of stimulation. However, with moderate cortisol increase, GR occupation increases, supporting synaptic long-term potentiation (LTP) important for learning and memory consolidation (de Kloet et al., 1999; Erickson et al., 2003). Significantly, regions of the PFC and ACC important for fluid cognitive functions and learning and memory (Allman et al., 2001; MacDonald et al., 2000; Paus, 2001) have been shown to contain a relatively high density of GRs (Diorio, Viau, & Meaney, 1993; Roozendaal et al., 2004).

Abnormally high or low levels of cortisol, indicating increasingly high or low GR occupation, however, are associated with fluid cognitive and

behavior regulation deficits. In contrast to the association between moderate levels of GR occupation and LTP, high or low levels of GR occupation are associated with synaptic long-term depression (LTD) rather than LTP. This relation between cortisol and cognition has been demonstrated in cortisol infusion studies conducted with adults in which moderate increases in cortisol are associated with higher levels of performance on tasks assessing fluid cognitive functioning (Lupien, Gillin, & Hauger, 1999; Lupien et al., 2002). In contrast, highly elevated cortisol levels have been associated with impairments in declarative or long-term memory function in adults, as well as deficits in fluid cognitive functioning, in many (see Erickson et al., 2003 for review) but not all studies (Monk & Nelson, 2002). Similarly, hypocortisolism, that is, persistently low level of diurnal cortisol, has been associated with cognitive and behavior regulation deficits both among children experiencing severe trauma and neglect but also children experiencing low levels of caregiving sensitivity within what would be considered the normal range of care (Gunnar & Vasquez, 2001).

In children it has been assumed that levels of cortisol would be linearly and positively related to attentional and behavioral difficulties (Gunnar, Tout, de Haan, Pierce, & Stansbury, 1997; Gunnar & Donzella, 2002). However, in keeping with the inverse U relation between cortisol and fluid cognition and behavioral self-regulation, moderately higher levels of cortisol have been associated with increased performance on measures of executive function and behavioral self-regulation in children (Blair, Granger, & Razza, 2005; Davis, Bruce, & Gunnar, 2002). Very high or very low levels of stress reactivity and difficulty with the up- or down-regulation of this reactivity are likely to be associated developmentally with problems with fluid cognition and self-regulation, particularly for children facing early stress. Furthermore, given the principle of use-dependent synaptic plasticity and the fact that the PFC is relatively slow in maturing (Gogtay et al., 2004), it is likely that reciprocity between prefrontal and limbic brain structures in the up- or down-regulation of the glucocorticoid response to stress may become biased toward either emotional reactive or effortful cognitive types of responding fairly early in life. Particularly in young children, fluid cognition and the many aspects of self-regulation to which fluid cognition is related may be driven to some extent by early experience effects on emotional arousal and difficulty with the regulation of this arousal.

A. Genes and Environments

A focus on the stress response and developing emotionality and emotion regulation in the study of fluid cognition provides for a systems approach to understanding developing competence, one that integrates genetic and

constitutional aspects of the child with early caregiving experience in a holistic and person-oriented approach to development. Of particular interest are a number of studies indicating the relation of specific genetic polymorphisms associated with neurotransmitter regulation to fluid cognitive abilities. Single nucleotide polymorphisms (SNP) associated with neurotransmitter and neurotrophin activity in the hippocampus and PFC are related to variation in fluid cognitive function and perhaps susceptibility to early stress effects on cognition. Individual differences in one aspect of fluid cognitive ability, the amount of information that can be actively maintained in working memory, have in part been related to an SNP of the catechol-*o*-methyl-transferase (COMT) gene, the val$^{108/158}$met polymorphism. This polymorphism determines the enzymatic action of the gene in inactivating dopamine. Individuals with the valine (val) allele of this polymorphism exhibit more rapid dopamine inactivation and reduced working memory ability. Furthermore, the methionine (met) version of the allele is unique to humans and those who carry this allele inactivate dopamine in the PFC less rapidly. The action of the gene is due to the relatively low presence of synaptic dopamine transporters in the PFC. The effect of the polymorphism on learning and memory associated with PFC function has been demonstrated in rodents and humans. COMT knockout mice display enhanced learning and memory and COMT inhibitors improve working memory performance in both mice and humans. Humans with the met/met genotype display high levels of performance on tests of working memory function while individuals with the val/val genotype perform worse. Individuals who are heterozygous display performance intermediate to those who are homozygous (Egan et al., 2001).

Similarly, study of an SNP related to the action of the human brain-derived neurotrophin factor (BDNF) gene in the hippocampus, the BDNF val^{66}met polymorphism indicates that individuals with the methionine allele of this polymorphism exhibit memory deficits. Presence of the met version of the allele is associated with reduced levels of *n*-acetyl-aspartate (an indicator of neuronal integrity) in the hippocampus and reduced episodic memory ability (Egan et al., 2003). Furthermore, brain imaging indicates that individuals with the met version of the allele exhibit reduced hippocampal activity in the encoding phase of working memory and that the interaction of met genotype with mean left hippocampal activity during encoding, along with mean left hippocampal activity during retrieval, account for substantial variance in working memory performance (Hariri et al., 2003).

Work on genetic polymorphisms associated with neural functioning helps to identify meaningful sources of variation in fluid cognitive abilities in humans and highlights the role of hippocampal and prefrontal cortical function in fluid cognition. These findings also help to underscore the

complexity of identifying determinants of variation in human cognitive functioning. They emphasize the need for studies that combine work of the type investigating genetic background with a clear characterization of early experience and developing physiology, cognition, and behavior in the prediction of developmental outcomes in diverse samples. Imperative in this approach is the understanding that genetic and physiological predisposition to high or low emotionality or fluid cognitive ability are only meaningfully understood in relation to experience. Whether general intellectual ability is high or low, there are multiple factors that can influence cognitive and social-emotional function and impact the quality of life.

The most compelling demonstration of the relation of early care experience to the development of the stress response and social and cognitive self-regulation is provided by a model of early experience in rodents. Brief maternal separation/handling of rat pups has been shown to increase levels of maternal behaviors upon reunion, namely, licking and grooming and a style of nursing known as arched back nursing, that promote hippocampal synaptogenesis and the development of effective stress regulation. This synaptogenesis in turn has been associated with advantageous learning, memory, and social behavioral developmental outcomes associated with the effective regulation of the stress response (Caldji, Diorio, & Meaney, 2000; Liu, Diorio, Day, Francis, & Meaney, 2000; Liu et al., 1997). In contrast, while high levels of maternal competence have been associated with positive outcomes in rats, both extended maternal separation and low level of maternal competence have been associated with adverse cognitive and behavioral outcomes. Furthermore, rat dams who are more stress reactive engage in a maternal style that tends to pass on the highly reactive profile. The mechanism of transmission of this maternal style through maternal behavior has been demonstrated through experimental cross-fostering manipulations (Liu et al., 1997). Of potentially wide ranging implication in this work is the finding that the effect of maternal behavior on later developmental outcome can be found at the level of the gene. In the rat model, high levels of maternal licking and grooming and arched back nursing chemically alter the expression of a segment of the genome important for the development of GRs and the regulation of the HPA axis, leading to stable and potentially long-term effects of early care on physiology and behavior (Weaver et al., 2004).

Notably for human populations, nonhuman animal models of early stress also indicate that environmental enrichment can offset negative outcomes associated with disadvantageous early care experiences. For example, enriched care is associated with the reversal of the effect of early stress on later stress responsivity and learning and memory in rats (Francis, Diorio, Plotsky, & Meaney, 2002). These studies, however, suggest a functional reversal of the effects of neonatal stress in which early environmental

enrichment leads to compensatory neurobiological mechanisms that alter the phenotypic expression of increased glucocorticoid reactivity and altered hippocampal function resulting from early stress (Bredy, Humpartzoomian, Cain, & Meaney, 2003).

IV. THE STUDY OF FLUID COGNITION IN RESEARCH ON MENTAL RETARDATION

The developmental neuroscience approach to fluid cognition highlights the role of emotional reactivity in fluid cognitive functioning and calls attention to processes and experiences that are distinct from overall level of intellectual functioning. In this, the approach may offer a perspective on developing competence and self-regulation in persons with MR that has not really been examined in much detail. For individuals with MR, as with typically developing individuals, fluid cognitive functioning is important for self-regulatory behavior. However, very little research on cognition in MR has focused specifically on fluid cognition, the stress response, or their role in self-regulation. Compounding this problem, the research that does exist on cognition in MR has employed a between-groups approach, examining cognitive abilities in persons with MR relative to chronological and mental age-matched controls. Furthermore, individual differences in nearly all cognitive abilities, fluid cognition in particular, among persons with MR have received scant attention.

Be this as it may, the work that is available is consistent with the idea that fluid cognition can serve as a valuable focus of work on cognition in mild MR, operating independently of general intelligence to influence competence and adaptive behavior. For example, in a sample of Finnish adults with MR of unknown cause and an average estimated IQ of 64, measures of phonological short-term memory processes characteristic of fluid cognition were unrelated to IQ but were significantly related to reading, writing, and sentence comprehension abilities (Numminen et al., 2000). Furthermore, although measures of working memory were significantly correlated with IQ in this sample, as would be expected, differentiation of the sample by patterns of working memory ability as indicated by a variety of working memory measures demonstrated unique positive associations between working memory and sentence comprehension, reading, writing, and mathematics over and above variance accounted for by IQ.

Similarly, among a sample of children with MR of mixed etiology undifferentiated by IQ, phonological-processing ability examined through measures of phonemic awareness and phonological short-term memory was associated with greater word decoding and progress toward literacy acquisition (Conners, Atwell, Rosenquist, & Sligh, 2001). Intelligence as estimated

by a short form of the WISC-III did not differ between groups characterized by strong and weak decoding abilities, again suggesting the role of fluid skills independent of measured IQ in developing competence in individuals with MR. Although measures of phonological ability are not specifically measures of fluid cognition, fluid abilities play an important role in developing phonological skills. As such, the results are consistent with the idea that fluid cognitive skills independent of general intelligence will be associated with progress toward literacy acquisition in children with MR.

A. Fluid Cognition and Development Versus Difference Approaches to the Study of MR

Although fluid cognitive abilities appear to be an important individual difference characteristic among persons with mild MR, it is interesting to note that somewhat paradoxically fluid cognitive ability is one of the few, if not only, aspects of cognition in which individuals with mild MR have been found to be deficient relative to mental age (MA)-matched controls. Such a difference in cognitive ability in persons with MR relative to typically developing individuals of equivalent MA is discrepant with the lager body of findings indicating that the cognitive development of individuals with idiopathic MR exhibits a structure and sequence appropriate for MA (Bennett-Gates & Zigler, 1999). MA appropriate structure and sequence in persons with idiopathic MR means that these individuals exhibit a level of functioning across a range of cognitive abilities that would be expected at a given developmental age and that the developmental progression of cognitive abilities is essentially the same as that of typically developing individuals, albeit at a slower rate of change. In the study of cognition in MR, however, disagreement has centered on the extent to which this developmental hypothesis should be supplanted in favor of a difference or deficit hypothesis. In the difference perspective, the cognition of individuals with idiopathic MR is thought to be characterized by a core deficit or deficits that result in mental functioning and development in individuals with MR that is substantially different from that of typically developing individuals.

Numerous reviews have detailed the considerable evidence in favor of the developmental perspective (Hodapp, Burack, & Zigler, 1990). Oddly enough, however, evidence in favor of the difference perspective is found in examinations of performance on what can be considered a classic measure of fluid cognitive abilities, the Tower of Hanoi. In the Tower of Hanoi, participants must move disks on pegs from an initial state to a specified goal state while following rules that constrain the movement of the disks. In the completion of this task, individuals with idiopathic MR have been shown to

exhibit levels of performance that are lower than expected for MA in one study (Spitz, Webster, & Borys, 1982) but not in another (Numminen, Lehto, & Ruoppila, 2001). Performance differences in idiopathic MR and MA matched controls on this task in the study showing a difference (Spitz et al., 1982) were associated with limited depth of search in problem solving, operationalized as failure to plan more than one move ahead when attempting to complete the goal state. Similarly, despite equal level of performance in the study showing no difference (Numminen et al., 2001) individuals with MR committed a greater number of rule violations and needed more trials to solve the problems than did the MA controls.

Additional insight into the developmental versus difference distinction in the study of fluid cognition in persons with MR can also be found in a direct comparison of individuals with idiopathic MR and MA-matched controls on various working memory measures (Numminen, Service, & Ruoppila, 2002). In this study, lower than expected working memory ability was observed among individuals with MR; however, this difference was observed only on measures involving nonsemantic information. On measures of working memory involving semantic information, that is, some crystallized knowledge component, performance in the two groups was equivalent, and on measures of skill and knowledge base, vocabulary, reading, writing, and math, individuals with MR outperformed the MA controls (Numminen et al., 2002). These results suggest a qualitative difference in the working memory of individuals with MR such that, according to the authors of the study, individuals with MR may be more dependent on crystallized knowledge when performing working memory tasks whereas MA controls may simply be more efficient in the utilization of working memory processes. In light of the developmental neuroscience approach to fluid cognition, however, individuals with mild MR would be expected to perform less well on working memory tasks involving unfamiliar stimuli as unfamiliarity could lead to increased stress and anxiety and therefore to reduced level of performance relative to MA controls.

Although firm conclusions regarding a developmental or difference perspective on fluid cognitive ability in individuals with MR cannot yet be reached, further work on this issue is important for conceptualizing mild MR. To a large degree, evidence has favored the developmental position on most if not all aspects of cognition, indicating similarity of both structure and sequence among persons with idiopathic MR relative to MA matched controls (Bennett-Gates & Zigler, 1999; Hodapp & Zigler, 1997). Although deficiency in fluid cognitive abilities might seem a likely candidate for a core deficit in MR given the apparent similarity of fluid cognition to g observed in typically developing samples, examination of the motivational approach to the study of cognition in MR combined with a developmental neuroscience

approach to fluid cognition provides an alternative perspective on MA-MR discrepancies in fluid cognitive abilities.

B. Motivational Approach

An alternative explanation for the apparent difference in the fluid cognitive skills of individuals with MR and MA matched controls is one in which the difference relates more to aspects of emotion and motivation than to any inherent cognitive deficit. In particular, given the neuroscience of fluid cognition outlined earlier, factors that heighten negative emotionality and stress would be associated with reduced fluid cognitive functioning and decreased motivation on a given task, leading to lower than expected levels of performance on tasks measuring fluid skills in individuals with MR relative to MA controls. As articulated by Zigler, a motivational deficit associated with repeated failure on cognitive tasks is thought to lead to specific personality characteristics and lowered expectancy of success that can be shown to account for some variation in the cognitive performance of individuals with MR (Bybee & Zigler, 1998). The same is true of children without MR for which little support and encouragement for learning is available (Seitz, Abelson, Levine, & Zigler, 1975; Zigler, Abelson, & Seitz, 1973). This chapter suggests that such a motivational deficit would be largest on fluid cognitive measures because of the close interrelation between aspects of emotional arousal and fluid cognition.

Outerdirectedness and positive and negative reaction tendencies are the terms that describe the emerging motivational deficit and lowered expectancy of success in individuals with MR thought to influence cognitive task performance. As conceptualized and empirically examined by Zigler, Switzky, and others (Harter & Zigler, 1974; Switzky & Haywood, 1991), these constructs are central to a model in which performance on cognitive tasks is understood to reflect a combination of atypical motivation tendencies as well as deficits in intellectual abilities. As originally proposed by Zigler, these personality characteristics of children with MR at least partially result from social deprivation, particularly in the instance of institutional rearing (Zigler, 1969). Individuals who exhibit outerdirectedness imitate the answers of others, even if they know a given answer to be incorrect, rather than independently generate possible unique solutions. Similarly, the positive and negative reaction tendencies describe temperamental inclinations of persons with MR to become either overly dependent on adults or to exhibit a general wariness of adults in ways that adversely affect the development of self-efficacy and intrinsic motivation (Hodapp, Burack, & Zigler, 1990). The positive reaction tendency is perhaps to a large extent the temperamental

characteristic of extroversion or surgency. Similarly, the negative reaction tendency, the inclination to avoid interacting with others in situations requiring assistance, may reflect an underlying temperamental predisposition toward wariness and fearfulness.

As temperamentally based characteristics, positive and negative reaction tendencies may represent the role of constitutional predispositions in individuals with MR just as they do in typically developing individuals (Rothbart & Ahadi, 1994). However, in individuals with MR, levels of general cognitive ability and prior experience that might work to offset temperamental and behavioral tendencies that interfere with cognitive task performance are limited. It is not that individuals with MR differ constitutionally from typically developing individuals but that constitutionally based approach and withdrawal tendencies may play a larger role in developing competence for individuals with MR than for nondevelopmentally delayed individuals. Characteristics are likely to become accentuated developmentally over time with experience, such that a positive or negative reaction tendency is more than simply predisposition. Similarly, the emergence of learned helplessness may be more common among individuals with MR as a history of failure in cognitive tasks may lead to increased likelihood of passive acceptance of failure and the absence of perseverance in response to cognitive challenge (Weisz, 1999). Individuals with MR are more likely to develop an external locus of control and failure-avoiding motivational expectancies as a result of the repeated experience of failure in cognitively challenging situations (Switzky, 1999). It is important to recognize that these aspects of personality and motivation are not in any way unique to individuals with MR and are not simply determined by genetic and constitutional factors but develop in response to experience in expected ways with predictable effects on cognition and behavior.

V. IMPLICATIONS FOR RESEARCH IN MR OF THE DEVELOPMENTAL NEUROSCIENCE APPROACH TO FLUID COGNITION

The developmental neuroscience approach to fluid cognitive function in which levels of negative emotion and stress may interact with and inhibit fluid cognitive functioning is consistent with the work of Zigler and others demonstrating the importance of motivation and personality variables in the study of cognition in individuals with MR. In particular, the developmental neuroscience approach helps to explain why cognitive deficits in individuals with MR relative to MA controls are more likely to be seen on measures of fluid cognition.

One overarching implication of a developmental neuroscience approach to the study of MR relates to the need for an increased research emphasis on the differentiation of cognitive abilities in persons with mild MR. Although assumptions about the general factor of intelligence have perhaps discouraged work on differentiation and on individual differences in patterns of performance on various cognitive measures, researchers have increasingly noted that the full scale IQ score, although perhaps a good indicator of g, by design provides little information on specific aspects of cognition and in some instances may actively obscure information on an individual's relative cognitive strengths and weaknesses (Detterman & Thompson, 1997). Differentiation of fluid cognitive abilities from g is just one example of an area in which it may be valuable to identify relative strengths and weakness in cognitive abilities in persons with MR. For example, the differentiation of fluid cognitive functions from general intelligence and other second-order aspects of cognition, such as speed of processing, would be valuable in the study of developmental disabilities and for designing innovative curricula and teaching approaches to meet the needs of diverse groups of children. In MR research it could also prove valuable in the refinement and definition of the adaptive behavior construct. Fluid cognitive performance higher or lower than expected from measures of crystallized intelligence or speed of processing or estimates of g derived from a Wechsler battery might be expected to be a robust indicator of adaptive functioning.

A second implication of the developmental neuroscience approach to research on MR concerns the developmental interaction between cognition and emotion and the role that the early caregiving environment may play in this interaction. From an organizational systems perspective on development (Cicchetti & Tucker, 1994; Werner, 1957), it is necessary to consider the ways in which an advance or a lag in one developmental domain, such as cognition, may affect and be affected by advance or lag in another such as emotional development. Furthermore, it is necessary to consider the role of environmental factors in providing for and shaping early experiences that may promote or attenuate developmental gains in one or more domain. For example, as shown in the study of the development of children with Down syndrome, cognitive delays that are characteristic of the disorder are associated with a particular pattern of emotional development. Similarly, differences in levels of emotional arousal and arousability may influence the nature and types of cognitive processes that the individual experiences (Cicchetti & Schneider-Rosen, 1984). The structuring of early experience by caregivers will both influence and be influenced by the organizational pattern of cognitive and emotional development. The developmental neuroscience approach to fluid cognition emphasizes the importance of early experience and early relationships with caregivers in providing the foundations both

physiologically and psychologically upon which cognitive and emotional developmental organization can proceed. Early difficulty in the parent–child relationship, for typically developing and developmentally delayed children can be thought to bias the developmental organization of cognitive and emotional functioning in ways that lead to maladaptive, highly reactive responses to the environment that can limit and interfere with normative socialization processes.

A third implication of the developmental neuroscience approach to the study of MR concerns its consistency with a general characterization of mild MR as a disorder of self-regulation as much as a disorder of cognitive impairment (Whitman, 1990; Whitman, O'Callaghan, & Sommer, 1997). Importantly, this characterization emphasizes the role that problems with the regulation of emotion may play in the etiology of MR rather than viewing problems with emotion and emotional development in individuals with MR only as sequelae of intellectual deficits. This approach is consistent with a bidirectional relation between cognition and emotion in which cognition is seen not only to regulate but also to be regulated by emotion. This distinction between appraisal and arousal approaches to the study of relations between cognition and emotion is long standing in psychology. Much of the focus in research on MR has been from an appraisal standpoint (i.e., that in the absence of fully developed cognitive abilities emotion and personality are essentially static). In contrast, however, arousal theories highlight the ways in which emotional arousal, particularly that associated with fear and anxiety but also anger and frustration, can interfere with cognitive functioning, particularly fluid cognitive functions. Advances in neuroscience have demonstrated the action of limbic structures, the amygdala in particular, in response to emotional arousal and highlight the role of subcortical and nonconscious processing in response to and regulation of emotional arousal (Bechara, 2004; LeDoux, 1996). This work clearly suggests the need for an expanded research base on the development and adaptive functioning of individuals with MR that takes into account the neurobiology and psychophysiology of emotional arousal and emotion regulation. Examination of indicators of parasympathetic and sympathetic autonomic nervous system function as well as the HPA response to stress should provide valuable additional information that can help to advance understanding of individual differences in cognitive function and personality development among persons with MR.

A fourth implication of the developmental neuroscience approach to MR concerns the possible trainability of fluid cognitive functions and the generalizability of training to aspects of competence and real world functioning. Training studies of fluid cognitive ability with typically developing children and adults and children with ADHD indicate promising effects on fluid

cognitive abilities and also on the neural substrate that supports fluid cognition. In a controlled randomized study, training of approximately 30 min/day, 5 days/week, for 5 weeks on several simple working memory and inhibitory control tasks in children with ADHD was associated with increased performance on all tasks as well as pretest–posttest treatment related increases in performance on the Raven progressive matrices test and parental reports of inattention, impulsivity, and hyperactivity (Klingberg et al., 2005). These training effects on working memory were also seen in a sample of young adults and shown using fMRI to be observable in the neural substrate that supports working memory. Findings indicated that utilization of the training program was associated with training-related increases in magnetic resonance signal intensity in frontal and parietal cortices during performance of a fluid cognitive task. These training-related increases in signal intensity were not observed in a control task (Olesen, Westerberg, & Klingberg, 2004).

Although training studies are at a very early stage, effects have been seen in adults and children and with different types of training programs and outcomes (Rueda et al., 2005). An important direction for future work, however, concerns the replication of effects with diverse samples of children and adults and the examination of the generalizability of training effects to aspects of functioning with which improvement in fluid cognition might be associated. Studies of fluid cognitive training would seem to be particularly applicable to the study of MR as it may prove to be an aspect of functioning that is amenable to improvement through training and might reasonably be expected to have some advantageous consequences for various aspects of adaptive functioning.

VI. CONCLUSIONS

In conclusion, this chapter suggests that the measurement of fluid cognitive abilities has been a neglected aspect of cognition in research on MR. Somewhat ironically, fluid cognitive abilities can be generally characterized as central to higher order cognitive processing and therefore deficient in persons with MR. Fluid cognition, however, is highly integrated with and to some extent dependent on lower level limbic and autonomic aspects of brain function and information processing. It is this emotion and arousal-related aspect of fluid cognition that makes it a particularly useful aspect of cognition to examine in research on MR. By focusing on the relation between emotion and fluid cognition, the developmental neuroscience approach to the study of fluid cognitive abilities is consistent with a holistic approach to

the study of MR and with the characterization of MR as a self-regulatory disorder. It is likely that further work on fluid cognition in MR that utilizes a developmental neuroscience approach can contribute to a growing and increasingly comprehensive understanding of idiopathic MR.

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Dietary Supplementation with Highly Unsaturated Fatty Acids: Implications for Interventions with Persons with Mental Retardation from Research on Infant Cognitive Development, ADHD, and Other Developmental Disabilities

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I. ATTENTION-DEFICIT/HYPERACTIVITY DISORDER AND MENTAL RETARDATION

Attention-deficit/hyperactivity disorder (ADHD) is the most common neurodevelopmental disorder of childhood with prevalence estimated at between 2% and 18% in the general population of children (Rowland, Lesesne, & Abramowitz, 2002). This syndrome is characterized by a range of cognitive and behavioral symptoms that are shared with a number of other mental health conditions including learning disorders, mental retardation, oppositional defiant disorder, conduct disorder, anxiety, obsessive–compulsive disorder, depression, bipolar disorder, and adjustment disorder (Rappley, 2005). Moreover, ADHD can coexist with many of these same disorders. Szatmari, Offord, and Boyle (1989) reported a large epidemiological survey indicating that up to 44% of ADHD children had at least one other psychiatric disorder, 32% had two others, and 11% had three others. A growing body of researchers have been producing evidence that dietary supplementation can improve learning and behavior of children with ADHD and other developmental disorders. Because ADHD-related symptoms are frequently observed in people with mental retardation, nutritional interventions may

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therefore hold promise also for modifying maladaptive behaviors and cognitive functioning of persons with mental retardation.

The specific symptoms of ADHD include a range of difficulties associated with both cognition and behavior control. These symptoms are used as the basis for describing three subtypes of ADHD: the inattentive subtype, characterized in the classroom or at home by difficulty sustaining attention, not listening or following through, losing and forgetting things; the hyperactive or impulsive type, where the child shows problems controlling his or her own behavior, taking turns, sitting still, and doing things quietly; and a third type that includes both sets of behaviors. The final type accounts for 80% of all diagnosed cases of ADHD.

As indicated by the description of symptoms, the behaviors associated with ADHD overlap significantly with the symptoms associated with the diagnoses of learning disorder and mental retardation. Consistent with this observation, Rappley (2005) described ADHD as both mimicking and coexisting with a range of other mental health conditions. This observation also highlights the potential for difficulty in diagnosing ADHD particularly when mental retardation co-occurs. The extent to which this difficulty creates errors in diagnosis and in establishment of comorbidity cannot be estimated. Nonetheless, many children with ADHD are likely to experience subaverage intellectual functioning or learning difficulties, with Ishii, Takahashi, Kawamura, and Ohta (2003) indicating, that 34% of participants in their study of clinically treated Japanese ADHD sufferers had IQs ranging between 51 and 84. Miller, Fee, and Netterville (2004) suggested that the prevalence of ADHD was up to three times higher in children with mental retardation. Other American studies suggest that the prevalence of ADHD in special classes falls somewhere between 9% and 33% (Das & Melnyk, 1989; Hunt & Cohen, 1998). It is not clear why this prevalence is so high, but biological deficits have been suggested as a possible contributor (Edelstein & Glenwick, 1997).

Co-occurrence of ADHD and mental retardation means that individuals diagnosed with both disorders face long-term difficulties in learning and social adjustment. These difficulties in normal IQ children with ADHD are frequently treated with CNS stimulant medications, such as methylphenidate and dextroamphetamine, with claims that 70–80% of such children will show improved attention with the use of one of these medications (Jadad, Boyle, Cunningham, Kim, & Schachar, 1999). Although the mechanism through which improvement is achieved is not known, research using positron emission topography (PET) has indicated that administering normal doses of methylphenidate to healthy adult men increased their dopamine levels (Volkow, Fowler, Wang, Ding, & Gatley, 2002). These same drug treatments have been used with both children and adults with mental retardation to relieve symptoms of ADHD, with reviews of trials showing

them to be efficacious (Aman, Buican, & Arnold, 2003; Pearson et al., 2003). However, it is important to note that when children with mental retardation have received stimulant medication, improvements in attention and behavior are more variable and subtle than those obtained with children from the general population (Aman, Kern, McGhee, & Arnold, 1993).

Longitudinal follow-up of children with mental retardation and ADHD who were treated several years earlier indicated that many of these children still had behavioral and emotional problems that persisted into late childhood and early adolescence (Aman, Pejeau, Osborne, Rojahn, & Handen, 1996; Handen, Janosky, & McAuliffe, 1997). These difficulties included problems making and keeping friendships, legal difficulties, and psychiatric hospitalization (35% of the Aman et al. sample and 22% of the Handen et al. sample had been hospitalized in the intervening period). As Pearson et al. (2000) pointed out, this high rate of hospitalization of individuals with comorbid mental retardation and ADHD has far-reaching implications for mental health care.

