



Mental Health and Well-being in Adolescent and Adult Siblings of Persons with Severe Mental Illness: A Scoping review

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Abstract

Systematic knowledge about siblings of persons with mental illness is needed. The aim of this literature review was to gain knowledge about the mental health and well-being of siblings of individuals with severe mental illness (i.e., bipolar disorder, schizophrenia, eating disorders, psychosis) and factors associated with sibling outcomes. We reviewed quantitative studies published between 1990 and 2022 where siblings aged >12 years were compared to healthy control individuals. Eleven studies were included (combined sibling $n = 1736$). Seven of the 11 studies reported solely more negative outcomes for siblings than for healthy control individuals. Two studies reported both more negative and more positive sibling outcomes, with one study reported solely more positive outcomes and one no difference between siblings and healthy control individuals. The studies reporting negative outcomes had the highest methodological quality. The following factors were associated with sibling outcomes; demographic factors, i.e., age, sex, and number of siblings in the family, contextual factors, i.e., having contact with health care professionals due to own mental health problems, and characteristics of the sibling with severe mental illness, i.e., premorbid adjustment. The findings show that a family system approach with interventions addressing the entire family is indicated when a person is treated for severe mental illness. Further research on sibling outcomes should address methodological challenges such as poor representativeness and a wide age range in samples and use well-validated outcome measures. Factors associated with sibling outcomes should be examined further to increase the understanding of risk and protective factors in siblings.

Keywords Severe mental illness · Siblings · Wellbeing · Mental Health · Literature review

Highlights

- Most studies showed more negative mental health outcomes for siblings compared to healthy control individuals.
- Studies showing negative outcomes had the highest methodological quality.
- We identified several moderating factors, including sibling sex, age, health service contact, and illness characteristics.
- The findings can inform intervention and support programs for families of persons with severe mental illness.

Severe mental illness (SMI, i.e., bipolar disorder (BD), schizophrenia, eating disorders, and psychosis) has been associated with mental health problems and the well-being of persons close to those who are diagnosed with such ill-

nesses (Sin et al., 2014). A large group of next-of-kin persons to those with SMIs are siblings. Mixed results have been found regarding outcomes for siblings of persons with SMI. The first literature review on the topic comprised 56 studies on siblings aged 6 to 81 years of persons with various diagnosed mental health disorders (Shivers and Textoris, 2021). This review showed large variations in outcomes among siblings. Thirty of the 56 studies in the Shivers and Textoris (2021) review included between-group control data. Of these 30, seven studies found more negative mental health outcomes for siblings, three studies found more positive outcomes for siblings, and five found no difference. Eleven studies found both positive and negative

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outcomes depending on the variable and/or informant. The remaining four comparator studies used intervention groups or other next-of-kin groups as controls. The positive outcomes reported for siblings in Shivers and Textoris (2021) were more sensitivity to others, greater use of problem-solving coping, and less sibling rivalry. The negative outcomes reported for siblings were increased behavioural and internalizing symptoms, and social problems. A later meta-analytic review focused more specifically on depression and anxiety outcomes in siblings of persons with mental illness (Jayasinghe et al., 2023). This review included 15 studies with siblings aged >10 years of persons with various mental disorders ranging from anxiety/depression to schizophrenia. The meta-analytic findings showed higher depression (moderate effect size) and anxiety (small effect size) among siblings than among controls (Jayasinghe et al., 2023).

These two previous reviews on mental health in siblings have provided important insights. However, both had some embedded methodological challenges, including a wide range of disorders across a wide age range, which provides limited specific knowledge. Importantly, Shivers and Textoris (2021) included studies with very heterogeneous control groups, ranging from norm populations, siblings of persons with different mental or neurodevelopmental disorders or other family members (next-of kin), as comparisons to siblings of persons with mental health disorders. Comparison of findings across very different control groups is tricky. In the review also studies with no control group were included, which further complicate the interpretation of the findings. A limitation of Jayasinghe and colleagues' (2023) meta-analysis is the focus on narrow outcomes (anxiety and depression) across a wide range of disorders in the ill sibling. Hence, albeit these two reviews have provided an overview of the field, there are still knowledge gaps.

Herein, we present an updated review that applies three distinct features that are in contrast to previous reviews, aiming to provide a more succinct overview of the state-of-the-art for sibling outcomes. First, we narrowed the focus to severe mental illness (i.e., bipolar disorder (BD), schizophrenia, eating disorders, and psychosis). The focus on severe mental disorders is important because these illnesses are associated with millions of healthy living years being lost (Walker et al., 2015; McIntyre et al., 2020; Hoeken and Hoek, 2020). Further, a systematic review and meta-analysis showed that the risk of mortality rate for people with a severe mental illness, are higher than for people with a milder form of mental illness, like depression or anxiety disorder (Walker et al., 2015). It can be hard to define what is considered “severe”, but we used chronicity and impact as guiding features.

Second, we applied a resilience perspective and included positive, as well as negative, sibling outcomes. This was

done by including search words from a resilience-based framework, such as coping, self-efficacy, and support. In their review, Jayasinghe et al. (2023) also included positive outcome search words, but the current review is the first to do so with a parallel SMI focus.

Third, we narrowed the comparison groups down to healthy control individuals, i.e., either siblings with a healthy brother or sister, a random population comparison group sample, or matched samples recruited voluntary from the general population free of any mental health disorders. The review by Jayasinghe et al. (2023) also addressed the issue of heterogeneity in control groups by including comparison groups with siblings without mental illness. However, they also included studies without control groups

The World Health Organization (2022) has defined mental health as more than the absence of mental disorders or disabilities, and as “a state of well-being in which an individual realizes his or her own abilities, can cope with the normal stress of life, can work productively and is able to make a contribution to his or her community”. When assessing mental health, the presence of disorders or symptoms (e.g., depression, anxiety) are often used as operationalizations, whereas positive aspects of psychological adjustment comprise different variables of psychological well-being. Thus, mental health comprises a range of dimensional (e.g., symptoms levels), and categorical (e.g., disorders) problems and includes the absence of mental health problems.

