

SIBLINGS OF INDIVIDUALS WITH AUTISM SPECTRUM DISORDERS ACROSS THE LIFE COURSE

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In this article, we review the literature on siblings of individuals with autism spectrum disorders (ASD) from a lifespan developmental perspective, from infancy through adulthood, focusing on the sibling relationship and sibling well-being. We situate this review within the larger body of research on siblings of individuals with developmental disabilities (DD) across the lifespan. We then consider the genetic aspects of ASDs and their implications for siblings. We conclude that there is an evidence of atypical social and communication development in some siblings of children with an ASD during infancy. During childhood and adolescence, siblings describe both positive and negative aspects of their sibling relationship and there is some evidence that siblings of children with an ASD may be at heightened risk for social and behavioral adjustment problems. The limited research on adulthood suggests that lack of closeness in the sibling relationship and social and emotional difficulties may continue. We encourage more attention focused on developmental issues, specifically with respect to samples in narrower age groups and in longitudinal research. Finally, we note the variability in sibling outcomes, and suggest further examination of potential moderating and mediating factors, including genetic predispositions.

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MRDD Research Reviews 2007;13:313–320.

Key Words: siblings; lifespan; sibling relationship; well-being

In this article, we provide a developmental perspective on the sibling relationship and well-being of siblings of individuals with autism spectrum disorders (ASD). We use the term ASD to include children with autism, Asperger syndrome, and pervasive developmental disorders (PDD). All of these disorders share impairments in communication, social reciprocity, and the presence of restricted and repetitive behaviors [APA, 2000]. We organize this review from a developmental perspective, discussing the literature on two topics, sibling relationships and sibling well-being, within each life stage, from infancy through adulthood. The term “sibling relationship” includes the physical actions and interactions between siblings, as well as the more subjective, cognitive, and affective components of the relationship [Cicirelli, 1995]. Research, therefore, has focused on the dimensions of companionship, affect, and caregiving in the sibling relationships. The term “sibling well-being” includes outcomes that represent well-being according to the age of the sibling, such as social engagement and communication in infancy, behavioral adjustment during childhood, and mental health in adolescence and adulthood.

Among all human relationships, sibling relationships typically last the longest. The sibling relationship is also unique in that it is ascribed rather than achieved, and typically one of relative egalitarianism [Cicirelli, 1995]. Nurture and conflict in the sibling relationship provide siblings with experiences that foster the development of emotional understanding, self-regulation, and a sense of belonging and comfort [Brody, 2004]. Siblings begin as play partners and sources of support through intimate daily contact during childhood. During adolescence and young adulthood, siblings experience decreased contact and intimacy as they establish their independent lives. Intimacy in the sibling relationship during adulthood is maintained through telephone and other forms of communication and through periodic visits. Thus, the sibling relationship in adulthood is more susceptible to change due to factors in the individual siblings’ lives [Cicirelli, 1995]. Finally, during middle and later adulthood, sibling relationships often become more salient, especially when other family and social supports are less available [White, 2001].

The literature on siblings in the general population indicates that sibling relationships show considerable continuity throughout childhood and adolescence [Dunn et al., 1994], although more recent research has focused on the personal, interpersonal, and ecological variables that influence sibling relationships [Kramer and Kowal, 2005]. Overall, girls tend to report more affection and intimacy in their sibling relationships than boys [Akiyama et al., 1996; Kim et al., 2006], although the sex constellation may also make a difference. In a recent study, sibling intimacy was relatively stable in same-sex dyads from childhood through adolescence, but in mixed-sex dyads intimacy declined from middle childhood through early adolescence and then increased in middle adolescence [Kim et al., 2006]. With respect to parental influences, considerable

Grant sponsor: National Institute on Aging; Grant number: R01 AG08768; Grant sponsor: National Institute on Child Health and Human Development; Grant numbers: P30 HD03352, R03 HD039185; Grant sponsor: Graduate School of the University of Wisconsin–Madison.

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Received 20 August 2007; Accepted 22 August 2007

Published online in Wiley InterScience (www.interscience.wiley.com).

DOI: 10.1002/mrdd.20171

research supports a positive association between positivity in the parent–child relationship and positive affect and prosocial behavior in the sibling relationship [Brody, 1998], although the relative importance of the mother–child versus the father–child relationship remains debated [Kim et al., 2006]. Furthermore, children and adolescents who perceive that they receive less warm and more negative treatment from their parents than their siblings have greater emotional and behavioral difficulties, especially if they perceive that treatment to be unjust [Brody, 2004].