Research by Pearson et al. (2000) further confirms that individuals with both mental retardation and ADHD tend to have a poorer prognosis than those with ADHD alone following treatment with stimulant medications. Higher levels of both emotional and cognitive disturbance characterize them and, although medication may ameliorate some symptomology, it is not a cure nor can it deal with the full range of debilitating behaviors over the longer term. This latter outcome is consistent with the more general findings from research on children with ADHD, the National Institute of Health (NIH) Consensus Statement on the Diagnosis and Treatment of Attention Deficit Hyperactivity Disorder (1998) noting that "stimulant medications may not 'normalize' the entire range of behavior problems." The statement went further by emphasizing that medication appeared to achieve little improvement in academic performance or social skills. There are implications that, if lower IQ is involved, the prognosis for medication appears to be poor. Pearson et al. (2000) have argued that the improvement in ADHD achievable by medication is moderated by patient IQ and that individuals with ADHD and mental retardation require special monitoring in order to discern those symptoms that can be lessened by medication and those that cannot. Moreover, there is a need to focus attention on discerning whether current treatments primarily influence cognitive (i.e., attention, concentration) or behavioral outcomes (i.e., irritability, hyperactivity, conduct disorders).

Concerns about the possible overdiagnosis of ADHD and associated overprescription of stimulant medications, concerns about the drugs' long-term efficacy, and the lack of ability to enhance academic achievement have moved attention to alternative treatment regimes, including nutritional interventions (Schnoll, Burshteyn, & Cea-Aravena, 2003). This work parallels research that

has studied more generally the link between micro- and macronutrient intake and mental health outcomes, including low average IQ and mental retardation (Benton, 2001). A brief summary of this work is described in the next section, followed by a discussion of the possible role of highly unsaturated fatty acids (HUFAs), together with a brief description of research that has tried to explain the mechanism by which HUFAs impact on brain development. The chapter continues with a review of research that has linked various cognitive and behavioral outcomes, including ADHD, to HUFA intake and discusses a possible role for HUFA supplementation as a therapy for ADHD-related behaviors specifically, and for developmental disorders more generally.

II. NUTRITION, DIET, AND NEUROLOGICAL FUNCTIONING

Adequate nutrition is required for healthy brain development and functioning (Black, 2003; Bryan et al., 2004; Dutta-Roy, 1997; Tacconi, Federico, & Mario, 1997; Wachs, 2000). Growing evidence suggests that not only can nutritional deficits impact on central nervous system (CNS) functioning and thereby on learning and behavior, but that, to varying degrees, supplementation may be able to reverse problems caused by inadequate nutrition.

Much of the early interest in nutrition and developmental disorders focused on the role of food additives in developmentally aberrant behaviors such as hyperactivity. Although there was much initial enthusiasm around Feingold's Kaiser-Permanente diet (Feingold, 1975) for moderating hyperactivity in children, evidence was largely anecdotal with only two comprehensive studies undertaken. The Feingold diet was based on the elimination of all artificial colors and flavors as well as all foods containing salicylates. Data from the major empirical studies and a number of challenge studies indicated that the specific elimination of food colors from the diet did not appear to be a major factor in the reduction of hyperactive behavior (Silver, 1986), although there was some evidence to suggest that preschool children responded more favorably to the diet (Lipton & Mayo, 1983). However, as Schnoll et al. (2003) pointed out there are serious methodological limitations to conducting effective large-scale experiments to test the Feingold diet due to the complex dietary requirements, for example, elimination of 3000 additives from the diet. Furthermore, they suggest that a subset of children did appear to respond favorably to the diets with a large reduction in hyperactive behavior. It is possible that children react differently to different foods and food additives, for example, some may react to wheat, others to specific food additives, food colorings, or salicylates. These

individual differences would be very difficult to test effectively with large experimental groups.

Other foods that have attracted attention for their possible role in hyperactivity include refined sugars. In summarizing the results of several studies, Schnoll et al. (2003) concluded that consumption of refined sugar produces hyperactive behavior, although they also highlighted the significant design flaws in most of the research. These have extended from small sample sizes, the dosage levels of sucrose used, and the way in which participants were selected.

Attention has shifted to a possible role for micronutrient supplementation in the alleviation of some of the behaviors associated with suboptimal intellectual performance. Benton (2001) reviewed the effects of supplementation with a range of micronutrients on the intelligence of children, focusing his attention on double-blind placebo-controlled studies. He concluded that in 10 of the 13 studies included in the review, a benefit from supplementation had been demonstrated with nonverbal measures of cognition in at least a subsection of the experimental sample. He went on to point out that "... not all children respond to supplementation, rather there is a minority who benefit, whose diet offers low amounts of micronutrients" (p. 297). It is important to note that the Benton (2001) review indicates that it is unclear which micronutrients improved cognition because most studies involved a cocktail of ingredients.

Among the essential nutrients believed to be required for optimal neurological functioning, HUFA [also known as long-chain polyunsaturated fatty acids (LCPUFA)] from the omega-3 and omega-6 groups of polyunsaturated fatty acids (PUFA) have received increasing attention from researchers interested in a range of mental health conditions, including schizophrenia (Horrobin, 1997, 1998; Peet, 2003), bipolar disorder (Chiu et al., 2003; Stoll et al., 1999), and depression (Adams, Lawson, Sanigorski, & Sinclair, 1996; Hibbeln, 1998; Su, Huang, Chiu, & Shen, 2003; Tanskanen et al., 2001), as well as developmental disorders such as autistic spectrum disorder (Bell, Sargent, Tocher, & Dick, 2000; Bell et al., 2004), dyslexia (Baker, 1985; Richardson, Cox, Sargentoni, & Puri, 1997; Richardson et al., 2000; Stordy, 2000; Taylor et al., 2000; Taylor & Richardson, 2000), and ADHD (Richardson, 2003a).

Specific attention has not been given to mental retardation in lipid research to date. However, mental retardation and other neurodevelopmental disorders are all correlated with low body weight, which may be indicative of nutritional deficiencies (Tacconi et al., 1997). Additionally, developmental disorders that include mild mental retardation have been included in fatty acid research. Moreover, psychopathology in people with mental retardation is common (Bertelli & Cabras, 1997; Johnson, Handen, Lubetsky, & Sacco, 1995; La Malfa, Notarelli, Hardoy, Bertelli, & Cabras, 1997; Moss, 2001;

Zimmerman, Jinnah, & Lockhart, 1998) with comorbidity estimates ranging from 10% to 80% (Edelstein & Glenwick, 1997), suggesting that research identifying treatments that impact on these psychopathologies may have considerable relevance for the treatment of people with mental retardation.

III. WHY ARE LIPIDS IMPORTANT? THE ROLE OF HUFAs IN NEUROLOGICAL FUNCTIONING

Research indicates that diets in Western societies have become increasingly deficient in omega-3 fatty acid intake (Holman, 1998; Leaf, 2001; Ratnesar, Tapsell, Meyer, Calvert, & Storlien, 2000), probably due to the overconsumption of processed foods, saturated fats and *trans*-fatty acids, reduced fish intake, and misguided emphasis on diets with "no fat," all resulting in a depletion of omega-3 fatty acids. This has impacted adversely on the metabolism of omega-3 fatty acids (Simopoulos, 2002b) and resulted in increased omega-6 (n-6) to omega-3 (n-3) fatty acid ratios (Simopoulos, 2002a), thereby altering the fatty acid composition of neural membranes (Tiemeier, van Tuijl, Hofman, Kiliaan, & Breteler, 2003). Although the ideal n-6 to n-3 ratio is not clear (Riemersma, 2001), recommended ratios range from 3 to 10:1 (Haglund, Wallin, Wretling, Hultberg, & Saldeen, 1998). In Western diets, however, current ratios are unduly high and have been estimated at 15–16:1 (Simopoulos, 2002a).

The n-6 and n-3 groups of PUFA are termed "essential" fatty acids (EFAs) because they must be regularly provided via limited dietary sources. Humans can obtain LNA, the parent omega-3 fatty acid, through their diet via green leafy vegetables, nuts, and seeds. Long-chain n-3 HUFA (EPA and DHA) are made by phytoplankton and transferred via the food-chain to marine organisms, particularly cold-water fish and marine animals. Therefore, humans can also obtain elongated omega-3 HUFA via algal or fish sources.

Essential fatty acids have 18 carbon atoms with double carbon bonds and are termed PUFA. These chains undergo further processes of elongation (lengthening of fatty acid chain) and desaturation (insertion of double bonds) to form HUFA. The n-6 and n-3 families of polyunsaturated fatty acids and their metabolic pathways can be viewed in (Fig. 1). Unlike saturated fats, which are sluggish and chiefly used for insulation and generating energy, HUFA are chemically active lipids that play vital roles in biological processes. A large body of research has linked EFA deficiencies to physical health problems, such as cardiovascular disease, and they have also been linked to infant brain development, cognitive performance, and mental health.

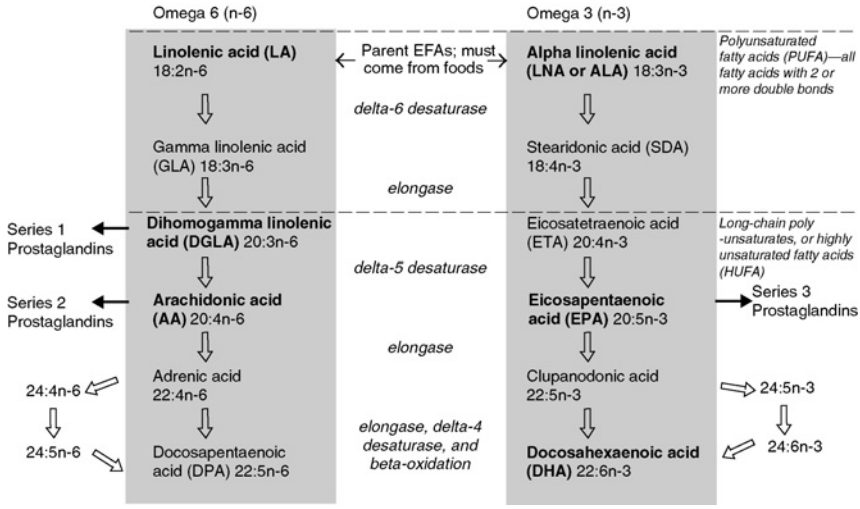

FIG. 1. Metabolic pathways of omega-3 and omega-6 essential fatty acids (EFAs). (Sources: Erasmus (1993), Haag (2003), Ley-Jacobs (2000). Note: Desaturase enzymes insert double bonds; elongase enzymes add two carbons. Fatty acids depicted as number of carbon molecules: number of double bonds; n-3 and n-6 refer to position of first double bond from the omega end in the chain).

Sixty percent of the dry weight of the brain is composed of fats or lipids. Omega-3 HUFA are selectively concentrated in the brain and nervous system, indicative of their role in the structure and function of neural cells (Zimmer et al., 2002). The main PUFA in the brain are the longest chain omega-3 HUFA: docosahexaenoic acid (DHA) and arachidonic acid (AA) from the omega-6 group. Omega-3 HUFA eicosapentaenoic acid (EPA) and omega-6 HUFA dihomogamma linolenic acid (DGLA) also seem to play important roles in brain function (Richardson, 2003a). Importantly, DHA levels in neuronal membranes vary according to dietary oil intake (Hamilton, Greiner, Salem, & Kim, 2000; Tacconi et al., 1997; Yehuda, Rabinovitz, Carasso, & Mostofsky, 1998; Youdim, Martin, & Joseph, 2000) and there is evidence that low HUFA and high cholesterol levels reduce neural membrane (phospholipid) fluidity (Yehuda, Rabinovitz, Carasso, & Mostofsky, 2002; Yehuda et al., 1998). Animal studies and human infant autopsies have also shown that when insufficient omega-3s are available during early neural development, there is a decrease in the DHA content of the brain.

Research in the last decade has found a link between HUFA and CNS functioning (Leaf & Kang, 2001), pointing to their role in healthy brain

functioning (Bell et al., 2000; Richardson, 2003a; Richardson & Puri, 2002), neural cell signaling (Haag, 2003; Hamilton et al., 2000; Horrobin, 1998; Richardson et al., 2000; Yehuda et al., 1998, 2002; Youdim et al., 2000), and neurotransmitter processes (Chalon, Vancassel, Zimmer, Guilloteau, & Durand, 2001; Youdim et al., 2000). Neurotransmitters are believed to have a profound influence on mental health, and importantly, Lauritzen, Hansen, Jørgensen, and Michaelsen (2001) reported that the transmission of neurotransmitters, such as dopamine and serotonin, has been found to vary in relation to n-3 content of neural membranes.

Increasing awareness of the role of lipids in the brain has stimulated research investigating the impact of EFA deficiencies on learning and behavior, although this area is in great need of further attention by behavioral scientists (Mostofsky, 2001). Animal studies have found abnormalities in dopaminergic systems consistent with behavioral abnormalities associated with n-3 deficiency. Dopamine pathways are associated with the frontal lobes, and are believed to influence motivational behavior, emotional functions, and cognitive functions such as working memory (Chalon et al., 2001; Zimmer et al., 2002). It is interesting to note that stimulant drug treatments for ADHD are hypothesized to function by increasing dopamine levels. A number of animal studies have demonstrated a link between n-3 HUFA and cognitive functioning (Gamoh et al., 1999; Kaplan & Greenwood, 1998; Lim & Suzuki, 2000), and studies on animals with reduced maternally-derived DHA have revealed impaired performance on various neurological tasks (Salem, Litman, Kim, & Gawrisch, 2001). Following a review of controlled animal studies, Lauritzen et al. (2001) concluded that there is evidence for the role of omega-3 HUFA in improving learning and behavior. Human studies have provided additional indications of their role in behavioral and cognitive outcomes. In the following section we turn to consideration of the effects of omega-3 deficiency on human infant development.

IV. HUFA AND INFANT DEVELOPMENT

HUFAs are necessary for intellectual growth and development in the developing neonatal brain and in early childhood (Broadhurst et al., 2002). During the first year of life, the brain uses 60% of the infant's total energy intake for neural membrane development (Tacconi et al., 1997). The third trimester of pregnancy and early postnatal period are also periods when AA and DHA rapidly increase in the infant brain (Wainwright, 2000), and DHA is especially important in synaptogenesis, or synaptic formation, during brain development (Haag, 2003). However, fetal and infant n-3 HUFA levels are influenced by maternal dietary fish oil intake (Dunstan et al., 2004; Gibson,

Neumann, & Makrides, 1997; Uauy et al., 1996), rendering the developing brain vulnerable to HUFA deficiencies.

A number of longitudinal studies comparing breast-fed to bottle-fed infants have found significant cognitive benefits from breastfeeding at various ages (in one study up to 15 years of age) after controlling for social and demographic variables (Uauy et al., 1996). A rigorous meta-analysis by Anderson, Johnstone, and Remley (1999) of 20 clinical studies looking at cognitive outcomes of breast-fed versus formula-fed infants showed a high, consistently significant difference between breast-fed and formula-fed infants, with an average IQ difference of 3.2 points after adjusting for possible confounding variables such as SES status and parental education. This difference was maintained throughout childhood and adolescence, and breast-fed children were less likely to develop emotional and behavioral problems. Low-weight infants showed a greater cognitive advantage from breastfeeding (5.18 points) than normal weight infants (2.66 points), supporting conclusions drawn in individual studies. These differences are attributed to the omega-3 content of breast-milk because infant formulas do not contain DHA or AA, the most prevalent HUFA in the brain. In further support of this supposition, it has been found that blood DHA concentrations are higher in infants who were breast-fed compared to those given formulas (Anderson et al., 1999; Gibson, Neumann, & Makrides, 1996).

Intervention studies with infants have had mixed results. Of six randomized studies with term infants, three found no significant effect on the Bayley Scales, a global test of infant development, whereas three other studies found effects with global tests and specific performance tasks (Lauritzen et al., 2001). In another study with term breast-fed infants, the mothers were given varying amounts of DHA supplementation (Gibson et al., 1997). The results were significantly correlated with DHA milk and infant plasma levels at 3 months of age but no effect was found between the groups on the Bayley Motor Development Index (MDI) at 12 months of age. It was concluded that there were no effects of infant DHA status on long-term neurodevelopment. However, all these infants were breast-fed and from middle class families. Therefore, this study shows no comparison with formula-fed infants from lower SES backgrounds who might be more likely to suffer from HUFA deficiencies. A review of randomized, controlled experiments found beneficial neural outcomes for infants with breast-milk or DHA-supplemented formulas (Salem et al., 2001). In these studies the quantities of DHA were relatively small, comprising 0.1–0.35% of total fatty acids, suggesting that even small quantities of DHA can have an important impact on neurological functioning.

Lauritzen et al. (2001) have warned that studies comparing breast-milk with formula should be interpreted cautiously because there can be many other

differences between breast-milk and formula apart from HUFA content, including immune factors, leukocytes, growth factors, and hormones. In addition to the nutritional content of breast-milk, in these nonrandomized studies other maternal variables associated with the choice to breastfeed might influence outcomes also (Birch, Garfield, Hoffman, Uauy, & Birch, 2000).

Thus, controlled experimental studies comparing the effect of formula with or without HUFA supplementation on cognitive outcomes have been undertaken. A randomized clinical trial with 56 infants found significant motor and cognitive advantages, including an average seven-point increase on the MDI of the Bayley's Scales of Infant Development, in two groups with formula supplemented with DHA or DHA+AA compared to a control group with DHA-free formula (Birch et al., 2000). However, Willats and Forsyth (2000) reviewed this and other controlled trials with formula supplementation and found that they have produced inconsistent results. They concluded that tests assessing specific cognitive outcomes were possibly more sensitive to changes in HUFA status than global measures of development. Studies using visual information processing tests (not related to visual acuity but positively correlated with childhood IQ) showed consistently higher scores in HUFA supplemented infants. Problem solving ability, also positively correlated with childhood IQ, was higher in infants given supplemented formula. The ability to solve problems involves processes that have been linked to prefrontal cortex activity, suggesting that HUFA assists in development of the prefrontal cortex. It was concluded again that low-weight infants demonstrated greater cognitive benefit from HUFA supplementation (Willats & Forsyth, 2000). Importantly, as pointed out earlier, these infants are at greater risk of suffering from mental retardation.

In extreme cases, some infants are believed to have inherited defects in PUFA metabolism that may contribute to the profound retardation observed in peroxisomal disorders (Uauy et al., 1996). These disorders are associated with abnormally high levels of long-chain *saturated* fatty acids (Moser & Moser, 1996), which could impact on CNS functioning via brain inflammation, decreased myelination of nerve axons, and reduced membrane fluidity. In children with Batten's disease, which is associated with mental retardation and severe motor impairment, HUFA levels were reported as lower than in controls, and improvements recorded in five patients following supplementation (Bennet, Gayton, Rittey, & Hosking, 1994, cited in Martinez, 1996). Zellweger syndrome, the most severe of these disorders with profound mental retardation and persistent seizures, is associated with a drastic deficiency of DHA in the brain 20–30% below normal levels (Martinez, 1996). DHA therapy, combined with AA supplementation, was given to six patients with Zellweger syndrome. The infant with the worst,

classical case of Zellweger syndrome was very ill when the treatment commenced and was only treated for 3 months before he died of acute bronchopneumonia. Clinical neurological and visual improvements were observed in the other five patients—four infants and one 5-year-old child—and were associated with normalization of DHA levels (Martinez, 1996). Moser and Moser (1996) reported that clinical results with DHA intervention in peroxisomal disorders so far have been more positive with patients who have not manifested neurological symptoms at the time of therapy. They suggested that this could be due to the difficulty in reversing neurological impairment once it has already occurred. Alternatively, autopsies revealed that it could have been due to a failure to reduce long-chain saturated fats in the brain (Moser & Moser, 1996). PUFA metabolism has also been reviewed in phenylketonuria (PKU), with suggestions that this might be a major causal factor in the microcephaly and mental retardation that accompanies PKU and that supplementation of AA and DHA for mothers and PKU patients might have therapeutic benefits (Infante & Huszagh, 2001). These authors have suggested that defective synthesis of AA and DHA is implicated in several neurodevelopmental disorders including mental retardation.

V. HUFA AND COGNITIVE FUNCTIONING

There is evidence that a range of macro- and micronutrients can influence children's cognitive development (Black, 2003; Bryan et al., 2004). Among these nutrients, omega-3 deficiency has been linked to reduced cognitive functioning (Broadhurst et al., 2002).

A number of animal studies have demonstrated the effect of HUFA deficiency on cognitive performance in rats. In one study, test performance was superior on a spatial memory test by rats on a soybean oil diet (containing n-3) compared to rats on a lard diet (negligible n-3; high in saturated fat). These effects could not be explained by other physiological variables such as basal body temperature variations, energy consumption, body weight, or a specific enzyme related to brain activity (Kaplan & Greenwood, 1998). Further studies reported by Kaplan and Greenwood examined this effect on specific aspects of learning and memory that rely on different regions of the brain. These studies found a difference on three different cognitive tasks. The animals on a lard diet showed deficits in spatial memory, short- and long-term temporal memory, and in learning of various maze problems. It was concluded that the effects of EFA deficiency are generalized across different brain regions, rather than being specific to certain areas of the brain. However, it was suggested that impairments in cognitive performance could also be attributed to high saturated fatty acid (SaFA) intake and that

the functions of EFAs may be influenced by overall FA dietary composition, ratios of FAs, oxidation of FAs, or other mediating factors, such as the influence of SaFAs or SaFA:HUFA ratios on insulin levels, considering that variations in blood sugar level are known to affect cognitive performance (Kaplan & Greenwood, 1998).

One animal study was conducted to test learning ability with rats that were fed fish oil deficient diets over three generations (Gamoh et al., 1999). Half of these rats were given DHA supplementation over 10 weeks, and the two groups were tested on reference memory and working memory. Reference memory refers to information that is retained for later use, while working memory relates to information that is only retained for short-term use, and they both involve different brain regions. The DHA supplemented group showed significantly less reference memory errors than the control group, although working memory results were inconclusive. It is interesting to note that working memory is an executive function that involves the frontal lobes in the brain. The supplemented group had a significant increase in DHA and AA concentrations in the hippocampus and cerebral cortex regions of the brain, which are both important for memory formation and therefore learning. This increase was not found in the cerebellum and brain stem, and DHA uptake into the brain therefore appeared to be selective.

In another study, DHA intake improved maze-learning performance in young and old mice compared to control groups (Lim & Suzuki, 2000). However, old mice performed less well, and also had lower brain DHA levels. Improvement of maze-learning ability following DHA supplementation was replicated in a later controlled trial, and the effects were measured over various time periods (Lim & Suzuki, 2001). It was observed that learning only improved 2 weeks after DHA levels increased in mice brains, indicating that DHA needs time to be incorporated into neural membranes before learning outcomes are influenced.

Therefore, there is evidence that omega-3 HUFA deficiency can impact on neural development and cognitive outcomes, such as memory and learning in animals, and on information processing and IQ in infants. Furthermore, it appears that omega-3 supplementation may be able to replenish omega-3 levels in the brain and reverse unfavorable cognitive outcomes. There are some inconsistencies in this research although, underscoring the need for further investigation. A critical review highlighted possible interpretative flaws in animal and infant research examining the effects of omega-3 fatty acids on learning and cognition (Carlson & Neuringer, 1999). Further research is called for to clarify these interpretations and to elucidate the mechanisms by which HUFA influence neural membranes. It is also important that future research attempt to distinguish the various different aspects of cognition (e.g., speed of information processing, working memory,

attention) and behavior (e.g., impulsivity, distractibility) associated with omega-3 deficiency and that these results be linked backed to possible neural mechanisms. Methodological issues, which might be responsible for some of the inconsistent results reported in lipid research, are considered later in this chapter.

To summarize, to this point the literature reviewed has highlighted the possible impact of HUFA on normal development by analysis of animal and infant research, by exploration of links between nutrition and cognition in children and by reference to a range of possible biological mechanisms through which HUFA may impact on cognitive functioning. In the following section, attention is turned to the role of HUFA in abnormal development, with a focus on the symptoms and behaviors implicated in, or existing comorbidly with, ADHD and/or mental retardation. Summaries suggest that there is growing evidence pointing to a biological basis for neurological disorders (Horrobin, 1998; Richardson & Ross, 2000), with many of these focusing on a critical role for neurotransmitters. The roles that HUFA are believed to play in the brain provide a theoretical connection to the current neurotransmitter theories for various mental illnesses, for example, the role of serotonin in depression (Maes & Smith, 1998) and dopamine in schizophrenia (Horrobin, 1997) and ADHD (Biederman, 1997). Accordingly, HUFA deficiencies have been reported in people with a range of psychiatric problems (Chalon et al., 2001; Pomerantz, 2001). In addition, a possible role for HUFA in conduct, behavioral and psychiatric disorders, all of which have a higher prevalence in individuals with ADHD and mental retardation, as well as cognitive difficulties including attention and problem solving, has been highlighted. This work is summarized later.

VI. HUFA AND ADHD SYMPTOMS

Increasing attention has been given to the role of essential fatty acids in childhood developmental disorders. Given that brain development continues throughout childhood (Thatcher, 1991), HUFA supplementation may have a beneficial effect on developmental milestones, particularly in children with developmental disorders that might be related to problems with HUFA metabolism and/or deficiency.

As discussed in an earlier section, ADHD is one of the most common disorders affecting school-aged children, and has a high comorbidity with a number of other developmental disorders including mental retardation. There is substantial evidence for a neurological basis for ADHD (Chabot, di Michele, Prichep, & John, 2001; Lubar, Swartwood, Swartwood, & Timmermann, 1995; Mann, Lubar, Zimmerman, Miler, & Muenchen, 1992;

Riccio, Hynd, Cohen, & Gonzalez, 1993). Research over the past 50 years or so has identified slower neocortical brain wave activity in children and adolescents with ADHD, particularly in the frontal and temporal lobes. This is consistent with noradrenergic and dopaminergic hypotheses, relating to neurotransmitter abnormalities that may contribute to symptoms of ADHD. ADHD has also been associated with micronutrient deficiencies (Bilici et al., 2004; Konofal, Lecendreux, Arnulf, & Mouren, 2004; Schnoll et al., 2003; Toren et al., 1996) food additives and food allergies/sensitivities (Schnoll et al., 2003), and fatty acid deficiencies.

A link between fatty acids and ADHD-related symptoms was proposed just over 20 years ago by Colquhoun and Bunday (1981), working with hyperactive children. They gathered evidence that implicated EFA deficiency as an underlying factor, including symptoms such as excessive thirst, eczema, and allergies, as well as zinc deficiencies.[1] They then conducted a small exploratory trial giving 25 children 2–3 g of evening primrose oil (containing n-6 DGLA precursors LA and GLA) per day, and reported impressive anecdotal improvements in symptoms for about half of the sufferers. Similar results were found in a placebo-controlled intervention trial using omega-6 PUFA with 18 boys suffering from ADHD (Arnold et al., 1989). Significant effects were only observed on teachers' ratings of the children on Conners' hyperactivity index after supplementation with evening primrose oil. In another group of 23 maladjusted children, DGLA levels were also found to be lower than in controls, as well as slightly lower levels of LA and AA (Mitchell, Lewis, & Cutler, 1983). It is not clear whether this study measured n-3 HUFA levels, although DPA (an n-6 HUFA) levels were higher in the maladjusted children, which has since been found to be indicative of DHA deficiency in the brain (Chalon et al., 2001; Hamilton et al., 2000; Lauritzen et al., 2001; Moriguchi, Loewke, Garrison, Catalan, & Salem, 2001; Wainwright et al., 1999; Youdim et al., 2000).

In 1986, researchers compared hyperactive children with controls matched on age and sex (Mitchell, Aman, Turbott, & Manku, 1987). They reported that hyperactive children displayed symptoms of fatty acid deficiency

[1]Zinc deficiency is commonly associated with ADHD (Bekaroglu et al., 1996; Toren et al., 1996), possibly due to its roles in enzyme activities required for PUFA metabolism and serotonin synthesis. Zinc deficiency was also found in hyperactive mentally retarded children (Bekaroglu et al., 1996). Zinc supplementation in a double-blind, placebo-controlled trial alleviated symptoms of impulsivity, hyperactivity, and poor socialisation, but not attention deficits (Bilici et al., 2004). Low free fatty acid levels were correlated with low zinc levels in the zinc group, indicative of biological interactions between zinc and PUFA. Bekaroglu et al. (1996) also found that free fatty acid levels were lower in children with ADHD, and these were significantly correlated with the low zinc levels.

(FADS) such as excessive thirst and urination, as well as symptoms of eczema, asthma, and other allergies. They also had more auditory, visual, language, reading, and learning problems than controls. These children were assessed by parents on the Revised Behavior Problem Checklist (RBPC) and by teachers on Conners' Teacher Questionnaire. Conners' subscales include hyperactivity, impulsive-hyperactive, psychosomatic, attention/learning, conduct, and anxiety scales. Investigation of fatty acid profiles revealed that the 44 hyperactive children tested had significantly lower plasma levels of DGLA, AA, and DHA compared to 45 controls. The hyperactive children also had significantly lower birth weights, which according to the authors supports the influence of prenatal factors on hyperactivity. Arnold (1994) investigated EFA metabolism in children with ADHD, and found that behavior problems were correlated with lower GLA levels but not with LA, its metabolic precursor. This is suggestive of ineffective metabolism. Although this study investigated omega-6 fatty acids, the same delta-6 desaturase enzyme that is involved in n-6 metabolism is required for metabolism of n-3 HUFA.