The concept “psychological well-being” has been suggested to include the dimensions self-acceptance, purpose in life, environmental mastery, positive relationships, personal growth, and autonomy (Ryff and Singer, 2008). According to Ryff and Singer (2008), the dimensions involve awareness and acceptance of personal strengths and weakness, and describe how the individual will have a better life if s/he manages to find a meaning and direction in life, if s/he chooses or creates environments suitable to his/her mental conditions, and if s/he self-actualizes and evaluates his or herself by personal standards. As the literature on outcomes in siblings of persons with SMI is limited, we need to examine studies with a range of different mental health outcomes. It is too early for an examination of more limited mental health outcomes (e.g., specific disorders or type of symptoms). Thus, to understand sibling outcomes when a brother or sister have SMI, both mental health and positive psychological well-being are important aspects to consider.

Examining risk and protective factors associated with sibling mental health problems and well-being may provide increased understanding of the assumed variation in outcomes among siblings, as well as enhance the possibility of preventing negative outcomes for siblings of persons with SMI. Different variables have been examined. In their review, Shivers and Textoris (2021) found that female sex,

degree of belief in the sibling with SMI's ability to control his/her behaviour, and greater severity of SMI symptoms were variables associated with more negative sibling outcomes. However, studies of sibling outcome predictors have been limited and findings are somewhat inconsistent, suggesting a need for examining factors related to sibling outcomes, comparing studies including more homogeneous control groups. Jayasinghe et al., 2023 explored several correlates that modified the magnitude of the group differences between siblings, yielding mixed findings. In summary, the authors found that age, sex, psychological characteristics (e.g., low sense of coherence, rumination, and external locus of control), negative evaluation about the impact of mental illness on social relationships, and stronger perception of stigmatization were associated with more psychological distress (i.e., depression and anxiety symptoms). Further, poorer social relationships, adverse childhood experiences, neurocognitive functioning, and negative evaluation of the sibling relationship were associated with more negative sibling outcomes (Jayasinghe et al., 2023).

The Present Study

In this literature review we examine the sibling resilience perspective by including studies on mental health and psychological well-being, as well as factors associated with these outcomes, in siblings of persons with SMI. We posed the following research questions: 1) Is mental health and well-being of adolescents and adults associated with having a sibling with SMI? and 2) Which factors are associated with mental health and well-being of adolescents and adults of siblings with SMI?

Method

Inclusion and Exclusion Criteria

Studies were identified through systematic electronic searches, using the PRISMA guidelines (Page et al., 2020), as well as supplementary hand searches in references of identified studies and literature reviews. Studies were included if they were (a) published between January 1990 and December 2022, (b) published in peer-reviewed Scandinavian or English language journals, (c) included typically developing siblings (i.e., persons with no known mental illness) aged 12 years or older of individuals with SMI, (d) addressed how siblings are coping by including studies on mental health and/or well-being of siblings of individuals with SMI, (e) included sibling with severe mental illness according to one of the following diagnoses: BD (any type), schizophrenia, eating

disorder (any type), or psychosis, and (f) compared siblings of individuals with SMI to healthy control individuals (i.e., siblings with a healthy brother or sister, a random population comparison group sample, or matched samples recruited voluntary from the general population free of any mental health disorders). Books, dissertations, and other publications that had not undergone peer review were excluded, as well as literature reviews and qualitative studies. Studies on individuals with physical illnesses, neurodevelopmental disorders, intellectual disabilities, learning difficulties, and developmental disabilities were also excluded. Previous reviews on sibling outcomes have excluded siblings of persons with eating disorders, even though these disorders satisfy criteria of being SMI, often having long duration and high level of disability. Thus, in addition to BD, schizophrenia and psychosis, the current review included eating disorders.

Literature Search

Searches were conducted through PsycINFO, Cumulative Index to Nursing and Allied Health Literature (CINAHL), and the Web of Science databases. Three databases were chosen to increase the likelihood of identifying relevant studies and were considered sufficient to be able to find studies in this research area.

The literature searches were done at two time-points. The first search was conducted on the 2nd of May 2021. A second updated search was conducted on the 4th of January 2023. The updated search applied the same procedure as the first and was done to include studies published between 2021 and 2022. The searches were conducted in cooperation with two University librarians.

Keywords related to sibling coping and the resilience perspective were applied. Terms for siblings were combined with terms for sibling's mental illness, and terms for adjustment, coping, and psychological adaptation. Studies regarding suicide were first included in the searches, but later removed because they did not have results regarding specific mental disorders in the ill siblings. Also, studies regarding autism spectrum disorder (ASD) were first included in the searches, but removed in the revision process after consideration that ASD is not necessarily considered a SMI. All keywords applied were combined by the Boolean term AND, and OR. Truncation was used to get more hits. A full presentation of search terms and combinations is available in the supplementary material.

Searches in PsycINFO, CINAHL and Web of Science generated 3129 hits. These were exported to EndNote, with duplicates removed either automatically by EndNote or manually when screening through titles and abstracts of the studies. A total of 2423 hits were screened by reading titles and/or abstracts. The majority of the studies

included for further review were then screened by two authors (in pairs). After the initial screening the authors screened about one third of the selected articles each. First, 16 studies were included. After handsearching the reference lists, 19 more studies were included. Thus, a total of 35 studies were read in full, systematically abstracting the design, recruitment setting, methodology, sample size, definition of the population, findings regarding sibling mental health and well-being, and factors associated with sibling outcomes. During the coding process, more studies were excluded as they turned out not to fulfil the inclusion criteria, i.e., not referring to specific mental health disorder, or including younger siblings (<12 years). In the final revision process five studies were excluded due to being studies of siblings of people with ASD. Eleven studies were finally included. For an overview of the search strategy and inclusion process, see Fig. 1.

Coding of Scientific Quality

The studies were coded for methodological quality. The first author coded the scientific quality of all the included studies. Half of these studies (50%) were then double coded by either the second or the third author, obtaining 93.7% agreement between scorers. The scientific quality of the studies was evaluated using the Mixed Methods Appraisal Tool (MMAT), which is designed for critical appraisal of research studies with a range of different designs (Hong et al., 2018). The items regarding quantitative descriptive studies were selected for the evaluation of studies in the present review. The following variables were coded, all as “yes” or “no”, or “can’t tell”, in line with the MMAT: 1) clear research question, 2) data address research question, 3) relevant sampling strategy, 4) representative sample, 5) appropriate measures, 6) low risk for nonresponse bias, and 7) appropriate statistical analysis. To judge the criteria, the authors needed to consider whether the source of the sample was relevant to the target population i.e., a match between the respondents and the target population; that the variables were clearly defined and the measurements justified i.e., appropriate regarding answering the research questions; whether the respondents and non-respondents were different on the variable of interest; and that the statistical analyses used were justified.

