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Siblings of Children With a Chronic Illness: A Meta-Analysis

Donald Sharpe, PhD, and *Lucille Rossiter,* MA
University of Regina

Objective: **To review the literature pertaining to the siblings of children with a chronic illness.**
Methods: **Fifty-one published studies and 103 effect sizes were identified and examined through meta-analysis.**
Results: **We found (1) a modest, negative effect size statistic existed for siblings of children with a chronic illness relative to comparison participants or normative data; (2) heterogeneity existed for those effect sizes; (3) parent reports were more negative than child self-reports; (4) psychological functioning (i.e., depression, anxiety), peer activities, and cognitive development scores were lower for siblings of children with a chronic illness compared to controls; and (5) a cluster of chronic illnesses with daily treatment regimes was associated with negative effect statistics compared to chronic illnesses that did not affect daily functioning.**
Conclusions: **More methodologically sound studies investigating the psychological functioning of siblings of children with a chronic illness are needed. Clinicians need to know that siblings of children with a chronic illness are at risk for negative psychological effects. Intervention programs for the siblings and families of children with a chronic illness should be developed.**

Key words: *pediatric chronic illness; siblings; meta-analysis; psychological adjustment.*

Between 5% and 40% of children suffer from a chronic illness (Newacheck & Halfon, 1998; Perrin & MacLean, 1988). Williams (1997) defined a chronic illness as a "medically diagnosed ailment with a duration of 6 months or longer, which shows little change or slow progression" (p. 312). In the United States, between 4 and 7 million children suffer from one or more chronic illnesses, and about one-half to one million of those children suffer from a severe chronic childhood disability (Newacheck & Halfon, 1998; Patterson, 1988). In all likelihood, these figures underestimate the number of children with a chronic illness (Lenton, Stallard, Lewis, & Mastroyannopoulou, 2001). Childhood chronic illness has long been thought to have a negative impact on the psychological functioning and behavior of the ill child, that "compared with healthy peers, children with ongoing health conditions are at greater risk of mental health problems . . . emotional disorders, abnormal behavioral symptoms, and school-related adjustment problems" (Cohen, 1999, p. 149). All family members could be affected by having a child with a chronic illness. This review focuses on the literature pertaining to the siblings of children with a chronic illness.

All correspondence should be sent to Donald Sharpe, Department of Psychology, 3737 Wascana Pkwy, University of Regina, Regina, Saskatchewan, Canada S4S OA2. E-mail: sharped@uregina.ca.

Siblings of Children With a Chronic Illness

In one of the first literature reviews of the impact of illness on the siblings of children with a chronic disease, McKeever (1983) concluded that these siblings were a "population at risk" (p. 210). Hannah and Midlarsky (1985) also found siblings to be a "population at risk to experience psychological difficulties" (p. 510), but there was the suggestion of some positive benefits to growing up with an ill sibling, such as greater compassion. More recent reviews (e.g., Faux, 1993; Packman, 1999; Williams, 1997) repeat these themes: negative outcomes but the suggestion of some long-term, positive effects. The most striking impression from these literature reviews, however, is the lack of consensus. "To anyone reading the literature reporting research studies of the psychological adjustment of the siblings of individuals with a disability, the overwhelming impression is one of contradiction and confusion" (Cuskelly, 1999, p. 111).

Quantitative Reviews of the Literature

A recent methodological advance to resolve discrepant findings across studies is meta-analysis. This quantitative review strategy is employed to assess factors both substantive and methodological that produce inconsistencies across studies (Schmidt, 1992). Howe (1993) employed a vote-count meta-analysis strategy to review 21 studies with control groups or normative reference groups that examined siblings of children with chronic illness. A vote-count meta-analysis is a simple tabulation of studies by their outcomes. Howe concluded that siblings of children with a chronic illness were at higher risk than other children for psychological problems, that neurological conditions produced more negative effects than nonneurological conditions, and that negative effects were most often manifested as internalizing behaviors.

