



# Non-psychopathology Related Outcomes Among Siblings of Individuals with Mental Illness: A Systematic Review

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## Abstract

The current study consists of a systematic review of the quantitative literature on siblings of individuals with mental illness (MI). Despite the prevalence of mental illness, little is known about how siblings are specifically affected in areas of psychosocial, emotional, and behavioral outcomes. The review yielded 56 studies that examined outcomes such as behavior problems, the sibling relationship, caregiving experiences, and knowledge of mental illness among siblings. The majority of studies from the initial search were focused on siblings-as-comparison group, examining siblings for risk factors for developing mental illness. In total, the study samples covered a sibling age range of 6–81 and a patient age range of 4–84. About half ( $k=27$ ) of the included studies had samples primarily composed of siblings of individuals with schizophrenia, leaving other MI diagnoses such as depression, anxiety, and mood disorders underrepresented. However, results from comparison studies were mixed—half found that the MI-Sibs had fewer negative outcomes than the comparison group, and half found that MI-Sibs had more negative outcomes. Multiple factors, including female sibling gender, greater severity of MI symptoms, and belief in the patient’s ability to control their own behavior, were all related to more negative outcomes for MI-Sibs. Future work will focus on expanding the representativeness of MI-Sibs samples and analyzing experiences of both the sibling and the individual with MI.

**Keywords** Mental illness · Siblings · Systematic review · Sibling relationship

Although many researchers have examined the impact of mental illness on the family unit, many studies focus on parents or caregivers (e.g. Corrigan and Miller 2004) rather than siblings of individuals with mental illness (MI-Sibs). Among studies of MI-Sibs, many contain analyses of genetic or environmental risk and sub-clinical symptoms of MI (e.g. Sariaslan et al. 2016) to better identify etiology and early signs of various diagnoses. There are far fewer studies examining the experiences of MI-Sibs who do not have a diagnosis themselves. The current review will summarize the existing quantitative literature on the social, emotional, and behavioral outcomes for siblings of individuals with mental illness across the lifespan. For purposes of the current study, classification of mental illness will exclude neurodevelopmental disorders, as defined by the DSM-V (e.g.,

intellectual disabilities, autism spectrum disorder, attention deficit/hyperactivity disorder, etc.; APA 2013).

## Families of Individuals with Mental Illness

### Families

The family literature shows that a diagnosis of MI can have broad, long-lasting impacts on non-diagnosed family members (e.g. Saunders 2003). Family members can experience increases in caregiving activities across the lifespan, stress from crisis situations, and stigma by association (Saunders 2003). Many studies focus on the challenges of mental illness, with families describing struggles and caution, coping and resilience (Flood-Grady and Koenig Kellas 2018; Zauszniewski et al. 2010). Most such studies use the framework of caregiving and provision of support, highlighting the different types of care and accommodation that family members provide for the individual with MI (McCann et al. 2015). From the perspective of the individual with

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MI, studies have also highlighted the importance of supportive family relationships on the health of the diagnosed individual (e.g., Waller et al. 2019). Together, the literature, on average, suggests that families are important sources of support for individuals with MI, especially when faced with a dearth of formal supports, but that caring for individuals with MI is a “difficult and demanding responsibility” (McCann et al. 2015, p. 203). Because many studies on families overall do not provide separate analyses for siblings, it is not clear to what extent MI-Sibs experience potential caregiving burden or provide support for the individual with MI. It is also not known if there are any benefits, perceived or otherwise, to having a brother or sister with MI.

## Parents

In terms of individual family members of individuals with MI, the majority of research thus far has focused on the experience of parents or caregivers, particularly mothers (Corrigan and Miller 2004). Studies have shown that, compared to mothers of individuals with intellectual and developmental disabilities (IDD), mothers of individuals with MI report higher levels of burden and depressive symptoms (Greenberg et al. 1997). However, mothers with higher levels of optimism and flexibility tend to have fewer negative outcomes, such as depressive symptoms (Greenberg et al. 2004; Seltzer et al. 2004), showing that numerous individual factors can impact outcomes for family members of individuals with MI. Specifically, mothers’ stigmatized appraisals of mental illness (that is, how negatively the mothers view their children’s mental illness) significantly predicted both the child’s symptom severity and life satisfaction over time (Markowitz et al. 2011). Collectively, the literature on parents of individuals with MI suggests that parents can face numerous challenges, but that supportive traits and strategies, including individual appraisals and coping styles, can contribute to more positive outcomes.

## Siblings

Although studies of parents and the entire family unit are important, siblings are likely to have different perspectives than the parents. The sibling relationship is typically the longest relationship a person will have in their lifetime (Cicirelli 1995), and siblings tend to navigate similar developmental stage of life concurrently (e.g. Goetting 1986). The life course perspective (e.g. Settersten 2003) therefore suggests that a large event, such as the diagnosis of MI, for one sibling, can result in life course alterations for all other siblings. These potential alterations are not well understood among siblings. Early literature focused on a deficit model, identifying increased risk for MI diagnosis among siblings in adulthood (e.g., Heston 1966; Lidz 1963) and increased

risk for emotional, social, and behavioral impairments (e.g., DeLisi 1987; Weismann and Seigel 1972, both adult samples).

However, like many areas of human research, more recent studies have examined a broader sample of sibling experiences, with several studies collecting qualitative data from siblings themselves, discussing their own stories and opinions. Many adult siblings feel reluctant in regards to providing care for their brother or sister with MI, and the sibs’ tolerance of the brother or sister’s behavior could be related to the siblings’ interpretation of said behavior (Johnson 2000). That is, if the siblings believe that the individual with MI is being lazy or stubborn, then the siblings are less sympathetic toward their brother or sister, as opposed to siblings who interpret the behavior as part of the illness (Johnson 2000). Reflecting on their childhood experiences, some adult siblings have described physically removing themselves from difficult situations with their brother or sister with MI when they were children—keeping busy through extracurricular activities or simply going to their room at home (Kinsella et al. 1996). Siblings report similar distancing strategies in adulthood, particularly if the family is disorganized in response to the brother/sister’s mental health crises (Graves et al. 2020).

Kovacs et al. (2019) propose the theory of relational dialectics (Baxter 2004) and the concept of ambiguous loss to help understand the varied experiences of MI-Sibs. The dialectical tensions between stressors that arise from a brother or sister’s mental illness and the adjustment and resilience MI-Sibs develop can contribute to varying descriptions and results in studies of MI-Sibs (Kovacs et al. 2019). Additionally, ambiguous loss can be used to describe experiences with a family member who is still living, but whose mental illness can contribute to the loss of former roles and relationships (Kovacs et al. 2019). The theories results from such qualitative studies are enlightening and beneficial. The current review will examine quantitative studies of sibling outcomes to determine how much quantitative results from studies of MI-Sibs’ emotional, behavioral, and social outcomes align with the way siblings speak about their own experiences in qualitative research.

## Factors that May Affect Sibling Experiences

### Diagnosis

“Mental illness” is, in and of itself, an incredibly broad category. Therefore, the experiences of siblings of individuals with schizophrenia may be very different than the experiences of individuals with anorexia. Further, the experiences of two siblings of two different individuals with schizophrenia can be substantially different from each other. In the

literature, many studies focus on “serious” or “severe” mental illness (SMI), categories that are defined in the United States as disorders that meet DSM criteria and result in impairment that interferes with at least one major life activity (National Institute of Mental Health 2013). Although the official definition does not exclude any specific diagnoses, studies of families of individuals with SMI often include individuals diagnosed with schizophrenia, schizoaffective disorder, or psychosis (e.g. Erickson et al. 1998; Greenberg et al. 1997). Thus, there seem to be fewer studies on family experiences of individuals with eating disorders, personality disorders, or major depressive disorder, despite the fact that such diagnoses can also result in impairment that meets the definition of “severe.” Studies may use the general definition of SMI as an inclusion criterion or may focus on families of individuals who have been hospitalized due to mental illness (e.g. Gerson and Rose 2012). Although such studies are certainly necessary to understand the sibling and family experience, they do not reflect the full spectrum of outcomes for siblings. Thus, it is important to also consider siblings of individuals with other MI diagnoses, including other symptoms and other levels of severity.