Stevens et al. (1995) measured fatty acid levels in 53 boys with ADHD and 43 controls aged 6–12 years, and compared EFA deficiency symptoms between the groups. Participants were allocated to the ADHD or control group according to their ratings on the Conners' Parent and Teacher Rating Scales. About half the boys in the ADHD group were taking stimulant medication, although there were no significant differences between medicated and unmedicated subjects with ADHD on variables measured, apart from "problems getting to sleep." Those in the ADHD group had symptoms of EFA deficiency such as thirst, frequent urination, dry hair, and dry skin. Total FADS scores were greater in the ADHD group than in the control group (39.6 and 9.3, respectively, $p < .001$). There were no significant differences between the groups on age, height, weight, body mass index, or SES status. Average polar fatty acid blood concentrations were significantly lower in the ADHD group, specifically AA, EPA, and DHA. DPA levels were again significantly higher in the red blood cells of the ADHD group. Unlike the study by Mitchell et al. (1987), there were no significant differences in DGLA levels. Average omega-3 levels were lower in the ADHD group. However, mean omega-6 concentrations did not differ between the groups, and therefore the ratio of total n-6 to n-3 fatty acids was significantly higher in the ADHD group. Further investigation revealed that a subgroup of children with higher rates of FA deficiency symptoms (40% of the ADHD sample) had significantly lower proportions of plasma AA and DHA than the other children in the ADHD group, whose plasma fatty acid proportions were not significantly different from those in the control group. Additionally, 81% of the control subjects had been breast-fed compared to 45% of ADHD

subjects, and the control subjects had also been breast-fed significantly longer (Stevens et al., 1995). Given that breast-milk has higher EFA content than most infant formulas, as outlined earlier, this is a possible contributing factor to EFA deficiency in these children.

These researchers then ranked the same 100 boys according to omega-3 status rather than behavioral symptoms (Stevens, Zentall, Abate, Kuczek, & Burgess, 1996). They also compared children on the Conners' Parent and Teacher Rating Scales and teacher evaluations of students' learning ability. EFA deficiency symptoms displayed by the children with lower omega-3 levels were frequent thirst and urination, and dry skin. Children with lower omega-3 levels had significantly more learning problems. They also had more frequent behavior problems, specifically hyperactivity, impulsivity, conduct, anxiety, temper tantrums, and sleep problems, that is, problems going to sleep and getting up in the morning.

Therefore, children with symptoms of ADHD who also have FADS might derive greater benefits from HUFA supplementation. Also, taken together, these studies implicate DGLA as a contributing factor as well as the two fatty acids now believed to be critical for neural functioning, AA and DHA. These three HUFA are reported to play important roles in various biological functions, including neurotransmitter uptake and synaptic transmission, which are necessary for healthy brain development and performance (Richardson, 2003a). Although DGLA supplementation was previously reported to alleviate some symptoms of ADHD, in the study by Stevens et al. (1995) plasma levels of DGLA did not differ significantly between ADHD children and controls. A study in Taiwan found significantly lower levels of LA, AA, and DHA in the red blood cells of children with ADHD compared with controls. There was no difference in total n-6 fatty acids between the groups but the ADHD children had significantly lower total n-3 fatty acids and higher n-6 to n-3 ratios (Chen, Hsu, Hsu, Hwang, & Yang, 2004). Burgess, Stevens, Zhang, and Peck (2000) have noted that n-3 fatty acid plasma proportions in their study were inversely related to parent and teacher ratings of behavior, whereas low plasma proportions of both n-3 and n-6 were related to a higher frequency of FADS. Therefore, it is not clear why previous studies reported an effect with evening primrose oil, as supplementation with n-6 alone would not be expected to improve behavioral outcomes. It is interesting that the physical symptoms of EFA deficiency, which are attributed to n-6 deficiency, are correlated with behavioral symptoms of n-3 deficiency. Whether the relationship between maladaptive behavioral outcomes and FADS is merely an artifact of n-6 deficiency in the presence of combined n-3 and n-6 deficiency remains to be determined.

Four randomized, double-blind, placebo-controlled studies have been completed with children who displayed ADHD-related symptoms. In the

first study, supplementation with 345 mg of DHA per day over 4 months in 6- to 12-year-old hyperactive children did not significantly improve objective or subjective measurements of ADHD symptoms, although there was a significant increase in DHA blood plasma levels in the DHA group (Voigt et al., 2001). In this study, stimulant medication was ceased for 24 hours before laboratory measures were administered. However, parents completed the subjective scales (Child Behavior Checklist and Conners' Rating Scales) while the children were receiving stimulant medication. This is likely to have influenced the results. Furthermore, children who had experienced ineffective treatment with stimulant medication or had previous diagnosis of mood, anxiety, thought, or bipolar disorders were excluded from the study. It is presumed that the rationale for this was to select a group of children with "pure" ADHD symptoms. As Richardson (2003b) points out, this may not be entirely practical due to the high comorbidity prevalent in ADHD, and as discussed later, the heterogeneity of ADHD and its overlap with other neurodevelopmental disorders is an issue that needs to be explored within a HUFA framework. Hirayama, Hamazaki, and Terasawa (2004) tested the effect of fish oil-enriched bread (supplying 3600 mg DHA and 700 g EPA per week) on symptoms of ADHD in a 2 month placebo-controlled, double-blind trial with 40 children aged 6–12 who were mostly drug free (34/40). The placebo bread contained olive oil. Again, there were no significant treatment effects, although paradoxically the control group had significant improvements on visual short-term memory and errors of commission, possibly due to practice effects. These improvements were not seen in the treatment group. Blood samples and FADS were not measured so it is not clear whether this sample had a deficiency in fatty acids. Given that the study was conducted in Japan, known to have high fish consumption, it is possible that they did not.

The other two intervention trials found improvements in symptoms with a combination of n-3 and n-6 HUFA. In the first study, 41 British children aged 8–12 years with ADHD-related symptoms and specific learning disabilities (dyslexia) were given a HUFA supplement or olive oil placebo over 12 weeks (Richardson & Puri, 2002). The HUFA supplement in this study provided a daily dose of 186 mg EPA, 480 mg DHA, 864 mg LA, and 42 mg AA, as well as α-tocopherol (vitamin E). These children all had above average scores on parent rating scales of ADHD symptoms and average general ability yet low reading achievement. Baseline measures on learning and behavior did not differ, but the HUFA group showed improvements on British Ability Scale (BAS) scores and ratings on the Conners' Parent Rating Scale following the 12 weeks supplementation, with medium effect sizes reported.

Similarly, the second study investigated the effect of PUFA supplementation, providing 80 mg EPA, 480 mg DHA, 40 mg AA, 96 g GLA, and 24 mg

α-tocopherol daily, or olive oil placebo over 4 months on American children with ADHD-like symptoms as well as thirst and skin problems indicative of FADS (Stevens et al., 2003). Plasma proportions of DHA, EPA, and α-tocopherol increased twofold in the intervention group and there was a significant drop in n-6 to n-3 ratios. The olive oil group had a small increase in ALA. For FADS, parents of children in both groups reported a large decrease in thirst, skin, and dry hair but significant HUFA treatment effects were only found for frequent urination scores. Parents of children from both groups reported significant improvements in hyperactivity, attention, and oppositional defiant behavior. However, significant treatment effects were found for only 2 out of 16 outcome measures: parent ratings of conduct and teacher ratings of attention. Oppositional defiant behavior scores significantly improved from clinical to nonclinical levels, and significant relationships were reported between the change in red blood cell HUFA and the magnitude of outcome measures.There were several methodological issues in this study, including the effect of olive oil on n-3 plasma levels in the placebo group and small sample size (total $N = 50$ with 17 dropouts mainly due to noncompliance or relocation) that probably did not provide enough power. There was also a confusing report of HUFA levels at baseline that contradicted previous findings: plasma levels of AA and DHA were lower in the ADHD group than in a healthy reference sample of children, yet red blood cell levels of AA and DHA were higher in the intervention sample. The implications of this finding are unclear, although the investigators suggested that EFA proportions in phospholipids (cellular membranes) might be more important than individual quantities.

Therefore, studies on HUFA and ADHD using only evening primrose oil or DHA have had mixed results, whereas there are indications from the two latter studies that a combination of n-3 and n-6 fatty acids is more effective. Richardson and Puri suggest that, as has been indicated with fatty acid trial results on symptoms of schizophrenia, EPA might be the important fatty acid for ADHD. EPA appears to have additional functions, other than its role as DHA precursor, which support DHA activity in cell membranes. Another difference in Richardson and Puri's (2002) study compared to Voigt's (2001) DHA study was that none of the children were receiving stimulant medication. Furthermore, participants had symptoms of ADHD in combination with dyslexia. Dyslexia has previously been associated with FADS (Baker, 1985; Richardson et al., 1997, 2000; Stordy, 2000; Taylor et al., 2000; Taylor & Richardson, 2000), which may help to explain the etiology for this subgroup of ADHD children that comprises 25–40% of children with ADHD (Richardson & Puri, 2002).

Clinical trials looking at the effect of supplementation with fatty acids in dyslexia (specifically on reading ability) and dyspraxia have been conducted

with children. These are as yet unpublished, but preliminary results are positive and have been reported in a review of intervention trials in fatty acid treatment of ADHD, dyslexia, dyspraxia, and the autistic spectrum by Richardson (2003b). The role of fatty acids in autistic spectrum disorder has been addressed by Bell et al. (2000, 2004). In the recent study, total n-3 and EPA were found to be significantly lower and AA/EPA ratios significantly higher in the red blood cells of patients with regressive autism and classical autism/Asperger's syndrome (ASP) than in controls. The clinical groups also had higher levels of RBC saturated fat and DPA. Regressive autism patients had lower levels of AA. DHA was slightly, but not significantly lower in both clinical groups. Further blood analysis revealed abnormal phospholipid metabolism and possible oxidation in the autistic groups. They also had significantly higher parent-reported FADS than the control group. Therefore, there is evidence that n-3 HUFA deficiencies might be implicated in autism, and intervention trials are warranted to determine the effect of HUFA supplementation on symptoms associated with autism and ASP.

Richardson and Puri (2000) reviewed research on the role of fatty acids in the brain with regard to ADHD. Importantly, they pointed out that the functions of fatty acids in neural membranes and synapses complement existing models of neurotransmitter actions in ADHD. Symptoms of ADHD are particularly related to known symptoms of dopamine malfunction such as problems with attention, motivation and reactivity to stimuli, and rewards. This is related to animal studies that have linked n-3 deficiency to impaired dopaminergic systems, particularly in the frontal cortex of the brain. Further indications that are difficult to explain with a neurotransmitter model alone include the higher prevalence of ADHD and other neurological disorders such as schizophrenia, dyslexia, and dyspraxia in males. Mental retardation also occurs more frequently in males (Castellví-Bel & Milà, 2001; Chechlacz & Gleeson, 2003). This is consistent with observations that males are more susceptible to fatty acid deficiencies (Richardson et al., 2000). Therefore, it has been hypothesized that the overlap between neurologically based disorders may be explained by phospholipid abnormalities (Bell et al., 2000; Horrobin, 1998; Richardson & Ross, 2000). This is further suggested by the finding that children with ADHD tend to have symptoms indicative of FADS such as excessive thirst, urination, dry skin and hair, soft brittle nails, sleep problems, allergies, and somatic complaints (Richardson & Puri, 2000; Stevens et al., 2003; Taylor et al., 2000). Symptoms that are consistent with EFA deficiency have also been observed in individuals with schizophrenia (Horrobin, 1998), dyslexia (Baker, 1985), and autism and Asperger's syndrome.

Considering the widespread prevalence of omega-3 deficiencies, it is possible that maternal and dietary deficiencies interact with an underlying

biological problem with EFA metabolism. Research to date suggests that genetic abnormalities in phospholipid metabolism may be exacerbated by environmental influences such as diet, stress, or pollution to create neurological deficits (Ward, 2000). This research could help to elucidate an important biological component of psychological functioning that is not widely addressed in the treatment of psychological disorders. Additionally, although the DSM-IV provides categorical criteria for diagnosis of mental illness, research has showed a correlation between FADS and learning and behavioral problems not only in boys with ADHD but also in controls. This is similar to other studies that have found linear relationships between the degree of HUFA deficiency and the severity of mental health outcomes (Adams et al., 1996; Edwards, Peet, Shay, & Horrobin, 1998), and suggests that these symptoms fall on a continuum (Richardson & Ross, 2000). Therefore, it may be more prudent to measure learning and behavior and HUFA levels dimensionally rather than categorically, which would enable children who may not fit into a specific category to be treated for problems with learning and behavior nonetheless. This biochemical approach to behavior problems has the advantage of treating symptoms without being encumbered by the issues and limitations inherent in diagnosis (Baker, 1985).

Problems with conduct disorders, including aggression, are commonly reported in ADHD, as they are among some people with mental retardation. Bihm, Poindexter, and Warren (1998) state that aggression in persons with mental retardation, with prevalence estimates in this group ranging from 8.9% to 23.4% or even higher, is a significant problem with regard to self-injury, treatment programs, safety of care workers, damage to property, and violent crime. Low concentrations of cerebrospinal fluid 5-hydroxyindoleacetic acid (CSF 5-HIAA) have consistently been associated with impulsive, violent, suicidal, hostile and aggressive behaviors, as well as early onset of alcohol dependence and personality disorders; and CSF 5-HIAA is linked to serotonin turnover in the frontal cortex in the brain (Hallahan & Garland, 2004; Hibbeln et al., 1998). As outlined previously, HUFA have also been associated with deficient serotonin neurotransmission and it is possible that CSF 5-HIAA mediates this interaction. A small number of studies have investigated plasma phospholipids in people with aggressive and related behaviors. An earlier study compared plasma phospholipid fatty acids between violent, impulsive offenders with intermittent explosive disorder; violent, impulsive offenders with antisocial personality disorder; and a control group (total $N = 50$) (Virkkunen, Horrobin, Jenkins, & Manku, 1987). Both offender groups had significantly higher levels of omega-6 fatty acids DGLA, adrenic acid, and DPA which as noted previously may be indicative of DHA deficiency in the brain. As might be expected therefore, the offending

groups both had lower plasma DHA levels, although only the antisocial group's levels reached statistical significance. Hibbeln et al. (1998) investigated the relationship between CSF 5-HIAA and DHA in blood plasma in a group of alcohol dependent subjects with a history of violent/impulsive behavior, compared with a control group of nonviolent subjects matched for their severity of alcohol dependence. Surprisingly, a medium negative correlation was found between CSF 5-HIAA and DHA in the violent group only, replicating a previous finding of similar magnitude. It is suggested that violent subjects might have a defect in the incorporation of DHA into neural membranes, resulting in a buildup of DHA in plasma and that supplementation with high doses of DHA may normalize serotonin function among impulsive, violent subjects. As the authors note, however, these relationships need to be further explored and experimentally tested before any conclusions can be drawn. A similar study compared plasma HUFA levels in cocaine addicts with a history of aggression and nonaggressive cocaine addicts on admission to an inpatient substance abuse unit. The aggressive group had significantly lower levels of DPA, DHA and total n-3 EFA, and a higher n-3: n-6 ratio. However, nearly 3 weeks after admission, n-3 levels had risen in the aggressive group and the differences became nonsignificant. This was indicative of poor dietary n-3 intake before admission, which could have contributed to their aggression levels (Buydens-Branchey, Branchey, McMakin, & Hibbeln, 2003). A study reported a cross-sectional analysis of fish and PUFA consumption, and its association with hostility in a group of 3581 young adults who participated in a large study investigating risk factors for heart disease (Iribarren et al., 2004). Consistent with the relationship between violence and low n-3 HUFA shown in the other studies, dietary intake of omega-3 rich fish was significantly associated with reduced hostility.

A small number of clinical trials have investigated the impact of HUFA supplementation on aggressive behavior. A double-blind, placebo-controlled study measured the effect of daily supplementation with oil containing 49.3% (1.5 g) DHA, 6.7% EPA, 7.3% oleic acid, 3.3% AA, and other oils for 3 months on cognitive outcomes and aggression in a group of 41 healthy Japanese students from two different universities (Hamazaki et al., 1996). The placebo capsules contained small amounts of LA and LNA. No differences between groups were observed on cognitive and dementia tests, but a significant difference between the groups was reported in aggression toward others (extraggression) at the end of the study, which ended in the middle of a highly stressful final exam period. Extraggression scores significantly increased in the control groups and decreased (although not significantly) in the DHA supplemented groups. Serum blood analyses confirmed a significant increase in EPA and DHA levels in the treatment groups. Thus, 3 months' supplementation with n-3 HUFA seemed to assist students in

controlling aggression toward others on a psychological test during a period of high stress.

Another double-blind study tested the effect of supplementation with 1 g ethyl-EPA daily or placebo over 8 weeks on moderate borderline personality disorder, characterized by emotional reactivity and impulsive aggression, in a group of women (Zanarini & Frankenburg, 2003). The treatment group had a significantly greater decrease in depression and aggression scores at the end of the study. A randomized, placebo-controlled trial was undertaken with 231 young adult prisoners to investigate the effect of supplementation with micronutrients and essential fatty acids over 4–5 months on antisocial behavior (Gesch, Hammond, Hampson, Eves, & Crowder, 2002). These authors cite increasing evidence that supports an effect of poor diet on antisocial behavior. The participants were given multivitamin/mineral capsules and fatty acid supplement providing omega-3 and omega-6 fatty acids (1260 mg LA, 160 mg GLA, 80 mg EPA, and 44 mg DHA daily) or placebos. Over the period of the trial, the treatment group received significantly fewer disciplinary actions, and the greatest effect was noted for violent behavior. Further investigation is clearly warranted into aggression and psychopathology with other disorders such as mental retardation.

VII. IMPLICATIONS FOR MENTAL RETARDATION

To date, scientists engaged in research on fatty acid have not focused specifically on the heterogeneous group of disorders that characterize mental retardation. Many people with mental retardation are likely to display comorbid symptoms including hyperactivity, impulsivity, aggression, anxiety, and DSM-IV diagnosed psychiatric disorders, such as bipolar disorder, schizophrenia (Zimmerman et al., 1998), and ADHD (Miller et al., 2004), and it has been suggested that the high rate of psychiatric disorders in children and adolescents with mental retardation might be a major problem in their functioning and adaptation (Phelps, Brown, & Power, 2002). The impact of HUFAs for the amelioration of learning, cognition, and behavior control problems certainly has implications for the treatment of these difficulties in people with mental retardation who have the same problems. This is particularly important, given that there are indications that some forms of mental retardation might be caused by defects in the structure and functioning of neural synapses (Chechlacz & Gleeson, 2003), which as noted previously, are highly connected with DHA. Case studies with peroxisomal disorders involving severe retardation, which is associated with fatty acid abnormalities in the brain (see infant development earlier in this chapter), have possible implications for the profound mental retardation

that is associated with these disorders. The extensive overlap between mental retardation and psychopathology points to the possibility that there is a common biological denominator underlying these problems. HUFA deficiency, which appears to impact on brain function in a variety of ways, provides a plausible link between them.

Intervention studies with HUFA suggest that infants with low birth weight can gain greater cognitive benefits from supplementation than infants within the normal weight range. This has important implications for developmental disabilities given that a gradient relationship has been found between low birth weight and poor developmental outcomes in childhood such as learning disabilities, low IQ, and mental retardation (Baumeister & Bacharach, 1996; Breslau, Chilcoat, DelDotto, Andreski, & Brown, 1996; Hollomon, Dobbins, & Scott, 1998). Data from a large-scale study reported average IQ differences between low birth weight (LBW) and normal birth weight (NBW) children of up to one standard deviation at 6 years of age, after controlling for maternal IQ and social class (Breslau et al., 1996). Another study of LBW and NBW children at age 6 found significant differences on verbal and performance IQ and on a range of neuropsychological tests, controlling for location, maternal IQ, education, and race (Breslau et al., 1996). There are also indications that maternal nutrient deficiency is related to low birth weight (Ramakrishnan, Manjrekar, Rivera, Gonzáles-Cossío, & Martorell, 1999), and with specific regard to HUFA, given that preterm infants also have low birth weights, intervention studies have found that supplementation with DHA significantly increases duration of gestation as well as birth weight (Smuts et al., 2003). Finally, in an exploratory placebo-controlled double-blind study that provided children with mental retardation with augmented dosages of specific nutrients (not including HUFA), the outcomes provide evidence that IQ and other symptoms of mental retardation might respond to nutritional supplementation (Harrell, Capp, Davis, Peerless, & Ravitz, 1981).

Therefore, notwithstanding the circumstantial nature of many of the arguments presented here, it is hypothesized that at least some forms and/or symptoms of mental retardation including maladaptive behaviors and/or co-occurring ADHD symptoms might respond to supplementation with HUFA, and possibly other nutrients. Children with mental retardation and with comorbid neurological problems are believed to respond differently to psychotropic medication than those without comorbid disturbances, evidenced by the adverse effects experienced by them (Phelps et al., 2002). Therefore, the possibility that psychiatric symptoms and/or symptoms of mental retardation might respond to HUFA supplementation has additional treatment implications for these individuals. Furthermore, if HUFA supplementation could assist in lowering aggressive behavior in some violent or

self-destructive people with mental retardation this could improve the success of treatment outcomes for those individuals. Behavioral scientists would be advised to investigate fatty acid status in people with mental retardation both with and without comorbid behavior and conduct problems, and the effects of nutritional and HUFA deficiencies and supplementation, focusing on neo- and postnatal infant brain development and breastfeeding as well as different phases of childhood and adulthood. This type of research might add more information to other known risk factors (Camp, Broman, Nichols, & Leff, 1998), thereby enhancing prediction and treatment of mental retardation.

VIII. PROBLEMS IN THE HUFA RESEARCH

It is difficult to compare clinical HUFA studies directly due to the different quantities and combinations of fatty acids employed. Additionally, very few if any studies have discussed the quality of the fatty acid supplementation that is being used. HUFA are highly unstable by nature and can be damaged by exposure to light, oxygen and heat; some fish may contain low HUFA, if any; for example, farmed fish tend to be fed with soy protein rather than algae; and fish may contain toxic levels of mercury and other pollutants (Friedland, 2003). Therefore, some inconsistent results in omega-3 research may be attributable to the quality of the oil or fish being used. Hu, Manson, and Willett (2001) have described failure to account for *trans*-isomers of unsaturated fats as a problem in lipid research, along with small sample sizes, poor dietary assessment techniques, and lack of adjustment for total energy and fat intake. Additionally, some studies have used semisynthetic EFA supplementation (Fenton, Dickerson, Boronow, Hibbeln, & Knable, 2001; Wainwright, Jalali, Mutsaers, Bell, & Cvitkovic, 1999), which may have different properties to natural EFAs. Furthermore, the bioavailability of nutrients depends on the presence of other nutrients in the diet (Wachs, 2000), and HUFA research does not generally consider the presence or absence of other nutrients that might impact on HUFA metabolism and brain function.

As well as micronutrient intake, other dietary factors may need to be considered; for instance, it is possible that high saturated fat and sugar along with low HUFA intake can contribute to symptoms of schizophrenia via similar mechanisms (Peet, 2004a,b). There is also evidence that oxidation in the brain can impact adversely on fatty acids and neural membrane structure and function (Tacconi et al., 1997; Youdim et al., 2000). It has even been recommended that vitamin E should always be used in association with HUFA because of its antioxidant properties (Tacconi et al., 1997). Some researchers have followed this recommendation by including an antioxidant

along with HUFA supplementation (Arvindakshan, Ghate, Ranjekar, Evans, & Mahadik, 2003; Richardson & Puri, 2002), with positive results.

Another reason for inconsistent results in psychological research may be attributed to the tests that are being used (Lauritzen et al., 2001). For instance, tests that have been used to assess infant cognitive development include various global measures as well as specific tests that tap into different areas of development. Therefore, the tests may show different results regarding relationships between n-3 and cognitive development, according to the area of the brain that is being targeted and the age of the infant in relation to stages of brain development. Lauritzen et al. (2001) suggested that specific rather than global tests appear to tap into DHA effects on the brain more effectively. Furthermore, it has been observed that tests of "learning ability" do not always ensure that cognitive performance can be distinguished from other factors that influence test performance such as sensory and motor abilities, motivation, and so on (Wainwright et al., 1999). It must also be remembered that many of the studies examining omega-3 and cognitive performance are based on rat models, which do not necessarily apply to humans.

Differences in n-3 intake may also influence research outcomes. For instance, Lauritzen et al. (2001) noted that in infant studies that showed an effect, even although the experimental groups had equivalent DHA intakes these studies had lower LNA in the control group, and therefore a larger difference between the control and the experimental group. Placebos should also be carefully selected. A number of studies have used olive oil, which has been reported to have active properties such as an association with serotonin signaling (Puri & Richardson, 2000). Another variable that needs to be carefully monitored is the possibility that participants, including those in placebo groups, may increase their intake of food containing n-3 fatty acids due to an increased awareness stimulated by involvement in an n-3 trial (see Horrobin, 2003; Peet & Horrobin, 2002).

Hwang et al. (1997) proposed that inconsistencies in n-3 research results may be due to variations in n-3 dosages, as well as the time periods over which they were administered. It can take 3 months of supplementation before the fatty acid composition of neural membranes is restored following long-term deficiency (Richardson, 2003b). The time period after which changes in neural membrane fatty acid composition become noticeable is likely to depend on the dosage level, and may also vary with age (Youdim et al., 2000). Conversely, brain plasticity may enable a certain degree of adaptation to biochemical changes before the effects of deficiency are observable (Wainwright, 2000); for instance in Parkinson's disease motor symptoms only become apparent when dopaminergic neuron deficiency in the brain exceeds 70–80% (Wainwright et al., 1999). Horrobin (2003) has

suggested that EPA dosages might produce a bell curve effect, whereby too much EPA might not be beneficial due to the resulting reduction in AA levels. A variable dose exploration trial found greatest improvements in schizophrenic symptoms with 2 g EPA per day (Peet & Horrobin, 2002). This supports the notion that optimal *ratios* of n-6 and n-3 fatty acids are of most importance for healthy phospholipid structure and function. Simopoulos (2002a) reviewed the effectiveness of different ratios in various diseases and concluded that the most effective ratio seemed to vary depending on the disease being treated. With regard to neural function it has been proposed that a ratio of 4:1 is best for optimal membrane fluidity (Yehuda et al., 2002).

Insufficient statistical power is another research issue that could be masking significant effects—a number of fatty acid intervention trials have been limited to small participant groups. It has also been suggested that studies should measure baseline EFA levels and exclude participants with EFA within normal range in order to avoid a Type II error (Hallahan & Garland, 2004; Richardson, 2003b). Inconsistent blood analyses (Stevens et al., 2003) suggest, however, that there needs to be further investigation of associations between biochemical measures of EFA levels, abnormal EFA metabolism, HUFA phospholipid ratios, and supplementation outcomes before researchers apply this research exclusion criterion.

There are also discrepancies in the type of omega-3 that is studied. Because of the logical importance of DHA due to its high concentration in brain phospholipids and neural synapses, many researchers have focused on this HUFA. However, some attention has also been given to EPA, the n-3 precursor to DHA, which might play roles in addition to synthesizing DHA that are critical for membrane phospholipid activity (Puri & Richardson, 1998; Richardson & Puri, 2002) and reducing inflammation in the brain caused by prostaglandin PLA_2 (Horrobin & Bennett, 1999). PLA_2 is produced by AA from the omega-6 series and can have adverse inflammatory effects if not controlled by the series 3 prostaglandins that are produced by EPA (see Fig. 1). In normal circumstances, fish oil provides both EPA and DHA. If EPA is uniquely important, this might have implications for the results of interventions that use DHA exclusively. Conversely, studies using EPA on its own may not derive potential benefits of increased DHA if there are obstacles (e.g., dosage, time, metabolic problems) to its conversion into DHA.

Additionally, blood measures of brain lipid status can vary. It is not possible to directly measure lipids in the brains of live organisms. Plasma, or serum, lipids have been widely used for assessing essential fatty acid status (Bryan & Hughes, 2002; Ward, 2000). However, Ward (2000) warns that plasma levels may be affected by dietary intake. Furthermore, in individuals who have poor cellular uptake of EFA, plasma levels might not accurately

reflect intracellular/phospholipid lipid levels (Mitchell et al., 1983). Peet et al. (1998) and Chiu et al. (2003) cite studies with human infants and primates that have established a good correlation between erythrocyte or red blood cell (RBC) measures of n-3 fatty acid status and brain n-3 HUFA. Following an extensive review, Lauritzen et al. (2001) concluded that circulating lipids are more prone to fluctuation according to dietary intake and that, apart from brain autopsies, RBC may be the biomarkers of choice for assessing brain DHA status.

Finally, research into fatty acids and mental health, although clearly important, necessarily crosses a number of disciplinary boundaries, connecting for instance nutrition, biochemical analysis, psychiatric diagnoses, psychological assessment, and so on. It is possible that some differences in research outcomes might stem from inadequate understanding of techniques and procedures that are not intrinsic to a particular research group's expertise. Pertinent limitations to randomized controlled clinical trials are addressed by Slade and Priebe (2001), and issues in fatty acid research with developmental disorders are critically reviewed by Richardson (2003b), with guidelines and recommendations given for future clinical trials.

IX. SUMMARY

Children with a range of developmental disorders including mental retardation and ADHD display problems in attention and learning, may be hyperactive and distractible, and may have difficulty with impulse control and aggression that result in the production of maladaptive social behavior. All of these problems may be linked to difficulties with neurotransmitter production and functioning. Consistent with this observation, CNS stimulant medication has been shown to alleviate some of these symptoms in the short to medium term. However, the effectiveness of this approach to treatment is variable and is frequently limited to behavior problems rather than cognitive and learning difficulties.