Sibling Outcomes

Effect size was applied to examine sibling outcomes. For three studies this was given, for five studies effect sizes (Cohens’ *d*) were calculated based on means, pooled standard deviations and sample size, whereas three

studies did not include sufficient information to calculate effect size.

Results

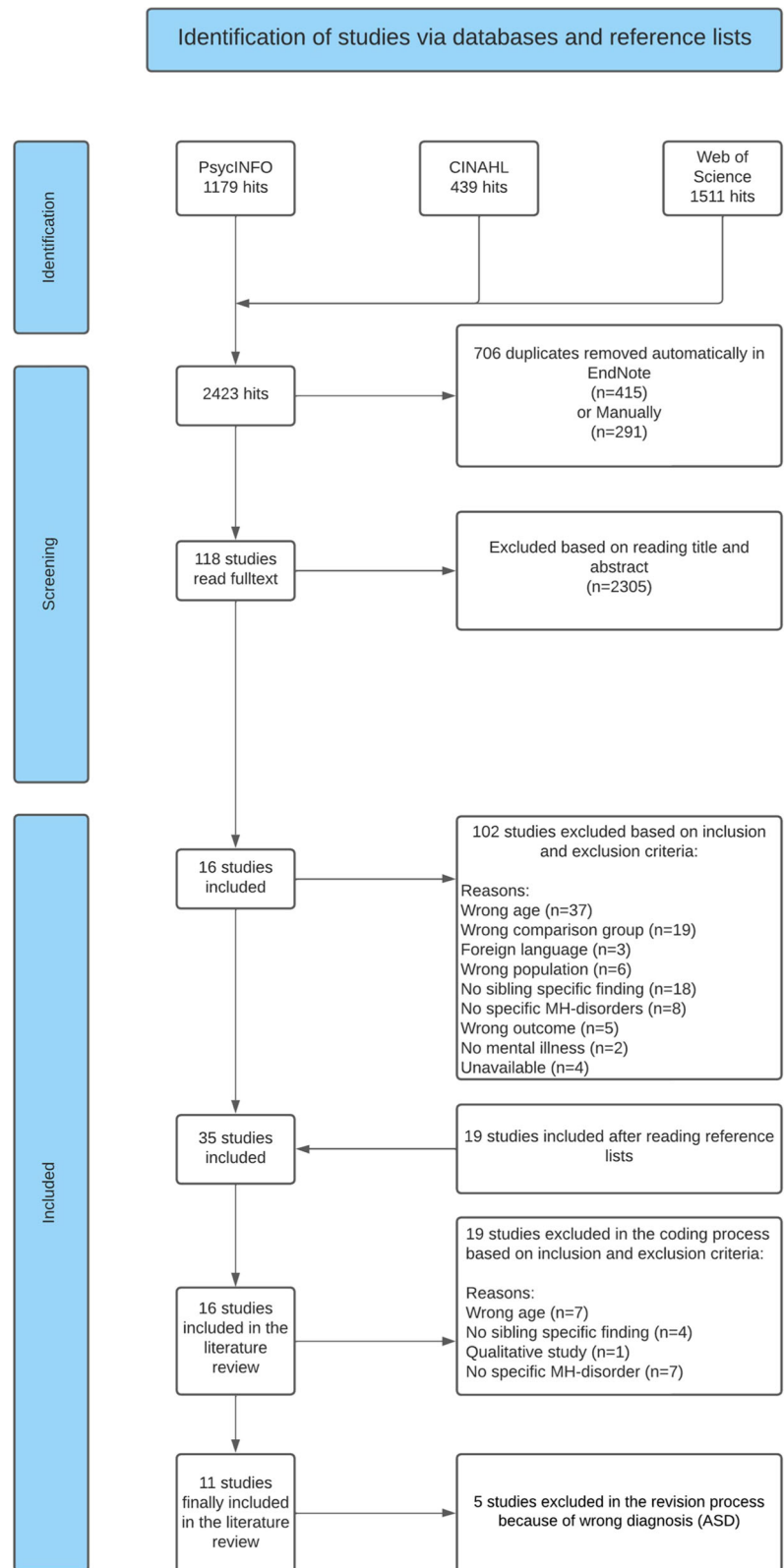
Study Designs and Sample Characteristics

Eleven studies met the inclusion criteria. An overview of the descriptive data from these studies are provided in Table 1. Nine studies were cross-sectional, of which one was a retrospective study. The two remaining studies were a cross-sectional registry study and a population-based longitudinal registry study. The sample sizes of siblings of persons with SMI ranged from 13 to 13923, with an age range from 15 to 72 years. The sex ratio across the studies could not be calculated since this ratio was not reported in all studies. See Table 2 for more details about the sibling sample.

The majority of the studies originated from Europe. The remaining studies were from Israel and USA. Different contexts and strategies were used to recruit siblings of persons with SMI, with some samples recruited from hospitals or academic centres, some from research institutes extracting data from genetic studies, and some applied baseline data in randomised controlled trials. Some studies applied a combination of different recruitment settings. Control groups were healthy control individuals (i.e., siblings with a healthy brother or sister, a random population comparison group sample, or matched samples recruited voluntarily from the general population free of any mental health disorders) mostly recruited through national registers, blood banks, research institutes, or being friends of the person with the SMI. Some of the included studies also compared mental health and well-being outcomes in the sibling group with outcomes in the siblings with a SMI, but only data on the outcomes and factors associated with the outcomes in the siblings (without a SMI) compared to the healthy control individuals were extracted for analysis in the present review. The diagnoses of the sibling with a SMI were most often established through clinical interviews (see Table 1).

In accordance with the inclusion criteria, all control groups comprised health control individuals. The control group sample sizes ranged from 21 to 278460, with a median of 200. In eight studies, the control group was recruited for the study in question. In two studies, control groups were population controls. In one study, the control group was drawn from another study of non-clinical samples. Five studies explicitly mentioned matching or balancing controls and siblings on at least one variable (i.e., age).