## Demographics

Beyond diagnosis and severity, demographic factors can certainly also play a role in sibling outcomes. Studies have shown that African-American or Latino siblings tend to be more involved in caregiving due to the more communal cultural expectations (Guarnaccia and Parra 1996; Horwitz and Reinhard 1995, both adult samples). Gender also plays a role, as adult female siblings report providing more emotional support than male siblings (Greenberg et al. 1999), and greater overall perception of burden than male siblings (Greenberg et al. 1997). The gender of the brother or sister with MI may also play a role, as adult siblings of sisters with MI reported higher levels of psychological well-being than siblings of brothers with MI, as compared to siblings of individuals with IDD or siblings of typically-developing individuals (Taylor et al. 2008).

## Interventions

Sibling support groups are one of the many areas in which services have outpaced literature. Across the country, variations on support groups and services for siblings of individuals with intellectual disability, chronic illness, or mental illness exist to provide information and opportunities for understanding from other siblings (e.g., Meyer and Vadasy 1994). Organizations such as the National Alliance on Mental Illness (NAMI) provide peer-led activities for family members of individuals with MI ([www.nami.org](http://www.nami.org)), and several organizations provide pamphlets and articles

online (Griffiths and Sin 2013). However, a recent review found that the vast majority of empirical studies on such support groups for younger individuals (i.e., under 18) focus on siblings of individuals with physical illness or disability and/or intellectual and developmental disabilities rather than siblings of individuals with mental illness (Smith et al. 2018). The review concluded that such interventions resulted in improvements in behavior and knowledge of illness and disability (Smith et al. 2018), suggesting that the presence of support programs or interventions may be beneficial to MI-Sibs, as well. The availability of these programs could have a positive impact on the overall sibling experience.

## The Current Study

The current study aims to review the extant quantitative literature on psychosocial and behavioral outcomes among typically-developing siblings of individuals with mental illness (e.g., siblings without a mental illness themselves), age 5 and older. The review will describe the demographics of the existing studies—gender and age of the siblings and individuals with MI, makeup of any comparison groups, nature of the brother/sister’s MI—as well as the outcomes being explored, covering all peer-reviewed studies available in English through 2019. The goal is to determine what is currently known about the experiences of typically-developing MI-Sibs and what are the gaps in the literature to help guide future sibling and family researchers.

## Methods

The present study utilized Preferred Reporting Items for Systematic Reviews and Meta Analyses (PRISMA; Moher et al. 2009) guidelines to conduct a systematic review of all peer-reviewed articles published up through December of 2019. Because no such review of MI-Sibs has been published to date, there was no limit on the earliest date of publication.

## Eligibility

Inclusion criteria for the current review were defined as follows: (a) peer-reviewed, published articles, (b) available in English, (c) articles describe quantitative results from empirical studies, (d) article describes results for non-diagnosed siblings of individuals with MI (if the article included a mixed sample of MI-sibs and non-MI-Sibs, at least one of the results had to be described separately—that is, exclusively for MI-Sibs), (e) at least one of the results describes psychological, behavioral, or emotional outcomes for the MI-Sibs that were not diagnostic in nature. That is, studies the solely examined MI-Sibs for the purpose of assessing

risk for developing MI or identifying sub-clinical symptoms or early signs of MI were not eligible. Studies were excluded if (a) the purpose of the study was to measure risk for developing MI among MI-Sibs, (b) the study did not report separate analyses for siblings (e.g. the sample included parents and/or other family members), or (c) “sibling” was only used as a single predictor variable in regression analyses. Several studies did include a subsample of siblings among the sample of family members in general, but exclusive reports of sibling outcomes were not given; “sibling” was only used as a categorical variable in regression analyses (e.g. comparing the predictive value of being a sibling as opposed to a parent, spouse, or child of the individual with MI).

## Literature Search

Three large, online databases were searched: PsychInfo, EBSCOHost, and Web of Knowledge. The search was limited to peer-reviewed articles published through December of 2019 and used the following Boolean search terms: (sibling\* OR brother OR sister) AND (“mental illness” OR “psychopatholog\*” OR depression OR anxiety OR bipolar OR schizophrenia OR “self harm” OR “personality disorder” OR “axis 1” OR “axis 2” OR “OCD”).

Titles were reviewed, yielding a total of 388 unique articles to be examined by abstract and/or full manuscript for further inclusion. Due to the broad range of potential MI diagnoses, the references of initially identified studies were examined for articles that may not have been included in the original search; this process yielded an additional 3 articles that were included in the review. Both authors reviewed the abstracts of the 391 total articles to determine if quantitative results were reported. This process narrowed down the list of eligible articles to 67; once coding was conducted, a further eleven manuscripts were excluded, either because “sibling” was only used as a predictor category in regression analyses or because the purpose of the study was to identify risk factors for MI, not understand the sibling experience. The final result was 56 eligible articles for inclusion. The flowchart for study inclusion can be found in Fig. 1.

## Data Extraction

Studies that met all inclusion criteria were read by the authors to extract information pertinent to the review. Data extraction was based on the PICOS method (Population, Intervention, Comparison, Outcomes, Study Design; Higgins et al. 2019); however, because the goal of the review was to synthesize all existing quantitative literature on MI-Sibs that did not exclusively assess MI risk, interventions (I) were not required. Coding manuals were completed for each of the 56 included articles with all available information on participant demographics (siblings, individuals with

MI, and any comparison groups), study methods, and results. Because of the wide range of study methods and purposes, results were split into two categories: between-group results (results comparing MI-Sibs to other samples) and within-group results (descriptive analyses or analyses describing statistical relationships among two or more variables; this category included regression analyses). Each category included a wide range of outcome measures and related variables. The two authors met frequently to compare codes, and any discrepancies were discussed and examined until the authors reached a consensus.

To assess study quality, the assessment rubric for quantitative studies developed by Kmet et al. (2004) was used. The rubric includes 14 assessment items (e.g., “Design evident and appropriate to answer study question,” “Analysis described and appropriate”) that are each rated on a 3-point scale (0 = no, 1 = partial, 2 = yes). Average quality scores are calculated by summing the total score and dividing by the highest possible score (number of relevant items  $\times$  2); thus, each quality score is represented as a decimal between zero and one, with higher numbers indicating greater article quality.

