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The Siblings of Individuals with Mental Retardation: A Quantitative Integration of the Literature

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The sibling relationship has recently become the focus of much research in developmental psychology. The family system perspective implies the presence of a sibling with mental retardation will impact on the psychological development and functioning of their typically developing siblings. Past reviews of the literature have found this impact to be negative but there is the suggestion of positive consequences to having a sibling with mental retardation. The present meta-analysis sought to quantitatively integrate 25 studies and 79 effect sizes from the literature on the siblings of individuals with mental retardation. A small negative effect for having a sibling with mental retardation was discovered that could not be attributed to a publication bias or some other artifact. This negative effect was greatest for direct observation measures, measures of psychological functioning, especially depression, and for children. Limitations to this meta-analysis and directions for future research are discussed.

KEY WORDS: siblings; mental retardation; meta-analysis; review.

Only recently has the significant role siblings play in the psychological and social development of their brothers and sisters been recognized and investigated by researchers (e.g., Banks & Kahn, 1982; Graham-Bermann & Cutler, 1994). This impact is likely to be magnified when one of the siblings is an individual with mental retardation. Many of the early studies investigating this issue (e.g., Farber, 1959; Gath, 1973) concluded siblings of persons with mental retardation are a "population at risk" (San Martino & Newman, 1974). It was believed that these siblings are disadvantaged because their parents' attention is consumed by their children with disabilities, their sibling interactions are impaired by having a

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brother or a sister with a disability, other children ostracize and isolate the siblings of children with disabilities, and because of excessive caretaking responsibilities and maturity demands placed on the typically developing siblings by their parents. More recent research has indeed identified some negative consequences to being the sibling of a child with mental retardation such as depression, loneliness, behavioral problems and low self-esteem (Bagenholm & Gillberg, 1991; Cuskelly & Gunn, 1993; McHale & Gamble, 1989; Rodrigue, Geffken & Morgan, 1993).

We reviewed the research pertaining to the psychological functioning and sibling relationships of siblings of individuals with mental retardation (e.g., Down's Syndrome). Studies of siblings of individuals with autism were reviewed because there is diagnostic overlap and frequent dual diagnosis of mental retardation and autism (American Psychiatric Association, 1994). We excluded studies for which the majority of participants were not the siblings of individuals with mental retardation. We also excluded from this review the substantial body of studies that pertain to the siblings of children with a chronic illness (for reviews of that literature, see Howe, 1993, Williams, 1997). Although it has been argued that type of disability, whether that be mental retardation or chronic illness, is not related to the psychological functioning of the typically developing sibling (Nixon & Cummings, 1999), we believe that conclusion is premature and thus this meta-analysis focuses exclusively on the siblings of individuals with mental retardation.

The psychological functioning and well-being of siblings of individuals with mental retardation have been investigated in a number of recent studies. McHale and Gamble (1989) found that siblings of children with mental retardation scored higher on measures of depression and anxiety, and scored lower on measures of social acceptance and conduct. Compared to children with typically developing siblings, Cuskelly and Gunn (1993) identified more conduct disorder problems in the female siblings of children with mental retardation. The psychological well-being of the typically developing sibling may be influenced by the severity of their sibling's disability. Fisman and colleagues (1996) found evidence for a greater risk of externalizing and internalizing behaviors in siblings of children with autism compared to siblings of children with mental retardation and siblings of typically developing children. Rodrigue and colleagues (1993) identified more internalizing and externalizing behavioral problems for siblings of children with autism than for siblings of children with Down's Syndrome and siblings of typically developing children. In their study, age of sibling and parental marital satisfaction were linked to the siblings' psychological functioning; older siblings experienced both more externalizing and more internalizing problems, and marital satisfaction was associated with better functioning for the typically developing sibling.

One area of research attention has been the amount of time siblings of children with mental retardation may be asked or required to spend in caretaking activities. Bagenholm and Gillberg (1991) found that siblings of children with mental retardation and with autism viewed themselves as having to work more often around the home than did siblings of typically developing children. McHale and

Gamble (1989) examined 62 children between the ages of 8 and 14, half of whom had siblings with mental retardation and half of whom had typically developing siblings. Home and telephone interviews were conducted of both the children and their mothers to account for time spent in caretaking activities. Compared to siblings of typically developing children, siblings of children with mental retardation reported spending more time in caregiving activities but not having more contact with their brothers and sisters with mental retardation. Stoneman, Brody, Davis and Crapps (1988) identified multiple caretaking roles for the siblings of children with mental retardation, and concluded these caretaking roles increase sibling conflict and restrict opportunities for peer contact and extra-familial activities.

A small number of studies have also examined the self-concept of siblings of children with mental retardation. Early research, such as that of San Martino and Newman (1974), found that younger siblings of persons with mental retardation over-identified with their older siblings with disabilities. These younger siblings appeared to have adopted the identity of being a sibling of a person with a disability rather than acquiring their own unique self-identity. In a more recent study, McHale and Gamble (1989) found lower self-esteem for female siblings of younger children with mental retardation when compared to controls.