Summers, White, and Summers (1994) conducted a vote-count meta-analysis of 13 studies of siblings of children with a chronic illness or an intellectual disability. These 13 studies were assessed for their methodological quality and research methodology, and study results were categorized as positive, negative, or nonsignificant. These researchers concluded that being the sibling of a child with a disability had both negative and positive consequences, that parent surveys and direct observation generated more negative findings than child self-reports, and that higher quality studies found fewer differences between siblings and comparison samples. Like Howe's (1993) review of the literature, the Summers et al. meta-analysis was constrained by the limitations to the vote-count review strategy: no estimation of effect size magnitude, no consideration of sample size, and no mechanism for evaluating systematically the impact of moderator variables.

This Study

A recent meta-analysis of 25 studies and 79 effect sizes from the literature on the siblings of individuals with intellectual disabilities (Rossiter & Sharpe, 2001) revealed a small negative effect for having a sibling with an intellectual disability that could not be attributed to a publication bias or some other artifact. This negative effect was most pronounced for measures of psychological functioning, especially depression, and adult reports versus child self-reports. This meta-analysis pertains to the siblings of children with a chronic illness. Based on the findings from traditional literature reviews and the vote-count meta-analyses, a negative effect was anticipated for having a sibling with a chronic illness. A number of hypotheses based on methodological and substantive issues were then derived.

Methodological Issues. The first methodological hypothesis was that studies published more recently would show fewer negative and more positive outcomes than earlier studies. Lamorey (1999) observed more recent studies to show fewer negative effects and more variation in outcomes. A second methodological hypothesis was that more negative effects would be found for parental reports than sibling self-reports (Summers et al., 1994). The third methodological hypothesis was that studies employing normative data for comparison to the sibling samples would produce negative effects of greater magnitude than found for studies that employed matched control groups (Lavigne & Faier-Routman, 1992).

Substantive Issues. A number of hypotheses were also made that related to substantive variables. First, a larger negative effect was expected for internalizing over externalizing behaviors. Howe (1993) found four of eleven studies of siblings of children with chronic illness showed a negative effect for internalizing behavior compared to only one of eight studies for externalizing behaviors. Second, sibling outcomes were anticipated to vary by the chronic disease and its features. One view is many chronic conditions of childhood produce similar psychological and behavioral effects (Vessey & Mebane, 2000).

Childhood chronic illnesses, however, vary on dimensions such as etiology, age of onset, impact on functioning, and prognosis (see Lobato, Faust, & Spirito, 1988). More severe chronic illnesses place greater restrictions on the child's activities (Newacheck & Taylor, 1992), and perhaps greater demands on parents, siblings, the family system, and the community (Patterson, 1988). Third, the interaction of sibling gender and birth order was considered (Howe, 1993; Williams, 1997).

Method

Fifty published studies from 1976 to 2000, representing over twenty-five hundred siblings of children with chronic illness, were identified from computer searches of databases such as PsycLit and MEDLINE, using key words such as "siblings" and "illness," from previous reviews of the literature and from the reference sections of located studies. Excluded from the meta-analysis were case studies, nonempirical or qualitative studies, or studies without an appropriate comparison group or normative data. Studies were also excluded that evaluated the reactions of healthy siblings to the illness or death of a brother or sister or pertained to the adult siblings of individuals with a chronic illness. Studies that employed no comparison group but that provided normative data were included in this meta-analysis.

Unpublished studies were not sought for inclusion in this meta-analysis. First, it is almost impossible to collect all published studies in all languages, much less all unpublished studies. Second, the peer-review process for published studies serves as an albeit imperfect form of quality control. Third, there is evidence that publication bias is less serious than once feared (Sharpe, 1997). Publication bias, the so-called "file-drawer" problem, is the belief that the failure to include unpublished studies in the meta-analysis might inflate the magnitude of effect sizes, given that published studies may overrepresent statistically significant findings. To ascertain the likelihood of such a publication bias, statistical and graphical analyses of effect sizes were conducted.