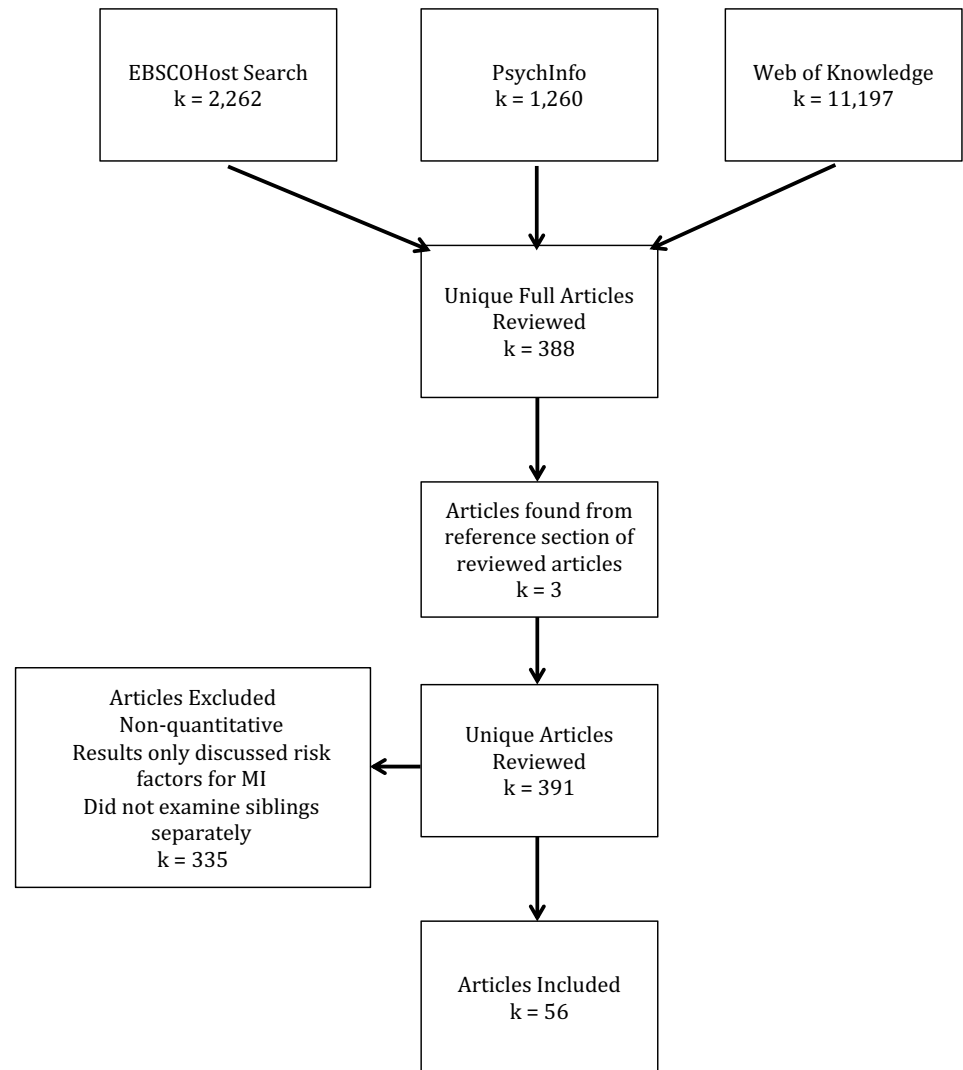
## Results

Our review yielded a total of 56 peer-reviewed, quantitative studies that analyzed social, emotional, and behavioral outcomes for MI-Sibs, reporting on a variety of outcomes for siblings.

## Population

The search yielded 30 studies that included a comparison group and 26 studies that had no comparison group (two of the 26 studies with no explicit comparison group compared the study sample to population norms of the included measures, and one compared its sample to data from a previously published study). Sample sizes for the MI-Sibs ranged from 9 to 746; sample sizes for the individuals with MI ranged from 11 to 746. The majority of studies ( $k=41$ , 73.2%) focused on adult siblings and the studies had more female siblings than male siblings (as is common in general sibling research). Four studies (7.8%) described multiple sibling samples, either from different study locations or in different treatment groups. These samples are described on separate lines in the demographics tables (Tables 1 and 2). The majority of studies (62.5%;  $k=35$ ) included adult samples; eight studies (14.3%) reported on child samples (i.e., every sibling participant was under the age of 18), and eight (14.3%) reported on samples that included both child and adult sibling. The remaining five studies (8.9%) did not include sufficient information about the age range

**Fig. 1** Flowchart of article search and inclusion



to determine whether individuals under 18 were included. However, the overall demographics of the total sample were difficult to obtain. Seven (12.5%) studies did not report the gender breakdown, and three (5.4%) of the studies did not report the age breakdown of the sibling samples; 18 (32.1%) did not report the gender of the individuals with MI, and 17 (30.4%) gave no information on the age of the individuals with MI.

In terms of the diagnoses of the individuals with MI, seventeen (30.4%) studies had samples focusing on a singular MI, while thirty-nine (69.6%) contained multiple MI diagnoses. The proportion of studies that were, at least in part, focused on siblings of individuals with schizophrenia was overwhelming. Nine (16.1%) reported schizophrenia as the singular MI in their study. Of those studies with mixed samples, eighteen (32.1%) reported schizophrenia as the majority sample in their article. Eleven (19.6%) reported schizophrenia as the minority or unspecified amount in their study, resulting in a total of 38 samples (67.9%) that included at

least some siblings of individuals with schizophrenia. Other diagnoses reported in the studies included eating disorders, personality disorders, mood disorders, obsessive/compulsive disorder, major depressive disorder, and generalized anxiety disorder.

## Methods

The included studies utilized a variety of methods, the most common of which was self-administered questionnaires ( $k = 42$ , 75.0%). Other methods included structured or semi-structured interviews (both in person or over the phone), clinical observations, use of data from medical records, neuropsychological test batteries, and secondary analysis of data from both longitudinal and cross-sectional studies. Four (7.8%) of the studies reported on interventions for MI-Sibs and family members, analyzing change in measured variables from pre-intervention to post-intervention. Possibly because so many studies focused on adult siblings (rather

**Table 1** Demographics for single-sample studies

	Siblings			Individuals with MI			
	N	Mean age (SD)	Gender (% Female)	N	Mean Age (SD)	Diagnosis	Gender (% Female)
Avcioglu et al. (2019)	103	37.14 (11.16)	42.7%	103	NR	Schizophrenia	NR
Bowman et al. (2014)	157	21.76 (4.38)	47.80%	123	21.45 (3.51)	First episode psychosis—schizophrenia (41.5%), schizophreniform (38.2%), schizoaffective (4.9%), bipolar affective disorder (10.6%), post-partum psychosis (0.8%)	29.30%
Dia and Harrington (2006)	65	9.9 (3.2)	58%	65	11.4 (2.5)	Anxiety disorder	45%
Friedrich et al. (2008)	746	39.7 (10.5)	74%	746	39.3 (10.0)	Schizophrenia	33%
Greenberg et al. (1997)	164	45 (nr)	70%	39		Schizophrenia or schizoaffective (68%)	50%
Hoover and Franz (1972)	57			30		schizophrenia	
Horwitz (1993)	108	40 (nr)	56.50%	85		Schizophrenia (80%), bipolar (15%)	40%
Horwitz (1994)	108	40 (nr)	56.50%	85	–	Schizophrenia (80%), manic depression (15%)	40%
Horwitz and Reinhard (1995)	70	37 (nr)	63%	70	35.5 (9.31)	Schizophrenia (51%), major depression (21.5%), bipolar (8.5%), psychotic disorder—unspecified (8%), “other” (11%)	41%
Jewell and Stein (2002)	111	38 (8.8)		111	37 (7.7)	Schizophrenia or schizoaffective (71%)	17%
Kageyama and Solomon (2019)	113	41.5 (11.7)	57.7%	113	40.9 (10.9)	Schizophrenia	33.6%
Landeen et al. (1992)	88	29	61%				
	22	29	61			Schizophrenia	
Laporte et al. (2011)	56	30.2	100%	56	28.7	Borderline Personality Disorder	100%
Leith et al. (2018)	242	40.7 (9.8)	81.80%	242	40 (8.94)	Schizophrenia/schizoaffective (77%)	33%
	103	38.9 (13.8)	78.6%%	103	38.4 (13)	Schizophrenia/schizoaffective (64%)	36%
Leith and Stein (2012)	103	38.9 (13.8)	78.60%	103	38.4 (13)	Schizophrenia (46%), schizoaffective (18%), mood disorder (18%), MDD (9%)	36%
Lively et al. (1995)	30	37	50%	22	37	Schizophrenia	31.80%
Lively et al. (2004)	752	39.7	73.70%	752	39.4 (10.1)	Schizophrenia	32.90%
Lohrer et al. (2002)	100	43.8 (14.2)	81%		44.6 (12.6)	Schizophrenia (80%)	31%
Lohrer et al. (2007)	156	44.5 (12.1)	76.30%	156	43.9 (11.3)	Schizophrenia or schizophrenia spectrum (84%)	34.60%
Sanders and Szymanski (2012)	30	43 (12.5)	53.30%	34		Schizophrenia (56%), bipolar (26%), anorexia, mental retardation, BPD, major depressive disorder, generalized anxiety disorder	
Sanders and Szymanski (2013a)	33	19.33 (9.26)	54.50%	37		Schizophrenia (60%), bipolar (21%), anorexia, BPD, major depressive disorder, generalized anxiety disorder	