One recent area of research consistent with the growing emphasis on the importance of sibling interactions is the impact a sibling with mental retardation may have on the quality of the sibling relationship. Three groups of children and young adults between the ages of 5 and 20 matched for gender, birth-order and socioeconomic status were interviewed by Bagenholm and Gillberg (1991). Siblings of children with autism were more negative with the respect to their perceptions of the sibling relationship compared to the siblings of children with mental retardation and typically developing children.

Other negative outcomes to being the sibling of a child with mental retardation have been less frequently investigated. One such outcome is less popularity and fewer friendships with other children. Andersson (1988) utilized sociometric friendship reports to discover siblings of children with mental retardation were more socially isolated than siblings of typically developing children. The siblings of children with mental retardation were also found to choose more solitary extracurricular activities such as reading than comparison children. Dyson (1989) found siblings of children with disabilities participated less frequently in extracurricular activities. Bagenholm and Gillberg (1991) similarly reported siblings of children with mental retardation and with autism were more lonely than those children with typically developing siblings and had more problems interacting with their peers.

In contrast to the somewhat bleak picture painted by the preceding review of the literature, many studies have failed to find negative consequences of being the sibling of an individual with mental retardation. Gath (1972) reported no difference in the number of behavioral problems exhibited by siblings of children with Down's Syndrome, Cleft Lip/Palate or typically developing children. Bischoff and

Tingstrom (1991) also failed to discover greater numbers of behavioral problems, lower social competence or lower self-esteem for siblings of children with disabilities. Auletta and DeRosa (1991) similarly did not find psychosocial adjustment difficulties or lower self-concept scores for adolescent siblings of individuals with mental retardation when compared to an age-matched control sample of adolescents. Roeyers and Mycke (1995) questioned siblings of children with mental retardation, siblings of children with autism and siblings of typically developing children about their sibling relationships. Children of siblings with mental retardation rated their sibling relationship somewhat more positively than did the siblings of typically developing children and scored higher on a measure of acceptance of their brother or sisters compared to controls. Gold (1993) failed to distinguish siblings of children with autism and siblings of typically developing children with regard to their social adjustment. There may also be some long-term positive aspects to having a sibling with mental retardation such as greater empathy and more appreciation for persons with disabilities (Grossman, 1972). In that vein, some studies have found adult siblings of individuals with mental retardation to be more inclined to seek careers in the helping professions (Cleveland & Miller, 1977).

Subject-related variables such as age and sex have been suggested as possible explanations for these contradictory findings. A number of studies, however, have not found any relationship between such variables and the functioning of siblings of children with mental retardation. For example, Senel and Akkok (1996) found that sex, family-size and education level did not influence the stress levels of siblings of children with disabilities. Similarly, McHale, Sloan, and Simeonsson (1986) reported no significant birth-order or gender effects. On the other hand, Dyson (1989) did report type of disability, age of the child with a disability, age-spacing, family size and mother's level of education were all significant moderator variables. Age of the child with a disability and the age-spacing between the child with a disability and his/her sibling were highlighted; there were more behavioral problems evident when the child with a disability was older and when the age-gap between the siblings with and without disabilities was smaller.

One recent methodological advance to summarize the findings from a body of studies and to resolve discrepant findings across studies is meta-analysis (Glass, 1976). Meta-analysis has been offered as a quantitative methodology to integrate the disparate findings from empirical studies and to assess factors both substantive and otherwise that produce inconsistencies across studies (Schmidt, 1992). This distinction between "study-generated evidence" and "review-generated evidence" (Hall, Rosenthal, Tickle-Degnen & Mosteller, 1994), the latter following from identification of significant moderator variables, reflects one advantage of meta-analysis over traditional literature reviews. Other advantages include a systematic and public procedure for reviewing research studies, greater weight being given to studies with larger sample sizes, and effect sizes to reflect the magnitude

of differences between conditions. Meta-analysis has become widely popular in psychology, medicine and education, but has also been widely criticized for combining studies that measure different things (the "apples and oranges" problem), for failing to capture all or a representative sample of studies conducted on some topic (the "file-drawer" problem), and for not discarding methodologically suspect studies (the "garbage-in/garbage-out" problem; see Eysenck, 1984). There are, however, statistical and procedural solutions to address many of these concerns (see Sharpe, 1997), and meta-analysis continues to grow in popularity and sophistication.

A meta-analysis of the literature on the siblings of children with a disability was published in 1994 by Summers, White and Summers. These authors coded 13 studies for their methodological quality and research methodology, and identified whether the findings from each study were positive, negative or non-significant. Their conclusion was that being the sibling of a child with a disability has some negative consequences such as aggression, but also positive consequences such as prosocial behavior. Parent surveys and observational techniques generated more negative findings than did child self-reports. Higher quality primary studies produced fewer differences between siblings of children with disabilities and comparison samples. The Summers et al. meta-analysis was limited, however, in a number of ways. The authors of the meta-analysis were able to collect only 13 studies that had an appropriate comparison group, and only eight of those studies investigated siblings of children with mental retardation. The other five studies examined the siblings of children with chronic disabilities. Effect sizes could not be calculated for 54 of 67 findings and for 4 of 13 studies. Furthermore, no studies more recent than 1989 were included in their meta-analysis.

