UNIVERSITAS BERGENSIS

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The mental health and well-being of siblings of individuals with severe mental illness: A systematic review

HOVEDOPPGAVE

profesjonsstudiet i psykologi

Ingvild Synnøve Lønning

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Forord

Jeg vil gjerne benytte anledningen til å rette en stor takk til mine to veiledere, Bente Storm Mowatt Haugland og Krister Westlye Fjermestad, som har stilt med god rådgivning og støtte underveis gjennom oppgaveskrivingen. Jeg er svært takknemlig for nyttige diskusjoner og refleksjoner rundt tematikken. Videre vil jeg takke bibliotekar Kjersti Aksnes-Hopland for flere gode møter og hjelp i litteratursøkingsprosessen, og min mann Christian Lønning for hjelp med figur og tabeller.

Haugesund, 08.12.2021

Ingvild Synnøve Lønning

Innholdsfortegnelse

Sammendrag

Hensikten med denne studien var å oppsummere kunnskap om psykisk helse og psykologisk velvære hos søsken til personer med alvorlig psykisk lidelse. Foreliggende studie er en litteraturgjennomgang av kvantitative studier om psykisk helse og psykologisk velvære hos søsken til personer med alvorlig psykisk lidelse publisert mellom januar 1990 og mai 2021, og hvor søsken til personer uten alvorlig psykisk lidelse blir brukt som sammenligningsgruppe. I foreliggende studie ble det undersøkt om psykisk helse og velvære hos ungdom og voksne (>12 år) var assosiert med det å ha et søsken med alvorlig psykisk lidelse, og hvilke faktorer som eventuelt var assosiert med dette. Det ble gjennomført søk i databasene PsycINFO, Cumulative Index to Nursing and Allied Health Literature (CINAHL) og Web of Science. Tretten artikler ble inkludert i studien. Søsken til personer med alvorlig psykisk lidelse viste en blanding av positive, negative og sammenfallende utfall på mål på psykisk helse og velvære sammenlignet med kontrollgrupper bestående av søsken til personer uten alvorlig psykisk lidelse. Det kommer også frem at noen demografiske variabler, faktorer ved familie relasjonen og helsevesenet, og karakteristikker ved broren eller søsteren med alvorlig psykisk lidelse er assosiert med søskens psykisk helse og velvære. Resultatene indikerer at det bør igangsettes forebyggende tiltak som omfatter hele familien når et medlem har en alvorlig psykisk lidelse. I tillegg trenger forskningsfeltet utarbeiding og testing av modeller som forklarer de komplekse sammenhengene og resultatene.

Abstract

The aim of this study was to gain knowledge about the mental health and the well-being among siblings of individuals with severe mental illness (SMI) and factors that are associated with the mental health and well-being of siblings. The current study is a literature review of quantitative studies on siblings of individuals with SMI conducted on studies published between January 1990 and May 2021 and where siblings of persons without SMI is used as a control group. The databases PsycINFO, Cumulative Index to Nursing and Allied Health Literature (CINAHL) and Web of Science were searched. Thirteen articles were included in the study. Overall, the studies indicated that siblings of persons with SMI have a mix of positive, negative, and similar outcomes compared to controls, and that there are some demographic factors, factors in the family relationship and health care, and characteristics with the sibling with SMI that are associated with sibling mental health and well-being. The findings indicate that preventive interventions should be initiated for helping the entire family when a member has a SMI. Also, different models should be tested for siblings of persons with SMI for further understanding the associations between SMI and the mental health and well-being of siblings.

Introduction

Siblings of Persons With Severe Mental Illness

Severe mental illness (SMI), e.g., bipolar disorders (BD), schizophrenia, eating disorders, psychosis, and autism spectrum disorder (ASD), directly affects persons who suffer from these illnesses. SMI may potentially also affect the mental health and well-being of persons close to him/her, such as parents, friends, and siblings. Siblings may for example be affected by the changes the illness poses to the sibling relationship, and/or the range of emotions they experience in response to their sister/brother’s SMI (Sin et al., 2014).

The sibling relationship is unique in that it typically is the longest lasting relationship in a person’s life (Orsmond & Seltzer, 2007), and that it is ascribed rather than obtained (Cicirelli, 1995). This relationship is also important as it may affect a child’s emotional development. For instance, a study found that a balance of nurturance and conflict in the sibling relationship fosters development of emotional understanding, self-regulation, and a sense of belonging and comfort in children (Brody, 2004). The presence of SMI in a sibling has been found to be a factor that can influence the life course trajectory of the sibling relationship by affecting the closeness between the siblings (Orsmond & Seltzer, 2007). It has also been found to put siblings at heightened risk for social and adjustment problems (Orsmond & Seltzer, 2007).

However, mixed results have been reported regarding the outcomes for siblings of a brother or sister with SMI. In a literature review with 12 included studies on siblings of persons with SMI, Meadan et al. (2010) found that some of the included studies reported positive and others reported negative outcomes among the siblings. The reported sibling outcomes were social/behavior and emotional adjustment, self-concept, perceived social support, and sibling relationship. However, the comparison groups in these studies varied widely. Some studies used normative data, e.g., children participating in large-scale studies on mental health, some included control groups of siblings of typically developing children, i.e., persons with no known mental illness, as comparison, some compared siblings of individuals with disabilities (e.g., Down syndrome, intellectual disabilities, or learning disabilities), whereas others had no comparison group (Meadan et al., 2010). This variety of comparison groups was already mentioned 16 years ago as one of four main methodological challenges in the field of sibling research (Hodapp et al., 2005). Other methodological challenges were the use of small convenience samples, limited ability to generalize the results to other siblings due to sampling procedures, and a lack of focus on genetic as well as environmental influences (Hodapp et al., 2005).

Also Shivers and Textoris (2021) reported mixed results regarding sibling outcomes in their literature review on outcomes of siblings of persons with SMI. The authors found that half of the 56 included studies in the review reported that siblings had fewer negative outcomes than the comparison groups, whereas half of the studies reported that siblings had more negative outcomes. The studies in this review addressed outcomes such as siblings' well-being, behavior problems, internalizing behavior, burden, and the needs of siblings. Also, this review reported a diversity of comparison groups in the included studies. In studies where siblings of persons with SMI had better outcomes (e.g., less emotional distress, and less sibling conflict), the control groups comprised parents of persons with SMI, typically developing siblings, community samples, or siblings of individuals with an intellectual disability. The comparison samples in the studies where siblings of persons with SMI had worse outcomes consisted mostly of community samples or siblings of typically developing individuals (Shivers & Textoris, 2021). The diverse set of comparison groups makes it challenging to interpret results about siblings' outcome and to compare findings between studies.

In a recent literature review on siblings of children with neurodevelopmental disorder, i.e., ASD, epilepsy, attention deficit hyperactivity disorder (ADHD), and cerebral palsy, the authors found four studies reporting reduced quality of life (QoL), and three studies reporting no reduced QoL among siblings compared to typically QoL measured using generic instruments (i.e., used across different clinical contexts making comparison possible). In addition to QoL, overall family functioning, self-reported anxiety and depression, and impact of the different neurodevelopmental disorders on health related QoL and well-being among the siblings were assessed (Lamsal & Ungar, 2021).