Studies by the same author(s) that appeared to examine the same participants (e.g., Breslau & Prabucki, 1987; Breslau, Weitzman, & Messenger, 1981) were treated as a single study for the purposes of this meta-analysis. Three of the primary studies (Faux, 1991; Stawski, Averbach, Barasch, Lerner, & Zimin, 1997; Wood et al., 1988) provided separate data for the siblings of children with distinctly different chronic illnesses. These subsamples were treated as separate studies. In total, 51 study-level effect size statistics were evaluated. Each study was coded for method of data collection (child self-report, parent report, or direct observation), chronic illness, age of siblings, gender of siblings, number of sibling and comparison participants, and dependent measure category: psychological functioning (e.g., Internalizing subscales of the Child Behavior Checklist), self-concept (e.g., Piers-Harris Self-Concept scale), caretaking, sibling relationship, peer activities (e.g., Social Competence subscale of the Child Behavior Checklist), cognitive functioning (e.g., intelligence test scores), and cognitive development (e.g., school performance). Parent and teacher reports were combined because only five studies asked teachers to complete a dependent measure. Four of the five comparisons based on teacher reports were not statistically significant. All codings were completed by the first author and checked independently by the second author. Disagreements were resolved by discussion.

Effect Size Calculations. An effect size statistic *d* (Hedges & Olkin, 1985) was calculated for each relevant outcome by subtracting the mean score for comparison participants from the mean score for siblings with a chronic illness and by dividing that sum by a pooled standard deviation. Normative data provided by the primary authors in the published studies were substituted for data from comparison participants when the latter were not provided. If means and standard deviations were not reported, effect sizes were calculated from summary statistics (e.g., *t* statistics, *p* values) by employing the meta-analysis software package *D-Stat* (Johnson, 1989). Effect sizes were weighted by the reciprocal of their variance as recommended by Hedges and Olkin (1985). When no data were reported in a primary study but the difference between the sibling and comparison groups was said to be nonsignificant, an effect size of zero was recorded. For all analyses, negative effect sizes reflect less positive functioning for siblings of children with a chronic illness relative to comparison children or normative data.

Effect sizes from the same study, chronic illness, dependent measure category, and method of data collection were combined and averaged. The resulting set of 103 outcome-level effect sizes was evaluated for their statistical significance (95% confidence interval around zero) and their homogeneity (Hedges & Olkin, 1985). The effect sizes from the 51

studies were also examined where appropriate to do so. The overall test for homogeneity (Q_T) assesses whether a set of effect sizes is internally consistent. For most meta-analyses, homogeneity of the set of effect sizes is not achieved without some combination of outlier analysis and partitioning of effect sizes into smaller clusters on the basis of moderator variables. The identification and removal of outliers are appropriate if homogeneity can be achieved by deleting no more than 20% of the effect sizes (Hedges & Olkin, 1985). Regardless of the outcome of the overall test of homogeneity, however, tests of moderator variables are justified when based on theoretical considerations (see Hall & Rosenthal, 1991).

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 (Q_W) and differences between mean effect sizes across clusters (Q_B) were calculated. A significant Q_B value implies differences in the mean effect sizes associated with the effect size clusters. Interpretation of such an outcome is less clear if there are significant differences in effect sizes within one or more clusters (the Q_W statistic for each cluster). When moderator variables were continuous (e.g., sample size), correlations between effect sizes and the moderator variables were calculated.

Results

The results are divided into three sections. The first section reports on tests of effect sizes: tests of the magnitude of mean effect sizes, tests for publication bias, and tests of homogeneity of effect sizes. The second section examines the role of methodological moderator variables, specifically, year of publication, method of data collection, and comparison group versus normative data. The third section considers substantive moderator variables, specifically, categories of dependent measures, differences by chronic illness, and effects of gender, birth order, and age of sibling.

Tests of Effect Sizes

Overall Effect Size. The weighted mean effect size for the 103 outcome-level effect sizes was $M_d = -.20$ (the equivalent of $r = -.10$), a negative value significantly different from zero (95% confidence interval = –23 to –.16). This effect size may be an underestimation of the true effect size magnitude. Thirty-two of the 103 effect sizes were conservatively coded as zero because the authors of those primary studies did not report statistics but stated differences were not significant. The weighted mean effect size after deleting those 30 observations was $M_d = -.26$ (95% confidence interval = –.30 to –.22). The weighted mean effect size for the 51 studies was $M_d = -.21$ (95% confidence interval = –.26 to –.16).