With these life course patterns and influences on the sibling relationship in mind, one additional factor that can influence the life course trajectory of the sibling relationship is the presence of disability in a sibling. Considerable research has examined how the sibling relationship is affected, and whether the well-being of the non-disabled sibling is impacted, when one sibling has a developmental disability (DD) other than an ASD [see Rossiter and Sharpe, 2001, for a review], but comparatively little research has focused on the sibling relationship when one sibling has an ASD. Therefore, we begin our review by summarizing the research on sibling relationships in DDs other than ASDs. This research is helpful in understanding how the presence of cognitive impairment affects sibling well-being and sibling relationships. Most individuals with ASDs have cognitive limitations, but they also have social and behavioral characteristics that might challenge the sibling relationship, such as impairment in social reciprocity and the presence of rigid behaviors [APA, 2000]. Thus, in understanding how ASDs affect the sibling relationship, it is helpful to contextualize our review within the larger body of research on siblings of individuals with DD other than ASDs.

SIBLINGS OF INDIVIDUALS WITH DEVELOPMENTAL DISABILITIES

In reviewing the research on sibling relationships when one sibling has a DD, Stoneman [2001, 2005] concluded that there are high levels of interaction when one sibling has a DD, especially during childhood, although some of this interaction is likely due to the expanded childcare roles that non-disabled siblings, especially sisters, often play. A meta-analytic review of the effects on the sibling relationship and sibling well-being came to a more negative conclu-

sion [Rossiter and Sharpe, 2001]. On the basis of 25 studies on siblings of individuals with DD, Rossiter and Sharpe concluded that there was a statistically significant but small negative effect on the psychological functioning of these siblings (depression, anxiety, and behavioral adjustment in terms of internalizing and externalizing behaviors). The effect varied at different stages of the life. The effects for siblings during childhood and adolescence were negative, although only the effect size for childhood was significant. The effect size for siblings during adulthood was positive but not significantly different from zero. Rossiter and Sharpe comment on these age-related findings, pointing out that only two studies were included in the analysis regarding adult siblings, and pondering whether the findings reflect effects due to self-report (which tend to be more positive than parental reports across ages) versus parental report (which is more common in studies on children and adolescents). Rossiter and Sharpe [2001] also speculate that the effect on siblings during adulthood may be less negative because of the decreased influence of the birth family on adults and also possibly because of the development of more mature and effective coping strategies. These observations of age-related differences in the effects of the sibling relationship underscore the importance of a life course perspective when assessing the impact of a sibling with a disability.

Our own research substantiates that sibling relationships are relatively positive during adulthood, and also provides considerable information regarding the factors that affect sibling relationships during this life stage [see Seltzer et al., 2005, for a recent review]. We have found that non-disabled siblings are often considered a member of the social support network of adult brothers and sisters with a DD [Krauss et al., 1992], and that the sibling who is most involved has a considerable amount of contact and relatively positive relationships with the brother or sister [Krauss et al., 1996], especially in comparison with siblings who have a brother or sister with mental illness (MI) [Seltzer et al., 1997]. Siblings of adults with DD generally provide higher levels of emotional rather than instrumental support [Seltzer et al., 1991], and provide more emotional support than siblings of adults with MI [Greenberg et al., 1999].

We have also found that sibling gender has important effects on sibling

caregiving and involvement. The most involved sibling is likely to be a sister [Seltzer et al., 1991], and she is likely to have an equally close relationship if she has a sister or brother with DD [Orsmond and Seltzer, 2000]. Brothers' sibling relationships are conditioned by the gender of their sibling; brothers with a brother with DD have closer relationships than brothers with a sister with DD. Furthermore, there is considerable variability regarding expectations of future caregiving roles [Krauss et al., 1996]. Although almost 20% of siblings expected to have their sibling with DD live with them in the future [Krauss et al., 1996], when followed up at a later point in time only 10% of siblings actually coresided [Freedman et al., 1997]. Finally, we found that when the sibling with DD moves out of the parental home to a community residential living arrangement, non-disabled siblings tend to be less worried about their brother or sister's future and have increased contact with their sibling, when compared with siblings when the brother or sister continues to live at home [Seltzer et al., 2001]. Thus, our research indicates that during adulthood, sibling relationships are relatively positive but are more likely to involve emotional support than direct caregiving. Furthermore, gender of both siblings and living arrangement of the brother or sister with DD (in the parental home, elsewhere) are important factors contributing to sibling closeness.