The Meadan et al (2010) review included 12 studies, whereas the review by Shivers and Textoris (2021) included 56 studies and the Lamsal and Ungar (2021) review included seven studies. Across these three previous reviews, there were no overlap in terms of included studies. The age ranges in the three reviews were also different. The study samples in Meadan et al.' (2010) review comprised a sibling range of 3-18 years. The age range of sibling in the Shivers and Textoris' (2021) review was 6-81 years, with the majority of studies comprising adult samples, whereas Lamsal and Ungar (2021) included studies with an age range of siblings between six to 30 years. Even with different age ranges all three reviews reached a similar conclusion: the large differences in measures and outcomes, research methodologies, age range, and comparison groups applied are serious limitations reducing the possibility of drawing any clear or consistent conclusions from the research studies (Lamsal & Ungar, 2021; Meadan et al., 2010; Shivers & Textoris, 2021). Thus, further research is needed where for instance more congruent (appropriate) comparison groups are applied. The current study is a literature review examining outcomes for siblings of persons with SMI as well as factors associated with sibling outcomes. The review includes studies that compare siblings of persons with SMI to siblings of typically developing persons, or alternatively to a normative sample.

Different theoretical models have been used to try to explain the effects on relatives when a family member has an illness. Powell and Gallagher (1993) proposed a theory that suggests a continuum where the influence a child with a disorder has on a sibling range from very negative outcomes at one end to very positive outcomes at the other end. According to the authors, factors such as the age of the siblings, the age of the child with the disability, the number of siblings, and the severity of the disorder, the adjustment and attitude of the parents, as well as dyad composition factors such as age and sex differences, and the context of the sibling activities may be associated with the sibling's adjustment (Powell & Gallagher, 1993).

Pedersen and Revenson (2005) developed a model where they suggest a family ecology framework for family functioning and adolescent well-being when a parent has a physical illness. The model has not been applied for siblings of individuals with a SMI. However, the four principles in the framework may be adopted also to examine sibling outcomes in families with persons with SMI. The four principles are: 1) individual behavior can only be understood within its social context; 2) individuals exist within a number of interdependent systems or contexts; 3) the reciprocal relationships between individuals and the social systems with which they interact are essential for understanding development and adaption; and 4) factors beyond the level of individual attributes (e.g., social and cultural factors) must be included to understand adaptational processes (Pedersen & Revenson, 2005). In the original framework, characteristics of the parental illness, such as type, and severity of illness is expected to affect adolescent well-being and family functioning indirectly through several individual- and family-level mediators. More specifically, illness severity is expected to impact appraisals of stigma and threat, physiological stress responses, the distribution of roles and responsibilities within the family, and daily hassles. These processes will in turn influence family functioning and adolescent well-being. The role redistribution mediational hypothesis in the framework implies that if a parent with an illness is incapable of fulfilling the roles s/he previously filled, other family members may adopt to these roles. Both models reviewed above offer possible ways of explaining the mixed results regarding positive and negative outcomes for the siblings of persons with SMI.

Examining risk factors and protective factors associated with sibling mental health and well-being may increase our understanding of the variation in outcomes in siblings, as well as enhance our ability to prevent negative outcomes for siblings of persons with SMI. Besides having a sibling with a SMI, there may be a range of other factors also affecting sibling outcomes. Findings regarding which factors influence the risk for negative social, emotional, and academic functioning among siblings have been mixed. Shivers and Textoris (2021) found that female sex, degree of belief in their sibling's ability to control their own behavior, and greater severity of mental illness symptoms in the sibling with SMI were factors associated with more negative sibling outcomes. Macks and Reeve (2007) found that male sex, being from a family with low socio-economic status (SES), only having one sibling, and being older than the child with the disability were associated with greater risk among siblings of children with ASD. Meadan et al. (2010) examined various factors associated with sibling adjustment in their review and found that some studies identified sisters to be more social and have fewer problems with prosocial behavior than brothers, whereas other studies showed that sibling sex did not affect socialization. In the same review other demographic factors such as birth order, sex-to-sex match of siblings, and family size were examined, but also here the findings were mixed.

Living with a child with ASD seem to have an increasingly negative effect on a typically developing sibling as the number of demographic risk factors e.g., being a male, being older than the child with disability, increase (Macks & Reeve, 2007). Mack and Reeve (2007) suggested that this may explain the lack of consistency among past studies. They propose that the studies reporting positive influences on siblings are using samples of children not exposed to multiple demographic risk factors compared to samples in studies reporting negative influences where children may have multiple demographic risk factors. Perhaps this may also be the case for other illnesses besides ASD, and for other age groups than children.

Besides demographic factors, also factors in the family relationship may be associated with sibling outcome. For instance, Meadan et al. (2010) found that level of family support and level of maternal stress affected siblings' social and academic adjustment in some studies. Because of these inconsistent findings the current review will focus on demographic factors and other associated factors as a part of a second aim of the study.

Sibling Outcomes in Different Phases of Life

Findings from different studies show that the sibling relationship is affected differently by a mental disorder in one sibling in various phases of life. For instance, Rossiter and Sharp (2001) found that developmental disorders in a sibling had a negative effect on sibling relationship during childhood and adolescence whereas in adulthood a small positive effect was found. According to the authors the changes in sibling relationship in adulthood may be a result of adults having better strategies to cope with the mental disorder of a sibling.

In a literature review, Orsmond and Seltzer (2007) examined siblings of individuals with ASD across the life span. During childhood, siblings were play partners and sources of support, whereas in adolescence and young adulthood they experienced decreased contact. According to Orsmond and Seltzer (2007) further changes seemed to occur in the sibling relationship during adulthood and later adulthood, with the sibling relationship getting more important when other family members and social supports became less available. Differences in the sibling relationship across the life course demonstrate the importance of studying sibling outcomes and factors associated with sibling outcomes across different stages of life. Adolescence is for example, a time of important changes in most areas; biologically, socially, emotionally, as well as in cognitive development (Sawyer et al., 2012). Transitions from childhood to adolescence and later to adulthood also affect intergenerational relations. During adolescence and adulthood both parents and siblings change the relational asymmetry that usually characterize childhood (Levitt et al., 2007). Little research has been done on the mental health and well-being of siblings of persons with SMI in a life course perspective from early adolescence and through adulthood (Orsmond & Seltzer, 2007). Consequently, the current review focuses on siblings of SMI during these important developmental phases.

Defining Severe Mental Illness, Mental Health, and Psychological Well-Being

When conducting research on siblings of individuals with SMI, clear definitions of SMI are needed. Type of psychiatric diagnosis, illness duration, and level of disability are criteria that have been suggested as definitions of SMI (Martínez-Martínez et al., 2020). In a review on the operationalization of SMI Martinez-Martinez et al (2020) found that all included studies were based on psychiatric diagnoses, but none applied criteria for inclusion or exclusion of psychiatric diagnoses. Furthermore, the authors found large variations in how SMI was defined across studies. Studies that used a disability criterion stated that the subjects needed to have a certain level of impairment due to mental illness, for example by receiving financial benefits or being unable to perform everyday activities. Regarding duration of the illness, this varied between the included studies, with some studies referring to two years since time of diagnosis or beginning of treatment, while others measured the days of hospitalization in the previous years (Martínez-Martínez et al., 2020).