**Table 1** (continued)

	Siblings			Individuals with MI			
	N	Mean age (SD)	Gender (% Female)	N	Mean Age (SD)	Diagnosis	Gender (% Female)
Smith and Greenberg (2008)	136	44.5 (8.7)	55%	136		Schizophrenia or schizoaffective disorder	
Smith et al. (2007)	137	44.5	56.20%	137	43.6	Schizophrenia/schizoaffective disorder	26.30%
Smith et al. (2016)	41	23.5 (4.6)	53.70%	41		Schizophrenia	
Tanaka (2008)	130			130		Schizophrenia (and spectrum), bipolar disorders, major depression	
Tanaka (2011)	130			130		Schizophrenia (and spectrum), bipolar disorders, major depression	
van Dijk et al. (2019)	309	35 (8.0)	55.4%	259	26.3%	Schizophrenia (78.4%), schizoaffective disorder (10.4%), other psychotic disorder (10.4%)	34 (8.0)

Laporte et al. (2011) compared sibling perceptions of parenting to those of their brother/sister w/MI

than child or adolescent siblings), the majority of studies used sibling self-report ( $k = 52$ , 92.9%). Other methods included parent/caregiver report and researcher observation.

## Study Results

### Between-Group Results

Of the studies that compared MI-Sibs to another sample, nine found the MI-sibs to be “worse” than the comparison group, that is, the MI-Sibs had significantly higher scores on measures of ill-being [e.g., more depressive symptoms (Latzer et al. 2015); poorer sibling relationships (Tschan et al. 2019)], while four studies noted the MI-sibs results were not significantly different than the comparison group on measures of expressed emotion (Moulds et al. 2000), temperament (Kelvin et al. 1996), internalizing or externalizing problems (Hudson and Rapee 2002), or quality of life (Tatay-Montiega et al. 2019). Nine studies found that the MI-sibs were had “better” results than the comparison group [e.g., less emotional distress (Zauszniewski and Bekhet 2014), less sibling conflict (Jacoby and Heatherington 2016)]. However, the composition of these comparison groups was widely varied. Of the comparison groups that reported “worse” outcomes than MI-Sibs (that is, the sibs were doing “better” than the comparison group), three were comprised of MI-Parents (that is, the studies found that MI-Sibs were less severely affected than MI-parents), four were comprised of typically-developing siblings or community samples, and one was comprised of siblings of individuals with intellectual disability. The final group was an intervention study, which found that siblings in the treatment group fared better than siblings in the “treatment as usual” group.

In contrast, the majority of comparison samples in which the comparison group was doing “better” than the MI-Sibs were composed of community samples (e.g. Barnett and Hunter 2012) or siblings of typically-developing individuals (TD-Sibs; e.g. Deal and MacLean 1995).

In terms of findings, the comparison results were decidedly mixed. Some studies found worse sibling relationships for MI-Sibs (Barak and Solomon 2005; Fox et al. 2002), while others found lower levels of negativity in the sibling relationship for MI-Sibs (Deal and Maclean 1995; Jacoby and Heatherington 2016). MI-Sibs were reported to have higher rates of behavior problems (Barnett and Hunter 2012; Deal and MacLean 1995) in some studies, but lower levels of internalizing behaviors (as reported by fathers, but not mothers) in another study (Barrett et al. 2005). Some studies found poorer emotional outcomes for MI-Sibs in relation to comparison groups (Latzer et al. 2015), while others found no differences on temperament (Kelvin et al. 1996). A full description of comparison results can be found in Table 3.

### Within-Group Results

The single most common result from the included studies is that female MI-Sibs have more negative outcomes than male MI-Sibs (e.g. Bowman et al. 2014; Greenberg et al. 1997). A total of 11 studies examined gender as a contributing factor, and every one reported more negative outcomes for female MI-Sibs and/or more caregiving responsibility for female MI-Sibs. Additionally, severity of the brother/sister’s symptoms was consistently found to be related to higher rates of caregiving (e.g., Horowitz 1993) and poorer outcomes for MI-Sibs (e.g., Bowman et al. 2014; Tanaka 2011). Additionally, increased MI-Sib belief in the patients’ ability to control

**Table 2** Demographics for studies with comparison samples

	Siblings		Individuals with MI		Comparison Group			Gender (% female)		
	N	Mean age (SD)	N	Mean age (SD)	N	Mean age (SD)	Group composition			
	Gender (% female)	Diagnosis	Gender (% female)	Diagnosis	Gender (% female)	Diagnosis	Diagnosis			
Amaresha et al. (2018)	34	29.42 (4.85)	34	30.78 (6.55)	29	30.63 (nr)	Siblings in "treatment as usual" group—waitlist	45%	68%	
Barak and Solomon (2005)	52	-	52	-	48	-	-	-	-	
Barnett and Hunter (2012)	75	11.35 (2.53)	75	11.52 (3.36)	75	11.52 (3.36)	ASD (5), disruptive behavior disorders (31), mood disorders (13), anxiety disorders (14), adjustment disorders (6), eating disorder (6)	*Community sample for CBCL, normative self-concept, normative sample for QoL	45%	
Barrett et al. (2005)	29	10.72 (2.81)	33	10.28 (1.73)	33	10.28 (1.73)	Anxiety disorder (GAD, 60.6%, separation anxiety, 24.2%; social phobia, 9.1%; specific phobia 6.1%)	"Non-clinical control children"	50%	77.7%
Barrett et al. (2004)	13	11.15 (2.38)	24	10.75 (2.54)	24	10.75 (2.54)	OCD (individual CBFT)	Siblings of individuals in the waitlist condition	50%	77.7%
"	10	12.36 (2.92)	29	12.90 (2.30)	29	12.90 (2.30)	OCD (group CBFT)		55%	
Chen and Lukens (2011)	32	42.38 (10.38)	137	34.9 (9.22)	137	34.9 (9.22)	Schizophrenia or affective disorder	41% (43) live with care recipient, recipients' duration of illness 10.64 years	40.10%	87.60%



Table 2 (continued)

	Siblings			Individuals with MI			Comparison Group				
	N	Mean age (SD)	Gender (% female)	N	Mean age (SD)	Diagnosis	Gender (% female)	N	Mean age (SD)	Group composition	Gender (% female)
Deal and MacLean (1995)	15	11.4 (2.6)	46.60%	15	15.2 (1.9)	Major depression (4), major depression/poly-substance disorder (3), intermittent explosive disorder (2), oppositional defiant disorder (2), major depression/PTSD (1), dysthymia (1), conduct disorder (1), adjustment disorder w/ mixed disturbance of mood and conduct (1)	33%	15	11.3 (1.8)	Younger sibs of adolescents who had not been psychiatrically hospitalized	46.60%
Fox et al. (2002)	36	10.4 (2.75)		36	10.1 (1.77)	GAD (55.6%), separation anxiety (25%), social phobia, (13.9%), specific phobia (5.6%)		15	10.3 (2.99)	Nonclinical sibs, 60% older than pair sibling	
Greenberg et al. (1999)	61	41.9 (nr)	54.10%	61	39.1 (nr)	Diagnosis of "serious mental illness", schizophrenia (72%)	24.60%	119	42.9 (nr)	Sibling of individuals with "mental retardation"	63.90%
Horwitz et al. (1992)	109	35 (nr)	61%	109	33 (nr)	"Severely mentally disabled"	38%	300	NR	Parents, spouses, or children of individuals with MI	NR
Hudson and Rapee (2002)	37			37		GAD (38%), Social phobia (32%), separation anxiety (19%), panic disorder (5%), OCD (3%), specific phobia (3%)		20		Non-clinical families	