We sought to extend and refine the Summers et al. (1994) meta-analysis. The first objective was to ascertain whether there is an overall negative or positive impact of having a sibling with mental retardation. The second objective was to determine whether these negative or positive effects were specific to selected domains; psychological functioning, self-concept, caretaking responsibilities, the sibling relationship, activities with other children, and attitudes toward individuals with disabilities. The third objective was to determine whether any of these effects can be attributed to specific aspects of the research methodology (e.g., method of data collection) or characteristics of the participants (e.g., age, sex).

METHOD

Twenty-five published studies from 1972–1999 representing over one-thousand siblings of persons with mental retardation were identified from searches of computer databases such as PSYC-LIT and ERIC using key words such as "siblings" and "mental retardation," from qualitative reviews of the literature (e.g.,

Boyce & Barnett, 1993), from the Summers et al. (1994) meta-analysis, and from the references of located studies. Case studies, non-empirical papers and studies without an appropriate comparison group (e.g., Gath & Gumley, 1987) were excluded from the meta-analysis as were studies for which none or only a small fraction of the sample had siblings with mental retardation or autism (e.g., Fernell, Gillberg & von Wendt, 1992). On the latter basis, we excluded studies that examined primarily siblings of individuals with other intellectual, developmental and learning disabilities, and siblings of individuals with chronic illness. Both child and adult studies were sought for inclusion in the meta-analysis although only two of the final 25 studies (Grossman, 1972; Konstam et al., 1993) had strictly adult samples. Studies included in the meta-analysis are marked by asterisks in the reference section.

Studies were evaluated for but not excluded on the basis of methodological quality except for the failure to utilize an appropriate comparison group. Methodological quality was evaluated on the basis of (a) an appropriate comparison group, (b) publication in a peer-reviewed journal, (c) adequate statistical information, (d) adequate sample size, (e) equivalence of experimental and control groups for age, sex and other sample characteristics, (f) recruitment strategy, (g) differential attrition, (h) reliability and validity of dependent measures, (i) specification of the disability and uniformity of the sample for that disability, and (j) other concerns. Studies by the same author(s) that appeared to examine the same sample of participants were treated as a single study for purposes of this meta-analysis (e.g., Stoneman, Brody, Davis & Crapps, 1987; 1988; 1989). Separate effect sizes were calculated for samples of participants with mental retardation and with autism although these effect sizes were combined at the study level.

Each of the 25 studies were coded for method of data collection (Self-Report, Parent Survey, Direct Observation) and nature of the dependent measures (Psychological Functioning, Self-Concept, Caretaking Responsibilities, Quality of the Sibling Relationship, Activities with Peers, and Attitudes toward the Disabled). Parent and teacher surveys were combined into the Parent Survey category because of the small number of teacher survey effect sizes. The label given to the disability of the siblings, age of participants, number of participants in the disabled and comparison samples, and a description of the dependent measures were also recorded. Coding was completed by the second author and independently checked by the first author. Disagreements were resolved by discussion.

An effect size statistic *d* (Hedges & Olkin, 1985) was calculated for each outcome by subtracting the mean score for participants with typically developing siblings from the mean score for participants who have siblings with mental retardation and dividing this difference by a pooled standard deviation. Otherwise, effect sizes were calculated from summary statistics (e.g., *t* statistics, *p* values). The meta-analysis software package *D-Stat* (Johnson, 1989) was used to derive these effect sizes and for calculating the meta-analysis statistics reported in the

results section. When no data were provided in a primary study but the comparison between samples was explicitly stated to be statistically non-significant, an effect size of zero was recorded. We did not attempt to contact authors of primary studies for additional statistical information because of the small likelihood such an undertaking would be successful (Orwin, 1994). Only a small number of relevant dependent variables could not be accommodated by following these procedures. For all reported analyses, negative effect sizes reflect less positive performance by the siblings of individuals with mental retardation compared to siblings of typically developing children.

After calculating effect sizes for all relevant dependent variables, all effect sizes from the same study, the same disability (mental retardation or autism), the same dependent measure category, and the same method of data collection were combined and averaged. The largest number of effect sizes contributed by one study was 8 (Bagenholm & Gillberg, 1991). Thus, a total of 79 unweighted effect sizes derived from 25 primary studies were collected for meta-analysis. These 79 effect sizes were then weighted by the reciprocal of their variance as recommended by Hedges and Olkin (1985), and the set of effect sizes were evaluated for their statistical significance (95% confidence interval around zero) and their homogeneity (Hedges and Olkin's (1985) homogeneity statistic Q_T). After the overall test for homogeneity, effect size clusters were created on the basis of moderator variables (e.g., method of data collection). The homogeneity of effect sizes within clusters (Hedges and Olkin's homogeneity statistic Q_W) and differences between mean effect sizes across clusters (Hedges and Olkin's homogeneity statistic Q_B) were statistically examined through a procedure analogous to the total, between- and within-group sum-of-squares in an analysis of variance. The independent effect sizes from the 25 studies were also examined where appropriate.