According to Martinez-Martinez et al. (2020), the different definitions of SMI can be classified into two major groups. The first was the State of New Jersey definition of “severe mental illness” (Mueser et al., 2015). According to this, SMI implies that you have: 1) a diagnosis of schizophrenia, schizoaffective disorder, major depression, BD, severe post traumatic stress disorder or borderline personality disorder; 2) significant functional limitations in major life activities within the past 3-6 months due to the mental disorder; and

3) during the past 2 years, either two or more treatment episodes of greater intensity than could be treated with out-patient services or a single episode lasting 3 months or more, or disruption in normal living situation to the point that supportive services were required to maintain the patient in that living situation, or law enforcement officials intervened.

The other major SMI definition emphasizes the impairments arising from having a psychiatric disorder (Martínez-Martínez et al., 2020). Here factors, such as disability, duration, safety, the number of hospital admissions, formal or informal support, and aggressiveness, were included. Most of the studies, however, applied one or more of the following criteria: psychiatric disorder, disability, and/or duration of the illness.

Despite the large variation in psychiatric diagnoses included in studies of siblings with SMI, siblings of patients with eating disorder and ASD are usually not included, even though these disorders satisfy criteria of being severe psychiatric diagnoses, often with long duration, and high level of disability. Thus, in addition to BD, schizophrenia and psychosis, the current review will also include eating disorders and ASD.

The present review applied the concepts of mental health and psychological well-being as concepts for evaluating outcomes among sibling of persons with SMI.

The World Health Organization (2018) defines mental health as the presence of mental disorders or disabilities, but also mental health beyond this as “a state of well-being in which an individual realizes his or her own abilities, can cope with the normal stresses of life, can work productively and is able to make a contribution to his or her community”. Mental health may be determined by a range of socioeconomic, biological, and environmental factors, with some probably associated with more negative outcomes than others.

The concept of psychological well-being comprise six dimensions: self-acceptance, purpose in life, environmental mastery, positive relationships, personal growth, and autonomy (Ryff & Singer, 2008). According to Ryff and Singer (2008), the self-acceptance dimension is a type of self-evaluation that involves awareness and acceptance of both personal strengths and weakness, whereas positive relations with others are highlighted as an important component of mental health. Personal growth is described as part of a self-realization process, involving a continual process of developing one’s potential.

The other three dimensions, purpose in life, environmental mastery, and autonomy, 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. When assessing mental health and psychological well-being, the presence of mental health disorders (i.e., depression or anxiety) often are involved in the operationalization of the term mental health, whereas it for the term psychological well-being rather is more a focus on the positive aspects of functioning. To get a broad perspective on sibling outcomes, the current review will focus on both terms.

Aim and Research Questions

The aim of the current study is to systematically review studies on mental health and well-being of persons aged 12 years and above who are siblings of individuals with SMI. In addition, the study aims to identify potential factors associated with the functioning of the siblings in the reviewed studies. The research questions are:

1. Is mental health and well-being of adolescents and adults associated with having a sibling with SMI?
2. Which factors are associated with mental health and well-being of adolescents and adults of siblings with a SMI?

Methods

Inclusion- and Exclusion Criteria

Studies on siblings of individuals with SMI were identified through systematic electronic searches, as well as supplementary hand searches in references of identified studies and literature reviews. The following criteria were deemed potentially eligible for inclusion: (a) studies published between January 1990 and May 2021, (b) studies published in peer-reviewed Scandinavian or English language journals, (c) studies on typically developing siblings aged 12 years or older of individuals with SMI, (d) studies including findings on the mental health and/or well-being of typically developing siblings of individuals with SMI, (e) the sibling with illness needed to have one of the following diagnoses: BD (any type), schizophrenia, eating disorder (any type), psychosis, or ASD, and (f) the studies had to compare siblings of individuals with SMI to either siblings of typically developing individuals or normative samples. Books, dissertations, and other publications that had not undergone peer review were excluded, as well as literature reviews and qualitative studies. Studies that included individuals with physical illnesses, intellectual disabilities, learning difficulties, or other developmental disabilities than ASD were also excluded.

Literature Search

Systematic electronic searches were conducted through the databases PsycINFO, Cumulative Index to Nursing and Allied Health Literature (CINAHL) and the Web of Science databases in which relevant research may be conducted. The searches were conducted on 2nd May 2021. Keywords related to the mental health and well-being of siblings of individuals with SMI were used. Terms for siblings were combined with terms for ill sibling’s mental illness, and with terms for adjustment, psychological adaptation, well-being, and coping in the well sibling. Keywords regarding suicide were used in the beginning of the search process, based on knowledge that SMI often is involved in suicide and suicide ideation (Li et al., 2021). Studies regarding suicide were later removed from the review because they did not have results regarding specific mental disorders. All keywords applied in the review 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 presented in Table 1.