Fig. 1 The search strategy and selection process (made by PRISMA-template)



Sibling Outcomes

The studies used a variety of methods for assessing sibling outcomes, the most common being self-administered

questionnaires. We were able, based on reported numbers, to calculate four effect sizes showing better outcomes for siblings compared to controls ($M d = 0.39$ across studies). We were able to calculate 16 effect sizes showing poorer

Table 1 Overview of Included Studies

Author (year)	Country	Recruitment setting	Design	Severe mental illness sample	SMI	How was illness established?
Arajärvi et al. (2006)	Finland	The Finnish Hospital Discharge Register.	Registry study, cross-sectional	$n = 124$ (65.3% males). Aged 31.2–64.9 yrs, M age 46 (SD 7.3).	Schizophrenia.	Registered diagnoses, based on a SCID interview diagnosis of schizophrenia or schizoaffective disorder.
Barak and Solomon (2005)	Israel	User association for schizophrenia + friends (controls).	Cross-sectional	$n = 52$. Sex and age not reported.	Schizophrenia.	Not reported.
Pierazzuoli et al. (2020)	Italy	Psychiatric facilities and psychiatric patient family associations.	Cross-sectional	$n = 30$ (76% males). Age not reported.	Schizophrenia spectrum disorder.	Not reported.
Plessis et al. (2020)	France	An online questionnaire.	Cross-sectional	$n = 201$ (80% males). Age not reported	Schizophrenia.	Diagnosed with schizophrenia by a healthcare professional.
Shapiro et al. (2009)	USA	Participants from the Sibling Study in the Genes, Cognition, and Psychosis Program at the National Institute of Mental Health.	Cross-sectional	$n = 286$ (76% males). Aged 16–64 yrs, M age 36.2 (SD 9.4).	Schizophrenia or schizoaffective disorder.	DSM-IV diagnosis, made by either a clinical psychologist or psychiatrist using a revised version of SCID.
Sin et al. (2016)	England	Baseline-data from RCT on internet-based psychoeducational intervention for siblings of individuals with first episode psychosis recruited via non-governmental organisations or services for early intervention for psychosis.	Cross sectional	$n = 89$ (62% males). Aged 15–57 yrs, M age 26.4 (SD 7.9).	FEP.	No information.
Stetved et al. (2022)	Denmark	Patients and siblings of patients from Copenhagen Affective Disorder Clinic. Healthy control individuals from the Blood Bank at Rigshospitalet Copenhagen, Denmark.	Cross sectional	$n = 382$ (34% males). Aged 18–64 yrs, M age 29.2.	BD.	Based on ICD-10 and DSM criteria.
Tatay-Manteiga et al. (2019)	Spain	University hospital.	Cross sectional	$n = 48$ (52% males). M age early stage BD: 43.4 (SD 10.3), late stage BD: 45.1 (SD 9.8)	BD.	Diagnosed with DSM-IV-TRBD type I.
Vedel Kessing et al. (2020)	Denmark	Combining data from Statistics Denmark (psychiatric diagnoses). Danish National Patient Register, Danish Psychiatric Central Research Register, Medicinal Product Statistics and Danish Medical Register on Vital Statistics.	Population-based longitudinal register study (from 1955 to 2017)	$n = 19$ 955. Median age 38 yrs (52% males).	BD (having a single manic episode or BD) during 1995 to 2017. BD main diagnosis at any contact.	ICD-8, ICD-9, ICD-10.
Vothknecht et al. (2013)	Netherlands	Academic centers (Amsterdam, Groningen and Utrecht) and their affiliated mental health care institutions.	Cross-sectional	$n = 600$ (80% males). M age 27 yrs (SD 7.1).	Schizophrenia.	No information.

Table 1 (continued)

Author (year)	Country	Recruitment setting	Design	Severe mental illness sample	SMI	How was illness established?
Walshe et al. (2007)	Ireland	Through voluntary support groups or direct psychiatric referrals, and through advertisements in local newspapers or from staff.	Cross-sectional, retrospective	$n = 50$ with personal and family history of schizophrenia (74% males). M age 32 yrs (SD 6.1) and 69 people with personal schizophrenia and “non-familial” schizophrenia (75% males). M age 31 yrs (SD 6.4). Age range both groups 16–69 yrs.	Schizophrenia ($n = 112$), schizo-affective disorder ($n = 6$) or psychotic disorder ‘not otherwise specified’ ($n = 1$).	According to DSM-IV from SADS-LA.

BD Bipolar Disorder, *DSM-IV* Diagnostic and Statistical Manual of Mental Disorders IV, *FEP* First Episode Psychosis, *ICD-8/9/10* International Statistical Classification of Diseases 8/9/10, *RCT* Randomized Controlled Trial, *SADS-LA* Schedule for Affective Disorders and Schizophrenia – Lifetime Version and clinical information, *SCID* Structured Clinical Interview for DSM-IV Axis I Disorders.

outcomes for siblings compared to controls ($M d = 0.56$ across studies). The results on sibling outcomes, measures used to assess the sibling outcomes, and effect size differences are summarized in Table 2.

Better mental health and psychological well-being compared to healthy control individuals

Two studies reported positive outcomes for siblings of persons with SMI compared to healthy control individuals. One study showed that siblings of persons with SMI used more problem-focused coping ($d = 0.71$) and were more involved in mental health arenas ($d = 0.65$) and in artistic activity ($d = 0.51$) (Barak and Solomon, 2005). Another study showed that the siblings had better mental health knowledge than the general population ($d = 0.19$) (Sin et al., 2016). Problem-focused coping is labelled a positive outcome because previous research has shown this coping strategy to be associated with better well-being in siblings of persons with schizophrenia (Avcioglu et al., 2019). No studies reported less mental health problems among siblings compared to healthy control individuals.

More mental health problems and lower level of well-being compared to healthy control individuals

Seven studies assessed mental health outcomes and well-being in siblings of persons with SMI compared to healthy control individuals. One study found that the siblings reported poorer academic functioning during adolescence (effect size $d = 0.39$) (deterioration in academic performance between childhood and adolescence) compared to healthy control individuals (Walshe et al., 2007). Another study found that siblings had impaired functioning in autonomy, occupation, cognition, interpersonal relationships, workability, practical housework, social activities, and relations (effect sizes d from 0.55 to 0.70) compared to healthy control individuals (Sletved et al., 2022).