Publication Bias. Given that only published studies were included in the meta-analysis, there is a risk of publication bias as studies that do not find statistically significant results may not be published and, therefore, may not be included in the meta-analysis. To investigate publication bias, four approaches were adopted. The first was funnel plots created by plotting sample sizes for the siblings against effect sizes at the study and outcome level. The plots were funnel-shaped. Data points were distributed across the lower left and right quadrants and were less frequent as sample size increased. This pattern is not consistent with a publication bias (Begg, 1994). Second, Wang and Bushman (1998) recommend a normal quantile plot over a funnel plot to assess publication bias. A normal quantile plot involves plotting the effect sizes against the quantiles or percentile ranks of the normal distribution. There was no gap in the effect sizes for the 51 effect sizes at the study level and a small gap around zero for the 103 effect sizes at the outcome level. Third, calculation of the fail-safe *N* statistic (Cooper, 1998) found that there would have to be an additional 566 nonsignificant studies to reverse the significant negative result from the 51 studies. This number is much larger than the cutoff value of 265 studies (five times the number of retrieved studies plus 10; Cooper, 1998). Fourth, one would expect a relationship between sample size and effect size magnitude if a publication bias were operating as larger effects, both positive and negative, would be found for studies with smaller sample sizes that do not have the statistical power to detect small effects. There was no significant correlation, however, between the number of sibling participants and the absolute value of the effect sizes at the outcome level (r [101] = .06) or study level (r [49] = .07).

Tests of Homogeneity. Heterogeneity of effect sizes was found for both the 103 outcome-level effect sizes (Q_T [102] = 354.7, $p < .0001$) and the 51 study-level effect sizes (Q_T [50] = 139.8, $p < .0001$). At the outcome-level, deletion of 11 outcomes (10.7% of the 103 outcomes) resulted in a homogeneous set (Q_T [91] = 112.6, $p < .12$). The mean effect size magnitude was

reduced from M_d = –.20 for the 103 outcomes to M_d = –.09 for 92 outcomes (95% confidence interval = –.13 to –.05). At the level of the 51 studies, homogeneity could be achieved by the exclusion of five studies (9.8% of the database) (Q_T [45)]= 61.4, $p < .10$). The mean effect size magnitude was reduced from M_d = –.26 for the 51 studies to M_d = –.09 for 46 studies (95% confidence interval = –.14 to –.02).

Methodological Moderator Variables

Year of Publication. There were modest, albeit nonsignificant, correlations between publication year and study-level effect sizes (r [49] = .21, $p < .14$). The interpretation for a positive correlation is that effect sizes were somewhat more positive for recent studies. There was also a modest negative correlation between year of publication and sample size at the study-level (r [49] = –.22, p <.12). This would suggest the sample size has declined over the past 20 years. More studies of siblings of children with a chronic illness were published in the 1990s (n = 27) than the 1980s (n = 19) and 1970s (n = 4). However, there were five large-scale studies (i.e., more than 100 siblings of children with a chronic illness) published in the 1980s compared to only two such studies published in the 1990s.

Method of Data Collection. Another methodological variable hypothesized to moderate effect size magnitude was method of data collection. Sixty-one outcomes were associated with parent report. Child self-report accounted for the remaining 41 outcomes. Only one outcome was the product of direct observation. After deleting the direct observation outcome for this analysis only, the difference between the mean effect sizes associated with child reports and parent reports was significant (Q_B [1] = 7.0, $p < .0001$, see Table I). Although both the child reports and parental reports mean effect sizes were significantly different from zero, the mean effect size for parental reports (M_d = –.23) was almost twice as large as that for child reports (M_d = –.13).