Table 1

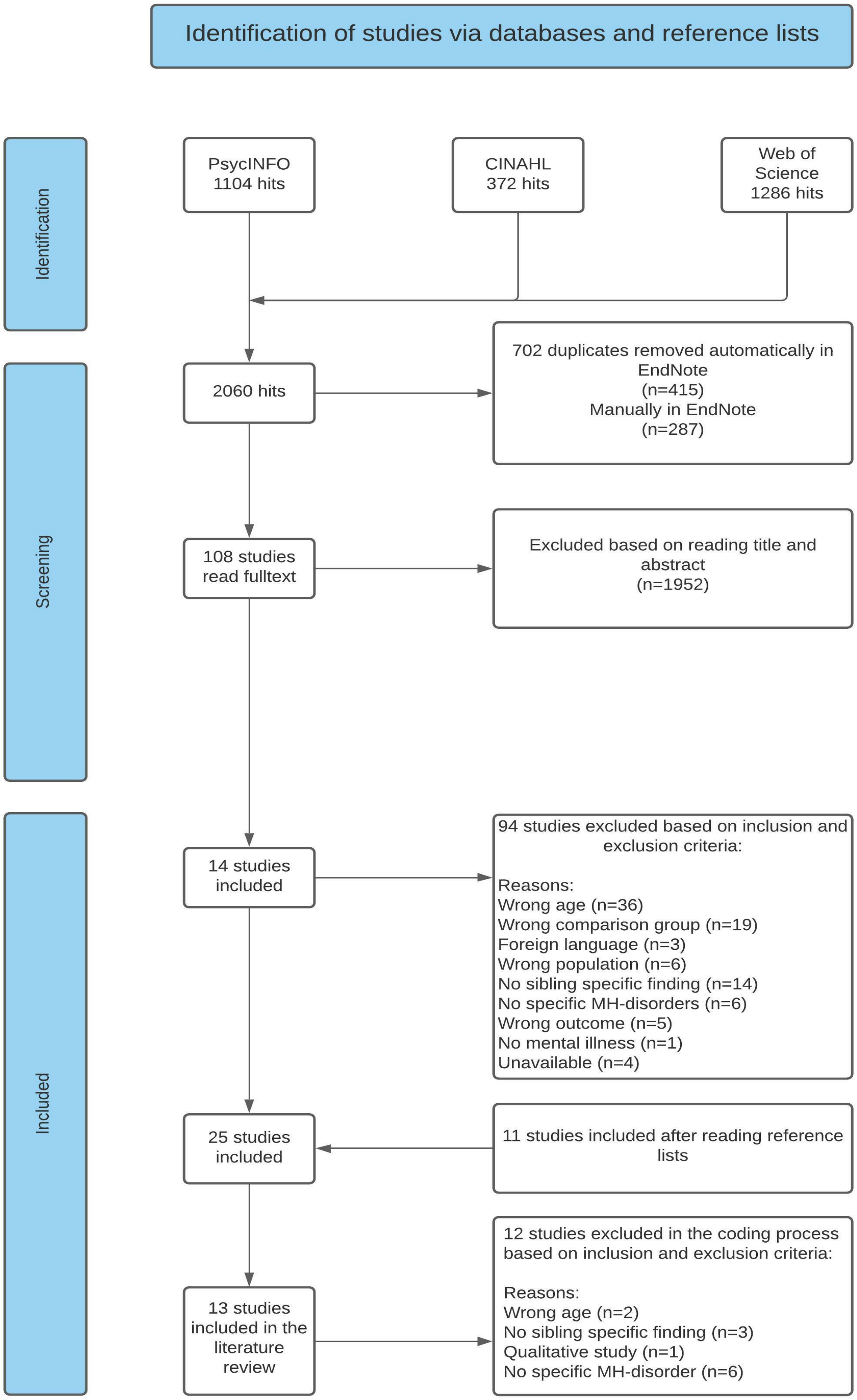
Search terms and combinations

		Database		
		PsycINFO	CINAHL	Web of Science
	Keywords	Results		
1	sibling*	24488	13516	61423
2	sister*	6527	4821	60116
3	brother*	6617	3214	25906
4	1 OR 2 OR 3	32537	19413	139425
5	mental* ill*	61392	30187	92645
6	mental* disorder*	178041	78097	161217
7	psychiatric ill*	8573	3764	43154
8	affective ill*	1403	257	11609
9	psychiatric car*	5632	12091	59559
10	psychiatric disorder*	40410	14498	124708
11	affective disorder*	35625	11557	38433
12	bipolar ill*	1305	651	15401
13	bipolar disorder*	42563	15561	59275
14	(manic depressive adj (ill* or disorder*))	1251	92	1998
15	psychosis	57269	17539	64943
16	psychotic	44662	18780	38565
17	schizophreni*	141784	34233	201102
18	suicid*	72741	43592	103927
19	"self-harm"	6928	5042	9205
20	5 OR 6 OR 7 OR 8 OR 9 OR 10 OR 11 OR 12 OR 13 OR 14 OR 15 OR 16 OR 17 OR 18 OR 19	458256	200660	639342
21	coping*	92532	60361	88792
22	resilien*	37680	16489	115269
23	self-efficacy*	52298	30781	62712
24	selfefficacy*	158	252	234
25	protective factor*	17292	9383	111722
26	adjustment*	119993	87034	323470
27	adaptation, psychological*	44498	31431	9891
28	support*	707002	562909	2686966
29	21 OR 22 OR 23 OR 24 OR 25 OR 26 OR 27 OR 28	938106	710219	3250623
30	4 AND 20 AND 29	1104	372	1286

Searches in PsycINFO, CINAHL and Web of Science generated 2762 hits. These hits were exported to EndNote, with 702 duplicates being removed either automatically as part of the export to Endnote or manually when screening through titles and abstracts of the studies. A total of 2060 hits were screened by reading the title and/or abstracts of the articles. This resulted in 246 articles included for further review. A team of three (the candidate, main supervisor, and co-supervisor) screened the 246 articles in pairs. Based on reading titles and abstracts, 108 studied were selected for further review. Applying teams of two evaluators at this stage was done to enhance reliability in the inclusion-and exclusion process. The candidate and the supervisors then read and screened 36 studies each. From this process 94 studies were excluded. A supplementary search in the reference lists of the included studies was done to include relevant studies not identified in the systematic search. This generated 11 studies. Thus, a total of 25 studies were included and read in full text. Based on the full-text reading twelve more studies were removed as they did not fulfil the inclusion criteria, mainly because they did not refer to a specific mental health disorder. The remaining 13 studies were coded for content and scientific quality. Final coding was based on comparison between the candidates scores and the scores of one of two other raters (supervisors). For an overview of the search strategy and inclusion process, see Figure 1.

Figure 1

The search strategy and selection process



Coding of Scientific Quality

As a part of the coding process, the scientific quality of the studies was evaluated, using the Mixed Methods Appraisal Tool (MMAT) (Hong et al., 2018). The MMAT is a critical appraisal tool designed for evaluating qualitative, quantitative, and mixed methods studies. Only the items regarding quantitative descriptive studies were selected for the evaluation of the studies in the present review. The following variables were coded, all as “yes” or “no”, or “can’t tell”, in line with the MMAT protocol: 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. The candidate coded the scientific quality of all the included studies, whereas 56% of the included studies were double coded by the supervisors.

Results

Thirteen studies met the inclusion criteria. An overview of the descriptive study data from these studies is provided in Table 2. This information is also presented briefly below.

Description of Studies

Study Designs

There were 11 cross-sectional studies, of which one was a retrospective study. The two remaining studies was a registry study and a population-based longitudinal registry study.

Sample Characteristics

The sample sizes ranged from 22 to 12923 siblings, with an age range from six to 72 years. Note that only findings concerning participants aged 12 years and above were included in the current review.

The majority of the studies originated from European countries. Two studies were from Italy, two from Netherland, one from Finland, one from France, one from England, one

Table 2

Descriptive study data

Author (year)	Country	Recruitment setting	Design	Comparison group	Instruments assessing sibling outcomes
Arajärvi et al. (2006)	Finland	The Finnish Hospital Discharge Register.	Registry study	Sample drawn randomly from Finnish general population.	SCID-I, SCID-II, and SANS
Barak et al. (2005)	Israel	User association for schizophrenia + friends (controls).	Cross-sectional	Siblings of persons without disorder.	BAS, a modified version of questionnaire assessing coping, feelings toward sibling, sibling relationship, life space and meaning.
Gold (1993)	Canada	Three agencies in Toronto area for children with autism and their families.	Cross-sectional	Siblings of nondisabled boys.	CDI, CBCL, Questions for Siblings, and Siblings' Domestic Work and Caregiving Work
Pierazzuoli et al. (2020)	Italy	Psychiatric facilities and psychiatric patient family associations.	Cross-sectional	Normal control siblings.	PBI, and MMPI-2
Plessis et al. (2020)	France	An online questionnaire.	Cross-sectional	Normal control siblings.	ASRQ-S, HAD, and SES
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	Sibling with a SMI, and healthy control participants.	PAS

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	Age-matched population norms from Health Survey for England & Attitude to Mental Illness Survey.	WEMWBS, MAKS, and ECI
Tatay-Manteiga et al. (2019)	Spain	University hospital.	Cross sectional	Patients with BD at early and late stage compared to well siblings (n=23) and healthy controls (n=21).	WHO-QoL BREF
Trubia et al. (2016)	Italy	Institute for Research on Mental Retardation and Brain Ageing (services of diagnosis, psycho-educational training and rehabilitation).	Cross sectional	Siblings of persons with typical/normal development AND siblings of persons with intellectual disablity (without ASD).	SD