Four studies reported on psychiatric disorders or psychological problems in siblings. Two studies demonstrated higher rates of psychiatric disorders among siblings of persons with SMI compared to healthy control individuals (8:1 for psychosis in Arajärvi et al., 2006; 2:1 for any disorder in Vedel Kessing et al., 2020). In one of the studies 43.3% of the siblings reported having previous psychological problems compared to the healthy control individuals where none reported having had any psychological problems (Pierazzuoli et al., 2020). Siblings also reported experiencing more emotional distress (effect size $d = 0.18$) (Plessis et al., 2020), and a higher rate of family history of psychiatric disorder (89.7% versus 10.3%) (Pierazzuoli et al., 2020).

Two studies assessed the sibling relationship. Overall, the quality of the sibling relationship was found to be poorer

Table 2 Outcomes and Factors Associated with Outcomes in Siblings of Persons with Severe Mental Illness

Author (year)	Siblings of persons with SMI	Instruments assessing sibling outcomes	Findings regarding sibling well-being	Factors associated with sibling well-being	Control group
Arajärvi et al. (2006)	$n = 183$. Psychotic sibling: (53% males). Aged 32–61 yrs, M age 44 (SD 6.3). Non-psychotic sibling: (48% males). Aged 24–72 yrs, M age 46 (SD 8.8).	SCID-I, SCID-II, and SANS.	More psychotic disorders than healthy control individuals (16.4% versus 2%).	Siblings who had contacted health care professionals for mental health problems, for alcohol or substance use problems or smoking had the highest odds ratios explaining the diagnosis of psychotic disorder.	Healthy control individuals from Finland selected from a population study ($n = 111$; 30–79 yrs)
Barak and Solomon (2005)	$n = 52$ (32% males). Aged 18–50 yrs.	BAS, a modified version of questionnaire assessing coping, feelings toward sibling, sibling relationship, life space and meaning.	More problem-focused coping ($d = 0.71$), objective ($d = 1.16$) and subjective burden ($d = 1.81$), negative feelings toward sibling ($d = 0.58$), and reduced quality of sibling relationship. More involvement in mental health arena ($d = 0.65$) and artistic activity ($d = 0.51$) than healthy control individuals.	More siblings = higher burden; higher education = less burden (higher correlation for the schizophrenia sibling group). For both groups: Single sibling = higher objective burden and greater fear of intimacy. Sisters reported higher subjective burden.	Healthy control individuals from Israel whose siblings had no illness ($n = 48$, 18–50 yrs)
Pierazzuoli et al. (2020)	$n = 30$ (16% males). Aged 36–63 yrs, M age 51 yrs.	PBI, and MMPI-2	Higher rate of family history of psychiatric disorder (89.7% versus 10.3%) and previous psychological problems (43% versus 0%).	Maternal care lower, but maternal favouritism and devaluation higher. Higher emotional trauma, emotional neglect, emotional abuse and physical abuse. Females oriented toward sexual harassment, while males oriented toward physical abuse, emotional abuse and physical threat. Males in caregiver group is oriented toward the higher values of symptoms.	Age and gender-matched healthy control individuals from Italy ($n = 30$, 37–65 yrs)
Plessis et al. (2020)	$n = 201$ (22% males). M age 37 yrs (SD 12.0).	ASRQ-S, HAD, and SES.	More emotional distress ($d = 0.18$) and a poorer sibling relationship compared to controls (warmth: $d = 0.44$; conflict: $d = 0.15$; rivalry: $d = 0.44$).	Antagonism, quarreling, overall rivalry, maternal rivalry and paternal rivalry associated with emotional distress.	Healthy control individuals from France with at least one sibling ($n = 1444$, M age 26 yrs)
Shapiro et al. (2009)	$n = 315$ (41% males). Aged 16–64 yrs, M age 37 yrs (SD 9.9).	PAS.	Poorer academic functioning in childhood ($d = 0.33$), early adolescent ($d = 0.34$), late adolescent ($d = 0.35$), and adulthood ($d = 0.43$) for siblings than for healthy control individuals.	Proband PAS scores predicted PAS scores of their own siblings in the Childhood and Late Adolescence subscales.	Healthy control individuals from the USA ($n = 261$, 16–64 yrs).
Sin et al. (2016)	$n = 90$ (15% males). Aged 16–58 yrs, M age 28 yrs (SD 8.4). Biologically related, step-or half-siblings, or related by adoption.	WEMWBS, MAKS, and ECI.	Poorer mental well-being ($d = 0.55$), but better mental health knowledge ($d = 0.19$) than healthy control individuals.	Siblings' education level associated with better mental health knowledge.	Age-matched healthy control individuals from surveys in England (ns 1100/ 2746, 16–58 yrs).

Table 2 (continued)

Author (year)	Siblings of persons with SMI	Instruments assessing sibling outcomes	Findings regarding sibling well-being	Factors associated with sibling well-being	Control group
Sletved et al. (2022)	$n = 129$ (47% males). Aged 15–63 yrs, M age 28 yrs.	Socioeconomic status outcomes (6 variables), FAST, and WSAS.	Poorer educational achievement and functioning ($d = 0.70$), and FAST subdomains: autonomy, occupation, cognition, interpersonal relationships, leisure, for siblings than healthy control individuals. Impaired functioning in WSAS total scores ($d = 0.55$), and WSAS subdomains: workability, practical housework, social activities.	No predictor analyses.	Healthy control individuals (blood donors) from Denmark; $n = 200$, 24–36 yrs).
Tatay-Manteiga et al. (2019)	$n = 23$ (30% males). M age 42 yrs (SD 11.8).	WHO-QoL BREF.	Siblings and healthy control individuals reported similar levels of QoL ($d = 0.12$).	No significant correlations between age and QoL.	Healthy control individuals with no first- or secondhand relatives with SMI ($n = 21$, M age 37 yrs).
Vedel Kessing et al. (2020)	$n = 13$ 923. M age 38 yrs.	Psychiatric disorders from registers.	Rates of ‘any psychiatric disorder’ constantly around twofold increased throughout lifespan (44.2% versus 27.6%).	A bimodal age distribution of hazard ratios of BD, unipolar disorder and use of alcohol or psychoactive drugs with highest hazard ratios up to age 20 and above 60 years of age.	Healthy control individuals from Denmark general population matched on year of birth and sex ($n = 278460$, M age = 39 yrs).
Vothknecht et al. (2013)	$n = 594$ (46% males). M age 28 yrs (SD 8.0).	SWN-K, WHOQOL, PANSS, and CAPE.	Siblings reported higher subjective well-being than healthy control individuals ($d = 0.20$).	All analyses with the predictors were non-significant.	Healthy control individuals from the Netherlands ($n = 295$, M age = 30 yrs).
Walshe et al. (2007)	A) 39 siblings of persons with personal and familial schizophrenia (36% males). Aged 16–69 yrs, M age 34 yrs (SD 7.8), and B) 67 siblings of persons with personal schizophrenic and non-familial schizophrenia (49% males). Aged 16–69 yrs, M age 35 yrs (SD 7.6).	PSA.	Poorer academic functioning during adolescence (A: $d = 0.52$ and B: $d = 0.25$).	No predictor analyses.	Healthy control individuals without personal or family history of psychotic illness ($n = 83$, M age = 31 yrs).