RESULTS

The weighted mean effect size for the 79 effect sizes was $M_d = -.06$, a negative value significantly different from zero (95% confidence interval $-.12$ to $-.01$). An effect size of $d = -.06$ is equivalent to a Pearson correlation coefficient value of $r = -.03$ (see Mullen, 1989). The weighted mean effect size for the 25 studies was $M_d = -.03$ (95% confidence interval .12 to $+.06$), a value that failed to differ from zero at the $p < .05$ level. Statistical significance aside, the magnitude of the difference between the siblings of persons with mental retardation and comparison siblings is small at best. One method for assessing the practical importance of a treatment effect is the Binomial Effect Size Display (Rosenthal, 1994; Rosenthal & Rubin, 1982). If one-hundred siblings were affected by the presence of a brother or sister with mental retardation and the Pearson correlation coefficient was -1.00, for example, one would find all one-hundred siblings to be

affected negatively. Conversely, a Pearson correlation coefficient of 0.00 would indicate that if there were some effect for having a sibling with mental retardation, fifty of the one-hundred siblings would show a negative effect and fifty of the one-hundred would show a positive effect. On this basis, one would anticipate from a correlation coefficient of $r = -.03$ that about 52 of one-hundred siblings of individuals with mental retardation should show some negative consequences and 48 of those siblings would show some positive effect.

On the other hand, this quite modest effect size may be an underestimation of the true effect size magnitude. Twenty-two of the 79 effect sizes were conservatively coded as zero. This was done because the authors of those primary studies stated that the difference between siblings of individuals with mental retardation and a comparison group was not statistically significant and provided no additional statistics (eight of the 22 effect sizes), or provided means or percentages but no standard deviations or summary statistics (fourteen of the 22 effect sizes). Approximately two-thirds of the effect sizes coded as zero came from three studies (Bagenholm & Gillberg, 1991; Lobato, Barbour, Hall & Miller, 1987; McHale, et al., 1986). Because one-and-a-half times as many effect sizes that could be calculated were negative rather than positive, we can infer that the true effect size is more negative than calculated. Furthermore, we repeated the preceding analysis deleting those 22 observations. The weighted mean effect size did become somewhat more negative, $M_d = -.08$ (95% confidence interval $-.14$ to $-.02$; $r = -.04$), a value significantly different from zero. All subsequent analyses reported below were repeated with and without these 22 observations, and no substantial differences between the two datasets were found. Thus, the dataset including the 22 effect sizes coded as zero was retained for subsequent analyses.

To assess whether the negative effect size could be attributed to a publication bias, in so far as we sampled only from published studies, three approaches were adopted. The first was a funnel plot created by plotting the sample size for each study against the effect sizes transformed into Z_{FISHER} scores (Mullen, 1989). Visual inspection of the funnel plot did not reveal evidence of a publication bias. Second, Wang and Bushman (1998) recommend a normal quantile plot over a funnel plot to assess normality of data and publication bias. A normal quantile plot is the plot of the standardized effect sizes against the quantiles or percentile ranks of the standard normal distribution. The distribution of the 79 effect sizes fell within the confidence interval for the expected line and provided no evidence for a publication bias. The same outcome was found when effect sizes were pooled to produce one effect size for each of the 25 studies. Finally, calculation of the fail-safe N statistic (Begg, 1994) for the 79 effect sizes found that there would have to be approximately 100 non-significant effect size values to reverse the outcome for the 79 obtained effect sizes. Taken together with the evidence from the funnel and quantile plots, we conclude that our finding of some small, negative effect for being the sibling of a person with mental retardation cannot be attributed simply to a publication bias.

Table I. Effect Sizes by Moderator Variables

Category	k	M_d	95% CI	Q_w
Method of Data Collection				
Self-Report	47	−.00	−.08/ + .07	83.6*
Parent Survey	30	−.13	−.21/ − .04	43.5
Direct Observation	2	−.61	−1.11/ − .10	0.5
Nature of Dependent Measure				
Psych. Functioning	29	−.16	−.24/ − .07	48.6*
Self-Concept	16	−.08	−.21/ + .04	4.6
Caretaking	6	−.22	−.43/ − .01	6.9
Sibling Relationship	20	+.19	+.06/ + .31	42.4*
Peer Activities	4	−.22	−.44/ − .00	1.6
Attitudes Disabled	4	+.25	−.00/ + .51	2.2
Psychological Functioning Subcategory				
Externalizing	16	−.15	−.26/ − .04	22.3
Social Functioning	9	+.10	−.06/ + .27	25.2*
Internalizing	8	−.19	−.37/ − .00	4.7
Depression	2	−.55	−.92/ − .17	0.0
Anxiety	3	−.20	−.39/ − .02	5.2
Age of Participant				
Child	59	−.11	−.17/ − .04	80.8
Teenager	18	−.00	−.11/ + .11	46.7*
Adult	2	+.26	−.02/ + .54	0.8

Note. k = number of effect sizes, M_d = weighted mean effect size, 95% CI = 95% confidence interval around zero, Q_w = within-cluster homogeneity test.
*$p < .05$.