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)	Randomly selected unaffected controls from general population matched on year of birth and sex.	Psychiatric disorders from registers
Verte et al. (2003)	Netherland	Treatment center for High Functioning Autism.	Case-control study, cross-sectional	Siblings of typically developing children matched on target child's age and sex, siblings' age and sex, birth order, age spacing and family size.	CBCL, MESSY, and SDQ-II
Vothknecht et al. (2013)	Netherland	Academic centers (Amsterdam, Groningen and Utrecht) and their affiliated mental health care institutions.	Cross-sectional	Patients and healthy controls.	SWN-K, WHOQOL, PANSS, and CAPE
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	Controls without personal or family history of psychotic illness.	PSA

SCID-I = Structured Clinical Interview for DSM-IV axis disorders (I) , SCID-II = Structured Clinical Interview for DSM-IV axis disorders (II) , SANS = Scale for the Assessment of Negative Symptoms, BAS = Burden Assessment Scale, CDI = Children's Depression Inventory, CBCL = Child Behavior Checklist, PBI = Parental Bonding Instrument, MMPI-2 = Minnesota Multiphasic Personality Inventory-2, ASRQ-S = Adult Sibling Relationship Questionnaire-short form, HAD = Hospital Anxiety and Depression, SES = Self-Esteem Scale, PAS = Premorbid Adjustment Scale, WEMWBS = Warwick-Edinburgh Mental Wellbeing Scale, MAKS = Mental Health Knowledge Schedule, ECI = Experience of Caregiving Inventory, WHO-QoL BREF = World Health Organization Quality of Life Scale, SD = Semantic Differentials, MESSY = Matson Evaluation of Social Skills with Youngsters, and SDQ-II = Self-Description Questionnaire II, SWN-K = The Subjective Well-being Under Neuroleptic Treatment Scale, WHOQOL = World Health Organization Quality of life, PANSS = Positive and Negative Syndrome Scale, CAPE = The Community Assessment of Psychic Experiences, PSA = Premorbid Social Adjustment Scale.

from Spain, one from Denmark, and one from Ireland. The three remaining studies were from Israel, USA, and Canada (Table 2).

Different recruitment settings were used in the studies. Four studies recruited samples from a combination of different settings (e.g., user associations and friends, psychiatric facilities and family organizations for psychiatric patient), two recruited from hospitals or treatment centres, two from national registers, one from a research institute, one from agencies which serve children with ASD and their families, one through web-based surveys, one from subjects participating in genetic studies on SMI patients and their siblings, and one from baseline data in a randomised controlled trial (RCT).

The included studies used a variety of methods assessing sibling outcomes, the most common of which was self-administered questionnaires.

The diagnoses of the sibling with a SMI were most often established through clinical interviews, and the mentally ill sample consisted mainly of men in the age range 9-69 years.

Sibling Outcomes

The findings regarding sibling outcomes have been grouped into three categories of better, worse, and same level outcomes compared to control groups. The results on characteristics regarding sibling outcomes are summarized in Table 3..

Better Psychological Well-Being Compared to Controls

Three studies reported on positive outcomes for siblings of persons with SMI compared to normal controls. The positive outcomes were problem-focused coping, involvement in mental health arenas and artistic activity, mental health knowledge, and the siblings' self-concept. Problem-focused coping is labelled a positive outcome because previous research has shown problem-focused coping in siblings of persons with schizophrenia to be associated with better well-being (Avcioglu et al., 2019).

Table 3

Mental health and well-being of siblings of persons with SMI

Author (year)	Mentally ill sibling sample	SMI	How was illness established?	Siblings of persons with SMI	Finding regarding sibling well-being
Arajärvi et al. (2006)	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.	n = 183. Psychotic sibling: (53.3% males). Aged 32-61 yrs, M age 44.4 (SD 6.3). Non-psychotic sibling: (47.7% males). Aged 24-72 yrs, M age 46.4 (SD 8.8).	The siblings had more psychotic disorders than comparison group. 37.7% of siblings had non-psychotic disorders, 16.4% had a psychotic disorder, 23.5% had other comorbid psychiatric disorders.
Barak et al. (2005)	n = 52. Age and sex not reported.	Schizophrenia	Not reported.	n = 52 (32% males). Aged 18-50 yrs.	More problem-focused coping, objective and subjective burden, and negative feelings toward sibling. Quality of sibling relationship declined after illness (improved with time in comparison group). 42.5% of siblings reported close relationship with their sibling with SMI compared to 95% in control group. Siblings more involved in mental health arena and artistic activity than controls.
Gold (1993)	n = not reported (100% males). Age not reported.	ASD	DCM criteria of ASD.	n = 22 (50% males). Aged 12-17 yrs, M age 13.52.	Siblings higher on depression.
Pierazzuoli et al. (2020)	n = 30 (76% males). Age not reported.	Schizophrenia spectrum disorder	Not reported.	n = 30 (16.7% males). Aged 36-63 yrs, M age 51.4.	Higher rate of family history of psychiatric disorder and previous psychological problems.
Plessis et al. (2020)	n = 201 (80.21% males). Age not reported	Schizophrenia	Diagnosed with schizophrenia by a healthcare professional.	n =201 (22.3% males). M age 37.9 (SD 12.08).	More emotional distress and a poorer sibling relationship compared to controls.
Shapiro et al. (2009)	n = 286 (75.5% males). Aged 16-64 yrs, M age 36.2 (SD 9.44).	Schizophrenia or schizoaffective disorder	DSM-IV diagnosis, made by either a clinical psychologist or psychiatrist using a	n = 315 (41% males). Aged 16-64 yrs, M age 36.94 (SD 9.96).	Academic functioning in childhood, adolescent, and early adulthood better for controls than for siblings.

			revised version of SCID.		
Sin et al. (2016)	n = 89 (62.2% males). Aged 15-57 yrs, M age 26.46 (SD 7.9).	FEP	No information.	n = 90 (15% males). Aged 16-58 yrs, M age 27.52 (SD 8.41). Biologically related, step-or half-siblings, or related by adoption.	Siblings, especially sisters, significantly poorer mental well-being, but better mental health knowledge than controls.
Tatay-Manteiga et al. (2019)	n = 48 (52.1% 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.	n = 23 (30.4% males). M age 41.5 (SD 11.8).	Siblings and healthy controls reported similar levels of QoL.
Trubia et al. (2016)	n = 31. Sex and age not reported.	ASD with ID	ICD-10 and according to ADOS.	n = 31 (45.2% males). Aged 12-42 yrs, M age 21.	No differences in perception of self (energy, positive affect and emotional stability) between siblings and controls.
Vedel Kessing et al. (2020)	n = 19 955. Sex and age not reported.	BD (having a single manic episode or BD) during 1995 to 2017. BD main diagnosis at any contact.	ICD-8, ICD-9, ICD-10.	n = 13 923. M age 38.7.	Rates of 'any psychiatric disorder' constantly around twofold increased throughout lifespan. Cumulative incidences from age 15 for any psychiatric disorder: 44.2% at age 80 with 27.6% for controls.
Verte et al. (2003)	n not reported (96.55% males). Aged 9-16 yrs, M age 11.83.	HFA (autism, Asperger's Syndrome or pervasive developmental disorder not otherwises specified).	According to DSM-IV.	29 siblings aged 6-17 and 29 control group siblings (no information on n or age in subgroup 12-16 yrs).	Sisters (12-16 yrs) had a more positive self-concept, and they were not more susceptible to adaptation problems than controls.