We now turn to the literature on siblings of individuals with ASDs, focusing on the sibling relationship and sibling well-being. However, first we begin with a brief review of the genetic basis of ASDs.

THE GENETIC BASIS OF ASDS

We need to consider both genetic and environmental effects when examining the impact of ASDs on siblings. There is a general consensus that there is a genetic basis to autism [see Rutter, 2000; Wassink et al., 2004; for reviews]. Siblings of individuals with an ASD are at greater risk for ASDs themselves as well as a range of related impairments. This is termed the “broader autism phenotype” (BAP) [Baurminger and Yirmiya, 2001]. Concordance rates for monozygotic twins range from 60 to 96% [Bailey et al., 1995; Ritvo, 1985] and from 0 to 24% for dizygotic twins [Bailey et al., 1995; Ritvo, 1985]. Recurrence rates (meaning the occurrence of an ASD in another immediate family member) for ASDs in siblings have con-

sistently been reported in the 3–5% range [Micali et al., 2004]. Therefore, any consideration of the impact on a non-diagnosed sibling who has a brother or sister with an ASD must take the BAP into account when interpreting the data, and acknowledge its potential to increase the vulnerability of siblings who do not have an ASD. Thus, it is ultimately not possible to fully differentiate BAP (i.e., genetic) factors from the reactive effects of growing up with a sibling with an ASD (i.e., environmental factors).

PRIOR REVIEWS OF THE WELL-BEING OF BROTHERS AND SISTERS OF INDIVIDUALS WITH AN ASD

There are no prior reviews of research on the sibling relationship when one sibling has an ASD. However, there have been prior reviews on the well-being of such siblings. The most recent review on the cognitive and psychiatric outcomes of siblings of individuals with an ASD is a meta-analysis and methodological critique of 18 studies by Yirmiya et al. [2001], which concluded that there is insufficient data to support the notion that siblings of children with an ASD are at increased risk for negative cognitive and psychiatric outcomes, with the exception of the increased genetic risk of ASDs (including the BAP). They note several limitations in the literature that preclude firm conclusions: the relatively small number of studies that are comparable to one another; the small samples studied, which makes it difficult to observe recurrence if rates are low; and the lack of comparison groups that also have genetic liabilities such as siblings of individuals with learning disabilities or psychiatric disorders.

Other reviews include a summary of the research by Bauminger and Yirmiya [2001] and a comprehensive review by Lainhart [1999] on the presence of psychiatric problems in siblings (and other relatives) of individuals with an ASD. Lainhart's review concluded that first degree-relatives of individuals with an autism (including siblings) are at increased risk of major depression, but the findings for other psychiatric disorders are mixed. The current review differs from these prior ones in that we examine both the sibling relationship and sibling well-being, and attempt to understand sibling outcomes in siblings of individuals with an ASD according to life stage, from infancy through adulthood.

SIBLINGS OF INDIVIDUALS WITH AN ASD IN INFANCY

A relatively recent development in the research literature on siblings of individuals with an ASD has been a focus on infant siblings. Although a primary purpose of this research is to identify atypical patterns of development that may be early markers for ASDs, these studies also provide information regarding the development of such siblings during the early years of life. In line with the skills of infants, these studies have focused on the development of social engagement and communication [Goldberg et al., 2005; Zwaigenbaum et al., 2005; Mitchell et al., 2006; Yirmiya et al., 2006].

At a very young age, 4-month-old siblings of children with autism did not differ from siblings of typically developing (TD) children on most measures of early social engagement, such as infant gaze, positive or negative affect when the mother stopped interacting with the infant, and their response to their mother when she started interacting with them again. However, their interactions with their mothers were less synchronous when they began the interaction, and they showed more neutral affect and were less likely to cry when their mother stopped interacting with them [Yirmiya et al., 2006]. Interestingly, more infant siblings of children with autism responded to their name being called than siblings of TD children. In another study, at 12 months of age, atypical language skills, imitation, and responsiveness to social and nonsocial stimuli were observed, which were later found to be related to the development of an ASD in some siblings [Zwaigenbaum et al., 2005].