Comparison Group vs. Normative Data. Eighty-two outcomes were evaluated against a comparison group compared to 21 outcomes contrasted to normative data. At the outcome-level, siblings of children with chronic illnesses appeared much worse off when compared to normative data (M_d = –.34), in lieu of comparison groups (M_d = –.09; Q_B [1] = 47.9, $p <$ .0001). Both mean effect sizes differed significantly from zero (see Table I). Caution should be taken in interpreting this outcome, given there were four-times as many comparison group studies as normative group studies.

Table I. Effect Sizes at the Outcome Level by Moderator Variables

Category	k	M_d	95% CI	Q_w
Method of data collection				
Child report	41	–.13	–.19/–.06	93.0*
Parent report	61	–.23	–.27/–.19	254.0*
Comparison data				
Control	82	–.09	–.14/–.05	147.3*
Normative	21	–.34	–.39/–.28	159.6*
Dependent measure				
Psych. functioning	47	–.22	–.26/–.17	129.1*
Self-concept	17	–.06	–.16/+.03	31.3*
Caretaking	4	–.14	–.37/+.09	4.5
Sibling relationship	6	+.12	–.06/+.30	7.1
Peer activities	22	–.29	–.36/–.22	140.8*
Cog. functioning	3	–.14	–.42/+.15	1.5
Cog. development	4	–.24	–.44/–.04	13.3*

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

Substantive Moderator Variables

Dependent Measures. Table I also presents the effect sizes at the outcome-level partitioned by category of dependent measure. The most frequently represented category of dependent measure was psychological functioning. Differences between effect size clusters were significant (Q_B [6] = 27.3, $p < .0001$). Psychological functioning, peer activities, and cognitive development effect size clusters produced negative mean effect sizes significantly different from zero. The sibling relationship category produced a positive effect size, although not significantly different from zero.

Internalizing vs. Externalizing Behavior. To test the hypothesis that more negative effects would be found for internalizing behaviors over externalizing behaviors, and in light of the heterogeneous effect sizes for the psychological functioning category, we further partitioned that dependent measure category. Studies that contributed to this category were examined first for dependent variables that reflected internalizing behaviors (e.g., anxiety, depression, the Internalizing subscale of the Child Behavior Checklist) or externalizing behaviors (e.g., behavior problems, aggression, the Externalizing subscale of the Child Behavior Checklist). The mean effect size for the 26 internalizing outcomes was M_d = –.41 (95% confidence interval = –.48 to –.34), a value signifi-

cantly larger than the mean effect size for the 24 externalizing outcomes (M_d = –.15, 95% confidence interval = –.23 to –.07; Q_B [1] = 21.5, p < .0001).

Chronic Illnesses. Table II presents the results at the study level for different chronic illnesses. Sixteen studies were not represented. Ten of those studies combined data from diverse chronic conditions and six studies examined unique chronic illnesses not considered by any other study of siblings of children with a chronic illness. Ten chronic illnesses were represented in the remaining 35 studies. Cancer was the chronic illness in 10 studies and diabetes in 6 studies. All other chronic illnesses were represented in two or three studies. Differences between effect size clusters were significant (Q_B [9] = 18.8, p < .03). All mean effect sizes were negative except for two studies evaluating cardiac disease that produced a positive effect size value not statistically different from zero.

Severity. A number of classification schemes for chronic illnesses have been employed in previous literature reviews. One variable we considered was prevalence (see Newacheck & Halfon, 1998), but all childhood chronic illnesses have low prevalence rates with the exception of asthma. Life expectancy is one imperfect measure of the severity of a chronic illness that has been shown to be related to family coping, achievement of maturational milestones, and expectations for the future (Patterson, 1988; Vessey & Melbane, 2000). This is in spite of the life expectancy for life-threatening chronic illnesses having risen substantially over the last two decades with advances in medical treatments (Jackson, 2000). For our purposes, there were data available on the mortality rates for all the chronic illnesses represented in our studies (see Newacheck & Halfon, 1998; Newacheck & Taylor, 1992; Patterson, 1988). Five studies could not be classified into greater or lesser severity because the studies combined chronic illnesses of different mortality rates or did not report the specific chronic illnesses of their participants' brothers and sisters.