We propose that consideration be given to further testing of dietary interventions, both as an approach to treatment and as a therapy for prevention in groups considered at high risk for the later display of developmental disorders (e.g., babies with low birth weight). We argue that this proposal is based on evidence that suggests that highly unsaturated omega-3 fatty acids have crucial structural and functional roles in the brain. They cannot be produced by the body and therefore must be provided via the diet or supplementation. Accordingly, it has been established that long-chain omega-3 fatty acids EPA and DHA levels vary in neural membranes proportionate to dietary intake and that many diets in Western societies are

deficient in n-3 intake. Although research findings are equivocal, there is theoretical and statistical support for the role of HUFA in neural functioning and for the impact of omega-3 deficiencies on brain development, mental health, learning, and behavior.

Considerably more research is required to determine optimal HUFA levels throughout pregnancy, early childhood, adolescence, adulthood, and senescence; optimal n-3 to n-6 ratios, particularly the relative importance and appropriate ratios of DHA, EPA, AA, and DGLA; specific mechanisms by which they influence brain and neurotransmitter functions; their interaction with genetic vulnerabilities, and so on. Within this context, more controlled clinical trials are necessary to explore further their influence on the spectrum of learning and behavioral difficulties. These trials need to allow for brain plasticity that may, to various degrees, moderate nutrient deficiencies, and at the other end of the spectrum, allow time for neural uptake of nutrients before any cognitive and behavioral outcomes become apparent. Some tests may be more sensitive than others to changes in HUFA status and more research is required to identify tests that tap into specific brain functions and behavioral outcomes that are affected by HUFA deficiency and supplementation. Dosages and quality of the fish oil are important considerations. There are also indications that some individuals may need higher than normal doses of HUFA or other specific nutrients to address deficits in brain function (Harrell et al., 1981; Laugharne, Mellor, & Peet, 1996; Pauling, 1995; Richardson, 2003c; Uauy et al., 1996), an issue that also needs to be addressed.

One limitation in HUFA and nutrition research to date is the focus on nutrients in isolation, without accounting for the interaction between micronutrients as they enhance or inhibit one another in their metabolism and functions (Bryan et al., 2004; Ramakrishnan et al., 1999; Tacconi et al., 1997; Wachs, 2000). The authors are currently addressing this deficit in a completed study with children who have learning and behavioral difficulties. The study compared the effects of HUFA supplementation alone with placebo and with HUFA combined with a multivitamin/mineral supplement to assess whether there are any additional benefits with the vitamin and mineral micronutrients.

Nutrition is only one component of a variety of interactive environmental and biological influences on developmental behavioral and cognitive outcomes, particularly in complex disorders such as mental retardation, and it is unlikely that all individuals will respond to supplementation. However, it is a variable that can be relatively easily and safely modified with minimal if any side effects, and there is enough evidence that a subgroup of individuals may be assisted by supplementation with fatty acids and other nutrients to justify in-depth investigations by scientists. Should mental

retardation researchers pursue research to investigate the effect of nutritional supplementation, particularly with HUFA, on symptoms of mental retardation, it is possible that this could provide a safe, preventative measure for at-risk groups and/or long-term clinical intervention to assist in the reduction of maladaptive behaviors and cognitive functioning associated with mental retardation and comorbid psychopathology. Behavioral nutritional and lipid research requires interdisciplinary collaboration to link the intrinsic physiological, chemical, and nutritional processes at the molecular level with the psychological, cognitive, and behavioral outcomes of brain function, with the ultimate aim of informing clinical diagnosis and treatment outcomes for psychological, cognitive, and behavioral disabilities.

AUTHOR NOTES

The studies referred to on pages 179 and 188 have since been published:

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Screening for Autism in Infants, Children, and Adolescents

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I. DIAGNOSIS IN AUTISM

It is generally accepted that the onset of autism is in infancy (Volkmar, Stier, & Cohen, 1985), within the first 30 months of life (Gillberg, 1989; Rutter, 1978). The age at which problems with development are first noticed or suspected has been asked of parents across a number of studies. In summarizing the results of these studies (Table I), the vast majority of parents suspect problems with their child's development well before the child is 24 months of age. Despite this, the average age at which parents first seek help for these problems has been reported as 27.6 months (range 1 month–38 years) (Howlin & Moore, 1997) and similarly 24.1 months (De Giacomo & Fombonne, 1998). Howlin and Moore (1997) found that on average parents tend to wait 6–7 months from when they first become concerned about their child to when they first seek help, while 23% of parents waited up to 12 months and 9.4% waited up to 2 years (Howlin & Moore, 1997). Similarly De Giacomo and Fombonne (1998) reported that on average parents waited 5.2 months before first seeking help.

There is a clear discrepancy between when parents first notice problems with their child's development and the age at which a diagnosis of autism is made. Table II provides a summary of the results of research examining the age at diagnosis, indicating that the vast majority of children are diagnosed with autism well after the age of 3 years. Comparison of this table with Table I reveals a marked contrast between the age at which parents recognize that their child has problems with development and the age at which a diagnosis of autism is given. A delay of 4.42 years has been reported, from the time at which families first began to be concerned about their child's

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TABLE I
AGE OF RECOGNITION/ONSET OF SYMPTOMS

Study	*N*	Age of recognition of problems
(Baranek, 1999)	11	Mean of 13.86 months
(De Giacomo & Fombonne, 1998)	80	Mean of 19.1 months
(Fombonne, 1995)	49	Mean age of 13 months
(Frith & Soares, 1993)	173	65% before 24 months
(Howlin & Moore, 1997)	1294	Mean age of 20.3 months
(Ohta, Nagai, Hara, & Sasaki, 1987)	129	57% between 18 and 30 months
(Ornitz, Guthrie, & Farley, 1977)	74	50% by 12 months of age
(Rescorla, 1986)	274	Before 30 months of age
(Short & Schopler, 1988)	1800	Mean of 20 months
(Siegel, Pliner, Eschler, & Elliott, 1988)	51	By 18 months of age
(Smith, Chung, & Vostanis, 1994)	127	Before the age of 18 months
(Volkmar, Cohen, & Paul, 1986)	50	Mean of 18 months
(Volkmar et al., 1994)	454	Mean of 12.7 months

TABLE II
AGE AT DIAGNOSIS

Study	*N*	Age at diagnosis
(Baranek, 1999)	11	Mean = 32.55 months
(Frith & Soares, 1993)	173	0–3 years: 30% 3–5 years: 46% 6+ years: 24%
(Howlin & Moore, 1997)	1294	Mean = 6.11 years (73.32 months)
(Siegel et al., 1988)	51	Mean = 4.5 years (54 months)
(Smith et al., 1994)	127	Young group mean = 3.63 years (43.5 months) Older group mean = 6.89 years (82.7 months)

development to the time they received a diagnosis (Howlin & Moore, 1997). A delay of 3.81 years from the time of first seeking professional help to receipt of a diagnosis has also been identified through this extensive survey of parents (Howlin & Moore, 1997).

One of the reasons for this delay is attributed to the various difficulties and challenges of diagnosing autism in young children. An accurate diagnosis of autism in infants and young children involves differentiating autism from a variety of developmental disorders including mental retardation, hearing impairments, speech and language disorders, and severe attachment and neglect problems (Rogers, 2001). Baron-Cohen, Allen, and Gillberg (1992)

and Stone and Hogan (1993) have highlighted the problem of using diagnostic criteria, which emphasize areas of social and communicative development which are in general, difficult to assess in preschool children. Behaviors can change rapidly during the early childhood period in all children (Lord, Storoschuk, Rutter, & Pickles, 1993). In young children with autism, symptoms can vary over time, and possibly present only intermittently (Ornitz, 1973), thus making careful and extended observation essential. This is often the case with repetitive and stereotyped behaviors, which are frequently not observed in short observation sessions in a clinical setting.

A follow-up study of children diagnosed with autism at 2 years of age found that a number of changes had occurred in the rates of behaviors by follow-up at 3 years of age (Lord, 1995). A number of items, which were found to differentiate the children with autism from those with speech and language delays without autism at 2 years of age, were found to be more prevalent in the children with autism at age 3. These included abnormalities in understanding gesture, sharing enjoyment, greeting, social reciprocity, and directing attention, all of which showed higher rates in the children with autism. A number of behaviors decreased in prevalence in the children with autism from age 2 to age 3 including abnormalities in the use of another's body as a tool, interest in children, and unusual sensory behaviors. This study clearly demonstrates the behavioral changes that can occur in young children with autism within a brief period of time.

Difficulties in distinguishing between autism and mental retardation (Vig & Jedrysek, 1999) and language disorders without autism in young children (Marcus & Stone, 1993) also complicate the diagnostic process. The differentiation of children with autism with a mental age of less than 18 months from those nonverbal children with developmental delay without autism, or with language impairment has been identified as particularly difficult, with a resulting over diagnosis of autism in these groups (Lord et al., 1993; Rutter, 1999). Difficulties potentially arise in this group of nonverbal, severely delayed children due to problems in differentiating deviance in their behavior from severe global delay (Lord et al., 1993).

A lack of specialized training of primary health care professionals has also been proposed as a reason for the reported delays in diagnosis (Baron-Cohen et al., 1992; Ornitz, 1973; Vostanis, Smith, Chung, & Corbett, 1994). The relative rarity of the condition also contributes to this problem (Baron-Cohen et al., 1992) by reducing the amount of exposure to and knowledge of autism compared to other, more prevalent conditions, as does the shortage of specialist services (Chung, Smith, & Vostanis, 1995; Vostanis et al., 1994). Early attempts to overcome this difficulty were made by authors, such as Prior and Gajzago (1974), who published a list of signs to aid medical practitioners in early detection.

II. SCREENING FOR AUTISM

A dual-level approach to the assessment and diagnosis of autism has been suggested (Filipek et al., 1999). Level 1 of the recommended approach suggests that primary care providers perform screening for developmental disorders on a routine basis. Level 2 involves assessment and diagnosis, performed by specialists in the evaluation and treatment of autism. However, the consensus panel (Filipek et al., 1999) acknowledged that we are a long way away from such an ideal standard practice, with fewer than 30% of primary care providers undertaking standardized developmental screening tests (Dworkin, 1992). Screening of at risk children who have developmental problems might more effectively channel referrals to the scarce resource at specialist assessment services for autism and improve the rate of early diagnosis. This chapter will review the reliability, validity, and utility of the currently available instruments designed to screen for autism.

Primary care professionals, such as general practitioners, community pediatricians, and child health nurses, are usually in the best position to implement screening procedures, but they have limited time and may not have trained in the identification of complex childhood disorders. It is therefore essential that screening be neither time consuming nor require extensive training. Ideally, a screening tool should be brief, be able to be completed by parents, and enable easy identification of those children requiring referral for specialist assessment. Screening instruments specifically developed for autism include measures for use with infants and preschool children and those for use with older young people. These two groups will be considered separately.

III. INFANTS AND YOUNG CHILDREN

Instruments developed specifically to discriminate infants and young children with autism from others include the Infant Behavioral Summarized Evaluation (IBSE) (Adrien et al., 1992), the Checklist for Autism in Toddlers (CHAT; Baron-Cohen, et al., 1992, 1996) the parent-completed modified version of the CHAT (M-CHAT) (Robins, Fein, Barton, & Green, 2001) the Screening Tool for Autism in Two-year olds (STAT) (Stone, Coonrod, & Ousley, 2000) the Autism Screening Questionnaire/Social Communication Questionnaire (SCQ) (Berument, Rutter, Lord, Pickles, & Bailey, 1999) the Pervasive Developmental Disorder Screening Test (PDDST), and an application of the Developmental Behavior Checklist (DBC; Einfeld & Tonge, 1995, 2002) described in this chapter.

Half of the CHAT is completed by the parent or carer while the other half is completed by a clinician. The M-CHAT is a modification of the CHAT,

which is completed by parents. Both the STAT and the IBSE are clinician-completed assessments. The SCQ (Berument et al., 1999) has a version for children of less than 6 years of age; at the time of writing there were no published evaluations of its psychometric properties with this age group. The PDDST has been designed to be used with children aged 2–3 years. It is a parent-completed questionnaire, consisting of 72 items. To the best of the authors' knowledge, there were no published evaluations of the PDDST at the time of writing.

A. Infant Behavioral Summarized Evaluation

The IBSE (Adrien et al., 1992) was adapted from the Behavioral Summarized Evaluation (BSE) in order to assess the behaviors of children with autism, aged 6–48 months. This was accomplished by adding 13 items to the original 20 items. Each item is scored on a scale of 0–4 (0 = the problem is never observed, to 4 = the problem is always observed) after 5 days of observation by a trained rater. Factor analysis produced two main factors which accounted for 59.4% of the total variance. One of these factors which comprised 19 items, accounted for 50.1% of the variance and was labeled the "Autism" factor (Adrien et al., 1992). Discriminant function analysis demonstrated that this factor was able to correctly classify 83.3% of intellectually disabled children with autism from those who did not have autism (Adrien et al., 1992). This 19-item version forms the IBSE for use with children aged 6 months to 4 years by a trained rater after at least 5 days of observation.

B. Checklist for Autism in Toddlers (CHAT and M-CHAT)

The CHAT (Baron-Cohen et al., 1992, 1996; Swettenham, 1996) was designed to be used by general practitioners or health visitors at a child's 18-month checkup. The CHAT consists of two sections. Section A contains nine questions (yes/no answers) which the clinician asks the parent and section B consists of five observations of behaviors/skills made by the clinician as present or absent. The CHAT can be completed by an experienced clinician in approximately 10 min.

Screening tests are evaluated in terms of their sensitivity, specificity, Predictive Value of a Positive test (PVP), and Predictive Value of a Negative test (PVN). Sensitivity (or true positive rate) is the probability of having a positive test result among those who have a positive diagnosis, while specificity (or true negative rate) is the probability of having a negative test result among those who do not have the diagnosis (Kraemer, 1992). Predictive Value of a Positive test is the probability of having a positive diagnosis among those who have a positive test (screen), and PVN test is the probability of not having the diagnosis among those who have a negative test (screen)

(Kraemer, 1992). A 6-year follow-up study has established that using a one-stage administration the CHAT has a sensitivity of 0.38, specificity of 0.98, and PVP of only 0.047 (Baird et al., 2000). Low sensitivity in a screening tool will result in cases with the condition being missed in the screening process. A low Positive Predictive Value will result in a number of cases screening positive who do not actually have the condition. Repeating the CHAT 1 month after the first administration resulted in an increase in the Positive Predictive Value to 0.75 but at a cost to sensitivity (0.18). Specificity was extremely high (0.99) indicating a very low false positive rate. This increase in the Positive Predictive Value indicates improved accuracy when the test is repeated 1 month later. That is, of those who screen positive, a significant number will have the diagnosed condition (in this case autism). However, this results in a significantly lowered sensitivity indicating that many cases who do have autism will not be identified by the screen. Data is lacking on the reliability of the CHAT.

Robins et al. (2001) developed a 23 item, parent-completed modified version of the CHAT (M-CHAT). Children were screened at well-baby clinics, indicating high sensitivity (0.87) and specificity (0.99) in a sample of 39 children with autism/PDD from a total sample of 1293 screened. It also has good internal consistency (Cronbach's α of 0.85). These results indicate a promising screening tool; however, the M-CHAT has not yet been evaluated specifically in terms of its ability to detect autism in children with developmental delay.

C. Screening Tool for Autism in Two-Year Olds

The STAT has been designed for use in children 24–35 months of age and consists of 12 items assessing 3 areas of behavior, namely imitation, play, and communication (Stone et al., 2000). The STAT is a 20-min interactional assessment, requiring a trained clinician to administer. A sample of 40 children aged 27–35 months was used to establish the scoring algorithm for the STAT, and a sample of 33 children aged 24–35 months was used to validate the screening algorithm (Stone et al., 2000). Sensitivity of 0.83 and specificity of 0.86 were obtained. Although this study provides preliminary support for the STAT as a screening tool, the very small sample sizes (only 7 and 12 children with autism respectively in the two samples) warrant caution and indicate the need for replication with a larger sample.

D. Developmental Behavior Checklist Early Screen

The authors took an existing parent-completed measure of behavioral and emotional problems in children with intellectual disability, the DBC (Einfeld & Tonge, 1995, 2002), and evaluated its efficacy as a screening tool for autism in children with developmental delay aged 18–48 months (Gray &

Tonge, 2005). This study involved identifying those items of the DBC which differentiated a sample of 18–48 months old children with autism and developmental delay from a control group of children with developmental delay who did not have autism.

The subjects with autism were obtained from a number of regions which were representative of the broad range of Australian social class, urban and rural communities and ethnic mix. All of the subjects in the autism sample had received a diagnosis of Autistic Disorder by specialists using DSM-IV (Diagnostic and Statistical Manual of Mental Disorders, 4th Edition) (American Psychiatric Association, 1994) criteria. The diagnosis was reviewed by either one of two clinicians experienced in the assessment and diagnosis of autism, and for whom interrater reliability had been established (calculated on a sample of 107 cases of Autistic Disorder (Cohen's $\kappa = 0.95$). The subjects with developmental delay without autism (control group) were recruited through representative early childhood intervention services in Victoria and New South Wales, Australia.

All the children in this study had a developmental delay defined by a score of 2 or more SDs below the mean on a developmental assessment tool such as the Bayley Scales of Infant Development (Bayley, 1969), Griffiths' Mental Developmental Scales (Griffiths, 1954, 1970), McCarthy Scales of Children's Abilities (McCarthy, 1972), Merrill-Palmer Scale (Ball, Merrifield, & Stott, 1978), Gesell Developmental Schedule (Ames, Gillespie, Haines, & Ilg, 1979; Gesell, Ilg, & Ames, 1974), Leiter International Performance Scale-Revised (Roid & Miller, 1997), or Psychoeducational Profile-Revised (Schopler, Reichler, Bashford, Lansing, & Marcus, 1990). Children with physical disabilities only were not included in the control group.

The autism group consisted of 60 children, of whom 49 (81.67%) were male with a mean age of 40.31 months (SD = 5.96, a range of 23–48 months). The developmental delay control group consisted of 60 children of whom 40 (66.67%) were male with a mean age of 35.92 months (SD = 7.57, a range of 19–47 months). The autism group was significantly older than the control group, $t\ (111.88) = -3.53$, $p = .001$.

Parents completed the DBC which consists of 96 questions regarding behavior and emotions using a 0–2 scale, where 0 = "not true as far as you know," 1 = "somewhat or sometimes true," and 2 = "very true or often true." The DBC was developed for use in children and adolescents aged 4–18 years; therefore those items which were given a score of zero ("not true as far as you know") in 75% or more cases, were excluded from further analyses. These items were excluded from further analyses because they were considered developmentally inappropriate for the age of the sample.

The remaining 65 DBC items were individually entered into a series of univariate logistic regressions to determine which items differentiated the

autism and control groups. Thirty DBC items were found to significantly discriminate between the groups ($p \leq .01$). Confirmatory factor analysis was then used to extract a single common factor from these 30 items. Loadings of the DBC items were permitted to load freely on the common factor, while the loading of the group membership variable was constrained to 1.00 in order to align the factor with autism. Polychoric correlations were used to ensure that any differences in the proportion of respondents endorsing each of the items did not affect the magnitude of the correlation coefficient (McDonald, 1985; Muthén, 1989). Using a conservative factor loading cut point of 0.60 or greater, 17 DBC items were selected to create a DBC autism screening algorithm. The decision regarding a cut point is arbitrary, however, it was decided that a cut point of 0.60 produced a sufficiently short screening tool but retained items which had face validity and were felt to be clinically important in the diagnosis of autism in young children.

The sum of these 17 items produces a screening score ranging from 0 to 34. An ROC curve was generated for the screening algorithm in order to evaluate the performance of the screen and assign a cutoff score. This resulted in an area under the curve (AUC) of 0.871 (SE = 0.033, 95% CI: 0.806–0.936), indicating good overall performance of the screening algorithm.

Cut points can vary according to the purpose of the screening. For example, in order to identify all or as many cases of autism as possible, setting the cut point low will maximize sensitivity, although at the expense of specificity. Conversely, if the resources to conduct many assessments with false positives are not available, setting the cut point high will increase the specificity, while reducing the sensitivity. In this study a cut point was chosen in order to optimize both sensitivity and specificity.

In a study of 111 children with developmental delay (autism = 56 and controls = 55) a cut point of 11 was applied to parent-completed DBC Early Screen (DBC Early Screen) data and a good screening profile was obtained [sensitivity = 0.86, specificity = 0.69, efficiency (correct classification rate) = 0.77, PVP = 0.74, PVN = 0.83]. Therefore, the DBC early autism screen has a satisfactory performance as a screening tool, with the added advantage of being brief and completed by parents.

E. Summary

There are relatively few instruments that have been evaluated as screening tools for autism with infants and young children. Considering the current emphasis on early diagnosis and subsequent early intervention, further work is needed to evaluate and develop the screening and psychometric properties of the above and other potential tools. However, perhaps the initial use of a parent-completed screen, such as the DBC Early Screen,

followed by a clinician-completed assessment, such as the CHAT, at least in at risk children with developmental delay, offers a reasonable prospect of channeling likely cases to scarce autism assessment services.

IV. CHILDREN, ADOLESCENTS, AND YOUNG ADULTS

Since Kanner first described a group of children with an "inborn autistic disturbance of affective contact" (1943, p. 250), a wide range of instruments and assessment tools have been developed for children aged 4 years and above. Some of these have been specifically developed as screening tools, while others have subsequently been used for this purpose. This section will first review those early instruments and then cover more recently developed instruments including those which have been specifically designed to screen for autism.

A. Early Instruments

Over the past 50 years a large number of instruments have been designed for use with children and adolescents with autism. Early tools included the BRIAAC (Ruttenberg, Dratman, Fraknoi, & Wenar, 1966), the Rimland Diagnostic Forms (Rimland, 1971), the BOS (Freeman, Ritvo, Guthrie, Schroth, & Ball, 1978), the Autism Behavior Checklist (ABC) (Krug, Arick, & Almond, 1980), the Childhood Autism Rating Scale (CARS) (Schopler, Reichler, DeVellis, & Daly, 1980), and the Real Life Rating Scale (RLRS) (Freeman, Ritvo, Yokota, & Ritvo, 1986). Although many were not necessarily designed as screening tools per se, they provide measures of a range of symptomatology of autism.

1. BEHAVIOR RATING INSTRUMENT FOR AUTISTIC AND ATYPICAL CHILDREN

The Behavior Rating Instrument for Autistic and Atypical Children (BRIAAC) (Ruttenberg et al., 1966) is made up of four core scales that measure the child's behavior in different areas, namely: (1) nature and degree of relationship to an adult as a person, (2) communication, (3) drive for mastery, and (4) stage, modulation, and expression of instinctual drives. It includes three supplementary scales that evaluate intellectual development, speech development, and social skills. Scores on the BRIAAC cannot be summed to arrive at a total score, and its psychometric properties have not been well described, thus limiting its potential as a screening tool.

2. THE RIMLAND DIAGNOSTIC FORM FOR CHILDREN WITH BEHAVIOR DISTURBANCES

Rimland's Form E-1, and the revised version Form E-2 (Rimland, 1971), was designed to identify autism as described by Kanner (1943). It is a parent-completed questionnaire which consists of 80 multiple choice questions covering the areas of social interaction and affect, speech, motor and manipulative ability, intelligence and reaction to sensory stimuli, family characteristics, illness development, and physiological and other biological data. A plus point is obtained for each question answered as characteristic of early infantile autism, and a minus point is scored for each question answered in the nonautistic direction. A total score is calculated which is the difference between the child's autism (+) and nonautism (–) scores. Rimland (1971) reports scores as ranging from −42 to +45 (based on a sample of 2218 children). A cutoff score of +20 or above is regarded by the author as indicative of classic Early Infantile Autism (Rimland, 1971). Neither evaluation has been made of its reliability and validity, nor how scores on the instrument compare to independent clinical diagnosis.

3. BEHAVIOR OBSERVATION SCALE FOR AUTISM

The Behavior Observation Scale for Autism (BOS) (Freeman et al., 1978) consists of 67 behaviors derived from a review of the clinical literature in autism, which are rated on a 0–3 scale (0 = did not occur to 3 = occurred continuously) during nine 3-minute rating intervals. Administration of the scale requires training; observers in one study participated in 2 months of training prior to data collection (Freeman & Schroth, 1984). Interrater reliability has been reported as good for 55 of the items (correlation coefficients greater than 0.84), but information is lacking on the reliability of the remaining 12 items (Parks, 1983). No information is available on its efficacy as a screening tool.

4. AUTISM BEHAVIOR CHECKLIST

The ABC (Krug et al., 1980) consists of 57 items, rated by a clinician, which are grouped into five areas: (1) sensory, (2) relating, (3) body and object use, (4) language, and (5) social interaction and self-help. The items were grouped into subscales on the basis of face validity. A score is calculated based on a rater's dichotomous scoring of the weighted items. Ranges are provided and they suggest that scores of 67 and above indicate a high level of autism, and scores under 53 indicate a low probability of autism. Studies have concluded that use of the suggested cutoffs provides significant levels of misclassification (Nordin & Gillberg, 1996; Volkmar et al., 1988; Wadden, Bryson, & Rodger, 1991). In one study evaluating the ability of the

ABC to differentiate between DSM-III-R (American Psychiatric Association, 1987) diagnosed autism and non autism, a correct classification rate of only 0.50 was obtained (Sevin, Matson, Coe, Fee, & Sevin, 1991). However, the sample size of this study was very small ($n = 24$), and there were only three children in the non autism group. A further study of 184 children and adolescents (aged 4–18 years, 95 of whom had a DSM-IV diagnosis of a PDD) with intellectual disability found an AUC of 0.76, sensitivity of 0.71, and specificity of 0.70 (de Bildt et al., 2003).

While the internal consistency of the total score has been reported as satisfactory (Cronbach's α of 0.873), it has been recommended that due to the low levels of internal consistency (Cronbach α's ranging from 0.38 to 0.79) of the subscale scores, only total scores should be used (Sturmey, Matson, & Sevin, 1992). The ABC has been shown to be uncorrelated with either the CARS or the RLRS (Sevin et al., 1991), indicating that it is measuring different behaviors than those measured by these two scales. Factor analysis has failed to support the division of the checklist into five subscales (Wadden et al., 1991) and concern has been raised regarding its reliability (Parks, 1983).

5. CHILDHOOD AUTISM RATING SCALE

The CARS consists of 15 scales: (1) impairment in human relations, (2) imitation, (3) inappropriate affect, (4) bizarre use of body movement and persistence of stereotypes, (5) peculiarities in relating to nonhuman objects, (6) resistance to environmental change, (7) peculiarities of visual responsiveness, (8) peculiarities of auditory responsiveness, (9) near receptor responsiveness, (10) anxiety reaction, (11) verbal communication, (12) nonverbal communication, (13) activity level, (14) intellectual functioning, and (15) general impressions. Each scale is given a score ranging from normal to severely abnormal (1 = behavior within normal range for child's age, to 4 = severely abnormal behavior) based on a clinical interview with the parent and direct observation of the child by someone trained in its use. It is stressed that for each behavior the age of the child being rated must be taken into account (Schopler et al., 1980). Total scores can range from 15 to 60. A cutoff score of 30 or above is indicative of a diagnosis of autism (Schopler et al., 1980). The CARS is reported to have good interrater reliability (correlation coefficients ranging from 0.55 to 0.93, average 0.71) (Schopler et al., 1980) and good internal consistency (Cronbach's α of 0.75, 0.85) (Garfin, McCallon, & Cox, 1988; Sturmey et al., 1992). Higher CARS scores have been found for adolescents with autism compared to those with intellectual disability without autism (Garfin et al., 1988), although sample sizes were small and it was not clear whether or not the clinicians rating the CARS were blind to the adolescents' diagnostic status. Another study of

40 children found that the CARS was able to correctly classify those with autism from those with intellectual disability only with 100% accuracy (Teal & Wiebe, 1986). Sevin et al. (1991) also found high levels of agreement between scores on the CARS above cutoff and DSM-III-R (American Psychiatric Association, 1987) diagnoses of autism, with 92% of a sample of 24 cases aged 2–22 years who met diagnostic criteria meeting the CARS cutoff criterion for autism. However, in addition to a small sample size, only three of the cases did not have autism.

More recently, studies have examined how CARS scores relate to the ADI-R (Lord, Rutter, & LeCouteur, 1994) and DSM-IV (American Psychiatric Association, 1994) diagnoses. One study found an observed agreement of 66.7% (Cohen's $\kappa = 0.40$) between the CARS and the ADI-R in 54 children aged 22–114 months referred for possible autism (Saemundsen, Magnusson, Smari, & Sigurdardottir, 2003). Pilowsky, Yirmiya, Shulman, and Dover (1998) obtained agreement of 85.7% (Cohen's $\kappa = 0.36$) in 70 cases aged 20 months–34 years. Research using a Japanese version of the CARS (CARS Tokyo version; CARS-TV) found that a total score of > 25.5 was able to differentiate young people (aged 25 months–25.5 years) with a DSM-IV diagnosed PDD from those with mental retardation (sensitivity of 0.86, specificity 0.83) (Tachimori, Osada, & Kurita, 2003). The need to conduct a full diagnostic assessment in order to score the CARS means that its main role is as a quantitative component of diagnosis rather than as a screening tool.