At age 14 months, Yirmiya et al. [2006] found no differences between siblings of children with an ASD when compared with siblings of TD children on cognitive or motor development, but siblings of children with an ASD showed delayed requesting behaviors. Goldberg et al. [2005] reported similar findings regarding language and early social communication behaviors. At 14–19 months, infant siblings of children with an ASD showed significantly less eye contact, fewer gestures, less turn-taking, less pointing and showing behaviors, and fewer requesting behaviors than siblings of TD children. Goldberg and colleagues also found that siblings not later diagnosed with an ASD performed more similarly to siblings later diagnosed with an ASD on these social and communication behaviors

than they did in comparison to siblings of TD children.

In contrast, Mitchell et al. [2006] found that at 12 months, siblings not later diagnosed with an ASD were similar to siblings of TD children on language and communication skills, but that by 18 months siblings of children with an ASD produced fewer play-related gestures than the TD group. Differences across these studies on infant siblings of children with an ASD likely arise because of the small samples, which may not be representative of the population, and differences in the types of measures used.

These infant sibling studies suggest some evidence of difficulties in social and communication behavior at a young age, even in siblings who are not later diagnosed with an ASD. The authors of these infant sibling studies argue that these developmental differences are due to genetic liability, although Yirmiya et al. [2006] acknowledge that the findings may be due in part to environmental effects. Social relationships and mother–child interactions in the family could be affected by the presence of the sibling with an ASD or the presence of BAP characteristics in mothers, independent of the BAP characteristics in siblings.

SIBLINGS OF INDIVIDUALS WITH AN ASD IN CHILDHOOD AND ADOLESCENCE

Sibling Relationships

During childhood, siblings of children with an ASD spend a considerable amount of time with their brother or sister. In an observational study of siblings ages 2–12, siblings spent on average 40 minutes out of every hour together when observed at home, and engaged in a variety of activities together, including pretend and rough and tumble play [Knott et al., 1995]. However, the siblings of children with autism spent less time together than siblings of children with Down syndrome (DS), and their brother or sister with autism responded less positively to initiations by the sibling. Another study using self-reports from siblings found that siblings of brothers and sisters with autism ages 7–12 expressed primarily positive feelings about their sibling relationship [Rivers and Stoneman, 2003].

The majority of studies on siblings of children with autism have grouped together siblings spanning the childhood and adolescent years, obscuring differences between these stages of

life. These studies agree that siblings describe the relationship with the brother or sister with autism in positive terms, although some negative aspects are noted, such as embarrassment or and disruptive behavior by the brother or sister with an ASD. Several studies show the similarity between sibling relationships when a sibling has an ASD when compared with a DD other than an ASD in terms of qualitative descriptions of the sibling relationship by siblings [Fisman et al., 1996, 2000; Pilowsky et al., 2004].

Roeyers and Mycke [1995] found no differences between siblings ages 8 and 14 of children with autism when compared with siblings of children with DD, but siblings of children with autism reported more acceptance and less verbal aggression compared with siblings of TD children. In a study by Kaminsky and Dewey [2001], siblings ages 8–18 of children with autism reported greater admiration of and less competition and conflict with their brother or sister than siblings of TD children, and were similar to siblings of children with DS in these aspects of the sibling relationship. However, they reported lower levels of closeness and intimacy than siblings of children with DS.

In terms of activities that siblings do together, in a qualitative study, almost half of the siblings ages 7–20 mentioned the good nature of their brother or sister with autism and that they played and had fun together, although feelings of embarrassment were also mentioned as well as concerns for the future of their brother or sister [Mascha and Boucher, 2006]. The most frequent activities they engaged in were playing together, watching television, and spending time outside.

Negative aspects of the relationship have also been identified by siblings. Some siblings reported greater feelings of embarrassment than siblings of children with DD or no disability [Roeyers and Mycke, 1995]. In a study by Ross and Cuskelly [2006], 84% of siblings (ages 8–15) reported aggression by the brother or sister with autism in their sibling interactions, which provoked feelings of anger. Bågenholm and Gillberg [1991] found that siblings ages 5–20 of children with autism played and interacted less with their brother or sister than siblings who had a brother or sister with DD. Furthermore, siblings of children with autism reported less favorable attitudes toward their brother or sister and saw their brother or sister's role in the family more negatively than

siblings who had a brother or sister with DD. More than half of the siblings of children with autism were unable to explain their brother or sister's disability, and one-third said they could only talk with someone outside their family about their brother or sister's condition. They also reported problems with destructive behavior in their brother or sister, and were concerned about their brother or sister's future.