Effect sizes Cohen's d reported as in papers, or calculated based on N, M, and SD when available. Control group age reported as mean if range was not reported in original papers

ASQ-S Adult Sibling Relationship Questionnaire-short form, *BAS* Burden Assessment Scale, *BD* Bipolar Disorder, *CAPE* The Community Assessment of Psychic Experiences, *DD* Developmental Disorders, *ECI* Experience of Caregiving Inventory, *FAST* Functioning Assessment Short Test rating scale, *HAD* Hospital Anxiety and Depression Scale, *MAKS* Mental Health Knowledge Schedule, *MMPI-2* Minnesota Multiphasic Personality Inventory-2, *PANSS* Positive and Negative Syndrome Scale, *PAS* Premorbid Adjustment Scale, *PBI* Parental Bonding Instrument, *PSA* Premorbid Social Adjustment, *SANS* Scale for the Assessment of Negative Symptoms, *SCID-III* Structured Clinical Interview for DSM-IV axis disorders (I/II), *SWN-K* The Subjective Well-being Under Neuroleptic Treatment Scale, *WEMWBS* Warwick-Edinburgh Mental Wellbeing Scale, *WHO-QoL BREF* World Health Organization Quality of Life Scale, *WHOQOL* World Health Organization Quality of Life, *WSAS* The Work and Social Adjustment Scale. *Yrs* years

for the sibling group than for the healthy control individuals (effect size $d = 1.16$ for objective burden and $d = 1.81$ for subjective burden) in Barak and Solomon (2005). The effect sized ranged from $d = 0.15$ to $d = 0.44$ for warmth, conflict and rivalry) in Plessis et al., (2020). The quality of the relationship reported by the sibling declined after the debut of the severe mental illness, whereas during the same period the sibling relationship improved for the normal comparison group. Further, the siblings reported more negative feelings towards their sibling with SMI compared to the healthy control individuals (effect size $d = 0.58$) (Barak and Solomon, 2005).

Comparable level of well-being as healthy control individuals

One study found similar outcomes for siblings compared to healthy control individuals. Overall, siblings of individuals with SMI reported comparable levels of quality of life as healthy control individuals (effect size $d = 0.12$) (Tatay-Manteiga et al., 2019).

Factors Associated with Sibling Mental Health and Well-being

The findings regarding factors associated with sibling outcomes are presented according to demographic factors, family relationship and health care, and characteristics of the person with SMI. The results are summarized in Table 2.

Demographics factors in siblings of persons with SMI

Three studies reported on demographic factors associated with sibling outcomes. These factors were either collected from population based registers (Vedel Kessing et al., 2020), or from self-reported questionnaires (Barak and Solomon, 2005; Sin et al., 2016). One study found associations between sibling age and ratio of psychiatric disorders in siblings. The results from this study showed highest hazard ratios for onset of bipolar disorder for siblings to persons with bipolar disorder with a hazard ratio of 20.24 (95% CI: 5.86–69.90) below age 20 years and a decrease to a HR around 3.5 in age 40–50 and an increase to around 4 to 7 for age 60–80 years (Vedel Kessing et al., 2020).

One study found association between sibling sex and outcome, with sisters reporting higher emotional burden than brothers (i.e., reporting pain and strong feelings of loss, anxiety, embarrassment and guilt evoked by the siblings' SMI) (Barak and Solomon, 2005). Barak and Solomon (2005) also demonstrated that having more siblings was associated with higher self-reported burden, and that unmarried siblings reported higher objective burden (i.e., the family's need to deal with the mental health system, financial problems, and the

disruption of proper household management), and greater fear of intimacy compared to married siblings.

Two studies showed that higher education in siblings was associated with less emotional and objective burden (Barak and Solomon, 2005) and better mental health knowledge among the siblings (Sin et al., 2016).

Characteristics of the sibling with SMI

One study found that premorbid adjustment, measured as the “degree of achievement of developmental goals” in the person with SMI predicted premorbid adjustment of the siblings without SMI on different developmental aspects, such as sociability, social withdrawal, peer relationships, and scholastic performance (Shapiro et al., 2009).

Contextual factors

Two studies examined contextual factors associated with sibling outcomes, and found that siblings of persons with schizophrenia who had contacted health care professionals due to their own mental health problems had the highest odds for having a psychotic disorder (Arajärvi et al., 2006). Further, perceived maternal care in their family of origin was reported to be lower with more experienced devaluation (i.e., the tendency to debase and humiliate the child) among siblings of persons with SMI than for healthy control individuals (Pierazzuoli et al., 2020).

Scientific Quality of the Reviewed Studies

An overview of the scientific quality of the included studies is provided in Table 3. Some studies were regarded as having several methodological weaknesses, especially regarding the representativeness of the samples. Most of these studies used non-probability sampling, where participants may represent a subgroup of siblings, making the results difficult to generalize. The majority of studies did not include sufficient information to allow the evaluation of risk of nonresponse bias, e.g., reasons why eligible individuals did not participate was not described, and inclusion and exclusion criteria were unclear. All in all only two studies were rated as having good scientific quality, i.e., used relevant sampling strategy, had a representative sample and appropriate measures (Arajärvi et al., 2006; Vedel Kessing et al., 2020). These were register studies, including large sample sizes, and also applied assessments of psychiatric disorders in siblings of persons with SMI.