The homogeneity statistic Q was calculated to assess the homogeneity of the 79 effect sizes (Hedges & Olkin, 1985). The 79 effect sizes were not a homogeneous set, $Q_T(78) = 136.5$, $p < .0001$. Such an outcome suggests further partitioning of those 79 effect sizes by moderator variables to attempt to locate the sources of that heterogeneity. The results for the first moderator variable, method of data collection, are presented in Table I. Direct Observation, albeit contributing only two effect sizes, produced a larger negative mean effect size compared to Self-Report and Parent Survey measures, $Q_B(2) = 9.0$, $p < .01$. Both Parent Survey and Direct Observation mean effect sizes ($M_d = -.13$ and $M_d = -.61$, respectively) were significantly different from zero, and the Self-Report effect size cluster was heterogeneous, $Q_W(45) = 83.6$, $p < .001$.

The results for partitioning of the 79 effect sizes by a second moderator variable, the nature of the dependent measure, are presented in Table I. Mean effect sizes for the six categories of dependent measures significantly differed from each other, $Q_B(5) = 30.7$, $p < .0001$. The negative mean effect sizes for Psychological Functioning ($M_d = -.16$), Caretaking ($M_d = -.22$), and Peer Activities ($M_d = -.22$), and the positive mean effect size for Quality of the Sibling Relationship ($M_d = +.19$), were all significantly different from zero, and post-hoc tests revealed the Psychological Functioning and Caretaking clusters differed

from the Sibling Relationship cluster. Effect size clusters were heterogeneous for Psychological Functioning and Quality of the Sibling Relationship, $Q_w(28) = 48.6$, $p < .02$, and $Q_w(19) = 42.4$, $p < .003$, respectively.

For one of the heterogeneous dependent measures categories, Psychological Functioning, effect sizes were further partitioned into five sub-categories: Externalizing Behavior (e.g., measures of conduct problems, aggressiveness), Social Functioning, Internalizing Behavior (e.g., measures of shyness, inhibited-anxious personality type), Depression, and Anxiety (Table I). This recategorization was accomplished by returning to the original unpooled effect sizes, and pooling those effect sizes from the same study and the same sub-category. Differences between the mean effect sizes for these five sub-categories was significant, Q_B (4) = 13.5, $p < .01$. Post-hoc tests revealed this significant difference can be attributed to greater Depression ($M_d = -.55$) for the siblings of persons with mental retardation when compared to their more positive Social Functioning ($M_d = +.10$). Negative mean effect sizes significantly different from zero were found for Depression and also for Externalizing Behavior, Internalizing Behavior and Anxiety. The Social Functioning cluster was statistically heterogeneous but further partitioning was precluded by the small number of effect sizes within that cluster.

Two additional analyses of the 79 effect sizes were conducted to ascertain whether sex and age of the siblings of individuals with mental retardation might explain some of the variability in effect sizes. Unfortunately, many studies did not provide separate data for male and female participants. In the same vein, many studies sampled participants over a range of ages and few studies examined adult siblings. In spite of these limitations, some effort was made to address sex and age differences. First, each study was coded for proportion of male siblings of individuals with mental retardation. These proportions served as the predictor variable in a weighted least squares regression analysis (Hedges & Olkin, 1985) with the dependent measure being the effect sizes from the 25 studies. Effect sizes from 23 of the 25 studies could be included in this analysis because the authors of those studies specified the number or proportion of male and female participants. The resulting test statistic $Q_R = .01$ clearly failed to exceed 3.84, the chi-square distribution critical value for 1 degree of freedom, indicating that proportion of male subjects did not significantly relate to effect size magnitude for those 23 studies.

Second, each effect size was coded according to whether the reported age of the sample of siblings of individuals with mental retardation was less than 13 years (child subjects; 59 effect sizes), 13 years or older but less than 19 years (teenage subjects; 18 effect sizes), and adult (2 effect sizes). These results are presented in Table I. The mean effect size for the Child ($M_d = -.11$) and Teenager clusters ($M_d = -.00$) were both negative although only the former effect size was significantly different from zero, and the mean effect size for the Adult cluster was positive ($M_d = +.26$) albeit marginally not different from zero. The Adult cluster differed significantly from the Child cluster, $Q_B(2) = 8.1$, $p < .02$. Both the

Child and Teenager clusters were heterogeneous, $Q_w(57) = 80.8$, $p < .06$ for the Child cluster, $Q_w(17) = 46.7$, $p < .0001$ for the Teenager cluster, suggesting these clusters need to be further partitioned by other moderator variables.