In summary, siblings during childhood and adolescence describe positive aspects of their sibling relationship, including siblings as activity partners, greater admiration, and less competition and conflict. There are also significant indications, however, that sibling relationships are less close when the brother or sister has an ASD than when they have DS or a DD other than an ASD. Siblings also are concerned about embarrassment, destructive behavior, social isolation, and their brother or sister's future.

Sibling Well-Being

Studies examining sibling well-being have either included siblings spanning the childhood and adolescent years, or only focused on adolescent siblings. Notably, no studies of sibling well-being specifically during childhood have been conducted.

From a genetics perspective, several researchers have studied social and emotional skills associated with the BAP. Dorris et al. [2004] found that siblings ages 7–17 of children with Asperger syndrome showed impairment in facial emotion recognition compared with siblings of TD children. Constantino et al. [2006] found evidence for subtle social impairments in male siblings ages 4–18 of children with autism. Using his Social Responsiveness Scale, siblings from multiple-incidence families were rated as having the highest degree of social impairment, followed by siblings with only one brother or sister with autism. Siblings of children with a variety of other psychological or behavioral disabilities had the lowest scores.

Further evidence of social difficulties comes from a study by Bågenholm and Gillberg [1991] wherein 35% of the siblings age 5–20 reported feeling lonely and that they had no friends. In contrast, sibling of children with mental retardation in this study did not report the same degree of loneliness and siblings of children without disabilities reported no feelings of being lonely. Other researchers have found that siblings whose brother or sister had ASD

are equally or more socially competent than siblings of children with DS or no disability [Gold, 1993; Rodrigue et al., 1993; Kaminsky and Dewey, 2002; Verte et al., 2003]

The research examining psychological well-being in this age range has focused on the presence of clinical psychiatric diagnosis as well as behavioral adjustment. Pilowsky et al. [2004] found no differences in rates of clinical diagnoses (including ADHD, ODD, and anxiety) between siblings ages 6–15 of children with autism, MR of unknown genetic etiology, and developmental language disorder. Gold [1993] studied siblings of boys with autism and found that they scored significantly higher on a screening measure of clinical depression than siblings of TD children.

A relatively large number of studies have examined sibling behavioral adjustment during childhood through adolescence using parental report, frequently examining internalizing and externalizing types of behavior problems with the Achenbach Child Behavior Checklist [CBCL; Achenbach and Rescorla, 2001]. Studies of children and adolescents tend to report higher levels of adjustment problems in siblings of children with an ASD than in siblings of children with no disability, with mixed findings as to how siblings of children with an ASD compare with siblings of children with other DDs such as DS.

Smith and Perry [2005] reported that 36% of siblings ages 6–16 of a brother or sister with an ASD had borderline to clinically significant internalizing behavior problems and 20% had externalizing problems. Similarly, Verte et al. [2003] reported that siblings ages 6–16 of high functioning children with autism had significantly more internalizing and externalizing behavior problems than siblings of TD children. Fisman et al. [1996] reported that siblings ages 8–16 of children with PDD had more internalizing behavior problems according to both parent and teacher report on the CBCL than siblings of children with DS or no disability. Externalizing behavior problems were elevated only according to parent report. Three years later, siblings of children with PDD no longer scored higher in internalizing behaviors according to parental report, but continued to exhibit higher levels of externalizing behaviors [Fisman et al., 2000].

Similarly, Rodrigue et al. [1993] found that siblings with a mean age of 9 years had significantly more internalizing and externalizing behavior prob-

lems than siblings of TD children, but not more than siblings of children with DS. Hastings [2003a,b] reported on the adjustment of siblings ranging in age from 4 to 16 years using the Strengths and Difficulties Questionnaire (SDQ). Compared to normative data, the siblings of children with autism had more peer problems, more overall adjustment problems, and lower levels of prosocial behavior. Bågenholm and Gillberg [1991] used the Rutter scales with siblings ages 5–20 and found that siblings of children with autism scored higher on the inattention/hyperactivity and conduct problems scales (externalizing types of behavior problems) than did siblings of TD children, but similar to siblings of children with DD (including DS). Almost all of these authors emphasize that although scores on the measures of behavior adjustment may be elevated in siblings of children with an ASD, they are generally within the non-clinical range.