Chronic illnesses of higher mortality rates, and thus greater severity, were HIV/AIDS, cancer, cystic fibrosis, renal failure, sickle cell anemia, and liver disease. Diabetes, cerebral palsy, rheumatic disease, bowel disease, craniofacial anomalies, cardiac disease, epilepsy, infantile hydrocephalus, spina bifida, hearing impairments, and asthma were disorders of lower mortality rates and thus considered less severe. The difference in the mean study level effect sizes between the two clusters was not significant (Q_B [1] = 2.2). The siblings of children with more severe chronic illnesses were no more at risk (M_d = –.17), compared to the siblings of children with less severe chronic illnesses (M_d = –.26).

Table II. Effect Sizes at the Study Level by Chronic Illness and Severity

Category	*n*	M_d	95% CI	Q_w
Chronic illness				
Cancer	10	–.28	–.39/–.17	31.6*
Diabetes	6	–.23	–.46/–.00	18.8*
Cystic fibrosis	3	–.00	–.13/+.13	0.0
Anemia	3	–.26	–.50/–.02	2.1
Bowel	3	–.32	–.62/–.02	21.7
Kidney	2	–.15	–.43/+.14	0.3
Seizure	2	–.11	–.56/+.34	0.1
Hearing	2	–.27	–.56/+.02	0.6
Spina bifida	2	–.26	–.58/+.06	2.1
Cardiac	2	+.20	–.10/+.50	0.9
Severity				
Greater	20	–.17	–.25/–.10	47.3*
Lesser	26	–.26	–.34/–.18	84.4*

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

Empirical Classification Approach. Lavigne and Faier-Routman (1992) adopted an empirical approach to evaluating differences across disorders by classifying chronic illnesses post hoc on the basis of their outcomes. Three categories of disorder were identified on that basis. We employed an analogous strategy by partitioning effect sizes at the study level by disease into three categories: (1) negative and statistically different from zero, (2) negative but not statistically different from zero, and (3) positive albeit not significantly different from zero. We then focused on those disorders in categories 1 and 3. In the former category were cancer, diabetes, anemia, and bowel disease. These four diseases can affect day-to-day functioning by requiring intrusive treatment regimes and by restricting school and play activities. In the latter category were cardiac, craniofacial anomalies, and infantile hydrocephalus. These chronic childhood diseases are often treated by surgical intervention and do not necessarily affect daily functioning to the same extent as those illnesses in category 1.

Gender, Birth Order, and Age of Sibling Effects. We attempted to determine whether gender and birth order influenced sibling psychological and social functioning. Approximately half of the studies provided some information relevant to gender effects. Unfortunately, with a few exceptions (e.g., Sahler et

al., 1994), those studies did not provide separate data for male and female participants, reported data selectively, or provided only summary statistics not amenable to meta-analysis. Primary authors often provided a statement of no significant effects for gender. Even less frequently presented was information pertaining to birth order or the combination of gender and birth order.

In the meta-analysis of siblings of children with mental retardation, Rossiter and Sharpe (2001) coded studies for the proportion of male siblings and assessed the relationship between those proportions and effect sizes. We adopted the same strategy in this meta-analysis. The authors of six primary studies did not provide sufficient information to determine the number of male and female siblings. The percentage of male participants in the remaining studies ranged between 30 and 61% (average 47%) with two exceptions; all siblings in Israelite (1986) and Silver and Frohlinger-Graham (2000) were female. The resulting correlation between the proportion of male participants and effect sizes at the study level was not significant (r [42] = .04).

When available, the mean age and the age range of siblings were recorded. When the authors of a primary study failed to provide a mean age for siblings, the midpoint of the age range was used. Across all studies, the youngest siblings were 2 and the oldest were 20 with an average age range of 9.9 years (SD = 3.4). The mean age of participants was 10.8 years (SD = 2.1). There were no significant correlations between the mean age and study-level effect size values (r [47] = –.11),and between the age range and study-level effect size values (r [39] = –.16).