Given the small number of effect sizes associated with adult participants, it is difficult to ascertain the robustness and the source of this age difference. One possibility is that the age difference effect may be attributable to adults giving self-report and children being assessed by parents and by direct observation. To the contrary, 33 of the 59 Child cluster observations were Self-Report—24 of the Child cluster observations were Parent Reports, only 2 Direct Observation. The weighted mean effect size for the 33 Self-Reports by children was $M_d = -.05$, and for the 24 Parent Reports on children $M_d = -.14$. The pattern of results for teenagers parallels the findings for children; the mean effect size for the 12 Self-Reports for teenagers was $M_d = +.03$, and for the 6 Parent Reports on teenagers was $M_d = -.07$. Thus, we find somewhat more positive results for Self-Reports compared to Parent Reports for both children and teenagers. Additional primary studies of adult siblings of individuals with mental retardation are needed to determine whether self-reports by adults are also more positive.

One additional variable that might influence the psychological functioning of the siblings of individuals with mental retardation is the level of mental retardation of the sibling with a disability (mild, moderate or severe/profound). Nine of the 25 studies did not provide any information in their description of the sample as to the level of mental retardation. In two of the studies, the level of mental retardation was said to vary from mild to severe, and in three studies the percentage of individuals with severe mental retardation varied from 17% to 30%. In contrast, four studies reported the degree of mental retardation varied from moderate to severe, and six studies reported severe mental retardation for most or all of their participants. Because for over half of the studies no information on level of severity of the mental retardation or a range of functioning was reported, it was not possible to ascertain statistically the impact level of functioning had on sibling functioning or the quality of the sibling relationship.

Finally, each study was evaluated for methodological quality using the criteria outlined in the method section. We reached the same conclusion as Summers et al., (1994) that the methodological quality of studies investigating siblings of individuals with mental retardation is high. All 25 studies of our studies were published, with 24 of the 25 studies published in journals. Grossman (1972) was published in book form. All 25 studies provided sufficient statistical information to permit effect sizes to be calculated. The study with the smallest number of participants, 12 in the experimental group and 13 in the comparison group, was Lynch, Fay, Funk and Nagel (1993). We purposely excluded case studies from the analysis because those studies would not provide sufficient sample size for analysis and typically do not provide statistical data. Twenty-three of the 25 studies had approximately equal numbers of participants in their experimental and comparison

groups. More importantly, 18 of the 25 studies matched their experimental and comparison groups on sex of respondents, and 14 of the 25 studies explicitly matched their experimental and comparison groups on age of respondents. Another 8 studies compared their experimental and comparison groups after-the-fact for age differences or reported the groups were similar on age, and 5 studies reported equivalent numbers of males and females in the two groups. The authors of 10 studies reported matching their experimental and comparison groups on a variety of other variables such as birth-order, socio-economic status, and family size. Most studies that reported how participants were recruited indicated their experimental and comparison participants had been drawn from comparable sources. Few studies reported differential attrition for their experimental and comparison groups. Eighteen of the 25 studies employed standardized dependent measures. All 25 studies in our meta-analysis examined the brothers and sisters of individuals diagnosed with mental retardation. Five of the 25 studies more broadly sampled from other disabilities although we excluded studies for which individuals with mental retardation were not the majority.

DISCUSSION

The present meta-analysis found a statistically significant but small negative effect for having a sibling with mental retardation on the functioning of the typically developing sibling. This finding is consistent with the results from the Summers et al., (1994) meta-analysis that counted the number of positive and negative study outcomes. The magnitude of this negative effect in the present meta-analysis, however, suggests the generalized concern about the social and psychological development of the siblings of individuals with mental retardation has been overstated. Family system theory (Minuchin, 1988; Seligman & Darling, 1989) implies that any difficulties experienced by one member of a family will impact negatively on all family members. One might presume that the impact of a sibling with a disability would be negative but this may not necessarily be so. Much would depend on the many factors both within and outside the family that determine successful development for the typically developing sibling: parents, other siblings, the extended family, the peer group, socioeconomic status, the educational system, the community and so on (Bronfenbrenner, 1979). Furthermore, the nature of the disability and the level of functioning of the sibling with disabilities would undoubtedly play a role on the impact of the disability on the typically developing child. Stoneman (1998) identifies personality and temperament, level of competence, health problems and secondary disabilities as etiological factors that will influence the relationship between the child with disabilities and their typically developing sibling.