In several other studies, siblings of children with an ASD were not found to be at greater risk of adjustment problems. Ferrari [1984] found no differences in behavior problems on the CBCL between siblings ages 6–13 of brothers with PDD, diabetes, and no health or developmental problems. Both Gold [1993] and Kaminsky and Dewey [2002] found no differences in adjustment problems (as measured by the CBCL) between siblings ages 7–18 of children with autism and siblings of TD children (in both studies) and DS [Kaminsky and Dewey, 2002]. Two additional studies did not use comparison groups, but examined adjustment problems in siblings in comparison to normative data. Mates [1990] found that home, school, and academic adjustment in siblings ages 5–17 of children with autism were not significantly different from normative samples. Ross and Cuskelly [2006] reported that mean scores of internalizing and externalizing behavior problems as measured by the CBCL by parent report were within the non-clinical range for a sample of siblings ages 8–15. Forty percent of the siblings were experiencing adjustment problems. With a larger sample of families participating in ABA therapy, Hastings [2003b] reported that siblings were reported as having fewer problems on the SDQ compared with the normative data.

Positive outcomes have been consistently noted in the domains of sibling self-concept and self-competence. Several authors have found that siblings of

children with an ASD report positive self-concept similar to norms [Mates, 1990] or comparison groups of siblings of TD children or children with DD or DS [Ferrari, 1984; Bågenholm and Gillberg, 1991; Rodrige et al., 1993]. Similarly, Verte et al. [2003] found that self-concept did not differ for younger siblings ages 6–11, but that adolescent female siblings age 12–16 of children with high-functioning autism (HFA) had a more positive self-concept than adolescent females of TD siblings.

Some of these studies spanning the childhood and adolescent years have examined age-related differences in sibling well-being. These studies are mixed in their findings. Gold [1993] found that siblings ages 7–12 reported significantly fewer depressive symptoms than siblings ages 13–17. Similarly, Rodrigue et al. [1993] reported that siblings younger in age (mean age was 9.11 years) had fewer internalizing and externalizing behavior problems than siblings older in age. In contrast, Hastings [2003b] found that older siblings (late childhood and adolescence) had higher prosocial behavior scores than younger children, and Verte et al. [2003] found that siblings ages 6–11 who had a brother or sister with HFA had more internalizing and externalizing behavior problems than siblings ages 12–16.

In our own research, we have focused on the psychological well-being of siblings during adolescence [ages 12–18; Orsmond and Seltzer, 2006]. With a sample of 57 siblings, just over one-third (36%) of siblings reported depressive symptoms at or above the clinical cut-off score of 16 on the Center for Epidemiological Studies Depression Scale [Radloff, 1977] and 8.5% of siblings reported clinically relevant anxiety symptoms on the Reynolds Child Manifest Anxiety Scale [Reynolds and Richmond, 1978]. Notably, these levels of depressive and anxiety symptoms are comparable to levels found in adolescent community samples [Radloff, 1991; Dierker et al., 2001].

To summarize, the literature yields mixed results regarding whether there is a negative impact on siblings when they have a brother or sister with an ASD, perhaps both due to a genetic (i.e., the BAP) and an environmental influence. There are also some indications that siblings are at greater risk of adjustment problems during childhood compared with siblings of TD children, with the risk possibly increasing during adolescence, although study findings are not consistent. The variability in these

findings is possibly due to small sample sizes, differences in age ranges of siblings who participated, and different measures used to examine well-being and adjustment. The research, however, has been consistent in noting that self-concept and self-competence do not appear to be negatively affected in siblings who have a brother or sister with an ASD and some studies observe comparable levels of depression and anxiety as age peers without a sibling with a disability.

SIBLINGS OF INDIVIDUALS WITH AN ASD IN ADULTHOOD

Sibling Relationships

There are few studies examining the sibling relationship during adulthood when one sibling has an ASD. Holmes and Carr [1991] included siblings ages 16 years and older who had an adult brother or sister with autism or DS. The two groups did not differ in their involvement in direct caregiving (and therefore the results were presented together for both groups). Over one-third of siblings helped their brother or sister during mealtimes. Both siblings who lived at home and those who lived away from the parental home often watched their brother or sister when their parents went out. Importantly, the mothers recognized siblings as a source of help in caring for the family member with autism or DS.