6. RITVO–FREEMAN REAL LIFE RATING SCALE

The Ritvo–Freeman Real Life Rating Scale (Freeman et al., 1986) consists of 47 behaviors, which are grouped into five areas: (1) sensory–motor, (2) social relationship to people, (3) affectual responses, (4) sensory responses, and (5) language. The basis of the development of the RLRS was the BOS (Freeman et al., 1978). The scale is completed after observing the person for at least 30 min in a natural setting. Each behavior is rated on a scale ranging from 0 (never demonstrates the target behavior) to 3 (target behavior is seen almost always). A total score and subscale scores can be calculated, but no cutoff scores for diagnosis are provided. Observers can be adequately trained to administer the scale in three training sessions (Freeman et al., 1986). Sevin et al. (1991) reported interrater observer agreement κ's ranging from 0.21 to 0.64 (mean of 0.31) and adequate subscale and total score interrater reliability ($r = 0.82$ for total score, subscales 0.70+ with the exception of sensory responses $r = 0.32$). A significantly high correlation ($r = 0.77$) has been reported between the CARS and the RLRS (Sevin et al., 1991) and good (Cronbach's α of 0.84) internal consistency for the total score but not the subscale scores (range of Cronbach's α from 0.42

to 0.68) (Sturmey et al., 1992). Like the ABC, the subscales of the RLRS were derived purely on the basis of face validity. In light of these considerations it has been recommended that only the total score of the RLRS be used (Sturmey et al., 1992).

More recently, the RLRS has been examined in terms of its correlation with the ADI-R (Lord et al., 1994). The scale was completed by parents on 226 children with a mean age of 8.3 years (Lecavalier et al., 2006). A low correlation was found between the RLRS and ADI-R total scores (Spearman's rho of 0.07) indicating that the RLRS may be measuring a different construct from that measured by the ADI-R. The only significant correlation was between the ADI-R Social domain and the RLRS Language subscale (Spearman's rho of 0.28). These results may reflect conceptual changes in the definition of autism from the time when the RLRS and the ADI-R were developed; DSM-III (American Psychiatric Association, 1980) to DSM-IV (American Psychiatric Association, 1994). These findings do not support the use of the RLRS as either a diagnostic or screening tool in contemporary clinical practice.

B. Recently Developed Instruments

In the past 10–15 years a range of measures have been developed to describe and measure symptoms of autism, along with instruments which were designed specifically as autism screening tools. In addition to these measures, general measures of behavioral and emotional disturbance namely the Child Behavior Checklist (CBCL) and the DBC have been evaluated in terms of their ability to identify children with autism. Each of these instruments is described in turn.

1. BEHAVIORAL SUMMARIZED EVALUATION

The BSE (Barthelemy et al., 1990) is a French rating scale designed for use with older children and adults and consists of 20 items. Each item is scored on a scale of 0 to 4 (0 = the problem is never observed, to 4 = the problem is always observed) after 5 days of observation by a trained staff rater. The total score of the BSE has been shown to have good interrater reliability across trained raters; for the total score a κ of 0.96 was obtained, 5 items had a κ above 0.60, and 5 ranged from 0.40 to 0.59 (Barthelemy et al., 1990). The BSE has also been shown to discriminate between intellectually disabled children with and without autism (Barthelemy et al., 1992). A correct classification rate of 0.90 was obtained with a group of 58 children (aged 2–8 years) with DSM-III diagnosed autism and a group of 58 with only intellectual disability (sensitivity of 0.84, specificity of 0.96). It was unclear whether

the trained raters who completed the BSE were blind to the diagnostic status of each child.

Nine items have been added to the BSE, resulting in the Revised Behavior Summarized Evaluation Scale (BSE-R) (Barthelemy et al., 1997). Interrater reliability was found to be good for the total score, although low for some individual items (total score κ of 0.97, 13 items κ above 0.60, 12 items 0.40–0.59, and 4 items below 0.40) (Barthelemy et al., 1997). Factor analysis produced six factors, with 2 accounting for 48.6% of the total variance (Factor 1 "Interaction Disorder" accounted for 38%) (Barthelemy et al., 1997). ROC analysis of Factor 1 resulted in a cutoff score of 27 (AUC = 0.80), which provides reasonable sensitivity and specificity (0.74 and 0.71, respectively) (Barthelemy et al., 1997). The requirement for 5 days observation by trained staff greatly limits its use as a practical screening instrument.

2. PERVASIVE DEVELOPMENTAL DISORDERS RATING SCALE

The Pervasive Developmental Disorders Rating Scale (PDDRS) is 51-item rating scale with three subscales: arousal, affect, and cognition (Eaves, Campbell, & Chambers, 2000). Items were developed based on the research literature on autism, DMS-III-R diagnostic criteria, and clinical files of children with autism. The PDDRS is completed by teachers and scored on a 5-point Likert scale. It has demonstrated test–retest reliability over an average period of 9.5 months (α coefficients ranged from 0.75 to 0.89) (Williams & Eaves, 2002) and excellent internal consistency (coefficient α of 0.89) (Eaves et al., 2000). The total score of the PDDRS was found to correlate significantly with the total score of the ABC ($r = 0.80$) (Eaves et al., 2000). Exploratory and confirmatory factor analysis has validated the three-factor structure of the PDDRS in children with autism (Williams & Eaves, 2005). A study with 104 children with autism (mean age 8 years) and without autism (mean age 10.5 years) demonstrated good sensitivity (0.88) and specificity (0.88) when completed by teachers and other professionals (Eaves et al., 2000). Raters were not blind to the child's diagnosis.

3. GILLIAM AUTISM RATING SCALE

The Gilliam Autism Rating Scale (GARS) (Gilliam, 1995) consists of 56 items which make up four subscales (social interaction, communication, stereotyped behaviors, and developmental disturbances). It has been designed to be completed by parents or teachers. The items are rated on a 4-point Likert scale ranging from "never observed" to "frequently observed." Subscale scores and a total score [Autism Quotient (AQ)] are calculated. The AQ can be calculated from two to four subscales and measures the likelihood that an individual has autism. The subscale scores can be converted to standard

scores with a mean of 10 and SD of 3, and the AQ to a standard score with a mean of 100 and SD of 15. The normative sample consisted of 1092 children aged 3–22 years, who according to parent or teacher reports, had a diagnosis of autism. Norms are not broken down according to age, gender, or IQ range. An AQ of 131 and above is said to indicate a ''very high'' probability of autism, 121–130 ''high,'' 111–120 ''above average,'' 90–110 ''average,'' 80–89 ''below average,'' 70–79 ''low,'' and below 69 ''very low.''

While good to excellent psychometric properties have been reported (Gilliam, 1995), two studies have failed to replicate this earlier work. The first of these examined the GARS in a sample of 119 children (mean age 6.17 years) with DSM-IV diagnoses of autism (South et al., 2002). The second study evaluated the GARS in a sample of 284 children and young people (mean age of 9.3 years) with autism spectrum disorders (Lecavalier, 2005). Both of these studies found the mean AQ to be lower than that reported in the GARS manual (90 and 86, respectively).

South reported that an AQ cutoff score of $\geq$90, resulted in over half (52%) of the children being classified as having a ''low'' or ''very low'' probability of having autism, equating to a diagnostic sensitivity of 0.48 (South et al., 2002). Similarly, Lecavalier (2005) found 62% of ratings scored $\leq$90. Low correlations with the subscales of both the ADOS and ADI-R were described (South et al., 2002). Exploratory factor analysis revealed three factors (covering the areas of social, communication, and repetitive and stereotyped behaviors), explaining 37.6% of the variance (Lecavalier, 2005). While the majority (74%) of the items loaded on the factors as originally proposed by the author of the GARS, a large number of items (48%) loaded on to the repetitive and stereotyped behaviors factor.

Interrater reliability between teachers and parents was calculated for 63 children, with an average intraclass correlation coefficient of 0.40 across the three subscales (Lecavalier, 2005). No significant differences were observed between parent and teacher ratings. Although good internal consistency was reported for the three subscales (coefficient α's of 0.82–0.85), the internal consistency was significantly lower for the Developmental Disturbance subscale (coefficient α of 0.68) (Lecavalier, 2005). In sum, the results of these studies suggest that the use of the GARS as a screening tool for autism should be undertaken with caution.

4. SCALE OF PERVASIVE DEVELOPMENTAL DISORDER IN MENTALLY RETARDED PERSONS

The Scale of Pervasive Developmental Disorder in Mentally Retarded Persons (PDD-MRS) (Kraijer, 1997) was designed to detect Pervasive Developmental Disorders in people with intellectual disability aged 2–55 years. The PDD-MRS was first published in Dutch in 1990 (Kraijer, 1997).

It was then revised, the standardization sample was increased, and it was translated into English. The PDD-MRS consists of 12 items, which are differentially weighted based on the ability of each item to differentiate cases with Pervasive Developmental Disorders from those without. Items are scored as present or absent during an assessment time period of not less than the last 2 months and no more than the last 6 months. The scale is completed by a professional based on observations, structured parent interview, and other sources of information such as teachers and other professionals who have had contact with the person. Scores of 0–6 indicate non-PDD, 7–9 doubtful, and 10–19 is indicative of PDD.

The standardization sample consisted of 1096 subjects with intellectual disability (8% profound, 31% severe, 35% moderate, 17% mild) in the Netherlands. Sixty-five percent of the standardization sample consisted of an institutionalized population. Internal consistency is reported as good (coefficient α of .80) along with interrater reliability (correlation coefficients of 0.83 and 0.85) (Kraijer, 1997). Test–retest reliability on scales completed 6 months apart resulted in correlation coefficients of 0.81 and 0.86, indicating stability over time. High sensitivity and specificity are also reported (0.94 and 0.93, respectively). A recent study of 184 children and adolescents (aged 4–18 years, 95 with a DSM-IV diagnosis of a PDD) with intellectual disability found an AUC of 0.77, sensitivity of 0.92, and specificity of 0.42 (de Bildt et al., 2003). It is a useful diagnostic tool, but given the time and comprehensive assessment required for its completion, it does not have utility as a screening tool.

5. SOCIAL COMMUNICATION QUESTIONNAIRE

The SCQ (previously known as the Autism Screening Questionnaire) was developed as a screening tool for autism (Berument et al., 1999). It consists of 40 questions which were based on the ADI-R (Lord et al., 1994). The questionnaire is completed by parents and includes questions on reciprocal social interaction, language and communication, repetitive and stereotyped patterns of behavior, and a question on self-injurious behavior and current language functioning. There are two versions of the questionnaire, one designed for children aged less than 6 years, and one for children aged 6 years and above. The SCQ has been shown to correlate highly with the ADI-R domain and total scores (Berument et al., 1999), reflecting the source of its items, and has high internal consistency (Cronbach's α of 0.90).

In a total sample ($n = 200$) aged 4–40 years, at a cutoff of 15 the SCQ was found to be able to differentiate between PDD and non-PDD cases (AUC = 0.86, sensitivity of 0.85, and specificity of 0.75 Positive Predictive Value = 0.93, Negative Predictive Value = 0.55). It was also found to differentiate between those with autism and those with mental retardation without autism (AUC = 0.92, sensitivity = 0.96, specificity = 0.67). However,

in the case of the group with mental retardation, it is important to note that the sample size consisted of only 15 individuals. Further analyses using the domain scores concluded that the total score provides the most satisfactory differentiation (Berument et al., 1999). The factor structure of the SCQ has also been examined, with a four-factor solution accounting for 42.4% of the variation (Berument et al., 1999). Internal consistency was satisfactory, with an α coefficient of 0.90 for the total score, and 0.67–0.91 for the factors.

The SCQ appears to be a promising screening tool. However, further work is needed in terms of its psychometric properties, particularly with persons with intellectual disability and with young children as no information was provided on the psychometric properties for the separate version of the questionnaire for children aged less than 6 years. The parents who completed the questionnaire had already received the diagnosis of autism for their children and had also experienced an assessment with the ADI-R. As the authors noted, it is possible that this may have attuned them to the relevant behaviors of their child and the item content of the questionnaire (Berument et al., 1999).

6. CHILD BEHAVIOR CHECKLIST

The CBCL (Achenbach, 1991) has been evaluated for its ability to identify children with autism in Brazil (Duarte, Bordin, de Oliveira, & Bird, 2003). The parents of 101 children completed the CBCL in an interview format. Thirty-six of the children had autism (mean age 7.4 years) or a "related condition" not further defined by the authors, 31 with other psychiatric disorders (mean age 7.8 years), and 34 school children (mean age 7.0 years) randomly selected from two public schools. It was found that the Total Problems score of the CBCL did not differentiate those children with autism from those with other psychiatric disorders, nor did the internalizing or externalizing scores. The Thought Problems, Autistic/Bizarre, and Aggressive scales differentiated the children with autism from those with other psychiatric disorders. The Thought Problems scale was best able to differentiate the children with autism from the school-aged group (sensitivity = 0.94, specificity = 1.00), while the Autistic/Bizarre scale was best able to differentiate those with autism from those with other psychiatric disorders (sensitivity = 0.88, specificity = 0.80). No information was available on the cognitive ability of the children with autism, and it is not known how the CBCL would perform in differentiating children with autism from those with intellectual disability without autism.

7. AUTISM SPECTRUM SCREENING QUESTIONNAIRE

The Autism Spectrum Screening Questionnaire (ASSQ) was designed to assess symptoms of high-functioning autism and Asperger's Disorder (Ehlers, Gillberg, & Wing, 1999). It consists of 27 items, and is completed by lay informants (e.g., parents or teachers). Items are rated on a 3-point Likert

scale; ranging from 0 (indicating normality) to 2 (definite abnormality). An epidemiological study reported good test–retest reliability and interrater reliability between teachers (Pearson *r*'s of 0.90 and 0.79, respectively) (Ehlers & Gillberg, 1993). The reliability of the ASSQ was further evaluated in a clinic-referred sample of children with high-functioning autism and Asperger's Disorder (including children with mild intellectual disability). Similarly high levels of test–retest reliability for teacher ratings 2 weeks apart ($r = 0.94$) and parent ratings ($r = 0.96$) were obtained, although ratings of parent–teacher agreement ($r = 0.66$) were considerably lower. ROC analyses of both teacher ($n = 106$) and parent ratings ($n = 109$) resulted in recommended cutoff scores of 22 and 19, respectively. These cutoff scores resulted in a true positive rate of 70% for the teacher completed ASSQs and a true positive rate of 62% for the parent-rated forms. However, caution is warranted in interpreting these results due to the small numbers of cases with an "autism spectrum" disorder in the parent and teacher completed datasets (21 and 20 cases, respectively).

8. DEVELOPMENTAL BEHAVIOR CHECKLIST–AUTISM SCREENING ALGORITHM

The Developmental Behavior Checklist–Autism Screening Algorithm (DBC-ASA) was developed from the DBC (Primary carer version) (Einfeld & Tonge, 1995, 2002), which is a parent- or carer-completed checklist designed to measure behavioral and emotional disturbance in children and adolescents with intellectual disability. The DBC has been standardized on a representative sample of children and adolescents aged 4–18 years with intellectual disability in Australia. The checklist consists of 96 items which are scored on a 0–2 rating scale, where 0 = "not true as far as you know," 1 = "somewhat or sometimes true," and 2 = "very true or often true." Parents are asked to rate the items in terms of their child's behavior in the past 6 months. A total score (Total Behavior Problems Score) can be calculated along with scores on five factor analytically derived subscales (Disruptive/Antisocial, Self-Absorbed, Communication Disturbance, Anxiety, and Social Relating) (Dekker, Nunn, Einfeld, Tonge, & Koot, 2002). A cutoff score for psychiatric "caseness" has also been derived which reliably identifies those children with clinically significant levels of behavioral and emotional disturbance (Einfeld & Tonge, 1996).

The DBC has been shown to have high reliability between parents (intraclass correlation = 0.80) (Einfeld & Tonge, 2002). Internal consistency is also high ($\alpha = 0.94$). High correlations between the DBC Total Behavior Problem Score and other, professionally administered, measures of behavioral disturbance in children with intellectual disability have been reported, providing evidence of concurrent validity.

The DCB-ASA was developed as part of a study on 180 cases referred for assessment to five Pervasive Developmental Disorder specialist assessment services and were aged 4–18 years with a DSM-IV (American Psychiatric Association, 1994) diagnosis of Autistic Disorder. Interrater reliability (calculated on a sample of 107 cases of Autistic Disorder) was high (Cohen $\kappa = 0.95$). Subjects with autism were individually matched on age range, sex, and IQ range with a sample of 180 children and adolescents with intellectual disability who did not have autism.

Univariate logistic regression analyses identified 54 DBC items that discriminated between the autism and control samples. Following a principal components analysis, 29 of these items were used to create an ''autism scale score,'' the total of which ranged from 0 to 58. Using ROC analysis, the most efficient cut point (in terms of minimizing both false negatives and false positives) was 17 (AUC = 0.80, sensitivity = 0.86, specificity = 0.69). These statistics suggest that the DBC-ASA is a reliable and valid parent-completed tool to screen for young people who have autism.

In a validation study of the DBC-ASA, 84 children and adolescents from a Swiss national survey were identified with autism, and their parent completed the DBC (German translation) (Steinhausen & Metzke, 2004). Scores were compared with an age- and gender-matched sample of young people with intellectual disability without autism from Germany. Following a univariate logistic regression analysis a modified screening algorithm of 32 items was produced (DBC-ASAR2) which had a slightly improved screening performance (AUC = 0.85, sensitivity = 0.85, specificity = 0.67). The authors concluded that the DBC is a suitable instrument to screen German-speaking populations of young people with intellectual disability for autism.

V. SELECTING A SCREENING TOOL

A. Infants and Young Children

In Table III we compare the studies of the efficacy of autism screening tools which have been designed for infants and young children. Although the STAT (Stone et al., 2000) has adequate sensitivity and specificity, it requires a clinician to administer it, thus limiting its use as a screening tool for Pervasive Developmental Disorders in infants and young children. The IBSE requires a trained rater, thus also limiting its practicality as a screening tool. The CHAT (Baird et al., 2000; Baron-Cohen et al., 1992) has been shown to have good specificity, particularly if the administration is repeated 1 month later, although the sensitivity is low. However, the CHAT has been used as a population screening test, with a 6-year follow-up, and is the only instrument

TABLE III

SCREENING INSTRUMENTS FOR INFANTS AND YOUNG CHILDREN: SCREENING EFFICACY[a]

Instrument	Age	Items	Administration	AUC	Sensitivity	Specificity	PVP	PVN
Infant Behavioral Summarized Evaluation (IBSE)	6–48 months	19	Trained rater					
	Autism from developmental delay			–	0.85	0.82	–	–
	Autism from typically developing			–	0.92	1.00	–	–
Screening Tool for Autism in Two-year olds (STAT)	24–35 months	12	Clinician	–	0.83	0.86	–	–
	18 months	14	Parent and clinician					
Checklist for Autism in Toddlers (CHAT)	1-stage administration			–	0.38	0.98	0.047	–
	Repeat administration after 1 month			–	0.18	1.00	0.75	–
M-CHAT	18–24 months	23	Parent	–	0.87	0.99	0.80	0.99
DBC Early Screen	18–48 months	17	Parent	0.87	0.86	0.69	0.74	0.83

[a]AUC—area under the curve, PVP—Predictive Value of Positive test, PVN—Predictive Value of Negative test, PDD—Pervasive Developmental Disorder.

that has been tested in this way. It is not known if the use of the CHAT specifically in children with developmental delay might result in higher sensitivity. The CHAT requires a clinician to administer, thus limiting its application in large populations.

The M-CHAT has high sensitivity and specificity but has not yet been evaluated in terms of its ability to differentiate children with developmental delay without autism from those with autism. The DBC Early Screen has adequate sensitivity and specificity and shows promise as a screening tool for infants and young children with developmental delay. Importantly, it is able to differentiate children with developmental delay without autism from those with autism. Further research is needed in a larger sample to validate its screening properties.

B. Children and Adolescents

Table IV summarizes the results of instruments which have been assessed as potential screening tools for autism for older children and adolescents. While a number have good psychometric and screening properties, a clinician, trained rater, or teacher is required to administer them (e.g., CARS, BSE, PDD-MRS) thus limiting their practicality as screening tools. The ABC has been shown to have low sensitivity and specificity, and the CBCL has not been tested in terms of its ability to differentiate autism from intellectual disability. The ASSQ has only been evaluated in a small sample, and has been designed for use in young people with normal IQ to detect high-functioning autism and Asperger's Disorder. In contrast, the DBC has been shown to be a reliable and valid screening tool for autism in children and adolescents with intellectual disability in both English- and German-speaking populations and is practical to use as it is completed by parents or carers.

VI. CONCLUSIONS AND FUTURE RESEARCH

The ultimate utility of a screening instrument for autism will depend upon the degree to which pediatricians and other health professionals, such as speech pathologists, who come into contact with infants and children with developmental delay or intellectual disability decide to use it in their daily practice. Early childhood health services would also need to be convinced of its value and introduce it as part of other population-screening policies such as monitoring weight and height. Pediatricians and other primary care providers are often reluctant to undertake developmental screening, with fewer than 30% of primary care providers undertaking standardized developmental

TABLE IV

SCREENING INSTRUMENTS FOR CHILDREN AND ADOLESCENTS: SCREENING EFFICACY[a]

Instrument	Age	Items	Administration	AUC	Sensitivity	Specificity	PVP	PVN
ABC	2–22 years	57	Teacher/	–	0.48	0.67	0.91	0.15
	4–18	12	Clinician					
			Clinician					
	PDD from non-PDD			0.75	0.58	0.77	–	–
	Autism from nonautism			0.76	0.71	0.70	–	–
CARS	25 months–29 years	15	Clinician	–				
	PDD from MR			–	0.86	0.83	0.97	0.50
	Autism from PDDNOS			–	0.71	0.75	0.77	0.69
BSE	2–8 years	20	Trained rater	–	0.84	0.96	0.96	0.86
BSE-R (Factor 1)	24 months–11 years	13	Trained rater	0.80	0.74	0.71	–	–
PDDRS	1–12 years	51	Teacher	–	0.88	0.88	–	–
PDD-MRS	4–18 years	12	Clinician					
	PDD from non-PDD			0.74	0.81	0.48	–	–
	Autism from non-autism			0.77	0.92	0.42	–	–

Social Communication Questionnaire (SCQ)	<6 and >6 years	40	Parent					
	Autism from mental retardation			0.92	0.96	0.67	–	–
	PDD from non-PDD			0.86	0.85	0.75	0.93	0.55
CBCL: Thought Problems and Autistic/Bizarre scales	4–11 years	118	Parent (interview)					
	Autism from typically developing			–	0.94	1.00	–	–
	Autism from other psychiatric conditions			–	0.88	0.80	–	–
	6–17 years	27	Parents/Teachers					
ASSQ	Teacher-completed forms			–	0.70	0.91	–	–
	Parent-completed forms			–	0.62	0.90	–	–
DBC-ASA	4–18 years	29	Parents	0.80	0.86	0.69	–	–
DBC-ASA (German translation)	4–18	32	Parents	0.85	0.85	0.67		

[a]AUC—area under the curve, PVP—Predictive Value of Positive test, PVN—Predictive Value of Negative test, PDD—Pervasive Developmental Disorder.

screening tests (Dworkin, 1992; Rapin, 1995). Given the time constraints on a pediatric appointment and the apparent reluctance on the part of primary care physicians to undertake screening tests, a Pervasive Developmental Disorder screening tool must be short and require minimal clinician input. Clearly tools such as the DBC or the M-CHAT which are short, completed by parents, and can be easily scored fulfill these requirements. Given the recognized importance of early intervention, both the development and improved use of screening tools is paramount. However, it is apparent that improved professional education and changes in early childhood services policy is required to facilitate a broader use of screening tools by primary care professionals who are likely to be the first to see children at risk of Pervasive Developmental Disorders.

When screening for developmental problems in infants and young children sensitivity, specificity, and positive predictive values of 70–80% are regarded as acceptable (Aylward, 1997; Glascoe, 1997; Squires, Nickel, & Eisert, 1996). However, it is recognized that there is often a trade off in specificity if the sensitivity is high (Aylward, 1997). The detection of false positives is usually not a problem in developmental screening because such children usually have other developmental problems and benefit from specialist assessment and early intervention (Glascoe, 2001).

False negatives cases that are missed by a screening tool are of much greater concern. No screening tool can replace clinical judgment and regardless of the screening score, if it is felt that a child needs further assessment, an appropriate referral for assessment should still be made. One should never rely just on a single test, clinical judgment is still vitally important and no paper and pencil test can replace this.

The limitations of screening must also be recognized. The purpose of any screening tool, regardless of its field of use, is to indicate those in need of a more comprehensive assessment (Aylward, 1997). It is important to educate and train primary care physicians in the identification of symptoms of autism in infants and children and to equip them with screening tools to aid in this process. However, it is equally important to emphasize that although research has shown that the diagnosis of autism in young children is stable, these diagnoses have invariably been made by clinicians experienced in the assessment of infants and young children with autism (Rogers, 2001). A positive result from the use of a screening instrument is not the same as a diagnosis of a Pervasive Developmental Disorder. Rather, it identifies that the child at risk requires specialist assessment. Like any psychological assessment tool, screening tools cannot be used without informed clinical judgment and awareness of the limitations of such tools. Aylward (1997) wisely cautions that while a screening test yields a result, that result still has to be interpreted and followed up with an assessment.

It is also important to consider the potential limitations of a parent report measure as a screening tool. Such a measure is sensitive to both parental denial and over concern. Parents may under report developmental problems for a number of reasons including parenting experience, denial, cultural factors, and the presence of pressing medical issues (Filipek et al., 1999). However, the concerns of parents have been identified as accurate indicators of developmental problems (Glascoe, Altemeier, & MacLean, 1989), but in the case of Pervasive Developmental Disorders, parents also need to realize that there is something wrong with their child's development above and beyond developmental and/or language delays. Glascoe and Dworkin (1995) have identified a number of advantages in using parental report in screening measures. These include eliminating the need to obtain the cooperation of children, providing a more thorough sampling of skills or behaviors than is usually obtained with direct observation and flexibility with administration such as in the waiting room, or at home prior to an appointment.

It is important to note that in the area of screening tools for autism, regardless of the age of the children or adolescents, there is a lack of prospective screening studies. With the exception of the CHAT UK screening study (Baird et al., 2000), evaluations of autism screening tools have been done with the parents of children who have already taken part in the assessment process and received a diagnosis of autism. It is possible that parents who presumably have a good understanding of their child's symptomatology and have received a diagnosis may complete screening checklists in a different way to those parents of children who have not yet undergone assessment and diagnosis. It is therefore essential that more prospective studies of screening tools are undertaken, in order to better evaluate how these tools perform in the "real world."

In addition to undertaking community field trials of screening tools, it is essential to evaluate the reliability and validity of any proposed screening tool. Internal consistency, interrater and test–retest reliability are as important as establishing the discriminative and predictive ability of a screening test. Evaluations of screening tests also need to focus on differentiating autism from developmental delay/intellectual disability, and language disorders, rather than just differentiating between autism and typical development. Research also needs to consider who is going to complete the screening tool. For ease of administration and taking into account clinician time restrictions, a screening test needs to be completed by parent or carers. Therefore, the test needs to be brief, clear, and straightforward to complete. Future research needs to address these issues in relation to the range of screening tools that are available.

The key goal in the early identification and diagnosis of children with Pervasive Developmental Disorders is to facilitate access to early intervention

programs and alleviate parental and family distress. Much clinical research has now been done in developing and establishing interventions to help children with autism and their families. The development and use of screening tests to aid in the early identification of children who require specialist diagnostic assessment is an essential component in obtaining a diagnosis and access to intervention.

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People with Mental Retardation and Psychopathology: Stress, Affect Regulation and Attachment: A Review

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I. INTRODUCTION

Psychopathology and challenging behavior are widespread in people with mental retardation (MR), including a substantial number of individuals with both problems. Many persons with mild MR and challenging behavior are dually diagnosed with conduct disorders (45%) and depression (22%), for example, and aggressive behavior and sexual offending are not uncommon (Dagnan & Sandhu, 1999; Dykens, 2000; Gillberg et al., 1986; Maughan, Collishaw, & Picklel, 1999; McBrien, 2003). Challenging behavior may therefore not only be an educational and social problem but also part of the mental health problems of people with MR. Although precise estimates of the prevalence of challenging behavior in people with moderate and severe MR are still being debated (Deb & Bright, 2001), most figures vary between 30% and 60%. The more severe the retardation, the higher the prevalence of challenging behavior. This prevalence is judged to be about three to five times as high as in normative populations (Došen, 1990). This chapter is concerned with and tries to explain the inordinately high risk for people with MR of the often comorbid problems of challenging behavior and mental health, problems that severely limit their quality of life (Janssen, Schuengel, & Stolk, 2005) and which tend to pose limits on the potential for a normalized existence within the general community.

The high prevalence of challenging behavior and psychopathology in people with MR continues to call for explanatory frameworks offering additional options for intervention. Furthermore, the widespread acceptance of the claim that human development rests on biological, psychological, as

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well as social processes that are interrelated as they unfold over time calls for theorizing that takes multiple levels of functioning into account. As Emerson (2001) puts it:

> While there may be compelling evidence that both behavioral and biological processes may be involved ... models of development and maintenance of challenging behavior have been advanced within, rather than across, these potentially complementary frameworks (p. 156).