Vothknecht et al. (2013)	n = 600 (80% males). M age 27.3 (SD 7.1).	Schizophrenia	No information.	n = 594 (45.5% males). M age 27.9 (SD 8.0).	Siblings reported higher subjective well-being than their sibling with SMI.
Walshe et al. (2007)	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 (75.4% 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.	A) 39 unaffected siblings of persons with personal and familial schizophrenic (35.9% males). Aged 16-69 yrs, M age 34 (SD 7.8), and B) 67 unaffected siblings of persons with personal schizophrenic and non-familial schizophrenic (49.3% males). Aged 16-69 yrs, M age 35 (SD 7.6).	Poorer academic functioning during adolescence in siblings of people with familial schizophrenia (deteriorate in academic performance between childhood and adolescence).

ASD = Autism Spectrum Disorder, FEP = First Episode Psychosis, ID = Intellectual Disability, BD = Bipolar Disorder, HFA = High Functioning Autism, ICD-8/9/10 = International Statistical Classification of Diseases 8/9/10, DSM-IV = Diagnostic and Statistical Manual of Mental Disorders IV, ADOS = Autism Diagnostic Observation Schedule, PAS = Premorbid Adjustment Score, SCID = Structured Clinical Interview for DSM-IV Axis I Disorders, SADS-LA = Schedule for Affective Disorders and Schizophrenia - Lifetime Version and clinical information.

Across the studies, positive outcomes were measured using different self-report as well as questionnaires completed by significant others, e.g., parents.

Barak and Solomon (2005) assessed problem-focused coping in siblings by a questionnaire examining three types of coping: help-seeking, avoidance, and problem-solving. The siblings were also asked to indicate if they participated in various areas of activities: study, work, political involvement, social life, activities in the mental health field, artistic activities and addiction to alcohol or drugs.

Self-concept among siblings of persons with SMI was assessed with the Self-Description Questionnaire (SDQ) (Marsh, 1988; Verte et al., 2003). For siblings between 12 and 16 years, a version for adolescents was applied (SDQ-II). In Verte et al.'s (2003) study self-concept was operationalized as both academic abilities (general school ability, verbal ability, and abilities in mathematics) and non-academic abilities (physical abilities, physical appearance, peer relations, and parent relations).

Sin et al. (2016) reported on siblings' knowledge of mental health. This construct was measured using the Mental Health Knowledge Schedule (MAKS) (Evans-Lacko et al., 2010). Siblings' knowledge about mental health was in Sin et al.'s (2016) study operationalized as knowledge about mental health and level of recognition and familiarity with various conditions.

Overall, the included studies showed that siblings of persons with SMI used more problem-focused coping, were more involved in mental health arenas and in artistic activity (Barak & Solomon, 2005), and had a more positive self-concept compared to normal controls (Verte et al., 2003). The siblings were also shown to have better mental health knowledge than the general population (Sin et al., 2016).

More Mental Health Problems and Lower Level of Well-Being Compared to Controls

Nine studies assessed mental health outcomes for siblings of persons with SMI. The outcomes measured were psychosocial adjustment, quality of the sibling relationship, psychiatric disorders, or psychological problems, and family history of psychiatric disorder.

Psychosocial Adjustment. Two studies assessed psychosocial adjustment in siblings of persons with SMI. The Premorbid Adjustment Scale (PAS) or the Premorbid Social Adjustment Scale (PSA) (Cannon-Spoor et al., 1982) was used in these two studies (Shapiro et al., 2009; Walshe et al., 2007). These are measures assessing five areas of adjustment (academic achievement, school adaptation, socialization, peer relations and hobbies) during childhood and adolescence (Walshe et al., 2007).

Overall, the results from these two studies showed that psychosocial adjustment was better for the siblings themselves compared to the person with a SMI during childhood, adolescence, and early adulthood (Shapiro et al., 2009). However, the siblings reported poorer academic functioning during adolescence (deterioration in academic performance between childhood and adolescence) compared with controls (Walshe et al., 2007).

Sibling Relationship. Three studies assessed the sibling relationship, where one of the outcomes was negative feelings towards their sibling. Barak and Solomon (2005) asked their subjects questions about various aspects of the sibling relationship. The researchers also assessed negative feelings using a list of 20 emotional responses towards the sibling as well as rating of intensity of feelings on a five-point Likert scale. Plessis et al. (2020) used the Adult Sibling Relationship Questionnaire-short form (ASRQ-S) to assess the sibling relationship. The sibling relationship was operationalized as knowledge, intimacy, and emotional support.

Overall, the three studies found that the quality of the sibling relationship was poorer for the sibling group than for the comparison group (Barak & Solomon, 2005; Plessis et al., 2020). Barak and Solomon (2005) also found that for the sibling group the quality of the relationship declined after the debut of the illness, whereas during the same time period the sibling relationship improved for the comparison group. In one study 43% of the sibling group reported a close relationship with their sibling with a SMI compared to 95% in the control group (Barak & Solomon, 2005). Also, the siblings reported more negative feelings toward their sibling than the comparison group (2005).

Psychiatric Disorders or Psychological Problems. Five studies reported on psychiatric disorders or psychological problems in siblings, using different outcome measures. Arajärvi et al. (2006) applied a diagnostic interview for all participants (patients, siblings, and controls), the Structured Clinical Interview for DSM-IV axis disorders (SCID-I) (First, 1997). The diagnostic interview was used to assess outcomes of mood episodes, psychotic symptoms, psychotic disorders, mood disorders, substance use disorders, anxiety and other disorder. To assess negative symptoms, SCID-II and the Scale for the Assessment of Negative Symptoms (SANS) (Andreasen, 1983) was used. Andreasen (1983) operationalized the negative symptoms in five subscales (affective flattening or blunting, alogia, apathy, inattention and asociality).

Gold (1993) measured depression in siblings by the Children's Depression Inventory (CDI) (Kovacs, 1981). This is a self-report instrument for clinical and nonclinical samples of children (seven to 17 years of age).

Pierazzuoli et al. (2020) assessed psychopathology by the Minnesota Multiphasic Personality Inventory 2 (MMPI-2) (Hathaway & McKinley, 1989) including a range of different scales, e.g., depression, paranoia, schizophrenia, social introversion. Previous psychological problems and family history of psychiatric disorder were defined as clinical and environmental characteristics in Pierazzuoli et al.'s (2020) study. The authors did not describe how these characteristics were measured. Plessis et al. (2020) created an emotional distress indicator, using the Hospital Anxiety and Depression Scale (HAD) (Lepine et al., 1985).

Lepline et al. (1985) operationalized emotional distress as depressive and anxious symptomatology. In Vedel Kessing et al.'s (2020) study psychiatric disorders were assessed through population-based registers.

Overall, the included studies demonstrated higher rates of psychiatric disorders among siblings of persons with SMI compared to normal controls (Arajärvi et al., 2006; Gold, 1993; Vedel Kessing et al., 2020). In all five studies siblings reported history of previous psychological problems compared to none in the control group did (Pierazzuoli et al., 2020), and experienced more emotional distress (Plessis et al., 2020). They also had a higher rate of family history of psychiatric disorder (Pierazzuoli et al., 2020).