In our recent research, we have examined how sibling relationships are affected by an ASD during adulthood [Orsmond and Seltzer, 2007]. Compared with siblings of adults with DS, siblings of adults with an ASD had less contact with their brother or sister, reported lower levels of positive affect in the relationship, felt more pessimistic about their brother or sister's future, and were more likely to report that their relationships with their parents had been affected. Different factors were associated with the sibling relationship for each group. For siblings of adults with an ASD, a closer sibling relationship was observed when the sibling had lower education levels, lived closer to the brother or sister with an ASD, used more problem-focused coping strategies, and when his or her brother or sister with an ASD had higher levels of functional independence. In interpreting these findings, we speculate that greater similarity among siblings (i.e., in education level and functional abilities) might promote sibling closeness.

To summarize, there is only a little research describing sibling relationships during adulthood when one sibling has an ASD. The available data suggest relatively lower levels of engagement in the sibling relationship during adulthood, especially compared with siblings of individuals with DS. Given the increasing prevalence rates [Rice, 2007] and the large number of individuals with ASDs who will be transitioning to adulthood over the next several years, there is a great need for more research. At this point, we have only a limited understanding of the extent to which siblings are involved in their brother or sister's lives, and whether or not they are positioned to take on increased caregiving responsibilities as their parents age and ultimately die.

Sibling Well-Being

Similar to the research on sibling relationships in adulthood, only a few studies have examined sibling well-being exclusively in adults. Some studies have included siblings of various ages, spanning childhood through adulthood, and these are included in this section.

There is some evidence of social and emotional difficulties in siblings of individuals with an ASD during adulthood. Smalley and Asarnow [1990] found that siblings (mean age of 25) of individuals with autism showed greater impairments in facial emotion recognition than siblings of individuals with no disability, supporting impairments in line with the BAP. Similarly, Piven et al. [1997] reported higher rates of social impairments (33%) in siblings who had more than one brother or sister with autism compared with siblings of children with a brother or sister with DS (0%).

In another study, Piven et al. [1989] used family history methods with 67 adult siblings (average age of 33 years) and found that 15% of the sample had received treatment for depression or mania, which they reported to be considerably higher than rates reported in epidemiological studies (roughly 3% of the general population). Other studies also suggest higher rates of affective disorder than in the general population [DeLong and Dwyer, 1988; Smalley et al., 1995].

In our own research, we examined the incidence of diagnosed disabilities in a sample of 117 siblings (mean age of 21 years) in our longitudinal study of families of adolescents and adults with autism [Seltzer et al., 2003]. The most frequent disabilities in siblings

were attention and hyperactivity disorders (4.6%), ASDs (2.4%), and psychiatric disorders (2.1%) [Orsmond et al., 2007]. Only ASDs were elevated compared with the general population. We also found that the presence of disability in multiple siblings was associated with poorer maternal well-being [Orsmond et al., 2007].

To summarize, as in other age groups, the research presents mixed results as to whether siblings of individuals with an ASD have poorer well-being during adulthood as compared with their age peers. Although some researchers have found elevated rates of psychological distress, we have not found such in our research. These differences are likely attributable to different samples (e.g., community, clinical, multiple incidence families) and also differences in the measures and methods employed.

SUMMARY AND AGENDA FOR FUTURE RESEARCH

Reviewing the literature on siblings of individuals with an ASD from a developmental perspective helps us to understand at which points in the life course siblings may be at greater risk, and how the sibling relationship and sibling well-being may change over time. With regard to this latter point, there are very few longitudinal studies, and thus inferences about age-related differences based on cross-sectional comparisons must remain tentative. Some of the patterns observed at different life stages may be due to cohort effects. For example, the adult siblings reported in the studies to date grew up in a time when there were relatively fewer services and less developed educational programs for children with autism. Their experiences were likely different from those of younger siblings in recent studies. Thus, our first suggestion is for more longitudinal research. Cross-sequential designs may also be fruitful as they control for both time and cohort effects. Using such approaches will be the only way to truly understand how the sibling relationship and sibling well-being change over time when one sibling has an ASD. Understanding sibling relationships from a developmental perspective is complicated, however, as one needs to take into account and measure developmental changes in the sibling, his or her brother or sister with the disability, and other life course changes in the family.

Second, understanding the development of siblings of individuals with

an ASD is complicated by the inclusion of a wide age range in the samples of many studies. The risk of psychological and psychiatric problems generally increases in late adolescence and early adulthood [APA, 2000], and thus including siblings in samples ranging from childhood through adulthood may obscure true age-specific prevalence rates. Furthermore, there is considerable literature on the changes in sibling and family relationships from childhood through adulthood [White, 2001; Parke, 2004]. Thus, we recommend that future research on siblings should take a life course developmental approach, considering the context of the life stage of the sibling and the unique risks for the sibling relationship and sibling well-being associated with each life stage.