Discussion

This meta-analysis found a statistically significant and negative overall effect for having a sibling with a chronic illness. This finding is consistent with quantitative reviews of the relevant literature that employed vote counts of significant and nonsignificant effects (e.g., Howe, 1993; Summers et al., 1994) and traditional literature reviews (e.g., Faux, 1993; Hannah & Midlarsky, 1985; McKeever, 1983; Packman, 1999; Williams, 1997). The magnitude of this negative overall effect was an effect size of –.20. Cohen's (1988) widely adopted criteria for effect size magnitude places the magnitude of the effect for siblings of children with a chronic illness at the upper limits of a "small" effect size. This effect size was substantially larger than the mean effect sizes reported by Rossiter and Sharpe (2001) for the siblings of children with intellectual disabilities, but much smaller than the mean effect sizes calculated by Lavigne and Faier-Routman (1992) for the psychological adjustment of children with a chronic illness.

Efforts were made to show that the negative outcome for the siblings of children with a chronic illness could not be accounted for by restricting our meta-analysis to published studies. An examination of the pattern of effect sizes from funnel and normal quantile plots, and results from calculation of fail-safe Ns and correlations between sample size and effect size, all serve as evidence against the results being an artifact of publication status. Furthermore, a computer search was conducted of the Dissertations Abstracts computer database using keywords from our computer searches. From the reading of the abstracts, 9 dissertations generated negative outcomes, 15 showed no differences or mixed results, and only 1 dissertation (Gold, 1999) produced a positive outcome for siblings of children with a chronic illness.

To investigate some possible determinants for the negative effect of having a sibling with a chronic illness, we examined a number of potential moderator variables. Methodological moderator variables were examined first. With the correcting of methodological flaws in early studies (Faux, 1993), reduction in mortality rates and improvements in the quality of life for children with chronic illnesses (Jackson, 2000), and the development of effective psychological interventions for children with chronic illnesses (see Kibby, Tyc, & Mulhern, 1998), we anticipated fewer negative findings for siblings in more recent studies. A correlation between effect size and year of publication was modest but in the anticipated direction. What has changed most over the last 30 years is our attitudes toward individuals with disabilities. In contrasting early studies with more recent research, Lamorey (1999) noted "the educational, political, and medical context of disability in the 1960s and 1970s incorporated little of the advocacy, intervention and habilitation efforts, normalization, and inclusion that characterize more current views of disability" (p. 81). A second encouraging result relating to year of publication was that more studies investigating the siblings of children with a chronic illness were published in the last decade than in all previous decades.

The influence of two other methodological moderator variables was considered: parent reports versus

child self-reports and comparison data versus normative data. In the first case, parental reports were decidedly more negative than child self-reports. Children may not perceive any negative effects or may deny such effects until adulthood. Conversely, parents may be overprotective of their children or may be overly sensitive to negative outcomes. Collaborative data from fathers and mothers of the siblings and from unbiased observers are needed to address this question. In the second case, siblings of children with a chronic illness fared better relative to a control group than when compared to normative data. Some authors went to considerable effort to ensure equivalency between sibling and control participants. Silver and Frohlinger-Graham (2000), for example, recruited female sibling and control group participants from the same university medical center and matched for sibling age, gender, birth order, and age spacing.

Given heterogeneous effect sizes and negative effects after partitioning by methodological moderator variables, a number of substantive moderator variables were considered. Classification of dependent measures into discrete categories revealed psychological functioning, peer activities, and cognitive development were associated with negative mean effect sizes. Consistent with Rossiter and Sharpe's (2001) meta-analysis of siblings of individuals with intellectual disabilities, the sibling relationship was the one category associated with a positive though not significant effect size. The sibling relationship is paradoxical, incorporating both conflict and companionship. Although having a sibling with a chronic illness may be associated with difficulties across a number of domains, the sibling relationship may be resilient and perhaps even enhanced in the context of disability.

Consistent with previous reviews of this and related literatures (Howe, 1993; Rossiter & Sharpe, 2001), internalizing behaviors such as anxiety and depression were associated with larger negative effects than were externalizing behaviors. One can only speculate as to why the brothers and sisters of children with a chronic illness respond by internalizing their difficulties. A caretaker role involves the sibling as a quasi-parent, participating in such activities as feeding and dressing their sibling. There is evidence that the caretaking role is elevated when one sibling has a disability (Boyce & Barnett, 1993), and internalizing behaviors may be a response to these inflated caretaking demands (Gold, 1993). Frustrations arising from parental inattention or caretaking responsibilities may not be easily externalized by the healthy sibling into behaviors such as aggression, given the precarious health status of their brother or sister.