Developmental psychopathology is gaining prominence as an overarching approach to developing theoretical explanations for problem behavior and mental health problems in children as well as adults (Cummings, Davies, & Campbell, 2000). In this theoretical framework individuals are perceived to develop in transaction with their environment and research is done analyzing this developing transactional relation. Within this framework the abundant research into *attachment relationships* and into the development of physiological and biopsychological *stress* or *affect regulation* is found to be successful and may also be relevant for understanding the high risk for psychopathology in people with MR. In this chapter, the interrelated processes of affect regulation and attachment will be discussed, and research will be reviewed pertaining to affect regulation and attachment in persons with MR. This chapter is led by the hypothesis that people with MR are at risk for developing psychopathology because of exposure to chronic dysregulated affective arousal. Under stressful circumstances, they may have insufficient resources to deal with stressors on their own; the fall-back option of turning to attachment figures as a safe haven may however also be less available because people with MR may be more often insecurely attached.

The link between stress and psychopathology forms a basic premise of the model put forward in this chapter. This link is moderated by the personal as well as environmental resources that a person can command to deal with a given stressor. The functional limitations that characterize the group of people with MR form another premise of this chapter. Learning to solve everyday social problems is a difficult challenge even for people with mild MR, due to the complex functional abilities needed to process social information and to learn from experience. Handicaps in this area may be compensated by extra support from the environment. Attachment relationships are a basic species-wide adaptation to dealing with weakness and vulnerability in the face of threats and dangers. Furthermore, early attachment relationships provide a developmental context for the development of strategies for affect regulation. In this regard, answers are needed to questions such as: How do people with MR cope emotionally with the many stressful (social) situations? Can aggression, depression, destructive behavior, or

sexual offending also be interpreted as maladaptive ways of coping? Are children with MR at risk for developing insecure attachment, and how may this strengthen the association between exposure to stressors and the development of psychopathology? These questions will guide our review of the literature about stress, affect regulation and coping in people with MR, and the literature about the obstacles for environmental support and attachment. On the basis of this review, avenues for further research are suggested, and inferences are made concerning adequate interventions.

II. STRESS AND PSYCHOPATHOLOGY

Bradley (2000) has argued that a failure to regulate stress is an important, nonspecific factor in the development of psychopathology. Bradley did this by showing that the same stress-related risk factors are common to many different disorders. Loss, trauma, abuse, attachment difficulties, and sensitivity to expressed emotion or familial conflict, all lead to increased arousal and all are found to be strongly associated to many different disorders. Individuals differ in their reactivity to negative arousal states. The so-called stress reactivity would have a constitutional basis (akin to the reactivity concept of Rothbart, 1986) as well as an experiential basis. Prolonged states of stress involve feedback-loops of physiological, emotional, and behavioral stress responses; in the process, easily accessible pathways are etched in the brain, conditioning the brain to fear states and at the same time using working memory resources, thereby limiting cognitive abilities for adaptive functioning. This transactional process perspective, which is shared by theorists, such as Schore (2001a,b) and Panksepp (2001), may explain a common developmental pathway to increasing vulnerability to stress, which may engender increasingly maladaptive ways of regulating negative arousal as a result of this vulnerability. Gross (2002) and Gross and Levenson (1997) found emotion dysregulation to be a prominent feature of many forms of psychopathology. They found that half of the nonsubstance related Axis I clinical disorders and all of the Axis II personality disorders involve some form of emotion dysregulation.

Bradley (2000) furthermore has argued that affect regulation is not only relevant to understanding the development of psychopathology but also to understanding the effectiveness of psychotherapeutic interventions: (1) Many, not to say all, therapeutic interventions focus on general distress relieving elements, either by stress-reducing medication, or by changing client's stress-inducing internal working models, schemata, attributions, or defenses. (2) Most therapeutic interventions try to induce reduction of client's distress by using desensitization by means of step by step exposure to the stress events

or feelings. (3) The warmth, caring, and understanding by therapists all are meant to share client's feelings and to protect the client from distress. This conclusion further underscores the important links between psychopathology, affect regulation and the environment.

The specific type of psychopathology may depend on a biologically or experientially mediated diathesis, or, according to Bradley, may depend on the individual's biologically or experientially mediated coping strategies. It may not be dependent on the characteristics of the stressor. An avoidant coping strategy, for example, may ultimately result in depression if a person experiences chronic and/or severe stress; a resistant strategy may lead to aggression.

III. AFFECT REGULATION, STRESS, AND COPING IN PEOPLE WITH MR

Stress is defined as a feeling of inescapable helplessness in a situation in which the individual's well-being is under threat (Lovallo, 1997). Lovallo argued that coping with stress proceeds in two phases, which we tried to visualize in Fig. 1.

A. Primary Appraisal

In the first phase of coping, that of primary appraisal, individuals appraise in what way a situation may endanger their well-being by instantaneous referencing to their implicit memory, beliefs, and former experiences in

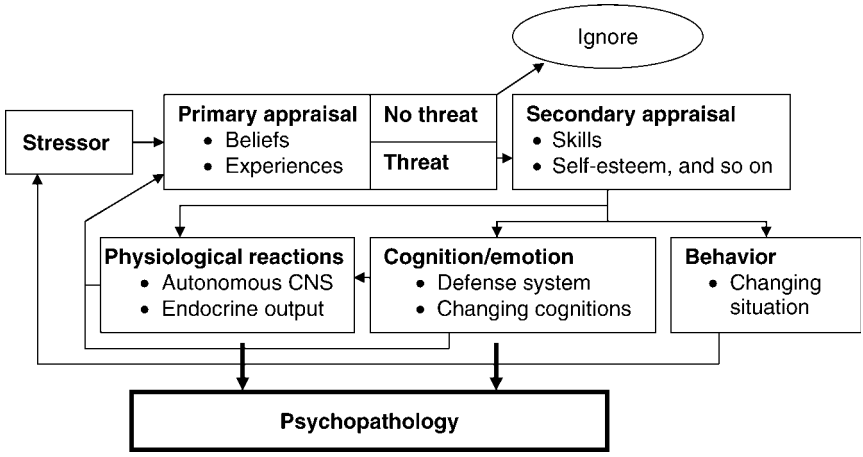

FIG. 1. Two phases of coping with stress.

comparable situations (Fig. 1). According to Porges (2003) and Bradley (2000) frightening former experiences are represented in the brain as easily accessible neural pathways. Stimuli and feelings bearing resemblance to these neurally represented frightening experiences instantly will activate these neural pathways, a phenomenon dubbed by Porges as neuroception of safety, danger, or threat. This is a basically automatic and unconscious response, mediated by the amygdala, the brain's generating center of emotions, leading to a feeling of undifferentiated arousal. If "no threat" is appraised the situation can be ignored and, according to Porges, the social engagement system can be used. Now the individual is free to engage and explore in social interactions. If on the other hand "threat" is appraised and the undifferentiated arousal felt, the body's physiological systems automatically prepare energy resources for a fight or flight response by sending neural and hormonal messages across the nervous system and across the bloodstream. The results are noticeable in the body's peripheral systems: accelerated heart rate, blood pressure, respiration, muscle tone, transpiration, and endocrine output. Clark and Wilson (2003), using the Reiss rating scale in people with Down syndrome, found levels of lacking self-control and impulsivity to be high, which may be an indication that primary appraisals (neuroception) of threat may be very common in this group. Because undercontrolled and impulsive responses to threat may actually exacerbate the threat and result in experiences of failure, proneness to primary appraisal of a stimulus as a threat may tend to increase.

B. Secondary Appraisal

In the second phase of managing stress, the secondary appraisal, the individual appraises the availability of problem-solving skills. The outcome of this appraisal is a function of both the actual skills the person has at his or her disposal as well as explicit memories about comparable situations, feelings, beliefs, self-esteem, cognitive internal working models, or schemata concerning these skills. For instance, the belief of learned helplessness, in which the individual thinks and feels totally out of control, will negatively influence the outcome of this secondary appraisal, independent of the actual level of problem-solving skills (Lovallo, 1997). Apart from learned helplessness, Bradley (2000) points to the additional concepts of maintenance of perspective, self-esteem, and self-efficacy influencing this secondary appraisal by adding meaning to the formerly undifferentiated affect. In this respect she describes the difference between affect, as a rather undifferentiated negative or positive state, and emotion, which is meaning coupled with affect. An undifferentiated negative affect may evolve into the meaningful emotion of anger and a positive affect may become the emotion of joyfulness.

The development of an emotion is a process in which the affect generating limbic centers of the brain, mainly the amygdala with its implicit memory of the mere association between situation and affect, interacts with emotions generating (pre)frontal circuits, cortical centers with their explicit memory of helplessness, perspective, and self-efficacy (Gross, 2002). In this process of secondary appraisal, the undifferentiated negative or positive affect of the primary appraisal can become a well-differentiated emotion like anger, sadness, or pride. Emotions are regarded as "time-tested responses to recurrent adaptive problems" (Gross, 2002, p. 281), therefore mediated by phylogenetically "old" parts of the brain, and the assignment of emotions to an aroused state proceeds largely outside of conscious awareness.

If the secondary appraisal turns out to be negative, that is, when the person draws the conclusion that the stressor exceeds the person's skills, the perception of threat will actually increase because now the self is perceived as vulnerable on a more conscious emotional level compared to the neuroception mechanism in the primary appraisal. Negative secondary appraisals are also associated with physiological responses preparing the organism for fight or flight, building up a state of hyperarousal. Actually, many forms of challenging behavior may require this hyperarousal. Especially prolonged states of hyperarousal are found to be strongly related to psychopathology because these prolonged experiential and physiological states are both a cause and a result of a deterioration of affect regulation in a circular loop, again etching in easily accessible maladaptive neural pathways.

People with MR have been noted to engage more frequently in fight or flight behavior (aggression, destruction, avoidance) than people without disabilities (Bihm, Poindexter, & Warren, 1998; Dykens, 2000). Under extreme circumstances hyperarousal can change into hypoarousal. This may manifest itself in the form of freezing or stilling in life threatening situations (Porges, 2003). In hypoarousal, corticosteroid stress hormones will continue to be elevated and the related increase of endogenous opiates causes blunting of sensory experiences like pain, a phenomenon that may lower the threshold for and facilitate continuation of self-injurious behavior in people with MR (Emerson, 2001). The hormonal chain of biological reactions in hyperarousal and hypoarousal, if prolonged, is held responsible for damaging the immune system in the long run, explaining the association found between psychosocial stress and health, ranging from asthma to cancer (Gross, 2002; Uchino, Cacioppo, & Kiecolt-Glaser, 1996). For example, the death rate in inhibited heart attack survivors with high scores on distress is found to be significantly higher than in the other attack survivors (Denollet et al., 1996). In the long run prolonged states of hyperarousal can even damage explicit memory, adaptive cognitive interpretations of situations, and former adaptive coping (Bradley, 2000). This may be the reason that depressive clients

frequently fail to remember things and have manic periods of maladaptive behavior.

C. Coping Strategies and Affect Regulation

If, on the other hand, the secondary appraisal is positive and the individual perceives his coping resources to be sufficient, the question becomes whether the problem is solved using an emotion-oriented or a problem-oriented strategy (Lovallo, 1997). In the emotion-oriented strategy the individual reappraises the stress situation by making use of defense mechanisms like sublimation, perseveration, or self-delusion. The use of these defensive emotional coping styles, frequently called avoidant coping, has been found to occur in people with mild MR, particularly in social situations. Hartley and Maclean (2005) found frequent avoidant coping in people with mild MR and noticed feelings of helplessness to be responsible for this coping style. Avoidant defenses seem to be the only coping strategies available when feeling helpless. The weaker positive relationship found by Elias (2005) between perceived competence and actual competence in children with mild MR, as compared to typically developing children matched on school grade, may be another illustration that emotion-oriented coping strategies (i.e., self-delusion) occur more often, because in spite of actual lower competence on several domains (as rated by teachers), perceived own competence was comparably high between the groups. Nothing about the actual stressor is actually changed by using emotion-oriented coping, which means that this form of coping has to remain active while the stressor itself may continue to exist and even increase. An emotion-oriented coping strategy may take initially less effort than problem-oriented coping, but if used indiscriminately will be more costly in the long run. Although some stressors may go away and leave not much harm, other stressors may pile up like unpaid bills, and self-deceptions used to avoid the stressors need to become increasingly elaborated and may hinder flexible functioning even more, while growing increasingly fragile. One example of such a process is the association between inflated self-concept and aggression (Baumeister, Smart, & Boden, 1996). Schuengel et al. (in press) found that in adolescents with cerebral palsy, perceived competence was not only significantly positively associated with actual competence as determined by medical assessment but also with aggressive behavior problems as reported by the parents. This suggests that some children and adults with disabilities may entertain positive self-concepts even with respect to aspects of functioning at which they objectively fail, at the cost of being easy to provoke, when others challenge that positive self-image.

A further potential negative by-effect of emotional coping may be that children using emotional coping let pass important opportunities for learning

how to cope with stressful situations. Bradley (2000) reviews experimental evidence that when in a state of hyperarousal people are unable to learn, because they are caught and incarcerated in that extreme experiential and physiological state, a phenomenon comparable to Porges' (2003) conception of a blocked social engagement system in situations of extreme stress. This latter conception has important implications for interventions. For instance, clients in a state of hyperarousal may be less amenable to cognitive behavioral interventions.

A problem-oriented strategy, on the other hand, means acting in the real world to remove the stressor. Gross (2002) delineates several methods of affect regulation in the real world. By means of *situation selection* people may select in advance low-stressful situations to engage in; in *situation modification*, for example, one may change the situation so as to make it less stressful, such as steering a conversation away from a painful topic; in *attentional deployment* one uses distraction of one's own attention to nonstressful actions or subjects; and in *cognitive change* one actually changes former beliefs, cognitive representations, or schemata by cognitive reappraisals. Gross refers to these four strategies as antecedent emotion regulation. He showed these antecedent strategies to be far more adequate in the regulation of emotions than problem-oriented strategies that are response-focused like *suppression of behavioral emotional responses.* In the case of bullying, for example, it turned out that a reappraisal of the perpetrator as someone to feel sorry for, is more effective and produces less psychological distress than suppression of the anger and grief responses. Hence, in a problem-oriented strategy antecedent-focused coping appears to be more effective than response-focused coping. It is more effective to avoid a potential-aggression evoking situation in advance, than to engage in that situation and count to 10. Again, this conception has important implications for interventions for people with MR.

D. Deficient Skills in People with MR

Which functional limitations may explain why people with MR may be confronted with stressful situations more, experience hyperarousal or hypoarousal more often, or develop more often maladaptive coping strategies? A number of those limitations will be reviewed later because of their obvious links to the stress-coping model outlined earlier, based on research findings. However, other limitations may play a role also, including limitations related to particular syndromes. The reviewed limitations with an obvious relevance to the model advanced here are: limited short-term memory, deficient executive functions, problems in perspective taking and empathy, and inadequate social skills.

1. LIMITED WORKING MEMORY AND DEFICIENT EXECUTIVE VERBAL FUNCTIONS

Limitations in working memory capacity may limit the range of verbal strategies to solve everyday problems (Hulme & Mackenzie, 1992; Jarrold, Baddeley, Hewes, Leeke, & Phillips, 2004; Mahler & Hasselhorn, 1990; Numminen et al., 2000). People with mild MR have been found to have limited working memory capacity (Van der Molen, Van Luit, Jongmans, & Van der Molen, in press). As pointed out by Bradley (2000) as well as Gross (2002), affect regulation is a great deal dependent on intact language. People need words to perceive, understand, and regulate emotions. The deficient executive verbal functions, for example, metaskills like self-monitoring, may make it difficult to monitor affects in the primary appraisal phase and to monitor and select adequate solutions in the secondary appraisal (Moffatt, Hanleymaxwell, & Donnellan, 1995). Besides, these deficient monitoring functions make it difficult to learn from failures.

2. PROBLEMS IN SOCIAL INFORMATION PROCESSING, PERSPECTIVE TAKING, AND EMPATHY

Problems in perspective taking and in empathy that may hamper accurate perceptions of the motives behind other people's social behavior can often lead to situations of conflict and stress (Benson, Abbeduto, Short, Nuccio, & Maas, 1993). Moffat et al. (1995) showed perspective taking and empathy to be associated with challenging behavior in people with MR. Van Nieuwenhuijzen et al. (2003) found problems in social information processing to be strongly associated with challenging behavior in people with mild MR.

3. INADEQUATE SOCIAL RELATIONS

Inadequate social skills of people with MR and the problems encountered by others to interact with people with MR may result in a lack of supporting friendships. The social network of people with mild MR has a mean of two persons, consisting of a professional caretaker (83%), family member (72%), or a peer (30%) (Robertson et al., 2001). Jarama and Belgrave (2002) concluded their thorough research among African-Americans with physical and learning disabilities that the balance between functional limitations, stressors, and the degree of social support and active coping emerged as a major determinant of adjustment. This appears to be in line with Groomes and Leahy (2002). Their study indicates that subjective meanings attributed by people with disabilities to stressful situations are related to coping problems and especially to levels of acceptance of the disability. In particular, social situations with peers and others appear to be stressful and have a greater frequency among people with mild MR compared to the general population

(Hartley & MacLean, 2005). People with even mild MR experienced more stress and less support from peers and this stress was associated with challenging behavior (Wenz-Gross & Siperstein, 1998). Chaney (1996), also studying the relation between stress and challenging behavior, found ordinary social situations to be stressful for people with severe MR. Most of the subjects showed extreme psychophysiological reactions in daily and common situations, and these reactions were found to be strongly related to challenging behavior. A strong relation has been found between the frequent social stress of people with mild MR and their depressive symptoms and physical complaints (Lunsky & Benson, 2001). There may be a circular loop: social situations are stressful for people with MR and the resulting social stress may also be the cause of inadequate social behavior. Porges (2003) explains the strong relation between perceived stress and problems in social behavior by pointing to the structure of human neurophysiology. Because of the specifics of the neurophysiological architecture, stress will obstruct the development and use of the social engagement system. This neurophysiological architecture is the reason, according to Porges, that social behavior can develop only in a context of minor distress.

In sum, we hypothesized that challenging behavior and psychopathology in people with MR may be explained by distress as a negative outcome of inadequate affect regulations and inadequate coping with the many (social) stressors encountered as a consequence of deficient skills. There is reason to hypothesize that in situations of excessive and prolonged distress the use of the cognitive skills available may be blocked and existing adaptive cognitive neural pathways may be even damaged.

Further, in addition to their problems in coping with stress, people with MR may lack the external buffering effect of support from significant others, especially support from attachment figures that are trusted by the person to be dependable, strong, and wise. In the next section, we review literature that indicates that many people with MR may frequently face stress without referring to a comforting, stress-buffering attachment figure. Furthermore, disorganized or disordered attachment may give rise to maladaptive developmental pathways of affect regulation.

IV. ATTACHMENT AND AFFECT REGULATION IN PEOPLE WITH MR

Attachment is the phenomenon that humans from infancy onward seek out a familiar, trusted person as a haven of safety in times of distress, and derive security from knowing the attachment figure or attachment figures are nearby, capable, and available (Bowlby, 1984). Bowlby argued that the

attachment behavioral system serves to restore homeostasis when a perceived threat or a separation leads to a negative emotion of fear or distress. The response of this goal-corrected system may consist of showing attachment behavior, such as looking, crying, or locomotion, to bring about more proximity to the attachment figure. However, the attachment system may also lead to other responses that at first glance do not seem to maintain or increase contact with the attachment figure but which do serve the goal of achieving a (relatively) calm affective state. Children may minimize their attention to their attachment figures or their absence, for example, if they believe that their call for attention will be rejected, which would lead to further upset. Children may also learn to delay their attachment behavior, for example, when they perceive that their attachment figures are not readily available but later will be an appropriate time to call attention to their emotional experience. These examples show that the attachment system is not regarded as a static system governing a fixed set of behaviors, but a dynamic system that adapts to the peculiarities of the caregiving situation as to optimize the attachment figures' effectiveness as a secure base from which to explore as well as a safe haven in times of danger.

Meaningful individual differences in patterns of attachment behavior have been demonstrated to exist across families (e.g., showing that quality of attachment is predicted by differences in parental sensitive caregiving behavior; De Wolff & Van IJzendoorn, 1997, for a meta-analytic review) as well as within families (e.g., there is only modest concordance within families between quality of attachment relationships between mother and child and between father and child; Van IJzendoorn & De Wolff, 1997, for a meta-analysis). Furthermore, attachment behaviors change with age, while the underlying organization of attachment behavior also reveals some significant continuity from infancy to adulthood (Fraley, 2002, for a meta-analysis). A considerable body of research has linked affect regulation to individual differences in quality of attachment in infancy (Hertsgaard, Gunnar, Erickson, & Nachmias, 1995; Nachmias, Gunnar, Mangelsdorf, & Parritz, 1996; Spangler & Grossmann, 1993; Spangler & Schieche, 1994, 1998; Van Bakel & Riksen-Walraven, 2004), adolescence (Kobak et al., 1994; Zimmermann, Maier, Winter, & Grossmann, 2001), as well as in adulthood (Dozier & Kobak, 1992).

Bradley (2000) is very explicit in showing the relevance of an attachment relationship for affect regulation. She emphasizes that in their early years children need the physical proximity of their parents to help them deal with demanding and stressful situations. The sensitive and responsive attachment figure buffers the stress felt by the child, for example, by an immediate protective response to the baby's gaze aversion when facing a stranger. Little by little the sensitive attachment figure while interacting with the infant

allows stress to enter the child's life, in a way using the principles of desensitization by gradual exposures to stress. Another frequently used parental response is mirroring. The parent imitates the infant's emotional behavior and reactions to stress, showing the child that emotions are shared and worthwhile. In later years more verbal strategies are used by parents in discussing and labeling the child's affects and emotions and in the teaching of coping skills. In this way, the child gradually learns to regulate affects.

In addition to research on attachment from a developmental behavioral perspective, such as described earlier, other research has approached attachment and bonding from a psychoneuroendocrinological perspective (Carter, 1998). This literature rests heavily on the study of animal models, which facilitates the integrative study of behavior, neuroendocrinological activity, and experimentally controlled environmental stimulation. This field of study is important because it suggests that processes underlying attachment formation and transactions between attachment relationships and emotional functioning are to a degree similar between humans and animals lacking the cognitive functions associated with the neocortex, such a rodents and priarie voles (Carter, 1998, for a review) as well as primates lacking the more advanced neocortical functions of humans such as rhesus monkeys (Suomi, 2005). Furthermore, studies in which animals are deprived of a mother figure or exposed to harsh mothering have shown that lower cognitive functioning can be a consequence of attachment experiences (De Kloet, Sibug, Helmerhorst, & Schmidt, 2005). That such findings also apply to humans is suggested by the low intellectual functioning of severely neglected children from orphanages, which recovers after adoption in families in most children but persists in a substantial minority of children (Rutter, O'Connor, & English and Romanian Adoptees (ERA) Study Team, 2004).

While documenting the importance of attachment relationships for the development of regulatory functions, these studies also suggest that caution should be exercised when interpreting studies on associations between quality of attachment and intellectual disabilities. This caution regards the direction of effects, as well as explanations for attachment risks based on a unidimensional conceptualization of intellectual disability as low cognitive capacity.

A. Empirical Studies of Attachment

Empirical studies on attachment in samples of persons identified with MR have predominantly been focused on young children, most often children with Down syndrome. Furthermore, almost all of these studies have used one or both of two instruments to assess individual differences in the quality of attachment relationships, the Strange Situation Procedure (SSP) with

its associated coding systems and the Attachment Q-Sort (AQS). Both instruments will be briefly described.

The SSP (Ainsworth & Wittig, 1969) is a laboratory-based protocol for children between 12 and 18 months old and their caregivers (most often parents). The procedure is conducted in an unfamiliar room with several toys, and consists of a series of episodes in which the child is with the caregiver, is joined by a stranger, and experiences two separations from the caregiver (first time left with the stranger, second time left alone) as well as two reunions. The mildly stressful conditions (unfamiliar room, unfamiliar person) as well as the two separations are assumed to activate the children's attachment behavioral system. Ainsworth, Blehar, Waters, and Wall (1978) discovered that individual differences in the patterns of children's behaviors during the SSP, especially immediately after the two reunions with the caregiver, were predicted by extensively observed interactions between mothers and infants in the year preceding the SSP assessment.

Ainsworth et al. (1978) developed a scoring and classification system for infant behavior during the SSP, permitting the classification of attachment relationships into three categories. The secure category is for children who may or may not be overtly distressed by the separation from their attachment figure, but who quickly reestablish contact on reunion, recover quickly from their distress, and recontinue with exploration of the toys. This pattern was predicted in Ainsworth's study by maternal caregiving at home characterized by consistent display of sensitive responsiveness. The insecure-avoidant category is for children who do not ostensibly show distress during separations from their attachment figure, and show little regard for the attachment figure on his or her return. The child may fail to greet, or may only give a cursory greeting and then focuses on the toys instead. This pattern was predicted in Ainsworth's study by maternal rejection and ignoring of the infant's emotional needs. The insecure-resistant category is for children who are focused on their attachment figure throughout the procedure, protest against his or her departure strongly, remain distressed throughout the separation (separation episodes are shortened if a child is more than mildly distressed), and remain distressed or angry throughout the reunion episodes as well. These infants do not instantly settle from distress if they are picked up by the attachment figure, unlike securely attached children. Ainsworth identified a history of inconsistent sensitive responsiveness for these children. Later studies have shown that the association between sensitive caregiving and attachment classification is robust, although the average effect size is fairly modest (combined $r = .25$; De Wolff & Van IJzendoorn, 1997). Furthermore, controlled intervention studies have yielded experimental evidence for the causal link between sensitivity and attachment by showing that experimentally induced effects on parental

sensitive responsiveness are related to effects on attachment security (Bakermans-Kranenburg, Van IJzendoorn, & Juffer, 2003).

In early studies with the SSP and the Ainsworth et al. (1978) classification system, not all children fitted the descriptions of the three categories, in particular children from at-risk populations such as maltreated children. A third insecure category was identified by Main and Solomon (1986) as disorganized/disoriented in directing attachment behavior to the attachment figure. Their attachment behavior during the SSP may reveal opposing attachment styles (i.e., avoidance and resistance) simultaneously or in sequence, may show interference of behavior incompatible with an attachment strategy (such as backward approach), may contain stereotypical behavior, stilling, freezing, or fright specifically during moments of reunion with the attachment figure (Main & Solomon, 1990). Antecedents of disorganized attachment include maltreatment and parental discord (Van IJzendoorn, Schuengel, & Bakermans-Kranenburg, 1999, for a meta-analysis) as well as direct observations of frightening as well as frightened maternal behavior (Schuengel, Bakermans-Kranenburg, & Van IJzendoorn, 1999) or hostile-helpless maternal behavior (Goldberg, Benoit, Blokland, Madigan, 2003; Lyons-Ruth, Bronfman, & Parsons, 1999). More so than for the other insecure patterns of attachment, disorganized attachment has been found to predict psychopathology directly, in particular externalizing behavior problems (Van IJzendoorn et al., 1999, for a meta-analysis).

Even with the addition of the disorganized category, there are still children who cannot be classified in one of the four attachment categories. Examples include children who do not display attachment behavior at all across the SSP, but do not particularly avoid contact with the attachment figure either. Also assigned to this category are children who would otherwise be classified as disorganized, except that the interpretation of infant behavior as disorganized *attachment* behavior is compromised by a physical or neurological condition that might produce behavior that is similar to behavior noted in the coding system as disorganized attachment behavior (Main & Solomon, 1990). Pipp-Siegel, Siegel, and Dean (2001) have listed several behaviors in the disorganized attachment coding system that may be confounded with neurological signs, such as freezing (could be indicative of petit mal seizures) or hitting at the face of the attachment figure (may result from dyspraxia). This list has been used by most researchers of disorganized attachment in populations such as children with Down syndrome, autism, or cerebral palsy, to exclude these confounded behaviors from consideration for classifying children with MR as disorganized. Nevertheless, only in the Vaughn, Goldberg, Atkinson, and Marcovitch (1994) study (described later) was the recommendation of Main and Solomon followed to assign children showing these confounded behaviors to the unclassifiable category; other

studies reserved the unclassifiable category for children not fitting the secure, avoidant, resistant, or disorganized patterns.

The strange situation paradigms and coding systems have also been adapted for children in the preschool age period (Cassidy, Marvin, & The Working Group of the John. D. and Catherine. T. MacArthur Foundation on the Transition from Infancy to Early Childhood, 1992). Categories for this age group are somewhat similar to the secure, avoidant, and resistant groups in infancy (although resistant attachment in infancy corresponds to ambivalent attachment in preschoolers), but the disorganized pattern gives way to a preschool classification of controlling attachment (with punitive and caregiving subgroups), and insecure-other (including disordered attachment subgroups). Evidence supports the validity of this coding system (Moss, Bureau, Cyr, Mongeau, & St. Laurent, 2004; Stevenson-Hinde & Shouldice, 1995). At age 6, a similar procedure and coding system has been developed and used, and the results have been found associated with longitudinal attachment assessment from infancy as well as concurrent measures of caregiving and parent–child relationships (Main & Cassidy, 1988; Solomon, George, & De Jong, 1995). For the infant as well as the preschool attachment classification systems, training opportunities exist for researchers to attain reliability with expert coders, and to demonstrate reliability on a standardized set of tapes. In addition to reliability between researchers working on the same study, researchers of attachment are required to also demonstrate reliability between researchers working in diverse research centers and different countries (Van IJzendoorn & Kroonenburg, 1990, for a review of cross-cultural consistency of coding attachment).