Other mental health outcomes assessed in the included studies were siblings' psychological well-being, and objective, and subjective emotional and practical burden. Sin et al. (2016) used the Warwick-Edinburgh Mental Well-being Scale (WEMWBS) (Tennant et al., 2007) to measure positive mental well-being. Barak and Solomon (2005) examined objective and subjective burden by the Burden Assessment Scale (BAS) (Reinhard et al., 1994). Objective burden is operationalized as the everyday practical demands stemming from the sibling's mental illness, and the subjective burden is operationalized as the negative feelings, such as anger, rejection, and depression, that stem from being close to the family member with a mental illness (Barak & Solomon, 2005).

Overall, the included studies showed that siblings of persons with SMI reported significantly poorer psychological well-being (Sin et al., 2016), and more objective and subjective emotional and practical burden.

Comparable Level of Well-Being as Controls

Three studies found similar level of outcomes for siblings compared to healthy controls, and one study reported on findings for siblings and healthy controls compared to patients. The outcomes found to be comparable for siblings and for healthy controls were quality of life (QoL), perception of self, social skills, self-concept, and subjective well-being. Subjective well-being was measured by the 20-item version of the Subjective Well-being Under Neuroleptic Treatment Scale (SWN-K) (Naber et al., 1994). Subjective well-being was found to be higher for siblings and healthy controls compared to the sibling with a SMI (Vothknecht et al., 2013). Other outcomes were measured using different self-report instruments across studies.

Tatay-Manteiga et al. (2019) used the self-reported World Health Organization Quality of Life Scale (WHO-QoL BREF) (Lucas-Carrasco, 2012) to assess global QoL. Tatay-Manteiga et al. (2019) operationalized global QoL as physical, psychological aspects, social and environmental aspects of quality of life. In a study assessing perception of self, participants were administered the Semantic Differentials derived by Osgood et al. (1957) (Trubia et al., 2016). The perception of self was operationalized as evaluation of self-image across different descriptions, e.g., strong, weak; calm, agitated; tender, hard; efficient, inefficient (Trubia et al., 2016).

Verte et al. (2003) reported psychological adjustment in siblings comprising behavior problems, social competence, and self-concept. To assess behaviour problems the Child Behaviour Check-List of Achenbach (CBCL) (Verhulst et al., 1988) was used. It was not described whether the self-report, parent report, or teacher report measure was applied. The Behaviour Problems scale in CBCL was used to assess behavior problems in the sibling of persons with SMI. To assess social competence the Matson Evaluation of Social Skills with Youngsters (MESSY) (Matson et al., 1983) was used. Here, social competence was operationalized as social skilled behavior and inadequate assertivity. Social competence between the siblings of children with high functioning autism and siblings of children with no disorder were evaluated by CBCL and total score on MESSY. SDQ-II was used to assess the self-concept of siblings of persons with SMI.

Overall, the included studies showed that siblings of individuals with SMI reported comparable levels of QoL as controls (Tatay-Manteiga et al., 2019). There were no differences between the two groups in perception of self (energy, emotional stability, and positive affect) (Trubia et al., 2016). Also, siblings of persons with SMI were no more susceptible to adaptation problems (e.g., behavior problems, social competence, or self-concept), than siblings of persons without a SMI (Verte et al., 2003).

Factors Associated With Sibling Mental Health and Well-Being

The second research question was related to factors associated with sibling outcomes. The results from the included studies were grouped in three categories: Demographic factors family relationships and health care, and characteristics of the sibling with SMI. The characteristics regarding factors associated with sibling outcomes are summarized in Table 4.

Demographic Factors of Siblings of Persons With SMI

Four of the included studies reported on demographic factors associated with sibling mental health and well-being. The demographic factors assessed were sibling age and sex, number of siblings, education, and marital status.

Overall, the included studies showed associations between the demographic factors of the siblings of persons with SMI and sibling outcomes. Two studies reported on sibling age. Both studies found associations between sibling age and ratio of psychiatric disorders (Gold, 1993; Vedel Kessing et al., 2020). Vedel Kessing et al. (2020) demonstrated a bimodal age distribution of hazard ratios for BD, unipolar disorder and use of alcohol or psychoactive drugs, with the highest ratios in individuals below age 20 and those above 60 years of age (Vedel Kessing et al., 2020).

Table 4

Factors associated with sibling mental health and well-being

Author (year)	Ill sibling sample	SMI	How was illness established?	Siblings of persons with SMI	Factors associated with sibling well-being
Arajärvi et al. (2006)	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.	n = 183. Psychotic sibling: (53.3% males). Aged 32-61 yrs, M age 44.4 (SD 6.3). Non-psychotic sibling: (47.7% males). Aged 24-72 yrs, M age 46.4 (SD 8.8).	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.
Barak et al. (2005)	n = 52. Not reported.	Schizophrenia	Not reported.	n = 52 (32% males). Aged 18-50 yrs.	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.
Gold (1993)	n = not reported (100% males). Age not reported.	ASD	DCM criteria of ASD.	n = 22 (50% males). Aged 12-17 yrs, M age 13.52.	Adolescent siblings scoring higher on depression than those under 12 years of age. Sisters do more domestic work than brothers.

Pierazzuoli et al. (2020)	n = 30 (76% males). Age not reported.	Schizophrenia spectrum disorder	Not reported.	n = 30 (16.7% males). Aged 36-63 yrs, M age 51.4.	Maternal care lower, but maternal favouritism and devaluation higher. Also higher scores in emotional trauma, emotional neglect, emotional abuse and physical abuse. Females is oriented toward sexual abuse, sexual trauma and sexual harassment, while males is oriented toward physical abuse, emotional abuse and physical threat. Males in caregiver group is oriented toward the higher values of symptoms.
Plessis et al. (2020)	n = 201 (80.21% males). Age not reported.	Schizophrenia	Diagnosed with schizophrenia by a healthcare professional.	n =201 (22.3% males). M age 37.9 (SD 12.08).	Antagonism, quarreling, overall rivalry, maternal rivalry and paternal rivalry associated with emotional distress.
Shapiro et al. (2009)	n = 286 (75.5% males). Aged 16-64 yrs, M age 36.2 (SD 9.44).	Schizophrenia or schizoaffective disorder	DSM-IV diagnosis made by either a clinical psychologist or psychiatrist using a revised version of SCID.	n = 315 (41% males). Aged 16-64 yrs, M age 36.94 (SD 9.96).	Proband PAS scores predicted PAS scores of their own siblings in the Childhood and Late Adolescence subscales.

Sin et al. (2016)	n = 89 (62.2% males). Aged 15-57 yrs, M age 26.46 (SD 7.9).	FEP	No information.	n = 90 (15% males). Aged 16-58 yrs, M age 27.52 (SD 8.41). Biologically related, step-or half-siblings, or related via adoption.	Siblings' education level associated with better mental health knowledge.
Tatay-Manteiga et al. (2019)	n = 48 (52.1% males). M age early stage BD: 43.4 (SD 10.3), late stage BD: 45,1 (SD 9.8).	BD (not currently hospitalized)	Diagnosed with DSM-IV-TRBD type I.	n = 23 (30.4% males). M age 41.5 (SD 11.8).	No significant correlations between age and QoL.
Trubia et al. (2016)	n = 31. Sex and age not reported.	ASD with ID	ICD-10 and according to the ADOS.	n = 31 (45.2% males). Aged 12-42 yrs, M age 21.	No predictor analyses.
Vedel Kessing et al. (2020)	n = 19 955. Sex and age not reported.	BD (identified having a single manic episode or BD at a psychiatric contact (as inpatients or outpatients) in the period from 1995 to 2017. BD main diagnosis at any contact.	ICD-8, ICD-9, ICD-10.	n = 13 923. M age 38.7.	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.