To investigate some possible determinants for the modest negative effect of having a sibling with mental retardation, a number of moderator variables were

examined. The first of the moderator variables was method of data collection in the primary studies; self-report, parent survey, and direct observation. Self-report produced the smallest negative effect size, direct observation the largest. The parent survey effect size fell between self-report and direct observation. This outcome might imply that the siblings of individuals with mental retardation and to a lesser extent their parents, view their development in a more positive light than is objectively the case. Because only 2 of the 79 effect sizes were coded as direct observation, however, this speculation is premature in the absence of additional studies employing direct observation of the siblings of individuals with mental retardation.

The second moderator variable that was evaluated was nature of the dependent measures. The most striking finding from this analysis was that psychological functioning was significantly more negative for the siblings of individuals with mental retardation compared to control participants. Because the psychological functioning effect size cluster was statistically heterogeneous and of particular interest given past research highlighting less positive psychological functioning for the siblings of persons with mental retardation, this cluster was further partitioned into five sub-categories. The resulting analysis revealed depression and to a lesser extent anxiety, internalizing behaviors and externalizing behaviors to all be problems for the siblings of individuals with mental retardation. Depression and anxiety have been highlighted in qualitative reviews of the literature (e.g., Hannah & Midlarsky, 1985) and elevated depression and anxiety scores might have mistakenly led earlier researchers in the area (e.g., Farber, 1959; Gath, 1973) to conclude the sibling of persons with mental retardation are maladjusted. We can only speculate as to why these problems are particularly salient for the siblings of persons with mental retardation. Gold (1993) suggested that siblings of children with disabilities may internalize problems and she implied those problems may be related to caretaking responsibilities. In the same vein, Gamble and McHale (1989) found other-directed cognitions to be related to elevated levels of depression and anxiety in siblings of persons with mental retardation, and McHale and Gamble (1989) reported differential treatment of disabled and typically developing siblings by parents to be related to depression and anxiety for the latter group of siblings. Research is needed to identify those specific aspects that contribute to the less positive psychological functioning of the typically developing sibling.

Two subject variables, sex and age of the siblings of individuals with mental retardation, were also evaluated. Sex differences, such as female children more often than male children being assigned caretaking responsibilities, have frequently been suggested in the literature. The proportion of male participants in the primary studies, however, was found to be unrelated to differences between siblings of individuals with disabilities and comparison siblings. This was a somewhat inappropriate comparison in so far as the number of male and female participants were equated in many of the primary studies, and because the proportion of male and female participants is an imprecise alternative to separate data for male and

female siblings. Furthermore, recent studies (e.g., Hannah & Midlarsky, 1999), have found sex differences such as male siblings of children with mental retardation having more difficulties at school and female siblings of children with mental retardation scoring higher on measures of internalizing problem behaviors. In regards to age of siblings, significant differences were found in our meta-analysis between child and adult siblings of individuals with mental retardation. Few studies have investigated adult siblings of persons with mental retardation, but one might anticipate more positive psychological functioning for adult siblings because the impact of a sibling with disabilities and the influence of the birth family should both diminish in adulthood, and because more advanced cognitive and social development might provide better coping mechanisms for dealing with sibling and familial stressors. The evidence for such positive outcomes is for the most part anecdotal aside from the four positive effect sizes for attitudes toward individuals with mental retardation identified in our meta-analysis. Finding more such positive outcomes would provide a more balanced perspective to the literature and some comfort to the parents of siblings of children with mental retardation. One might also anticipate that some positive aspects of having a sibling with mental retardation, such as greater empathy, are more likely to be exhibited in adulthood and thus some research efforts should be directed at long-term consequences of these sibling relationships.

Any meta-analysis is limited by the number and quality of primary studies, and the variables selected for investigation in those studies. All of the primary studies included in this meta-analysis were from published articles subject to peer review thereby providing at least some assurance of quality control. Furthermore, published studies that lacked control groups were by necessity excluded from the meta-analysis. In their meta-analysis of this literature, Summers et al., (1994) directly coded their primary studies for methodological quality using a coding scheme derived from Cook and Campbell's (1979) threats to internal and external validity. All of their primary studies were given ratings between 1 (high validity) and 3 (moderate validity). We examined the primary studies for quality in the present analysis but excluded studies only if there was no appropriate comparison group. Our strategy not to exclude studies on methodological grounds and to sample only published studies balanced an intentional publication bias against quality control. We did go to some efforts to determine our results could not be attributed to the exclusion of unpublished studies and dissertations, and we were satisfied that our body of studies was of acceptable methodological quality. Because most of the primary studies in our meta-analysis met our criteria for methodological quality and because it is notoriously difficult to construct coding schemes for quality (Sharpe, 1997), we did not assess whether there was a relationship between the effect size estimates and study quality.