**Table 2** (continued)

	Siblings			Individuals with MI			Comparison Group			
	<i>N</i>	Mean age (SD)	Gender (% female)	<i>N</i>	Mean age (SD)	Diagnosis	Gender (% female)	<i>N</i>	Mean age (SD)	Group composition
Jacoby and Heath-erington (2016)	64	19.31 (1.27)	64%	64	20.63 (4.29)	GAD (81%), OCD (22%), panic disorder (17%), panic w/ agoraphobia (11%), agoraphobia (9%), social phobia (47%), specific phobia (8%)	51.50%	309	18.89 (1.10)	participants who didn't endorse brother/sister anxiety
Kelvin et al. (1996)	31	58% > 12	41.90%	29	76% > 12	Major depression (MDD; 21) or anxiety (8)	65.50%	40	80% > 12	“Community sample” obtained from family practitioners; matched for age and sex with every 3rd proband w/ depression. Must be free of psychiatric disorder & physical illness
Latzer et al. (2015)	30	21.68 (4.17)	100%	30	20.67 (4.48)	Anorexia nervosa (10), bulimia nervosa (10), ED not otherwise specified (10)	100%	30	21.68 (4.17)	100%
Moulds et al. (2000)	19			19	20.9	Anorexia nervosa	100%	38		Mothers and fathers of patients/sibs
Oliver et al. (1987)	12	19.02 (2.31)	34%	12		Depression		16		Siblings of nondepressed individuals
*Östman et al. (2005)	80	NR	55%	623	40	Psychoses (42%), affective disorders (39%), “other diagnoses” (19%)	58%	339	NR	Spouses, parents, or children of individuals w/ MI

Table 2 (continued)

	Siblings		Individuals with MI		Comparison Group		
	N	Mean age (SD)	Gender (% female)	N	Mean age (SD)	Gender (% female)	Group composition
**Samuels and Chase (1979)	11	33.5	72.70%	11		81.80%	
**Sanders and Szymanski (2012)	30	43 (12.5)	53.30%	34			
Sanders and Szymanski (2013b)	33	44 (14.5)	54.50%				“Participants who did not experience trauma”
Sanders et al. (2014)	33		54.50%				“Participants who had at least one sibling, but no siblings diagnosed with mental illness”
Seltzer et al. (1997)	61	41.8	54.10%	61			Sibling of individuals with IDD

**Table 2** (continued)

	Siblings			Individuals with MI			Comparison Group				
	<i>N</i>	Mean age (SD)	Gender (% female)	<i>N</i>	Mean age (SD)	Diagnosis	Gender (% female)	<i>N</i>	Mean age (SD)	Group composition	Gender (% female)
Sin et al. (2016)	90	27.52 (8.41)	85.60%	90	26.46 (7.9)	First episode psychosis	37.80%			**All comparisons were made to external samples (listed in results)	
Tanaka (2010)	68	40 (12)	84%	130	42 (13)/46 (13)	Schizophrenia (and spectrum), bipolar disorders, major depression	31%/35%	62	47 (14)	Japanese siblings	68%
Tatay-Mantiega et al. (2019)	23	41.5 (11.8)	69.6%	48	44.21	Bipolar disorder (early- and late-stage)	52.1%	21	36.7 (10.9)	Healthy controls	66.7%
Taylor et al. (2008)	83	63.87 (4.42)	55.40%	83	62.18 (4.95)	Depressive disorder and/or anxiety disorder (85.5%), episodic mood disorder (8.4%), schizophrenic disorders (3.6%), alcohol dependence syndrome (2.4%)	66.30%	268	63.93 (4.76)	Siblings of individuals with Mild ID (IQ of 85 or below)	54.50%
" "								791	64.03 (4.42)	Siblings of individuals with no MI or ID	52.50%
*Tschan et al. (2019)	32	16.88 (4.02)	60.3%	89	16.18 (1.62)	Non-suicidal self injury (56), other MI (33)	100%	41	16.88 (4.02)	Siblings of individuals with no MI	60.3%
**van Langenberg et al. (2016)	46	16.4 (3.5)	60.90%	38	16.1 (1.8)	Anorexia nervosa or eating disorder not otherwise specified—AN type (36%)	92.1%				

Table 2 (continued)

	Siblings			Individuals with MI			Comparison Group			
	N	Mean age (SD)	Gender (% female)	N	Mean age (SD)	Diagnosis	Gender (% female)	N	Mean age (SD)	Group composition
Wolfe et al. (2014)	235	38.3 (4.4)	49%	97	37.0 (4.7)	Bipolar disorder (48%), schizophrenia (25%), major depression (24%)	52%	337	37.3 (4.7)	Siblings of individuals with developmental disabilities
" "								17,126	37.8 (4.5)	Population comparison sample
*Zauszniewski and Bekhet (2014)	14	46.28 (11.71)	100%	60	37.75 (13.96)	Anxiety disorder (1), bipolar disorder (27), depression (5), schizophrenia (27)		46	46.28 (11.71)	Mothers or other female relatives

\*Articles denoted with a single asterisk reported combined demographics (e.g., gender breakdown and age means for the combined MI-Sib and comparison groups)

\*\*Samuels and Chase (1979) compared their data to results from the Hoover and Franz (1972) study; Sanders and Szymanski (2012) and van Langenberg et al. (2016) compared their samples to measure norms

their behavior was related to more negative sibling outcomes (e.g. Greenberg et al. 1997; Smith et al. 2007).

Numerous outcomes were examined by within-group analyses, with the two most common being mental health of the MI-Sib and family functioning. Although the current review did not include studies that exclusively focused on rates of psychopathological diagnoses among MI-Sibs, many studies still included continuous measures of mental health outcomes, such as depression and anxiety (e.g. Barak and Solomon 2005; Lively et al. 1995). The MI-Sibs' own mental health was related to the severity of the brother/sister's symptoms (e.g., Lively et al. 1995), as well as the brother/sister's duration of illness (van Langenberg et al. 2016). Seven studies measured aspects of family functioning, with most finding that poorer family functioning was related to more negative MI-Sib outcomes, including greater levels of internalizing behavior problems and overall impairment (Dia and Harrington 2006; Hoover and Franz 1972). Full summaries of within-group results can be found in Table 3.

### Over Time Results

Most of the studies that reported results over time analyzed the impact of intervention programs, with one exception; Barak and Solomon (2005) had siblings report their own perceptions of how the sibling relationship had changed over time. MI-Sibs reported that their relationship with the patient had gotten worse, while TD-Sibs reported their sibling relationships had improved (Barak and Solomon 2005). Of the intervention studies, the results were mixed. Two studies reported positive outcomes of the interventions, with MI-Sibs in the treatment groups reporting increases in knowledge of MI (Amaresha et al. 2018; Landeen et al. 1992) and decreases in self-stigma (Amaresha et al. 2018). In contrast, the other two studies reported no effect of treatment (Barrett et al. 2004; van Langenberg et al. 2016), though both treatment and comparison groups in the Barrett et al. (2004) study reported decreases in depression and accommodation of the patient over time.