A second substantive variable that was considered was the nature of the chronic illness itself. Lavigne and Faier-Routman (1992) suggest that it is not the features of any specific disease that most affect psychological functioning, but rather features that vary across childhood chronic diseases, such as whether the disease is life-threatening. In this study, no difference was found in the functioning of siblings when their brother or sister had a more or less severe (i.e., mortality rates) childhood illness. However, siblings of children that have a chronic illness that affects their day-to-day functioning (e.g., bowel disease, cancer) are more negatively affected than siblings of children less in need of intense, daily assistance (e.g., craniofacial anomalies). Again, this alludes to the central role of caregiving demands and the amount of parental attention required by a child with a chronic illness. There are data available for broad categories of chronic illnesses on days of limited activity, proportion of children unable to engage in activities, number of school absences, and physician contacts (see Newacheck & Halfon, 1998). As better methods of quantifying disease severity are developed, future researchers should investigate further the impact of disease factors on psychological functioning of siblings.

Lavigne and Faier-Routman's (1992) meta-analysis of 87 studies of children with a chronic illness produced results strikingly parallel to our findings from the sibling literature. Lavigne and Faier-Routman found negative effects for overall adjustment and for measures of internalizing behaviors, externalizing behaviors, and self-concept. Larger effect sizes were found for internalizing behaviors over externalizing behavior, and for studies that employed normative comparisons over control groups. Lavigne and Faier-Routman also concluded the risk for psychological problems varied by disease.

Any meta-analysis is limited by the nature and number of primary studies, the data reported, the variables assessed, and the design of those primary studies. All the studies in this meta-analysis were published, so our results should not be generalized to unpublished research. We were unable to report on the effect of variables such as gender and birth order or other moderator variables such as family and

parental functioning (see Lavigne & Faier-Routman, 1993), as this information was not readily available in most reviewed studies. One last limitation is that our examination of moderator variables employed what is fundamentally a correlational technique to evaluate the results from primary studies that assessed preexisting groups. On that basis, we cannot conclude there is a causal relationship between adjustment problems and having a sibling with a chronic illness.

One fear often expressed regarding meta-analysis is that a quantitative review may inhibit future research by prematurely closing an area of inquiry (Boden, 1992). To the contrary, we believe this meta-analysis highlights the need for more, not less, research into the psychological functioning of siblings of children with a chronic illness. We hope that future research continues to employ comparison groups, but also direct observation, longitudinal, and qualitative research designs, nonreactive dependent measures, the reporting of gender and birth order data, and the assessment of parental/familial risk factors. We would also hope that more consideration will be given to features of specific chronic childhood illnesses. There is also the need for studies of adult siblings of individuals with a chronic illness and efforts to seek positive long-term consequences such as greater empathy and a better understanding of individuals with disabilities.

Family dynamics are an intriguing and often complex set of relationships and even more so when a child in a family is born with or develops a chronic physical illness. Families experiencing childhood chronic illness must adapt to caregiving burdens, stress, and anxiety demands. Clinicians working with the families of children with chronic illnesses need to be aware that siblings are at some risk for negative psychological effects. Information sessions and support groups have been shown to enhance children's psychological state, their knowledge of disabilities, and their understanding of the family situation (Wamboldt & Wamboldt, 2000). In a recent meta-analysis, Kibby et al. (1998) found psychological interventions for disease management and emotional/behavioral problems to be effective for children and adolescents with a chronic illness. These programs could be expanded to the siblings and families of children with a chronic illness. The results from this meta-analysis suggest that one focus for interventions should be internalizing behaviors such as anxiety and depression. Future research should explore the effectiveness of these interventions to assist the brothers and sisters of children with a chronic illness.

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*Denotes studies included in the meta-analysis.

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