The AQS (Waters & Deane, 1985) was developed for the observation of attachment behavior of children from age 12 to 48 months in their natural environment such as the home. The AQS is a set of items printed on cards (75, 90, or 100). Each item consists of a description of a particular behavioral characteristic that children may display in their home situation. A considerable proportion of the items describe so-called secure base behaviors, which are the behaviors children use to achieve felt security as well as to allow exploration of the environment (Vaughn & Waters, 1990). The items are sorted by an observer into a fixed number of piles labeled from ''most descriptive of the subject'' to ''least descriptive of the subject.'' A security score is calculated by correlating the resulting profile to a hypothetical profile based on expert judgments as to which ranking of the items corresponds to the prototypically secure child. On the basis of robust associations with classifications derived from the SSP as well as with parental sensitive responsiveness, a meta-analytic review by Van IJzendoorn et al. (2004) concluded that the AQS is a valid instrument to assess security of attachment. However, the AQS only yields (continuous) scores of attachment

along the dimension of security–insecurity. It does not identify disorganized attachment.

B. Empirical Studies of Attachment and Mental Retardation

Several studies have been conducted on attachment between parents and children with MR. The topics of these studies have predominantly concerned the validity of the SSP for children from atypical populations. The following review of studies focuses first on studies of children with Down syndrome and second on studies of children with MR who were not specifically selected on the basis of a genetic syndrome.

Early studies by Cicchetti and Serafica (1981) on 42 children with Down syndrome, aged 30–42 months, and by Thompson, Cicchetti, Lamb, and Malkin (1985) on 26 children with Down syndrome, aged 19 months, reported that Down syndrome infants responded differently to the SSP than other children. In a study by Thompson et al. (1985) they seemed less distressed by the separation from their mothers, took longer to become distressed but shorter to settle, and their range of responding was smaller in comparison to a group of 43 infants aged 12 and 19 months. Nevertheless, despite these differences, there was also similarity in the patterning or organization of responses across the SSP. For example, the intensity of distress was positively correlated with maintaining contact. Amounts of attachment-related behaviors during the SSP were similar. However, in both studies the authors refrained from reporting attachment classifications according to the Ainsworth et al. (1978) coding scheme because they deemed the support for the validity of these classifications in this population lacking.

A similar conclusion was reached by Vaughn et al. (1994), on the basis of an analysis of SSP classifications of 138 children with Down syndrome, from three separate samples. The ages ranged from 14 to 54 months: developmental age ranged from 8.5 to 36 months. The attachment distribution of the Down syndrome group was compared to the distribution of a comparison group of 146 nonhandicapped infants between 1 and 12 months of age. The SSPs were classified using the Ainsworth et al. three-category system, without using the disorganized category that was not yet regarded as an accepted category at the time of that study. Significantly fewer children with Down syndrome were classified as securely attached to their mothers than nonhandicapped children (46% compared to 60%), which was primarily due to the high percentage of unclassifiable cases in the Down syndrome group (42% versus 3%). Vaughn et al. interpreted these results as evidence that the SSP elicits less stress in children with Down syndrome than in nonhandicapped children. They observed that children with Down syndrome rarely became visibly distressed in the SSP, even during the separation episodes,

showed less maintenance of physical contact if they sought such contact, and required little or no comforting. These authors referred to earlier findings that children with Down syndrome display a "dampening of affect" (Emde & Brown, 1978) and concluded that the attachment behavioral system in children with Down syndrome may not fulfill the same function as in nonhandicapped children.

Ganiban, Barnett, and Cicchetti (2000) studied 30 infants with Down syndrome at 19 and 27 months of age in order to investigate whether negative reactivity might be a temperamental trait of children with Down syndrome that limits the activitation of the attachment behavioral system. Negative reactivity was assessed on the basis of structured observations of distress during each episode of the SSP. In contrast to the Vaughn et al. (1994) study, Ganiban et al. (2000) included the disorganized category. No consistent associations were found between negative reactivity and attachment behaviors scored within the SSP nor classifications of attachment quality. The authors therefore failed to find evidence in support of Vaughn et al.'s (1994) suggestion that negative reactivity might regulate the activation of the attachment behavioral system during the SSP. Secure attachment was found in 53% of the infants at 19 months and 43% at 27 months, whereas disorganized attachment was found in 23% at 19 months and 30% at 27 months. These distributions compare somewhat unfavorably to the normative distribution derived from averaging low-risk, community-based studies of attachment in the United States (62% secure, 15% disorganized; Van IJzendoorn et al., 1999).

Still, the important question remained whether attachment classifications of children with Down syndrome can be considered as a valid indicator of the quality of their attachment relationships. To demonstrate validity, attachment classifications should be associated with sensitive responsiveness of caregivers as well as with attachment quality rated through other means than the SSP, such as with the AQS. Atkinson et al. (1999) conducted such a study, using a longitudinal design, with 53 infants with Down syndrome, aged 14–30 months and mental age 12–23 months at the first assessment. In this study, infants were classified as secure, avoidant, resistant, disorganized, or unclassifiable. When assessed at 26 months of age, 40% of the infants were classified as secure, 2% disorganized, and 47% unclassifiable. The rest were classified as insecure-avoidant or insecure-resistant. At 42 months, the percentage secure had risen somewhat to 47.5%, 12.5% were classified as disorganized, and 32.5% as unclassifiable. Both distributions were significantly associated with ratings of maternal sensitive responsivity in the home as well as during laboratory tasks. Mothers of unclassifiable infants and mothers of the combined group of unclassifiable and insecure infants were significantly less sensitive than mothers of secure children. However, at 42 months unclassifiable infants and

the combined group of unclassifiable and insecure infants were also found more mentally delayed at 26 and 42 months. Both effects (of sensitivity and mental development) were qualified by a significant interaction effect, indicating that the combination of relatively high cognitive competence and maternal sensitivity was most strongly associated with secure attachment (as opposed to unclassifiable and insecure attachment). Furthermore, the study showed that secure infants obtained significantly higher scores on the AQS than unclassifiable infants and the combined group of unclassifiable and insecure infants. Mental development was also significantly and positively associated with AQS security, and the average AQS security score for infants classified as secure in the SSP was lower than typically found in low-risk samples. Taken together, the authors concluded that secure base behavior is less readily shown by children with Down syndrome, that this is not due to method artifacts and that this reflects important differences between attachment relationships of children with and without Down syndrome.

Finally, two other, somewhat smaller studies have been reported on infants with Down syndrome. Oates, Moore, Goodwin, Hobson, and George (2000) studied 10 infants with Down syndrome in the SSP and found that 4 out of 10 were classified as disorganized. Attachment classifications were, however, not associated with maternal sensitivity during a face-to-face interaction task. Rauh and Calvet (2004) studied 16 infants (mental age 13–15 months) with Down syndrome and found that secure attachment significantly predicted social-cognitive achievements on the Bayley test.

Research on attachment in other populations of children with MR than Down syndrome is more scattered. In their meta-analysis on autism and attachment, Rutgers, Bakermans-Kranenburg, Van IJzendoorn, and Berckelaer-Onnes (2004) discussed findings on the association between mental development and security of attachment in samples of children with autism or pervasive developmental disorder (PDD). They found that mental development (dichotomized on the basis of the quotient of mental to chronological age greater than or below .70) moderated the association between the presence of autistic symptoms and security of attachment such that only in samples with mentally delayed children, autism was associated with insecure attachment. Furthermore, they also reviewed multiple studies showing that within samples of children with autism or PDD, developmental delay was significantly and negatively associated with attachment security. Most of these studies did not use the disorganized attachment category. With respect to disorganized attachment, Willemsen-Swinkels et al. (2000) found that the percentage of disorganized attachment (but not unclassifiable attachment) was elevated in the group of children both with PDD and with MR (54%), whereas a normal percentage was found in the group of children with PDD only (16%). They were also able to show, using heart rate

measures during the SSP, that children with autism and/or MR showed a similar pattern of physiological arousal during separation and reunion as nonhandicapped children. Moran et al. (1992) reported a study on 19 infants who were developmentally delayed, ranging between 10 and 31 months chronological age and 4.5–22 months mental age. Diagnosis of children included genetic disorders, prematurity, and neurological problems. The average security score on the AQS was rather low, indicating considerable insecurity, but was significantly associated with observer ratings of maternal sensitivity. Finally, De Schipper, Stolk, and Schuengel (2006) reported on observations of secure base interactions between a small sample ($N = 6$) of children with MR and their professional caregivers in specialized day care centers. Attachment behavior was observed by independent researchers, using the AQS, as well as by the professional caregivers themselves, using a behavioral rating scale. Findings suggested that professional caretakers are a target for attachment behavior from children with MR in day care and that variations in the strength of attachment behavior can be reliably observed by both independent observers as well as professional caregivers themselves.

C. Conclusions on Attachment in Children with Mental Retardation

Taken together, studies on children with MR and Down syndrome reveal atypical attachment both at the level of attachment behavior as well as at the level of distributions across attachment categories. Children with Down syndrome were found less often to display clear attachment behavior, both in the natural setting as well as in the laboratory environment of the SSP, than nonhandicapped attached children. This was true even for children who could be classified as secure, in comparison to nonhandicapped securely attached children. Furthermore, studies found an underrepresentation of secure attachment and an overrepresentation of disorganized or unclassifiable attachment. On several grounds, these findings cannot be dismissed as artifacts from applying attachment measures developed for nonhandicapped children to children with MR. When attachment behavior was shown by children with MR, it followed similar patterns as in nonhandicapped children, albeit to a lower degree: distress on separation was lower and therefore less approach behavior was shown, accompanied by less affective signals resulting in less comforting by the attachment figure, and requiring less contact maintaining behavior. Differences in attachment quality have been found to be associated with maternal sensitivity in two studies (except in a small study of 10 children) and have been found to converge across measures of attachment quality as well as settings. Nevertheless, also

within samples of children with MR, attachment security was associated with measures of mental development, revealing the paradox that although most infants were advanced enough to develop attachment relationships on the basis of understanding of cause-and-effect as well as person permanence, intellectual disabilities still seemed to influence attachment.

These findings raise two important questions. The first regards the explanation of atypical attachment in children with MR. Atkinson et al. (1999) reviewed the possibility that affective signaling may be compromised in children with developmental disabilities. They pointed to a host of deficits found with respect to communication and attachment behaviors such as looking, eye contact, vocalizations, crying, smiling, turn taking, social referencing, initiative taking, approach, and language. When affective signals are fewer and harder to interpret, caregivers may also provide less affective feedback, hindering the development of smoothly patterned emotional communication such as found in secure attachment relationships. This may even mean that "the attachment system fails to achieve its set goal of contact and arousal reduction" (Vaughn et al., 1994, p. 105). Another explanation put forward by Atkinson et al. is motivational in nature. Especially for those children low in cognitive functioning within the population of children with Down syndrome, it may be very difficult to develop shared meanings with parents, which is regarded as a prerequisite for intimacy, which is the motivational basis for the partnership between parent and child. This may explain why reunion with a parent might not stimulate affective display in these children, which in turn explains why there are few observations of comforting or close physical contact within these reunion episodes. Of course, both explanations may be true, which could mean that a considerable proportion of children with MR are deprived not only from the affect regulatory function of attachment relationships but also from experiencing comforting and intimacy. Furthermore, both explanations suggest that the associations found between maternal sensitive responsivity and attachment quality might reflect causal pathways in both directions. On the one hand children with MR may develop atypical attachment as a result of insufficient sensitive responding from the part of the parents. On the other hand children with MR may show atypical attachment behavior that impedes sensitive responding on the part of the parents.

The second question regards the consequences of atypical attachment for the development of adaptive and maladaptive behavior. None of the studies on attachment in children with MR have reported follow-up associations with mental health outcomes. Nonetheless, the review of research in this chapter suggests that part of the association between MR and psychopathology and challenging behavior may be mediated by the development in infancy of atypical attachment to primary caregivers. The overrepresentation of

disorganized and unclassifiable attachment means that a considerable proportion of children with MR, or at least with Down syndrome, do not show patterns of interaction with their caregiver, which are organized around the function of regulating behavior and affect in stressful situations. In terms of the affect regulation strategies outlined by Gross (2002 see earlier section), these children seem to use less problem-oriented coping, such as modifying the situation by signaling the caregiver, to repair the alarming separation that occurred. These children use instead more response-focused coping, such as suppression of behavioral emotional responses, which if completed successfully would result in the absence of attachment behavior and if partly successfully would result in muted or unclear attachment behavior. Inhibition of attachment behavior under conditions of stress as well as indiscriminate display of attachment behavior (lack of a preferred figure for contact) are hallmark behaviors for disorders of attachment (American Academy of Child and Adolescent Psychiatry, 2005). These disorders are usually only found in children subject to extreme deprivation of attachment (such as Romanian orphanages; O'Connor & Zeanah, 2003; Rutter et al., 2004; Zeanah, Smyke, Koga, & Carlson, 2005) or maltreatment (Boris et al., 2004; Zeanah et al., 2004). Disorders of attachment are considered as mediators of influence on the development of childhood mental disorders as well as a form of psychopathology in itself (AACAP, 2005). The research reviewed here suggests that the threshold to develop disorders of attachment may be lower for children with MR.

Further research is needed to unravel the causes of atypical attachment, and to determine the role of caregiving. If ineffective affective signaling, lack of intimacy, and sensitive responsive caregiving are connected by a vicious circle resulting in atypical forms of attachment, interventions aimed at parenting may halt this deleterious process and may even contribute to improvement on these three areas. Bakermans-Kranenburg et al. (2003) have shown meta-analytically that short-term interventions focused on parental sensitive behavior not only improve caregiving but also attachment security. One controlled experimental study on adopted children has even reported that such an intervention resulted in less disorganized attachment (Juffer, Bakermans-Kranenburg, & Van IJzendoorn, 2005) but other studies have failed to find such effects (Bakermans-Kranenburg, Van IJzendoorn, & Juffer, 2005). This indicates that disorganized (and presumably unclassifiable) attachment should be prevented using specific methods geared toward the anteceding parental behaviors. Findings of lower sensitive responsive behavior and other behaviors conducive to secure attachment of parents of children with disabilities are equivocal, both regarding the issue whether these parents really are less sensitively responsive than parents of nonhandicapped children (Pelchat, Bisson, Bois, & Saucier, 2003) as well as regarding

the question of whether parents of children with disabilities display a different kind of sensitivity (McConachie, 1989). Nevertheless, children with MR may need extraordinary sensitively responsive caregiving to compensate for the risk factors they bring to the attachment relationship (Atkinson et al., 1999; Ganiban et al., 2000; Von Tetzchner, 2004), and they may be more vulnerable when confronted with inconsistent, frightening, hostile, or helpless caregiving. Dyadic interventions with caregivers of deaf–blind children have, however, shown promise in improving the quality of parent–handicapped child interactions using both video-based feedback and attachment interventions (Janssen, Riksen-Walraven, & Van Dijk, 2003).

For some parents, heightened sensitive responsivity may be more difficult than for others, and there are indications that these differences may be related to the parents' own attachment background (Oates et al., 2000; Pelchat et al., 2003). This situation is somewhat similar as the situation of foster parents, who have to build attachment relationships with children who enter with highly insecure expectancies of caregiving and insecure or atypical strategies of attachment. Dozier, Stovall, Albus, and Bate (2001) have shown that the foster parents' own mental representation of attachment determines the quality of attachment with the foster child, presumably by enabling some foster parents to be sensitive to the emotional needs underlying the often negative, disturbing, or unclear attachment behavior of the children. Another factor may be unresolved feelings and thoughts that parents hold about having a child with MR (Baxter, Cummins, & Polak, 1995; Stolk & Kars, 2000), and which may diminish their resources for sensitive caregiving and increase the risk for frightening, hostile, or helpless caregiving, especially during the important early years. For children with cerebral palsy, independent studies have shown that maternal resolution of diagnosis was significantly associated with security of attachment (Barnett et al., 2006; Marvin & Pianta, 1996). These findings have led to the development of an intervention program aimed at helping parents to adapt to their child with special needs, in order to support the development of secure attachment and family well-being (Barnett et al., 2003). In all, successful intervention to prevent the development of atypical attachment between parents and children with MR may require multimodal programs. Controlled studies might not only show the effectiveness of such preventive interventions programs but might also elucidate questions about the causality of caregiving-attachment associations in this population.

Further research on atypical attachment in children with MR should also address the mental health aspects of atypical attachment. In order for this research to move forward, atypical attachment may need to be studied more from a clinical perspective of developmental psychopathology than from the perspective of normal development. The high percentages of atypical attachment as well as the discrepancies across studies in assigning

atypical attachment behavior to the disorganized or the unclassifiable category suggest that the current coding system for the quality of attachment relationships may fail to comprehensively capture clinically relevant variation. Despite the many advantages of using standardized and empirically thoroughly tested instruments, such as the SSP and AQS, for the study of children at risk (Van IJzendoorn & Bakermans-Kranenburg, 2003), there are also important downsides, resulting in inconsistencies between studies and lack of discrimination of clinically meaningful patterns within the broad category of unclassifiable or disorganized attachment (O'Connor & Zeanah, 2003). Furthermore, the SSP and its current coding systems are ill-suited for longitudinal studies and experimental studies requiring pre- and posttests due to qualitative differences in patterns of attachment across age periods, and they are ill-suited for epidemiological studies and screening of large numbers of children. Promising developments are protocols, based on but not identical to the SSP, for diagnosing disorders of attachment as well as caregiver interview protocols for screening for disorders of attachment. These instruments enable the identification of different forms of disordered attachment, such as emotionally withdrawn/inhibited attachment, indiscriminately social/disinhibited attachment, and various other disordered patterns. Studies have emerged, which indicate that these instruments can be reliably used in research (Boris et al., 2004; Zeanah et al., 2004, 2005). Although low cognitive development did not impede the use of the screening instrument for disorders of attachment in a sample of children reared in social deprivation in Romanian orphanages (Zeanah et al., 2005), these instruments still await testing in the population of children with congenital MR.

More insight into processes determining attachment quality as well as into disorders of attachment may also be important for the study of adaptation to the challenge of living away from home for children and adults with MR (Došen, 2005a,b). Institutionalized people with mild MR have been found to display a more insecure attachment style than those who stayed in their original environment (Muris & Maas, 2004; it should be noted that while attachment *style* is conceptually related to attachment theory, it has not yet been longitudinally related to patterns of attachment in childhood). Although institutionalization may lead to seeking out new attachment relationships (Schuengel & Van IJzendoorn, 2001), stable alternative attachment figures within institutions may not be available because professional care often is discontinuous. Professional care is also given by many caregivers, each with different levels of sensitivity. This is an additional risk for adequate and well-organized attachment. It is believed that insecure attachment of people with even mild MR may be an important reason for their deficient exploration and learning in new (social) situations (Clegg & Lansdallwelfare, 1995). On the other end of the spectrum, Clegg and Sheard (2002) reported that in young

adults with severe MR, a form of insecure attachment called by the authors "overinvesting in one or a few relationships that become a source of jealousy" was associated with challenging behavior.

D. Conclusions on Interventions in Psychopathology in People with Mental Retardation

Promising clinical interventions have been developed for children with severe and multiple disabilities and severe challenging behavior, whose histories show an association between challenging behavior and histories of maltreatment, neglect, or caregiving instability (Došen, 2005a,b; Sterkenburg, 2004). These interventions await further empirical research, which might also reveal more about the supposed links of psychopathology in these children with attachment and affect regulation (Janssen, Schuengel, & Stolk, 2002). We hypothesized that interventions will be most effective if they build on a secure attachment relationship, because attention, trust, and motivation are needed by the person with MR to be able to take advantage of interventions aimed at developing adaptive problem-solving skills and coping styles. In our research (Sterkenburg, 2004; Sterkenburg, Janssen, & Schuengel, 2005) we found support for this hypothesis in a series of single case experiments with children with severe MR and a severe visual handicap and comorbid severe and persistent challenging behavior. All clients experienced early pathogenic care. In this intervention, behavior modification was given only after an intensive and systematic attachment-based therapy, based on Bowlby's (1984) description of the phases in the development of attachment. The hypothesis was that approval and disapproval given by an attachment therapist would be more effective than that given by a control therapist in the later phase of behavior modification. The success of the attachment-based therapy, the first phase, was shown by significant increases in attachment behavior over time, significantly more attachment behavior to the attachment therapist than to a control (only attention) therapist, and by a significant reduction of psychophysiologically assessed stress reactivity [the preejection period (PEP) and the respiratory sinus arrhythmia (RSA) of the heart rate] over the course of the therapy in the presence of the attachment therapist, not in the presence of a control therapist. We found a significant reduction and for some behaviors total fading of previously persistent challenging behaviors not only during the sessions of the therapy but also in the group home outside the therapy settings. We were surprised that the start of a significant fading of formerly persistent challenging behavior already took place in the first phase of building the attachment relationship, while no systematic attention was yet given to this behavior. This decrease of challenging behavior may be the result of the stress-buffering effect of an attachment relationship with the therapist. The cognitive representation of this comforting relationship may be the reason why we also found a

significant decrease in problem behavior in the group home. In the second phase of behavior modification only some residual challenging behavior had to be treated. These preliminary results show, at least in our atypical clients, that psychopathology and challenging behavior may be strongly related with the client's problematic affect regulation and may be solved by initial external affect regulation induced by a sensitive and responsive attachment figure.

In earlier sections of this chapter we revealed two more important principles for intervention in psychopathology. First of all, in clients in a state of hyperarousal therapists should not start interventions by teaching them new behavior because excessive arousal may block a person's capability to learn, and if prolonged, may even damage these cognitive systems (Bradley, 2000). This may imply that (cognitive) behavior modification or therapeutic or intuitive ways of teaching these clients new insight or beliefs are only effective after arousal reduction. This issue supports two-phased intervention, like the integrative therapy of Sterkenburg, described earlier.

Second, antecedent-focused coping appeared to be more effective than response-focused coping in preventing distress and arousal (Gross, 2002). Interventions aimed at earlier processes in coping are more effective. To give some examples, it may be more effective to avoid a potential-aggression evoking situation in advance, than to engage in that situation and count to 10. It may be more effective to change maladaptive beliefs than to invest in detailed coping skills for every different situation. These insights might find their way further into the care for people with MR.

Having acknowledged this all, it is striking that attachment issues receive only scant attention in the scientific literature and in the practice-based literature on people with MR. Even so, attachment behavior by people with MR is to be observed frequently, also within the context of professional care, for instance day care for children with MR (De Schipper et al., 2006). One of the topics for further research might be to clarify intuitive notions of professionals and parents with respect to attachment. Sometimes attachment theory is misunderstood as focusing on dependence instead of independence (Bowlby, 1984). On the contrary, the research literature is consistently showing that nothing is more empowering than venturing out in the world knowing that one may fall back on a trusted person as a safe haven.

V. CONCLUSIONS

We hypothesized some important risks that may be responsible for the high prevalence of psychopathology in people with MR: frequent and large amounts of distress, insecure attachment, and attachment-related problems in affect regulation. In this area of research in MR the focus of attention is mainly on behavioral dynamics with functional analysis of challenging

behavior and assessment of here-and-now contingencies as important key words. Contingencies and behavioral analyses are important to unravel in what situations challenging behavior occurs, but frequently this behavior may be driven and explained by a problematic affect regulation. Future research on psychopathology should focus more on developmental issues regarding affect regulation. The research so far suggests that attachment may underlie affect dysregulation from early on in development, except for children of extraordinarily sensitive caregivers. Further research in the area of attachment and MR may lead to important insights not only in intervention programes but also in prevention.

Research is needed as to the role of stress in challenging behavior and psychopathology. The use of nonintrusive ambulantory devices, measuring stress by means of sympathetic and parasympathetic control of heart rate is promising. Psychophysiological stress measurement may make it possible to do functional stress analyses of challenging behavior and to establish the relation between stressful situations, problematic affect regulation, and psychopathology. In this way ambulatory stress measurement may give nonverbal clients a ''voice,'' indirectly communicating what matters in their life.

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Diagnosis of Depression in People with Developmental Disabilities: Progress and Problems

ANN R. POINDEXTER

CONWAY, ARKANSAS

I. INTRODUCTION AND GENERAL INFORMATION

Depression affects millions of people in the United States and the rest of the world every year. Major depression is one of the most common conditions that primary care physicians are asked to diagnose and treat (Vollner, 2002) and is the second most common chronic disorder seen in primary care offices (Sharp & Lipsky, 2002). Nonetheless, there is evidence that it is substantially underdiagnosed. For example, in a study of the prevalence of depressive symptoms in primary care medical practice, Zung, Broadhead, and Roth (1993) collected data from a sample of over 75,000 persons who visited the offices of 765 primary care physicians for any reason from February to September 1991. They used an outcome measurement of the index score for the presence of depressive symptoms on the Zung Self-Rating Depression Scale. The overall prevalence of significant symptoms of depression was 20.9%, but less than 2% cited depression as the reason for their office visit.

Rowe, Fleming, Barry, Manwell, and Kropp (1995) reported very high prevalence rates in a study of 1898 patients from 88 primary care practices, using a self-administered health-habits questionnaire. They assessed depression both for lifetime incidence and for the past 30 days, using the *Diagnostic and Statistical Manual of Mental Disorders, 3rd Edition Revised* (DSM-III-R) (American Psychiatric Association, 1987) criteria for depression. Lifetime rates of depression were 36.1% for women and 23.3% for men. Barriers to diagnosis seemed to include a lack of knowledge of diagnostic criteria and the importance of diagnosis for general well-being of involved individuals, as well as the perception that depressive disorders represent a character weakness and therefore should be denied and/or covered up.

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A total of nine criteria for a major depressive episode are listed in the *Diagnostic and Statistical Manual of Mental Disorders, 4th Edition, Text Revision* (DSM-IV-TR) (American Psychiatric Association, 2000), from which at least five must have been present during the same 2-week period and represent a change from previous functioning. DSM-IV-TR criteria include depressed mood most of the day, nearly every day (either subjective report or observation made by others); markedly decreased interest or pleasure in most activities most of the day, nearly every day (either by subjective report or observation made by others); significant weight loss when not dieting or weight gain (a change of more than 5% of body weight in a month), or decrease or increase in appetite nearly every day; insomnia or hypersomnia nearly every day; psychomotor agitation or retardation nearly every day (observable by others, not self-report); fatigue or loss of energy nearly every day; feelings of worthlessness or excessive or inappropriate guilt nearly every day; diminished ability to think or concentrate; and recurrent thoughts of death (not just a fear of dying), recurrent suicidal ideation without a specific plan, or a suicide attempt or specific plan. For a diagnosis to be made at least one of the symptoms must be depressed mood or loss of interest or pleasure in daily activities. On occasion, symptoms of depressive disorder appear to follow a relatively consistent seasonal pattern (seasonal affective disorder).

Other depressive disorders described in DSM-IV-TR (American Psychiatric Association, 2000) include dysthymic disorder (at least 2 years of depressed mood for more days than not, with additional symptoms of depression but not meeting criteria for depressive disorder) and depressive disorder not otherwise specified (different disorders with depressive symptoms that do not meet criteria for depressive disorder completely). By definition, depressive disorder is not due to direct effects of a medical condition, drugs of abuse, or reactions to medications.

II. INCIDENCE AND PREVALENCE OF DEPRESSION IN PERSONS WITH MENTAL RETARDATION

Although a few isolated researchers have reported data that show that depression occurs at lower rates in individuals with mental retardation than in the general population (McDermott, Platt, & Krishnaswami, 1997), most experts in the field do not agree. In general, there is concurrence that psychopathology is higher in persons with mental retardation. For example, a chapter in a textbook of clinical psychiatry reports a two- to fourfold increase in psychopathology among persons with mental retardation (Popper, Gammon, West, & Bailey, 2003). This text notes that fully half of people

with mental retardation have an additional psychiatric diagnosis and psychiatric comorbidity seems to increase with severity of mental retardation. The frequency in their assessment seems to be the same in both children and adults.

The conclusion of higher psychopathology in individuals with mental retardation has been consistently reached for decades. For example, after noting the occurrence of severe depressive reactions in people with mental retardation, Gardner (1967) reviewed the then extant research on the incidence of vulnerability of people with mental retardation to severe depressive reactions. His studies revealed little or no definitive data, and he concluded that many studies had obvious defects. He stressed the need for systematic investigation of these issues. In another relatively early study, Berman (1967) noted that depression seemed to be associated with aversive antisocial behavior in some individuals with developmental disabilities. Berman also noted the lack of communication in the literature about the "depressed population" of people with mental retardation. Without attention to this issue, he concluded it was unlikely that progress would be made in the therapeutic realm.

Sovner and Hurley (1983) reviewed 25 published reports regarding the occurrence of affective illness, depression, and mania in people with mental retardation, using DSM-III criteria to assess the validity of both diagnoses. People with mental retardation were found to manifest the full range of affective disorders. Developmentally impaired social functioning and intelligence influenced the clinical presentation, but not the development, of affective symptomatology. They noted that these diagnoses can be made, even in people with severe/profound levels of mental retardation and/or those without language, by extended observations of behavior and vegetative functioning, including levels of motor activity, length and pattern of sleep, appetite and weight, and family historical data.

Investigators during the subsequent 20 years continued to report data that demonstrated higher prevalence. For example, using a community-based sample of 21 adults with mild mental retardation, Prout and Schaefer (1985) administered three self-report measures of depression. When they compared their subjects with the norms for adults without mental retardation, they found significantly higher frequencies of depression for the mildly retarded adults on two of the three measures. Moreover, approximately 50% of the adults with mental retardation had scores that met or exceeded recommended cutoffs for clinical depression. Although this sample was small, and there were undoubtedly many differences between it and the normed samples, the data are consistent with many other investigations and the conclusion that adults with mild mental retardation experience depression at a higher rate than do persons without mental retardation.