Discussion

The results from the current review showed that the strongest evidence points towards negative outcomes for

Table 3 Coding of Scientific Quality

Author (year)	Clear research question	Data address research question	Relevant sampling strategy	Representative sample	Appropriate measures	Low risk nonresponse bias	Appropriate statistical analysis
Arajärvi et al. (2006)	Yes	Yes	Yes	Yes	Yes	Can't tell	Yes
Barak and Solomon (2005)	Yes	Yes	Yes	No	No/Yes	Can't tell	Yes
Pierazzuoli et al. (2020)	Yes	Yes	No	No	No	Can't tell	Yes
Plessis et al. (2020)	Yes	Yes	No	No	Yes	Yes	Yes
Shapiro et al. (2009)	Yes	Yes	No	No	No	Can't tell	Yes
Sin et al. (2016)	Yes	Yes	Yes	No	No/Yes	No	Yes
Sletved et al. (2022)	Yes	Yes	Yes	Yes	Yes	Yes/No	Yes
Tatay-Manteiga et al. (2019)	Yes	Yes	Yes	No	No/Yes	Can't tell	Yes
Vedel Kessing et al. (2020)	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Vothknecht et al. (2013)	Yes	Yes	No	No	Yes	Yes	Yes
Walshe et al. (2007)	Yes	Yes	No	No	Yes/No	Can't tell	Yes

Both codes given when coders disagree

siblings, with 16 of 20 calculated effect sizes showing poorer outcomes for siblings than controls. A few studies showed siblings had more positive or similar outcomes as the healthy control individuals. It also seemed the studies with the highest quality reported negative sibling outcomes. In contrast to previous reviews, we focused on healthy controls only. This means the control groups used in the current study were similar in terms of psychiatric profiles. This provides a clear indication that adolescent and young adult siblings are at increased risk of negative outcomes compared to controls. In terms of associated variables, the present review showed that sibling age, sex, level of education, perceived quality of maternal care, premorbid adjustment of the person with SMI, as well as contextual factors (i.e., contact with health care professionals) were associated with sibling outcomes. None of the studies considered genetics and/or multiple level risk factors within the same study, which makes it tricky to systematically synthesize the state-of-the-art regarding predictors.

In terms of the negative sibling outcomes, we found higher rates of psychiatric disorders, poorer sibling relationship quality, higher rate of previous psychological problems, and more emotional distress among siblings of persons with SMI. Furthermore, the siblings reported impaired functioning in several domains, i.e., academic functioning, occupational functioning, cognition and autonomy, and more emotional and practical burdens, as well as higher rates of a family history of psychiatric disorder. Walshe et al. (2007) have suggested that academic problems found among siblings may reflect a genetic risk for SMI (e.g., schizophrenia). Genetic risk may also partly explain the increased prevalence of psychopathology found among siblings, such as in the two register studies included in the review (Arajärvi et al., 2006, Vedel Kessing et al.,

2020). The mental health problems and distress in siblings of persons with SMI demonstrated in the present review are in line with research showing increased risk of BD, as well as other psychiatric disorders, in first-degree relatives of patients with BD. This increased risk is explained by both psychological mechanisms and genetic factors (Lambert et al., 2016).

The family ecology model (Pedersen and Revenson, 2005) represents a model for understanding psychological mechanisms in families with chronic parental illness. However, this model may also explain some of the negative outcomes in siblings of persons with SMI. According to the model, a role redistribution mediational hypothesis is suggested for families with a chronic ill family member. That is, after the onset of SMI, siblings may have to take on extra chores, tasks, and/or responsibilities. This may reduce the time and energy available for e.g. recreational activities that have been proven to be beneficial for the mental health and well-being for the sibling of persons with SMI (Kinsella et al., 1996). The model has some limitations. It does not mention contextual factors beyond the family (e.g., community and cultural factors), and does not consider the genetic vulnerability contributing to negative outcomes in mental health and/or well-being of siblings.

In terms of similar outcomes for both siblings and healthy control individuals, the present review showed that siblings of individuals with SMI reported comparable levels of quality of life as healthy control individuals. The continuum model proposed by Powell and Gallagher (1993) represents a possible way of understanding how disability in a person is associated with a sibling's mental health and well-being in a more or less positive way, and why some outcomes may be shown to be similar for both siblings and control groups. Different factors that seem to contribute to

where siblings are placed on this continuum include family characteristics such as socioeconomic status, family size, and religion. Parental attitudes and expectations and characteristics of both the sibling with SMI and the sibling without SMI were found to contribute, such as gender, age, temperament, and the severity of the persons' illness.

The positive outcomes for siblings included more use of problem-focused coping, more involvement in mental health arenas, and better mental health knowledge than the general population. These results are in line with a previous review on sibling outcomes, (Shivers and Textoris, 2021), and supports the credibility of the findings, even though different inclusion criteria were applied in the reviews. Problem-focused coping strategies aim to solve problems and reduce the effects of stressful events (Orsmond et al., 2009), and are associated with appraising an illness/problem as manageable. In contrast, appraising the illness/problem as a threat or something outside their control may be associated with emotion-focused coping, strategies more closely associated with depression and reduced self-esteem (Dysvik et al., 2005). This may explain why problem-focused coping has been associated with mental health and well-being for siblings of persons with SMI (Avcioglu et al., 2019). Thus, siblings who appraise the SMI as manageable, something they have the resources to cope with, may be more inclined to apply problem-focused coping as opposed to emotion focused coping.

Siblings of persons with SMI participated more in mental health arenas, for instance by choosing a career in the helping professions. Previous research suggests that siblings who participate in a wide range of activities outside the home and establish healthy relationships have sources of satisfaction and may develop a feeling of autonomy (Hoover and Franz, 1972). The concept of "constructive escape" has been used in the sibling research field, indicating that the child in families with chronic illness in a family member may be temporarily relieved from stress in the environment (e.g., engaging in outlets or activities), either mentally or physically, through activities or relationships outside the family (Kinsella et al., 1996).