### Article Quality

The article quality varied greatly, ranging from 0.364 to 0.955, with an average quality score of 0.674 (SD=0.154). The middle 50% of values ranged from 0.556 to 0.788, and only 4 articles had quality scores greater than 0.90. Just over half of the studies (53.6%,  $n = 30$ ) scored full points for "study design is evident and appropriate," but only six studies (10.7%) adequately described "method of subject/comparison group selection." Additionally, 11 studies (19.6%) did not control for confounding at all (i.e., were rated 0 for that item), and four studies (7.1%) did adequately

**Table 3** Summary of results of included studies

Authors	Method	Reporter	Between-Group Results	Within-Group Results
*Amaresha et al. (2018)	Semi-structured interview	Sibling	Treatment group showed greater gains in knowledge and decreases in self-stigma	Sib well-being was related to burden (-); maternal warmth (+), rejection (-), and overprotection (-); father warmth (+), rejection (-), and overprotection (-), social support (+), problem-focused coping (+) and emotion-focused coping (-)
Aviciglu et al. (2019)	Questionnaires	Sibling		
Barak and Solomon (2005)	Questionnaires	Sibling	MI-Sibs reported more negative feelings toward their brother/sister, less sibling closeness, less parental closeness, and more shame than TD-Sibs. MI-Sibs reported more sensitivity to others and greater use of problem solving coping than TD-Sibs	
Barnett & Hunter (2012)	Questionnaires	Sibling and/or caregiver	Parents reported significantly more behavior problems among sibs than norms. Sibs reported lower QoL than norms	Sib behavior problems were related to family functioning (-), global self-worth (-), and family rigidity (+)
*Barrett et al. (2005)	Questionnaires, semi-structured interviews, & clinical observation	Sibling and parents	By father report (but not mother), MI-Sibs had lower levels of internalizing problems than nonclinic children	
Barrett et al. (2004)	Questionnaires & semi-structured interviews	Sibling	No main effect of treatment for any outcomes	
Bowman et al. (2014)	Questionnaires, medical file review	Sibling		Differences by sib age and gender—Older brothers reported greater QoL than younger sisters; younger brothers reported better psychological QoL than younger sisters. Sibs of patients with no history of suicide attempts or violence reported greater physical, psychological, social, and environmental QoL. Patient history of violence predicted sibling physical, social, and environmental QoL (-)
Chen and Lukens (2011)	Archival data from structured interviews	Sibling	Compared to MI-Parents, MI-Sibs reported less pride in the patient, but greater levels of well-being and fewer depressive symptoms	



Table 3 (continued)

Authors	Method	Reporter	Between-Group Results	Within-Group Results
Deal & MacLean (1995)	Questionnaires & semi-structured interviews	Sibling and parents	Parents of MI-Sibs reported more sibling total behavior problems, internalizing problems, withdrawal, somatic problems, anxious/depressed behavior, and social problems and less total competence and social competence than parents of TD-Sibs. MI-Sibs self-reported more general anxiety, physiological anxiety, and worry/oversensitivity than TD-Sibs, but less sibling rivalry and parent partiality	
Dia and Harrington (2006)	Questionnaires	Parent		MI-Sib internalizing behavior was predicted by family interference (+); total sib behavior problems were predicted by length of patient's treatment (+)
Fox et al. (2002)	Questionnaires & structured interview	Sibling, parent, researcher	MI-Sibs reported more conflict in the sibling relationship than TD-Sibs; in conversation, MI-Sibs demonstrated more controlling language toward their brother/sister than did TD-Sibs	
Friedrich et al. (2008)	Questionnaires	Sibling		MI-Sibs reported need for patient services, opened communication, and emotional support from loved ones. Understanding schizophrenia was reported as the most useful types of coping, while denial and distancing were reported as the least helpful. Communication and availability to answer questions were ranked as important needs from mental health services
Greenberg et al. (1997)	Phone interviews	Sibling		Global subjective burden was related to being female (+), education (+), sibs' own psychological symptoms (+), belief that the patient has control over their symptoms (+), age (-), and being younger than the patient (+)

**Table 3** (continued)

Authors	Method	Reporter	Between-Group Results	Within-Group Results
Greenberg et al. (1999)	Questionnaire	SSibling	MI-Sibs were less likely to plan to provide caregiving than IDD-Sibs	Current instrumental support was related to the patient living with parents (+), the sib having children of their own at home (-), geographic distance (+), and sibling closeness during adolescence (+). Emotional support was related to the sibling being female (+), the sib having children of their own at home (-), and sibling closeness during adolescence (+)
Hoover and Franz (1972)	Interviews & researcher ratings	Researcher		MI-Sibs reported low levels of functional impairment and moderate levels of family entanglement; impairment and entanglement were significantly correlated (+)
Horwitz (1993)	Phone interviews	Sibling		Total help to patient was predicted by severity of MI (+), distance from patient (-), having deceased parents (+), and number of sibs in the study (-). Black and Puerto Rican siblings reported more total help
Horwitz (1994)	Phone interviews	Sibling		Sib help for the individual with MI was related to reciprocity (+), symptom severity (+), having one living parent (+), and number of sibs in the study (-); sib willingness to help was related to reciprocity (+), distance from brother/sister (-), and number of sibs (-); hypothetical help was related to feelings of obligation (+), reciprocity (+), and how much disruption the patient's illness caused (-)
Horwitz and Reinhard (1995)	Interviews	Sibling		Black sibs reported more caregiving; white sibs reported more caregiving burden. Burden was also related to stigma (+) and caregiving duties (+)
Horwitz et al. (1992)	In-person or phone interviews	Sibling	Sibs report less involvement and assistance than parents, spouses, and children	Involvement was related to the sibling relationship (+) and perceived patient need (+)
Hudson and Rapee (2002)	Questionnaires & observation	Sibling and parents	No differences between MI-Sibs and nonclinic sample on any measures	

Table 3 (continued)

Authors	Method	Reporter	Between-Group Results	Within-Group Results
Jacoby and Heatherington (2016)	Questionnaires	Sibling	MI-Sibs reported less sibling conflict than TD-Sibs	Sibling warmth was related to the sibs' perceptions of their brother/sisters illness stability (-). Sibling conflict was predicted by the sibs' perceptions of their brother/sister's responsibility for the brother/sister's anxiety (+)
Jewell and Stein (2002)	Questionnaires	Sibling		MI-Sibs' current level of provided support for their brother/sister was correlated with willingness and intention to provide support in the future (-)
Kageyama and Solomon (2019)	Questionnaires	Sibling		About half of siblings experienced or witnessed violence or aggression; younger age of sib and patient were positively related to witnessing/experiencing violence, as was male gender of sib/patient
Kelvin et al. (1996)	Questionnaires	Parent	MI-Sibs without a diagnosis of their own did not differ from community comparison group on any scale of temperament	Emotionality was significantly related to siblings' psychopathology (+) in the MI-Sib group
*Landeem et al. 1992	Questionnaire	Sibling		MI-Sibs' knowledge of schizophrenia increased after a day-long workshop
Laporte et al. (2011)	Questionnaires & semi-structured interviews	Sibling	MI-Sibs reported lower levels of perceived maternal care than their brother/sister	Sibs reported low levels of depression and anxiety
Latzter et al. (2015)	Questionnaires	Sibling	MI-Sibs reported more depressive symptoms and more negative sibling relationship qualities than TD-Sibs	Psychological distress was related to depression (-), sibling relationship (-), and sense of coherence (-)
Leith et al. (2018)	Questionnaires	Sibling		Feelings of personal loss were related to being female (+), support group affiliation (+), and ambivalence (+). Stress-related growth was related to support group affiliation (+) and balanced care (+; i.e. balance between self-care and care for patient)
Leith and Stein (2012)	Questionnaires	Sibling		Overall personal loss due to MI in MI-Sibs was related to current caregiving (+), support group affiliation (+), being female (+), severity of MI (+), and dependence of brother/sister (+)