Third, another challenge to understanding continuities and discontinuities in the sibling relationship has to do with measurement. The methodologies used in the studies are obviously dependent upon the skills and development of the sibling. The research on siblings in infancy and young childhood is dependent upon observational measures and parent report. Self-report is often employed during adolescence or adulthood. As Rossiter and Sharpe [2001] observed, the findings varied by the measurement method employed. Moreover, the siblings' own understanding and ability to evaluate aspects of the sibling relationship and their well-being change over time.

Fourth, the variability noted in both the sibling relationship and sibling well-being at each life stage suggests that we need to pay greater attention to understanding factors that put siblings at greater risk or, alternatively, that serve as protective factors across the life course. Some of the inconsistent findings across studies may be due to different sample characteristics, methods for data collection, and measures. Attempts towards greater consistency in methods across studies will allow for more appropriate direct comparisons and more accurate information regarding the well-being and sibling relationship when one sibling has an ASD.

Moreover, this variability in findings suggests that there may be unmeasured factors that are influencing the sibling relationship and well-being. Paralleling the research on sibling relationships in TD individuals, the research on the impact of disability on the siblings has moved from descriptive to explanatory research. Researchers have begun to examine the important influences of

factors such as marital stress and coping on the sibling relationship when one sibling has autism [e.g., Rivers and Stoneman, 2003; Ross and Cuskelly, 2006]. Further research along these lines will help us better understand the influences that might be altered through support or intervention to help siblings.

Another factor that has the potential to influence sibling well-being and the sibling relationship is the genetic predisposition siblings may have to the BAP. As Bauminger and Yirmiya [2001] suggest, a genetic predisposition (BAP) may operate as a diathesis (vulnerability), which under certain stressful environmental conditions, may result in a particular outcome. In our research, we have conducted analyses investigating the interplay between genetic vulnerability (expressed as the BAP in siblings or the mother, family history of ASDs) and stressful family situations (e.g., behavior problems in the brother or sister with ASD) [Orsmond and Seltzer, 2006]. We found adolescent siblings with a family history of ASD reported significantly more depressive symptoms than those without a family history of ASD. Siblings also were at greater risk of depressive and anxiety symptoms when they had high BAP characteristics and experienced more life events in the past year. Finally, siblings were also at elevated risk for greater anxiety symptoms when their mothers reported BAP characteristics and higher depressive symptoms (which could act as both a genetic liability and an environmental stress). More research along these lines, examining how biological and environmental factors interact to influence sibling well-being and the sibling relationship, will be fruitful in terms of identifying siblings at risk for negative outcomes and for developing interventions that promote individual and family well-being.

Other mediating or moderating factors that have been explored in the research reviewed in this article include sibling characteristics (such as gender, birth order, social support, coping, self-concept, and social competence), characteristics of the brother or sister with an ASD (such as age, gender, level of functioning), and other family characteristics (such as family size, socioeconomic status, the parental marital relationship, parental preferential treatment of siblings). For almost all of these factors there are contradictory findings. Furthermore, it is impossible to determine, at this point, which factors are more important at which life stages.

Moreover, studies of siblings of individuals with an ASD have been conducted almost exclusively with Caucasian families in the Western countries. More attention paid to cultural influences will be important in understanding the sibling relationship and sibling well-being in this context.

Finally, as Bauminger and Yirmiya [2001] point out, a major impediment to reaching firm conclusions about the sibling relationship and sibling well-being when one sibling has an ASD is the use of different comparison groups in different studies. Many studies compare siblings of individuals with an ASD with siblings of individuals with DS, which controls for environmental effects (e.g., the stress of growing up with a sibling with a disability). Few studies have used comparison groups with known complex genetic liabilities, such learning disabilities or psychiatric conditions. In addition, few studies have sought to identify variables and mechanisms that put siblings of individuals with an ASD at elevated risk.

To conclude, the limited literature about siblings of individuals with an ASD across the life course suggests an elevated risk of negative well-being outcomes and poorer relationships. However, this pattern is tempered by findings suggesting positive relationship quality. Nevertheless, the well-being outcomes are danger signals that should be taken seriously, as these siblings are in line to inherit the responsibility for long-term family caregiving from their parents. These results call for not only more research but also more clinical and support services for siblings of individuals with an ASD. ■

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