One limitation to the present meta-analysis was the number of primary studies and their presentation of the data. We were successful in locating nearly twice as many primary studies as Summers et al., (1994) and deriving effect size estimates

for those studies. Nonetheless, we must echo Boyce and Barnett's (1993) statement at the conclusion of their review of this literature that "It is perhaps a pat answer to simply call for further research, but in this field much will be learned if the body of research continues to grow based on the thoughtful use of findings within the research field and changes in contexts outside the field of research" (p. 179). One of the most often expressed fears about meta-analysis is that a quantitative review may inhibit future research by prematurely closing an area of investigation (Boden, 1992). To the contrary, the present meta-analysis highlights the need for more studies investigating the impact of having a sibling with mental retardation and suggests future directions for that research. There is a need for a greater number of studies of adult siblings of individuals with mental retardation and studies that employ observational measures to collaborate self- and parent-reports. This meta-analysis and future meta-analyses of this literature would benefit from the reporting of statistical information for all dependent measures, participants' level of mental retardation, and separate data for male and female participants. The failure to report sufficient information to permit the calculation of effect sizes for all dependent measures is the bane of meta-analysts and is easily remedied by the authors of primary studies providing statistical information for all dependent measures. We adopted the common practice in meta-analysis of coding these non-significant outcomes as zero and found no evidence to suggest these missing effect size estimates would have substantially changed the findings from this meta-analysis.

A second limitation to this meta-analysis is that we cannot conclude there is a causal relationship between adjustment problems and having a sibling with mental retardation. Practitioners of research synthesis are constrained from making cause-and-effect statements both by the correlational nature of moderator variable analysis and by the internal validity of the primary studies that contributed to the meta-analysis (Hall et al., 1994). Meta-analysis cannot rule-out some third variable accounting for any differences identified through moderator variable analysis and all the primary studies in our meta-analysis employed a quasi-experimental design in their examination of pre-existing groups. In spite of the efforts of primary researchers to equate the experimental and comparison groups on confounding variables such as age, sex, birth-order, and socio-economic status, we cannot conclude that any differences found between the brothers and sisters of persons with mental retardation and typically developing individuals is the result of their siblings' disabilities, per se. A number of recent studies (e.g., Bristol, Gallagher & Schopler, 1988; Trute, 1995; Veisson, 1999) have found parents of individuals with disabilities to be at risk for psychological problems because of social isolation and caretaking responsibilities. In the same vein, there is evidence to suggest that autistic-like features and psychiatric disorders aggregate in families of individuals with autism (Bolton, Pickles, Murphy & Rutter, 1998). On that basis, any negative effects found for siblings of individuals with mental retardation could plausibly be attributed to their parents or to a genetic predisposition for psychiatric difficulties.

In her review of the research literature on siblings of children with mental retardation, Stoneman (1998) concluded that past efforts to seek pathology in the siblings of individuals with mental retardation were misguided, that there is now some agreement "having a sibling with mental retardation does not cause one to become maladapted, attack classmates, fail at school, or become clinically depressed," and that "some siblings benefit, some seem not to be affected, and a small group are harmed" (p. 685). The somewhat pessimistic view arising from past research in this area can be attributed, in part, to researchers limiting their search to negative and short-term consequences of being the sibling of a person with mental retardation. Future research must also evaluate potential positive consequences to being the sibling of a person with mental retardation, such as greater empathy and a better understanding of individuals with disabilities, and must recognize the possibility that some positive consequences may not appear until adolescence or adulthood. The positive impact of typically developing siblings on the child with a disability should also be considered. For those children for whom it appears having a sibling with a disability is associated with some difficulties, researchers and clinicians need to determine whether it was the presence of the sibling with a disability or rather some related factor that is the source of the problem.

If negative outcomes such as depression or anxiety arise in the brothers and sisters of children with mental retardation, both parents and clinicians must recognize the clinical implications for such outcomes. If these siblings are indeed at a greater risk for developmental and/or clinical difficulties, clinicians must be aware of this possibility so that preventative measures can be established to assist the sibling. Although little is known about the long-term effects of such interventions, information sessions and sibling support groups have been found to increase childrens' knowledge of their siblings' disabilities, facilitate understanding of themselves and their families' situation, and improve their emotional state (Lobato, 1993). We must be cautious, however, in assuming that all individuals who have sibling with mental retardation are at risk. There are likely many different reactions to having a sibling with mental retardation. Some children may well experience positive reactions to having a sibling with mental retardation. Positive outcomes need to be encouraged by caregivers and professionals. Researchers must continue to search for explanations as to why some children who have siblings with mental retardation have greater difficulties than others. To this point, the literature is only suggestive as to what those explanations might be. When specific risk factors are identified, early and appropriate interventions should be implemented.

Methodologically, we applaud and encourage the use of comparison groups of children and adult research participants with typically developing siblings. Direct observation and non-reactive dependent measures should be employed in combination with self-report and parent/teacher survey methodologies. Some recognition of the many different types of mental retardation and different levels of functioning of individuals with mental disabilities needs to be incorporated into

research designs. In that vein, a comparison of siblings of individuals with mental retardation to siblings of children and adults with other disabilities (e.g., learning disabilities, chronic illness) might provide some valuable insights. Many recent writers have commented that the sibling bond is often our most enduring relationship. The sibling bond is no less enduring nor less influential when one sibling has a disability.

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