**Table 3** (continued)

Authors	Method	Reporter	Between-Group Results	Within-Group Results
Lively et al. (1995)	Verbal administration of questionnaires	Sibling		Nearly all siblings reported that the patient's MI had an impact on the sibling relationship and their relationship with their parents. Two-thirds of sibs reported the patient's illness had an impact on their own mental health
Lively et al. (2004)	Questionnaires	Sibling		Female siblings and younger siblings reported more disturbing behavior from the patient. Disturbing behavior of the individual with MI was related to relationships for the MI-Sibs (-; family, sibling, spouse, friends), sib health (-) and self esteem (-), sib feelings about being a caregiver (-), and school and work performance (-)  Less than half of participants were familiar with the law, and only 5% correctly identified all three provisions. Planned future caregiving (+), education level (+), membership in a support group (+), more reading about mental illness (+), greater brother/sister problems with treatment (+) were all associated with awareness of the law
Lohrer et al. (2002)	Questionnaires	Sibling		Caregiver status was predicted by having deceased parents (+), being female (+), and having a male sibling (+)
Lohrer et al. (2007)	Questionnaires	Sibling		
Moulds et al. (2000)	Questionnaires	Proband and sibling	Siblings did not differ from parents on level of expressed emotion	
Oliver et al. (1987)	Questionnaires	Sibling	MI-Sibs reported less family cohesion, more conflict, and less intellectual-cultural orientation than TD-Sibs	
Östman et al. (2005)	Semi-structured interviews	Sibling	MI-Sibs were less likely than spouses, parents, and children to report having to give up leisure time or experiencing mental health problems of their own, but also less likely to report that their own needs for support were met	
Samuels and Chase (1979)	Semi-structured interviews	Sibling (rated by interviewer)		MI-Sibs reported low-to-moderate rates of mental illness, low levels of functional impairment, and moderate levels of family entanglement

Table 3 (continued)

Authors	Method	Reporter	Between-Group Results	Within-Group Results
Sanders & Szymanski (2012)	Questionnaires	Sibling	MI-Sibs performed better than norms on Experiential emotional intelligence (e.g., perceiving emotions), but worse on strategic EIQ (e.g. managing emotions)	Sibling involvement in caregiving was related to attachment with parents (–); sibs also reported lower levels of total attachment to mother, communication with mother, and non-alienation with mother after the patient's diagnosis as opposed to before
Sanders and Szymanski (2013a)	Questionnaires	Sibling		
Sanders and Szymanski (2013b)	Questionnaires	Sibling	MI-Sibs reported more posttraumatic growth than comparison group overall	Posttraumatic growth was related to sibling involvement in caregiving (–)
Sanders et al. (2014)	Questionnaires	Sibling	MI-Sibs reported more Hero and Lost Child classification factors than TD-Sibs and less Mascot and Scapegoat factors	
Seltzer et al. (1997)	Questionnaires	Sibling and parents	MI-Sibs were less likely than IDD-Sibs to report that their brother/sister affected the sibs' feelings about their career, choice to have children, romantic relationships, or plans for the sibs' future; MI-Sibs were less likely to report that their brother/sister affected the sibs' feelings about individuals with disabilities. MI-Sibs spent less time in person with wither brother/sister than IDD-Sibs and reported less understanding, trust, fairness, respect, and affection both for and from their brother/sister	Well-being for MI-Sibs was related to income (+), closeness of the sib relationship (+), and number of effects on them from the brother/sister's diagnosis (–)
Sin et al. (2016)	Questionnaires	Sibling	Female MI-Sibs reported lower well-being than age-matched population norms, but not male MI-Sibs. MI-Sibs had greater knowledge of mental illness than community samples and more positive appraisals of caregiving than MI-parents	

**Table 3** (continued)

Authors	Method	Reporter	Between-Group Results	Within-Group Results
Smith and Greenberg (2008)	Questionnaires	Sibling and primary caregiver		Sibling relationship was related to family cohesion in childhood (+), perceived gains (+), threats of harm from the patient (-), fears of the patient (-), and belief that the patient can control their behavior (-)
Smith et al. (2007)	Questionnaires	Sibling		Instrumental support was predicted by being married (+), family responsibility (+), earlier age of illness onset (+), reciprocity (+), perceived personal gains (+), control attributions (-), and geographic distance (-). Emotional support was predicted by sib being female (+), family responsibility (+), sib relationship quality (+), reciprocity (+), and geographic distance (-)
Smith et al. (2016)	Questionnaires & neuropsychological test battery	Sibling		36.6% of sibs reported high levels of depressive symptoms; symptoms were related to global neurocognition (-), impact of MI on social activities (+), parentification (-), and stigma (+)
Tanaka (2008)	Questionnaires	Sibling		Demographic factors (being a female sib, younger age of onset of MI) predicted sib experiences (-). Social support moderated the relationship between demographics and negative experiences
Tanaka (2010)	Questionnaires	Sibling	MI-Sibs in the US reported resources as being more useful than MI-Sibs in Japan	
Tanaka (2011)	Questionnaires	Sibling		Severity of MI (-), support since diagnosis (+), caregiver status(+), availability of resources (+), and social desirability (+) all predicted sibling positive outcomes
Tatay-Mantiaga et al. (2019)	Questionnaires	Sibling	No differences between MI-Sibs and controls on any scale of QoL	



Table 3 (continued)

Authors	Method	Reporter	Between-Group Results	Within-Group Results
Taylor et al. (2008)	Secondary data analysis from a longitudinal study	Sibling	MI-Sibs had less contact with their brother/sister, less similar outlook on life with their brother/sister, lower levels of extraversion, and lower levels of conscientiousness than TD-Sibs. MI-Sibs of brothers had lower agreeableness and higher neuroticism than TD-Sibs of brothers. MI-Sibs of sisters had lower neuroticism than TD-Sibs of sisters. MI-Sibs did not differ from IDD-Sibs on any measure	
Tschan et al. (2019)	Questionnaires	Sibling	NSSI-Sibs reported more coercion in the sibling relationship than other MI-Sibs and control sibs	For NSSI-Sibs, warmth (+), conflict (+), and empathy (+) in the sib relationship were related to sib internalizing problems Proactive action (+), seeking social support (+), palliative reaction (-), avoidance (-), passive reaction (-), and expression of emotion (-) were related to well-being
van Dijk et al. (2019)	Questionnaires	Sibling		
*van Langenberg et al. (2016)	Questionnaires & medical records	Sibling, mother, father, patient	MI-Sibs reported higher levels of emotional difficulties, hyperactivity, and total difficulties than norms	Sib emotional and total difficulties were related to the brother/sister's duration of illness (+); sib behavior did not improve following brother/sister treatment
Wolfe et al. (2014)	Secondary data analysis from a longitudinal study	Parent	MI-Sibs were less likely to be divorced than IDD-Sibs	
Zauszniewski and Bekhet (2014)	Secondary data analysis from a cross-sectional study	Sibling	Female MI-Sibs reported less overall emotional distress and depression than MI-Mothers or other female family members	

(+) and (-) indicate the direction of the correlational relationship with the outcome variable; (+) designates a positive correlation between the listed variable and the outcome, (-) designates a negative correlation. Studies designated with \* report interventions

define outcomes measures and/or report means of assessment measures.

## Discussion

The current study consists of a systematic review of the literature on outcomes for siblings of individuals with mental illness. The majority of the research on MI-Sibs involves risk studies; that is, studies conducted to determine how at-risk MI-Sibs are for developing mental illness themselves or to identify subthreshold symptoms of a given MI to use for future diagnostics. Although these studies certainly have value for etiological and treatment purposes, they do little to illuminate the experiences of typically-developing siblings and how these siblings are affected by their brother or sister's MI. The studies included in the current review identify several outcomes for MI-Sibs, but overall, the review highlights the numerous gaps in the sibling literature.

First, despite most reports of schizophrenia putting the prevalence below 1% (Moreno-Kustner et al. 2018), just under half of the included studies in our sample focused primarily or entirely on siblings of individuals with schizophrenia, with another eleven studies including a minority or unspecified percentage of schizophrenic patients. The impact of schizophrenia on the family unit and relationships in general can be profound and widespread, so it is important to understand the experiences of siblings. However, the current review revealed just how little is known about siblings of individuals with other diagnoses. Although symptoms of depression and anxiety may be less severe than those of schizophrenia and other psychotic disorders, it would be unwise to presume that the siblings of individuals with these diagnoses are unaffected. Additionally, far more research is needed about siblings of individuals with other less-prevalent diagnoses, such as eating disorders or personality disorders. We hope that this review will illustrate the importance of understanding siblings of individuals with MI and just how much work is still needed within this population.