Charlot, Doucette, and Mezzacappa (1993) surveyed two groups of institutionalized adults with mental retardation, one group with a prior diagnosis of affective disorder and one with a prior diagnosis of another psychiatric disorder. Seventy-four percent of their subjects had severe or profound mental retardation. Study findings showed that 13% of those in the affective disorder diagnosis group did not meet DSM-III-R criteria for depression or mania, but 20% of the group with other psychiatric disorders did meet criteria. Aggression was a frequent attendant of psychopathology in both groups. These findings supported previous reports that affective disorders may be under diagnosed in this population.

Despite some indications of progress in assessment, other studies still find evidence of substantial methodological problems. For example, Davis, Judd, and Herrman (1997) assessed the reported incidence of depression in adults with intellectual disability, both in community and in hospital-based studies, utilizing visual analogue scale measures of emotion/behavior, the CORE measure of psychomotor disturbance, and self-designed diagnostic criteria. They noted problems with interpreting studies, with the incidence in community data very low, and in hospitals just 5–10%. Outpatient studies exhibited a particularly wide range of findings, with some reporting no cases of depression. They concluded that well-designed studies to assess prevalence and evaluate treatment of depression in this group are urgently needed, particularly the melancholic features of the disturbance. A report from Rojahn and Esbensen (2005) concurs.

With regard to focus on particular age cohorts, Patel, Goldberg, and Moss (1993) examined the prevalence of psychiatric morbidity in 105 people over 50 years of age with mental retardation (called learning disability in the United Kingdom), using the PASS-ADD. When dementia was excluded, they found a prevalence of 11.4% ($n = 12$), most of which were depression and anxiety. Seventy-five percent of these cases were unknown to mental health services. Immediate care staffs were usually aware of the symptoms but were often unaware of their clinical significance. In another study from the United Kingdom the prevalence of psychiatric disorders was compared in aging people with developmental disabilities and that of young adults from the same population. Elderly people had a greater prevalence of psychiatric morbidity than younger controls (68.7% versus 47.9%), with high rates for depression and anxiety disorder. It was concluded that as people with developmental disabilities age, their need for psychiatric services increases.

At the other age extreme, Kroll et al. (2002) assessed the mental health needs of 97 children and adolescent boys in secure care for serious or persistent offending. Twenty-six (27.5%) of the boys had an intelligence quotient (IQ) of

less than 70. The need for psychiatric help was high on admission to a secure unit, with the most frequent disorders being depression and anxiety. There were, of course, high rates of aggression, substance misuse, self-harm, and social, family, and educational problems. The incidence of aggressive behavior and related problems decreased after admission because of the heavy supervision, but the rates of depression and anxiety remained high. The authors noted that many of these boys appeared to require psychiatric and psychological assessment, but this was accomplished for only a small percentage of them.

Very little is known about the occurrence of seasonal affective disorder in people with intellectual disability. Cooke and Thompson (1998) reported two case histories of winter incidence with good response to light therapy and recommended that further studies should be done in this population, including standardized diagnostic criteria and systematic severity of depression scores.

Kirn (2003) noted the frequent occurrence of depression in people with epilepsy, with survey data suggesting that major depression is more prevalent among people with epilepsy than among people with a nonneurologic chronic condition. Also, a PET study of patients with epilepsy detected temporal lobe dysfunction in association with depression. This would seem to be significant for a population of individuals with mental retardation, with their relatively high incidence of associated epilepsy.

Carvill (2001) examined the coexistence of sensory impairments, intellectual disability, and psychiatric problems, and noted that most studies agree that behavioral and developmental problems are more common among children who are blind or visually impaired, and probably among those who have hearing problems. Carvill notes that psychiatric assessment of individuals with sensory impairments is seldom easy and is especially difficult in those with intellectual disability. When no or limited communication skills are present, careful and close observation of behavioral manifestations may be the only means of examination, but the examiner should be aware of the mannerisms and stereotypies often seen in individuals who are blind and deaf–blind.

In sum, based on the consistency of research during the last 50 years, two related conclusions seem reasonable and important. First, it appears that persons with mental retardation are at higher risk for depression than persons without mental retardation. Second, this conclusion of higher prevalence is reached although depression tends to go undetected or unreported in individuals with mental retardation. Both these conclusions lead to the importance of focusing on a variety of diagnostic issues when considering depression in persons with mental retardation.

III. DIAGNOSTIC ISSUES IN PERSONS WITH MENTAL RETARDATION

In children and adolescents, and in persons with mental retardation, the mood in depression may be irritable rather than obviously depressed (Sovner & Fogelman, 1996). Depressed mood in a person with mental retardation may show up as a sad facial expression, withdrawal, vague physical complaints, onset of aggressive symptoms, and/or regression in behavior.

Other possible symptoms of depression include significant weight loss or weight gain, when not dieting, of 5% or more of body weight in 1-month period, or decrease or increase of appetite almost every day. Children with depression may not lose weight but may fail to make expected weight gains for their gains in height (body mass index [weight in kg/height in meters squared] will decrease). Changes in vital functions, such as a decrease in appetite or insomnia, are frequently the first manifestation of a mood disorder in adolescents with mental retardation (Masi, 1998).

People with depression often have sleep disorders—either insomnia or hypersomnia—nearly every day. Sleep disturbances in people with mental retardation who are depressed may show up as disruptive behavior at bedtime or during the night, or excessive sleepiness during daytime activities. Individuals with mental retardation who are also totally blind have a relatively high incidence of sleep disorders due to difficulty with entrainment to a 24-hour day, and, in fact, people who can see no light often have natural sleep rhythms nearer 25 hours than 24 hours (Poindexter, 2002). Therefore, use of sleep disorder as a criterion for depression in an individual who has mental retardation and total blindness probably is not valid in many cases. In another issue related to sleep and depression, a news article (Clinical capsules, 2003) noted that children with epilepsy have a higher than normal rate of sleep apnea, and the apnea appears to be associated with mood and behavior disturbances. Because a significant percentage of children with mental retardation have epilepsy, this may be appropriate to consider for this group. In a survey of 23 children with epilepsy with an average age of 10 years, 19 had abnormal polysomnographs with obstructive hypopneas or apneas that disrupted sleep and caused hypoxia. A statistical analysis showed a clear relation between higher scores on measurements of hyperactivity-impulsive/inattentive behavior and delayed REM sleep onset. Also, higher scores on measures of depression were related to increased length of apnea intervals.

Either psychomotor agitation or psychomotor retardation can be observed nearly every day in depressed persons. In some cultures depression may be masked by different acting-out behaviors representing psychomotor agitation. Both type and frequency are colored by local customs and determinants

(Lesse, 1979). Psychomotor agitation in a person with mental retardation may present as a behavior problem of onset, and psychomotor retardation may present as a change in productivity/performance in school or at work site. People with depression often complain of being tired a lot or having a loss of energy. Benson, Reiss, Smith, and Laman (1985) studied psychosocial correlates of depression in adults with mental retardation and found that depression was associated with informant ratings of poor social skills. Specific social skill deficiencies associated with depressed mood were identified in a group of 45 adults with mental retardation who had mixed or no psychiatric diagnoses by Laman and Reiss (1987). They observed that the subjects with depressed mood were withdrawn and interacted less with others, and that their social interactions were more inappropriate and ineffective. They suggested that some instances of antisocial behavior might be motivated by depressed mood.

Social comparison, the process by which we evaluate ourselves through comparison with others, is a thought process involved with presentation of symptoms of depression. Helsel and Matson (1988) studied a group of 99 adults with mild to severe mental retardation on a variety of measures, and found that depression and social skill measures correlated significantly with each other on self-report and informant reports. Manikam, Matson, Coe, and Hillman (1995) administered self-report measures of depression, general psychopathology, and social skills to adolescents ranging from moderate retardation to above normal intelligence. Adolescents with mental retardation reported more depression and general psychopathology symptoms than did the cognitively normal control group. Adaptive behavior functioned as a moderator variable that mediated the relationship between depression and intellectual functioning—those who could more readily adapt had less severe depressive symptoms. Dagnan and Sandhu (1999) examined the relations among social comparison processes, self-esteem, and depression in 43 people with mild and moderate intellectual disability. In their study, depression was significantly negatively correlated with social comparison on the social attractiveness and group belonging dimensions, and with positive self-esteem. Knowledge of these processes might improve cognitive behavioral interventions for this group of people.

Individuals with depression often complain of feelings of worthlessness or excessive, inappropriate guilt. This guilt may be as extreme as being delusional, and does not, by definition, involve merely self-reproach about being sick. Persons with mental retardation who are depressed may express these feelings in statements such as ''I'm dumb—stupid—no one likes me'' (Sovner, Hurley, & LaBrie, 1982). They may also have decreased ability to think or concentrate, and may appear more indecisive than usual. These symptoms may be reported by the individual or by others.

In persons with mental retardation, cognitive disturbances associated with depression may show up as a decrease in IQ or functional ability when retested, or by change in attention span while performing their usual activities. People with depression, including those with mental retardation, often have recurrent thoughts of death or dying, and may either think a lot about suicide, plan suicide, or make suicide attempts (Hurley, 1998; Patja, Livanainen, Raitasuo, & Lonnqvist, 2001).

Lunsky (2004) found that suicidal ideation and suicidal gestures are evident in adults with mental retardation, including people not receiving mental health services and that psychosocial correlates of suicidality are similar to those in the general population. She based her findings on structured interviews with 98 adults with intellectual disability, with corroborative information from charts and caregivers. One in three individuals reported that they think "life is not worth living" sometimes or a lot. Eleven percent reported previous suicide attempt(s). Twenty-three percent of informants were unaware of the suicidal ideation reported by their family member/client. Individuals who reported suicidal ideation reported more loneliness, stress, anxiety, and depression, and reported feeling they had less social support than others. Patja et al. (2001) noted that most suicide victims had mild mental retardation and were hospitalized for comorbid mental conditions. The suicide rate was about one-third that of the rate in the general population with similar risk factors, and suicide methods were passive. Alcohol was involved in only one case.

Although depression is at least partially of biologic origin, life events certainly still play a part in the development of the condition (Dubovsky, Davies, & Dubovsky, 2003). Ghaziuddin, Alessi, and Greden (1995) studied the role of life events in the occurrence of depression in a group of children with pervasive developmental disorders (PDD) and diagnosed depression as compared with a group of children with PDD and no diagnosed depression. Information was gathered about the occurrence of unpleasant life events in the 12 months prior to the onset of depression and from a comparable time period in the control group. Children with depression experienced significantly more life events, particularly bereavement. The authors strongly urged that future studies include exploration of the role of both biologic factors and environmental stressors in the onset of depression in this population.

As described in this section, persons with mental retardation frequently manifest symptoms of depression similar to those of persons without mental retardation. However, these symptoms are sometimes more difficult to detect because they may be masked by the mental retardation and other disabilities. Therefore, it is especially critical to examine the actual procedures and diagnostic instruments that are commonly used with individuals with mental retardation.

IV. TESTING TECHNIQUES AND INSTRUMENTS

A. Dexamethasone Suppression Test

Unfortunately, no valid laboratory test or other similar diagnostic procedure is available for the diagnosis of depression. Severe depression has been associated with an oversecretion of cortisol, and possibly a loss of the normal daily rhythm variation of cortisol secretion. Assessment of cortisol patterns via the dexamethasone suppression test (DST) has been one of the most extensively investigated neuroendocrine challenge tests used in psychiatric research. Despite many studies, the role of the DST in clinical psychiatry is unclear (Morihisa, Cross, Price, Precioso, & Koontz, 2003). In one study the DST was performed for three groups of institutionalized persons with mental retardation. The groups included persons with symptoms of depression, nondepressed persons with other problematic behavior, and control subjects. Results showed that depressed individuals more frequently, although not significantly, had positive DST tests compared with the other two groups. The group with problematic behaviors did not differ from the control group. Unfortunately, there was no mention made as to whether any of the persons in any group was receiving anticonvulsant drugs, such as phenytoin, carbamazepine, or barbiturates, since these drugs, as well as a number of medical conditions, interfere significantly with the DST (Morihisa et al., 2003).

Mudford et al. (1995) also studied this issue, administering the DST to 40 adults with severe and profound mental retardation. All participants were free from known conditions that may have given misleading results. Of nine participants who showed symptoms possibly indicating depression, the DST results concurred in two cases. However, there appeared to be four or five false-positive DST results, and no consistent behavioral profile was evident for positive DST responders. Mudford et al. note that their data do not suggest usefulness for the DST in this population.

B. Assessment Difficulties

There is evidence of lack of concordance among different measures of depression. Rojahn, Warren, and Ohringer (1994) investigated the association between various depression assessment methods in 38 adults with mild or moderate mental retardation, half of whom had relatively high and the other half relatively low depression screening scores. Utilizing measures that included a standard psychiatric interview, an informant rating scale, and a self-report measure, they found that association among measures was generally low, yielding discordant classification results.

Contributing to the difficulty of differential results with different diagnostic techniques is the influence of level of mental retardation. Meyers (1998) examined the symptomatology of 28 people with severe depression who had moderate to profound levels of mental retardation. More than 70%, 17 of 28, manifested five or more criteria for major depression, as classified by DSM-IV. Mean number of criteria met for major depression was 4.6, with 5.2 for the 15 with moderate mental retardation. This was significantly greater than 3.8 for the 13 with severe and profound mental retardation. Other symptoms included aggression, self-injury, or stripping behavior, which occurred in 40% with moderate and 54% with severe/profound mental retardation. She reported that all of the 28 cases responded to antidepressant drugs and/or mood stabilizers.

Depression is especially difficult to diagnose in persons with mental retardation at the severe or profound levels, primarily because of their inability to verbally express their feelings. Sovner and Hurley (1989) emphasize that a clinical interview alone is rarely diagnostic. They feel that the diagnostic process cannot usually rely solely on the interview, but an assessment process must include detailed behavioral observations collected by family members, other caregivers, and workshop personnel. This is particularly important in assessing sleep disturbances and changes in performance. Psychiatric assessment of individuals with even profound mental retardation can be carried out if longitudinal behavioral data have been collected on the basis of objective measurements of changes in mood and thinking.

Lowry (1998) noted that many factors can interfere with the diagnosis of mood disorders in people with intellectual disabilities. Diagnostic overshadowing is the tendency of mental health clinicians and others to attribute any signs of psychopathology to the person's mental retardation, not a coexisting mental health problem (Reiss & Szyszko, 1983). As a result, treatment programs may be slanted toward providing behavioral treatments and only considering psychiatric care if many of these interventions have failed. Even when access to psychiatric care is readily available, excessive use of antipsychotic medication in this population may mask underlying symptoms of mood disorders.

C. Modified and New Assessment Techniques and Instruments

Although both researchers and clinicians have sometimes used existing procedures and instruments in the diagnosis of persons with mental retardation, especially those functioning in the mild range, there have also been attempts at modification. For example, Clarke and Gomez (1999) assessed the utility of modified DCR-10 criteria in the diagnosis of depression in a

retrospective study of 11 inpatients with intellectual disability. Modifications were made by adding items found in earlier research to be behavioral equivalents of depression in people with severe intellectual disability to DCR-10 criteria. All of the 11 individuals had a remission of symptoms as described by the modified criteria within 5 weeks of starting antidepressant treatment. The authors point out the limitations of their study in that the study group was relatively small, other medications were being administered that may have influenced the mental state, and the difficulty in relating improvements seen with the prescription of an antidepressant medication to an actual diagnosis of depressive disorder.

A number of instruments have been designed over the years to assess depression and other psychiatric disorders in persons with developmental disabilities. Kazdin, Matson, and Senatore (1983) assessed 110 adults with borderline, mild, moderate, or severe mental retardation, utilizing a number of tests. The test battery included modified versions of the Beck Depression Inventory, the Zung Self-Rating Depression Scale, the MMPI depression scale, the Thematic Apperception Test, and the Psychopathology Instrument for Mentally Retarded Adults (PIMRA). Both clinical and direct service personnel rated the individuals on the Hamilton Rating Scale for Depression and an informant version of the PIMRA. They found that the measures correlated significantly with each other and were consistently related to the diagnosis of depression. Matson, Kazdin, and Senatore (1984) examined the psychometric properties of the PIMRA in the same sample as earlier. The psychometric properties of the scale were reviewed and/or evaluated, including internal consistency of items, test–retest reliability, and factor analysis. Balboni, Battagliese, and Pedrabissi (2000) investigated whether the PIMRA (Senatore, Matson, & Kazdin, 1985) could detect specific psychopathological disorders in 652 individuals with mental retardation living in the community or in residential facilities. The sample included people with different levels of mental retardation. As a part of their study they compared 55 persons with anxiety disorders and 49 with depression to 50 control subjects of the same age, intelligence level, and gender ratio but without dual diagnosis. Those with diagnoses had higher factorial scores, both on the overall scale and on the factors specifically related to their disorders.

Reynolds and Baker (1988) examined the psychometric characteristics of the Self-Report Depression Questionnaire (SRDQ), a 32-item self-report measure of depressive symptomatology in persons with mental retardation, in a group of 89 adults living in community-based settings. Although they concluded that their results showed the instrument to be helpful, they also pointed out the instrument should be viewed as one of several assessment perspectives that might provide valuable insight regarding affective status of

people with mental retardation. They cautioned that the SRDQ is not a diagnostic instrument; it does not provide a formal diagnosis of depression, but results may serve as an indicator that further studies should be performed.

Sturmey, Reed, and Corbett (1991) critically reviewed a group of instruments that are used for formal diagnosis from a psychometric perspective. They noted that although some psychometrically sophisticated measures were identified, the area was marked by an absence of important psychometric data for many measures. They concluded that the period of the 1980s had been marked by considerable research into the development of assessments of psychopathology in people with learning difficulties, but much of this research was of a preliminary nature and required further development and refinement. They recommended that further research should be done both for development and for validation of screening procedure and instrument formats and that further studies should include more individuals with severe and profound learning difficulties.

Reiss and Rojahn (1993) studied the relation between aggression and depression in 528 adults, adolescents, and children, who were rated on either the adult or child versions of the Reiss assessment instruments for dual diagnosis (Reiss, 1988; Reiss & Valenti-Hein, 1990). Criterion levels of depression were evident in about four times as many aggressive as nonaggressive subjects. The authors noted that aggression is sometimes associated with syndromes of psychopathology other than depression. They suggested that by evaluating when aggression is and is not associated with various categories of psychopathology, future researchers may be in a position to find ways of improving the effectiveness of clinical treatment—as in prescribing medications specifically for the underlying diagnosis rather than just for the aggression itself. This instrument has also been used internationally. Gustafsson and Sonnander (2002) performed a psychometric evaluation of a Swedish version of the Reiss Screen for Maladaptive Behavior and noted that this instrument can be used as intended by staff members as a primary screening device for the identification of mental health problems in people with intellectual disability in a Swedish setting.

Benavidez and Matson (1993) investigated the performance of 25 adolescents with mild to severe mental retardation and 25 adolescents with normal intelligence on self-report and informant versions of three childhood depression measures, the Children's Depression Inventory (CDI), the Bellevue Index of Depression, and the Reynolds Child Depression Scale. Strong correlations between total measure scores were found. Analyses of variance comparing adolescents with mental retardation and the control group differed significantly only on the Bellevue Index of Depression. The correlations between self-report and informant versions were not high, however, perhaps limiting the usefulness of the instrument.

The consistency of reports about feelings and emotions from individuals with intellectual disability was assessed by Lindsay, Michie, Baty, Smith, and Miller (1994) in a group of 65 people with mild or moderate intellectual disabilities. The test battery, all self-report measures, included the Zung Self-Rating Anxiety Scale, the Zung Depression Inventory, the General Health Questionnaire, and the Eysenck-Withers Personality Test. Results showed a significant amount of convergent validity in the subjects' emotional systems.

Groups of investigators from the United Kingdom have performed a number of studies on the Psychiatric Assessment Schedule for Adults with Developmental Disability (PASS-ADD) and Mini PASS-ADD instruments. Moss, Prosser, Ibbotson, and Goldberg (1996) investigated the ability of the PASS-ADD to detect symptoms that had been found to exist during routine clinical assessment of individuals. They found that this instrument was in good agreement with the information provided by referring psychiatrists. The mini PASS-ADD was developed to enable nonpsychiatrists to accurately recognize clinically significant psychiatric disorders in the people they serve. Prosser et al. (1998) investigated the internal consistency, inter-rater agreement, and validity in relation to clinical opinion, using a sample of 68 people with intellectual disability who were in contact with psychiatric services. The validity results (81% agreement on case recognition) appeared to be sufficiently good to anticipate that this instrument would be helpful. Deb, Thomas, and Bright (2001) used the Mini PASS-ADD to screen for presence of psychiatric disorders in 101 randomly selected adults with intellectual disability and their caregivers. Out of this group 90 had sufficient communicative abilities to make the administration of the test possible. The full PASS-ADD was then used for psychiatric interviews in 20 individuals who were picked up on the original screening by an interviewer blind to the original results. Final diagnosis was made according to ICD-10 criteria. Three individuals were felt to have depression on the initial screening (3.3%) and two were felt to have depression after full testing (2.2%). These figures were roughly comparable to percentages in other studies. Feasibility, reliability, and validity of the Spanish version of PASS-ADD (PAS-ADD-10) were assessed by Gonzalez-Gordon, Salvador-Carulla, Romero, Gonzalez-Saiz, and Romero (2002). They concluded that its overall feasibility was judged adequate by raters, and was considered extremely useful for training.

Utilizing the checklist of challenging behavior (CCB), Jenkins, Rose, and Jones (1998) surveyed the prevalence of challenging behavior in a group of 63 individuals with difficult behavior, utilizing the PIMRA, observer version for comparison. The relationship between nonaggressive behaviors as measured by the CCB and mental health as measured by the PIMRA showed significant overlap, but there was less consistency in the relationship between aggressive behaviors in the CCB and the PIMRA. Thus, there may

be difficulties in discriminating between challenging behavior and behavior resulting from the presence of mental health problems.

Ailey (2000) investigated the reliability and validity of the CDI using 27 adolescents with mild mental retardation attending schools in a large metropolitan area. Scores on the self-report CDI were compared with scores on an observer-completed inventory of psychopathology completed by teachers and other staff, the Reiss Scales for Children's Dual Diagnosis. For female students, statistically significant negative associations were found between CDI scores and scores on the Reiss Scales and its depression subscale. Practically, then, school nurses may find the CDI useful as a self-report screening tool for depression.

Masi, Brovedani, Mucci, and Favilla (2002) examined the concurrent validity of different informant and self-report assessment instruments of psychopathology, both general and specific for anxiety and/or depression, in referred adolescents with mental retardation and also diagnosed with depression and anxiety disorders, according to DSM-IV criteria. Their sample was a consecutively referred group of 50 adolescents with mild and moderate mental retardation, with a mean age of 15.1. They were assessed using standardized assessment techniques, including the PIMRA (informant version, total score, affective, and anxiety subscales), Child Behavior Checklist (CBCL) (informant version, total score, internalizing and externalizing scores, anxiety–depression scale), Zung Self-Rating Depression and Anxiety Scales. PIMRA and CBCL total scores were closely intercorrelated. Anxiety measures were positively correlated, with PIMRA and CBCL total scores, as well as with the internalizing score of CBCL. Depression measures were not correlated; their correlation with more general measures of psychopathology was weak, indicating assessment problems in this group of persons.

Lunsky (2003) assessed the role of gender in the presentation of self-reported depressive symptoms in people with intellectual disability. She based her findings on structured interviews with 99 men and women, with corroborative information provided from caregivers and chart reviews. She found that, overall, women reported higher levels of depression than men, 2:1 female-to-male ratio of diagnosed depression in the general population. Men and women with higher depression scores were more lonely and had higher stress levels than individuals with lower scores. Women with higher scores were more apt to report coming from abusive situations, to have poor social support from family, and to be unemployed when compared to women with lower scores. Similar differences were not found when comparing men with higher and lower depression scores.

Psychometric properties of the Beck Depression Inventory and the Zung Self-Rating Scale in adults with mental retardation were reported by Powell (2003). His findings suggest that these tests have good clinical utility with

adults with mental retardation and that depression in these individuals may be more severe than in others. The Beck Depression Inventory appeared to have clinical validity with this population, but the Zung seemed less useful, since there may be a tendency of these persons to minimize their distress, thus further complicating the diagnostic process.

A screening scale for anxiety, depression, and mood for people with mental retardation (ADAMS) was developed at Ohio State University to facilitate diagnostic efforts (Esbensen, Rojahn, Aman, & Ruedrich, 2003). The authors reported that this scale seems to be a psychometrically sound instrument for screening.

Use of the DSM system (American Psychiatric Association, 1987, 2000) is fraught with many difficulties primarily because of communication problems with the affected individual. Hurley (1996) explored major depressive episodes occurring in individuals with Down syndrome and described a framework for substituting "mental retardation equivalents" (symptoms that indicate the presence of a criterion rather than verbal report) for standard diagnostic criteria. In another study the Marston 30 Symptoms Checklist was used to determine whether or not the notion of "depressive equivalents" could provide a few of the core characteristics needed for the diagnosis of depressive disorders in people with severe/profound intellectual disability. It was found that the first six observable criteria of DSM-III-R, including irritable mood, were sufficient for diagnosing major depression in people with severe/profound intellectual disability. It was noted the particular importance of sleep and appetite disturbances and changes in the level of activity. Presently a companion volume to DSM-IV-TR for use with people with intellectual disability is in the last stages of development by the National Association for the Dually Diagnosed (Mental Health and Mental Retardation) (NADD) and American Psychiatric Association, with expected release sometime in 2006. This volume will include lists of observable equivalents to various DSM-IV-TR diagnostic criteria, including depressive disorders. Further information will be available on the NADD website, www.thenadd.org.

Considerable disagreement often exists between information obtained from affected individuals themselves and other informants (Moss et al., 1996). This clearly indicates the importance of interviewing both affected individuals and others to improve the sensitivity of case detection. As noted earlier, on occasion, individuals may deny symptoms in order to appear more competent and in control.

A published practice guideline for assessment and diagnosis of mental health problems in adults with intellectual disability (Deb, Matthews, Holt, & Bouras, 2001) may be of great help in working out some of the diagnostic difficulties related to depression. The authors clearly describe clinical features, biologic features, cognitive features, appearance, and the psychotic

features of severe cases. They outline guidelines for diagnosis and differential diagnosis and include an extensive evidence-based reference list.

V. PROGRESS AND PROBLEMS

Despite the progress in the almost 40 years since Gardner (1967) and Berman (1967) began to alert the field of developmental disability to the significance of depression in the population being served, much is left to be done. McBrien (2003) reported that the validity of the conceptual frameworks for depression is still in doubt, despite the present widespread acceptance that depression can occur in adults with intellectual disability. She noted that the difficulties encountered in both the assessment and diagnosis of depression have hampered the individual clinician, leaving unanswered questions of prevalence, treatment choice, and outcome.

Difficulties of diagnosis of psychiatric disorders in children and adolescents seem to be exceptionally worrisome. In a study examining factors related to psychiatric hospital readmission among children and adolescents who were wards of a midwestern state, Romansky, Lyons, Lehner, and West (2003) found that children who were readmitted within 3 months of discharge from the index hospitalization were significantly different in a number of ways from the children who were not readmitted. Those in the former group were rated as more learning disabled or developmentally delayed and had received fewer posthospital service hours than the children who were not readmitted. The highest rates of readmission were found among children who lived in congregate care settings before the index hospitalization and those who lived in a rural region. The authors noted that these findings indicate that prevention of readmission among these children must focus on community-based services.

Despite the obvious high incidence and prevalence of depression in populations of individuals with developmental disability and the efficacy of treatment for the condition described in the general population, information is extremely limited regarding treatment results in persons with developmental disability. This clearly seems to be an overlooked important research area, one that should be addressed. Recognition of this neglect prompted the convening of a workshop in late 2001 dealing with research on mental health problems in people with mental retardation. It was held in Rockville, Maryland, and cosponsored by the National Institute of Neurological Disorders and Stroke (NINDS), the National Institute of Child Health and Human Development, the National Institute of Mental Health, the National Institutes of Health Office of Rare Diseases, and the Joseph P. Kennedy Jr. Foundation (NINDS, 2001). The workshop was designed to identify barriers

to the inclusion of people with mental retardation and developmental disabilities in federally funded research in the United States. The goal was to define ways to increase inclusion of people with mental retardation in research in order to promote evidence-based treatment for this population. This certainly seems to be an important step forward in addressing the issues of clinical research in this population.

One area of concern that appears to be virtually unexplored in research is that of response of individuals with mental retardation and depression to various treatment modalities, including, but not limited to, medications. Other potential areas for significant research include use of the NADD/American Psychiatric Association companion to DSM-IV-TR to facilitate diagnosis and the impact of more precise diagnosis and subsequent rational treatment programs on the lives of the people concerned. Other research in the early diagnosis of depression in young children with developmental disabilities also appears important.

In sum, at this stage, it is clear that depression is a major mental health concern for individuals with mental retardation, just as it is for persons who do not have intellectual disabilities. Research needs to focus on epidemiology so that we know the degree to which this population is affected. Achieving the goal of good estimates of incidence and prevalence is dependent on the development of reliable and valid assessment procedures and instruments for individuals with mental retardation. Finally, research on treatment outcomes to advance evidence-based practice must be promoted in order to treat everyone with depressive disorders appropriately and successfully.

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