Siblings of persons with SMI had better mental health knowledge than the general population (e.g., Sin et al., 2016). This is in line with a more general finding that a relationship with a person with mental illness is associated with better mental health knowledge (Henderson and Thornicroft, 2013). It is plausible that better mental health knowledge, for some siblings, may function as a protective factor against negative outcomes, but this is an issue that needs to be examined.

The mixed sibling outcomes in the present as well as previous reviews demonstrate the need for continued research in this field. Mixed results regarding sibling outcomes have been explained as the result of studies applying

different measures, samples, and control groups (Jayasinghe et al., 2023; Shivers and Textoris, 2021). The current review addresses one of these issues by including only studies with healthy control individuals as comparison groups. However, even with this restriction, some sibling outcomes were still mixed. To better understand sibling outcomes, in addition to restricting type of control groups, future research should also include samples with a narrower age span and/or developmental levels, and more homogeneous categories of outcome (e.g., internalizing symptoms, self-concept),

Furthermore, future studies should focus more on identifying predictors and moderating variables, examining predictors of outcomes to identify possible sub-groups of siblings with difference regarding risk of negative outcomes. Theories explaining the development of psychopathology is of relevance since some of the siblings seem to struggle in life and may develop psychological problems or symptoms. Emotion-focused therapy (EFT) explain psychopathology by looking at how emotions are not being met in a regular basis in a person's childhood (Elliott et al., 2004). For siblings this may be of relevance since the parents often are engaged with the sibling with an illness and may have limited time to attend to the needs of the other/s sibling/s.

Regarding associated variables to sibling outcomes, sisters reported higher emotional distress than brothers. This is in line with more general research findings showing that females are more likely to provide care for relatives with chronic illnesses, and that these care responsibilities are associated with more mental health problems, insomnia, somatic symptoms, and lower life satisfaction (Haugland et al., 2020; Shivers and Textoris, 2021). In terms of age, the label "young adult carer" has been used to describe individuals between 18 and 25 years who provide support, assistance or informal care to family members with disability, chronic physical or mental illness, or substance misuse (Becker and Becker, 2008). This developmental phase, often termed emerging adulthood, may be particularly difficult, as the care-provider may struggle to balance their time between caring and being independent, being left with little energy to care for his/her own needs (e.g., completing an education, choosing a career, socializing with friends, establishing romantic relationships). Haugland et al. (2020) found several factors associated with poorer outcomes in young adult carers, for instance higher level of financial stress, single parent families, and coming from migrant families. The studies included in this review did not provide information about caring responsibilities among the siblings of persons with SMI. However, findings from the carer literature may apply to siblings of persons with SMI and the associated contextual factors identified in this research field need to be investigated in further studies on siblings of persons with SMI.

Finally, theoretical models are welcomed, to explain the negative outcomes as well as the variation in outcomes among siblings of persons with SMI. In addition to focusing on factors associated with sibling outcomes this may increase our understanding of how SMI in a person may influence siblings and the moderating and mediating mechanisms explaining such associations.

Limitations

This review has limitations. A major limitation, which also reflects challenges in the field, is that the findings are based on only 11 studies, each examining very different outcomes. Based on the methodological quality coding only two studies were identified as high on methodological quality, whereas the remaining nine had important weaknesses, especially regarding the representativity of the samples. This points to the need for methodological improvements in future studies on siblings of persons with SMI (e.g., larger, more representative samples, more restricted age spans, the use of more standard outcome variables and measures across studies). The limited methodological quality of many of the studies suggest that the findings in the present review must be interpreted with caution.

There were also limitations regarding the search process. First, the constructs “mental health” and “well-being” were not included as search terms, but were terms applied when summarizing the findings as the most optimal common terms for variables included. Although specific terms were included to capture these constructs (e.g., mental ill, adjustment, coping, psychological adaptation), this limitation could potentially have excluded relevant studies. Second, unpublished studies were not included, which may have biased the review towards conclusion of differences existing between siblings of persons with SMI and control groups.

Most of the included studies were cross-sectional. Therefore, it is not possible to conclude about causality concerning sibling outcomes and the SMI. Common genetic and/or environmental factors may affect both the person with SMI and his/her sibling. As the studies cannot provide answers regarding direction of associations between SMI in a person and siblings outcomes, future research should include longitudinal studies to help disentangle the direction(s) of these associations. This will ease the interpretation and increase the validity of findings. There seems to be an unbalance in the sibling sex represented in the studies, with more males in the samples with SMI, and more females in the sibling samples. Unfortunately, the relative percentage of women versus men in the different sample could not be calculated due to lack of information in several of the studies.

We relied on healthy controls only in our comparison analyses, which enhances interpretability of the sample differences. Nevertheless, less than half the studies used control groups that were matched on at least one variable, and there were differences in how the control groups were recruited across studies. The sample sizes of the control groups also varied considerably, which does challenge how clearly we can conclude regarding outcomes.

Practical Implications

The results from the current review indicate mostly negative, but also some positive outcomes in siblings of persons with SMI. This has several practical implications. First, the findings are relevant for interventions and support programs aiming to help families when a member has a SMI, for instance by focusing on early intervention focusing on all family members when a sibling has a SMI. A possible intervention could be EFT’s skill training program for parents, that can help parents learn how to meet the emotions and needs of the well-siblings (Hagen et al., 2019). Second, it may be of importance to focus more on female siblings, since it seems that they may be at risk for developing more negative outcomes.

Conclusion

The increased risk for mental health problems in siblings of persons with SMI needs to be acknowledged and receive appropriate support from authorities, as well as professionals working within health and educational systems. There is a need to develop specific interventions aiming to alleviate and/or prevent adjustment problems in the siblings. A family perspective is indicated when treating persons with SMI. Finally, limitations in research studies on sibling outcomes need to be addressed and improvement in methodological quality of the studies is warranted.

Data availability

Further data available upon request.

Material availability

Available upon request from the second author.

Code availability

Available upon request from the second author.

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Compliance with ethical standards

Conflict of interest The authors declare no competing interests. Note that a preliminary version of this review was submitted by the first author as a master thesis at the Faculty of Psychology, University of Bergen, Norway, in 2020. The search was updated in Jan 2023.

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