Second, although the review included over two-dozen comparative studies (studies that compared MI-Sibs to another population), the results of these studies varied quite a bit. Some studies found that MI-Sibs reported more negative outcomes than comparison groups, while some found that MI-Sibs reported fewer negative (or more positive) outcomes. Because these studies included such a diverse set of outcomes (sibling relationship, behavior problems, temperament, etc.) and comparison groups (siblings of typically-developing individuals, other relatives of individuals with MI, etc.), it is challenging to make any generalizations about the results. Due to the variability in family constellation, severity of MI, differences across diagnoses, and many other potential contributory variables, it is likely that

few conclusions overall can be drawn about MI-Sibs as an entire population. Therefore, while comparison studies are valuable and should be explored further, it is important to measure individual differences when researching experiences of MI-Sibs.

Despite the variability in measures and methods, the current review did yield a few consistent themes. In terms of outcomes, ten of the studies included continuous measures of sibling mental health. Although studies exclusively focusing on sibling diagnostic rates were not eligible for inclusion in the current review, studies that used quantitative measures of depression or anxiety to indicate more general fluctuations in mood were summarized. It is difficult to disentangle shared genetics from shared experience when it comes to analyzing mental health among MI-Sibs, but mental health is still a worthy concentration. Other family studies, including studies of families of individuals with IDD, assess mental health (e.g., Emerson 2003) as a way of understanding the impact of having a brother, sister, or child with a disability. Therefore, measures of mental health, especially continuous measures (i.e., not strictly diagnostic rates) should continue to be explored in MI-Sib studies as well.

More studies overlapped in their use of predictor or correlational variables, with multiple studies each examining the impacts of sibling gender, severity of MI, and sibling knowledge of mental illness. As is consistent with other populations, female MI-Sibs were more likely than male sibs to provide caregiving, and, when asked, female siblings reported more severe behavior problems for the brother/sister with MI than did male siblings (Lively et al. 2004). Additionally, studies that included measures of MI severity consistently linked a negative relationship between severity and sibling outcomes (i.e. greater symptom severity was related to poorer sibling outcomes). However, some studies also included measures of sibling knowledge of MI and sibling attributions of control over MI. The more siblings understood about the progression and etiology of mental illness, the less likely they were to make negative control attributions of their brother/sister's illness. As MI in general is still largely misunderstood globally (Rüsch et al. 2005), many siblings may not have a comprehensive idea of what their brother or sister is going through; therefore, educational interventions for siblings and other family members may help buffer against negative consequences for both the sibling and the individual with MI.

## Implications

The current review has numerous implications for research and practice. First, as mentioned above, the review clearly identifies gaps in the MI-Sib literature, the most prominent of which being the lack of studies on siblings of individuals with more commonly-diagnosed disorders, including

depression, anxiety, eating disorders, and mood disorders. Because of the significant variability in symptoms, treatment, and societal perception, it is important to study sibling experiences among families of individuals with varying MI diagnoses. Additionally, very few studies included the perspective of the individual with MI. To more fully understand the sibling relationship and the impact of MI on family members, including reports from the patient is essential. In terms of study methods, researchers should explore practices beyond self-report (though self-report is certainly very valuable). Because certain events related to mental illness can be traumatic—hospitalizations, suicide attempts, etc.—it is important to understand how such events may impact the sibling. Methods such as biofeedback can help pinpoint the physiological impacts of recollection of such events. Additionally, to better understand the impact of MI on siblings, it is important to assess siblings across the lifespan. Adult siblings, who may not live with the individual with MI may have very different outcome than child or adolescent siblings who still live at home and are thus more directly present to symptoms of MI.

For families and service providers, one of the most important recommendations is to include siblings in support activities. As mentioned above, knowledge of mental illness may help improve both sibling outcomes and the sibling relationship, and even one-day workshops (e.g., Landeen et al. 1992) can significantly improve sibling's understanding of MI. Media portrayals of mental illness are often skewed and inaccurate (e.g., Stuart 2006); therefore, promoting understanding among siblings may have to include debunking commonly-held, yet incorrect beliefs about people with MI. Additionally, simple awareness that siblings can be impacted by their brother or sister's illness can help families prepare to seek out support systems for the siblings. Again, this process may look different at different stage of the lifespan—young children not understanding why their brother or sister with MI acts the way they do to adult siblings considering whether or not to have their own children, given the genetic linkage of mental illness. Several of the included studies found that more severe MI symptoms were related to more negative sibling outcomes (e.g., Bowman et al. 2014; Horowitz 1994); therefore, siblings who are exposed to more severe and traumatic behaviors and events may be more at risk for negative outcomes. Families and clinicians should take special care to address potential sibling outcomes, especially if the individual with MI is hospitalized or otherwise engaged in rehabilitation treatment.

## Limitations

As with all literature reviews, the current manuscript is limited by the content of the included studies. A substantial barrier to general interpretation of the current review lies in

the variance in the information studies provided. Not only did several studies not include basic demographic information, such as age and gender (e.g., Hudson and Rapee 2002; Tanaka 2008), but many published studies did not report statistics in a way that permitted interpretation of individual variables. For example, Jewell and Stein (2002) reported model fit indices for their regression models, but not beta values for individual variables. Therefore, we cannot determine which variables (e.g. sibling affection, parent support) independently contribute to variance in sibling caregiving (Jewell and Stein 2002). While severity of MI was consistently found to relate to sibling outcomes, the majority of included studies did not assess symptom severity. Additionally, some studies (e.g., Chen and Lukens 2011; Tatay-Montiega et al. 2019) did not specify which individuals with MI were related to the siblings in the sample. That is, the samples were reported collectively (e.g., relatives were listed separately—mothers, fathers, siblings—but demographic information about the entire MI group was reported together), or listed MI groups separately, but siblings collectively. Such presentation of demographics prevents researchers from determining which characteristics of the individuals with MI are related to sibling outcomes. Finally, it is important to note that the current review excluded samples in which the MI-Sib had a mental illness themselves. Due to the heritability of mental illness, this exclusion criteria leaves out a likely substantial proportion of MI-Sibs and thus, the results are not generalizable to all siblings of individuals with mental illness. These limitations in reporting not only hinder interpretation of the individual study, but it limits the potential of such studies being included in future meta-analyses.

Additionally, the current review attempts to summarize a wide range of potential outcomes for siblings. Thus, the included categories may have some redundancy across descriptions. That is, the categories are not necessarily clear-cut. Finally, when discussing family impacts of MI, it is always important to acknowledge the possibility of shared genetic variance. Although the current review was limited to siblings without a diagnosis of MI themselves, it is impossible to know how many of these siblings experience subthreshold MI symptoms or perhaps qualified for, but have not yet received, an MI diagnosis. Therefore, the classification of “typically-developing” siblings is, in and of itself, somewhat artificial.

## Conclusion

The current study is the first review of its kind to summarize the quantitative literature on psychosocial and behavioral outcomes among siblings of individuals with mental illness. The included studies covered a wide range of measures and

outcomes, allowing for few consistent areas of interpretation. However, the review makes several important contributions to the literature. First, the study highlights the gaps in knowledge regarding siblings of individuals with less-severe MI, such as depression or anxiety. Second, the review identifies the importance of knowledge and understand of MI for both the siblings themselves and the sibling relationship. Finally, the review calls attention to the needs of MI-Sibs in general. We hope that future researchers, families, and service providers can continue to explore ways to best support siblings of individuals with mental illness and promote positive family relationships across the life course.

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**Conflict of interest** The authors have no conflicts to report.

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