Verte et al. (2003)	n not reported (96.55% males). Aged 9-16 yrs, M age 11.83.	HFA (autism, Asperger's Syndrome or pervasive developmental disorder not otherwises specified).	According to DSM-IV.	29 siblings aged 6-17 and 29 control group siblings (no information on n or age in subgroup 12-16 yrs).	Negative self-concept associated with lower social competence skill for both siblings and controls (NB results reported for the whole age group - no separate results for 12-16 yrs).
Vothknecht et al. (2013)	n = 600 (80% males). M age 27.3 (SD 7.1).	Schizophrenia	No information.	n = 594 (45.5% males). M age 27.9 (SD 8.0).	All analyses with the predictors were non-significant.
Walshe et al. (2007)	n = 50 with personal and family history of schizophrenia (74% males). M age 32 yrs (SD 6.1) + 69 people with personal schizophrenia and "non-familial" schizophrenia (75.4% 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.	A) 39 unaffected siblings of persons with personal and familial schizophrenic + B) 67 unaffected siblings of persons with personal schizophrenic and non-familial schizophrenic. A) (35.9% males). Aged 16-69 yrs, M age 34 (SD 7.8). B) (49.3% males). Aged 16-69 yrs, M age 35 (SD 7.6).	No predictor analyses.

ASD = Autism Spectrum Disorder, FEP = First Episode Psychosis, ID = Intellectual Disability, BD = Bipolar Disorder, HFA = High Functioning Autism, ICD-8/9/10 = International Statistical Classification of Diseases 8/9/10, DSM-IV = Diagnostic and Statistical Manual of Mental Disorders IV, ADOS = Autism Diagnostic Observation Schedule, PAS = Premorbid Adjustment Score, SCID = Structured Clinical Interview for DSM-IV Axis I Disorders, SADS-LA = Schedule for Affective Disorders and Schizophrenia - Lifetime Version and clinical information.

One study examined sex of the siblings of persons with SMI and association with outcomes, finding that sisters reported higher emotional burden than brothers (Barak & Solomon, 2005).

Barak and Solomon (2005) also demonstrated that number of siblings was associated with higher self-reported burden, and that unmarried siblings reported higher objective burden and greater fear of intimacy compared to married siblings.

Two studies reported on siblings' education level. Overall, the findings showed that higher education was associated with less burden (Barak & Solomon, 2005), and with better mental health knowledge in siblings (Sin et al., 2016).

Family Relationships and Health Care

Three studies examined family relationship and health care and the association with sibling outcomes. The variables assessed were contact with health care professionals, maternal care, and sibling relationships. Different measures were used to assess these variables. For instance, Plessis et al. (2020) used the Adult Sibling Relationship Questionnaire-short form (ASRQ-S) to assess the sibling relationship, and Pierazzuoli et al. (2020) used the Parental Bonding Instrument (PBI) to assess the relationship between the sibling and parental figures (Parker et al., 1979). Two dimensions from this instrument, and two additional dimensions suggested by Gilbert, Allan and Goss (1996) were included: the care dimension (evaluating the parental style on a continuum from affection and emotional warmth to coldness and neglect), the overprotection dimension (evaluating the parental style in a continuum from independence to control and intrusion), the devaluation dimension (assesses the tendency of parents to debase the child), and the favouritism dimension (the tendency to favour brothers or sisters to the detriment of the subject). Findings from the included studies showed that siblings who had contacted health care professionals for mental health problems had the highest odds ratios for psychotic disorder (Arajärvi et al., 2006).

They also showed that maternal care was lower and maternal devaluation was higher in siblings of persons with SMI than for the comparison group (Pierazzuoli et al., 2020). The following qualities of the sibling relationship were associated with emotional distress in siblings: antagonism, quarrelling, overall rivalry, maternal rivalry and paternal rivalry (Plessis et al., 2020).

Characteristics of The Sibling With a SMI

To assess characteristics of the sibling with SMI, one study used Premorbid Adjustment Score (PAS) to measure academic functioning. The findings from this study were that academic functioning in siblings with a SMI predicted PAS scores of their siblings without SMI on sociability and social withdrawal, peer relationships, scholastic performance, adaptation to school, and ability to form socio-sexual relationships (Shapiro et al., 2009).

One study addressed social competence in siblings of individuals with SMI and factors that could be associated with this. Verte et al. (2003) measured social competence using SDQ-II. They found that siblings with a more negative self-concept also had lower social competence skills. This was the case both for the siblings of ASD and the comparison group.

Scientific Quality of the Included Studies

An overview of the scientific quality of the included studies is provided in Table 5. There were several weaknesses in the scientific quality of the included studies, especially regarding the sampling strategies. Most of the studies used a non-probability sampling and did not provide a clear justification of the sample frame used. When using a non-probability sample the participants may represent a subgroup of siblings and the results get difficult to generalize. Also, often the reasons why certain eligible individuals chose not to participate were not mentioned in the studies, and any attempts to achieve a sample of participants that represented the target population were not done. In many of the studies there were no clear description of the inclusion and exclusion criteria either. From all the included studies there were only two that were shown to be of good scientific quality, with coding yes on all or all but one scientific variable (Arajärvi et al., 2006; Vedel Kessing et al., 2020). These were both register studies with large samples.

Table 5

Coding of scientific quality

Author (year)	Clear research question	Data adress research question	Relevant sampling stategy	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 et al. (2005)	Yes/Yes	Yes/Yes	No/Yes	No/No	No/Yes	Can't tell/Can't tell	Yes/Yes
Gold (1993)	Yes	Yes	No	No	No	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/Yes	No/Yes	No/No	No/Yes	No/No	Yes/Yes
Tatay-Manteiga et al. (2019)	Yes/Yes	Yes/Yes	No/Yes	No/No	No/Yes	Can't tell/Can't tell	Yes/Yes
Trubia et al. (2016)	Yes/Yes	Yes/Yes	No/Can't tell	No/No	Yes/Can't tell	Can't tell/Can't tell	Yes/Yes
Vedel Kessing et al. (2020)	Yes/Yes	Yes/Yes	Yes/Yes	Yes/Yes	Yes/Yes	Yes/Yes	Yes/Yes
Verte et al. (2003)	Yes/Yes	Yes/Yes	No/Yes	No/No	Yes/Yes	Can't tell/Can't tell	No/Yes
Vothknecht et al. (2013)	Yes	Yes	No	No	Yes	Yes	Yes
Walshe et al. (2007)	Yes/Yes	Yes/Yes	No/No	No/No	Yes/No	Can't tell/Can't tell	Yes/Yes

Discussion

The present study reviewed studies on the mental health and well-being of siblings of individuals with SMI, as well as factors associated with sibling outcomes. A systematic literature review was carried out comprising adolescent and adult siblings of persons with SMI. The current literature review revealed mixed results on outcomes for mental health and well-being of siblings of individuals with SMI. In addition, some factors such as sibling age, sex, level of maternal care and quality of sibling relationship seemed to be associated with siblings' mental health and well-being. A quality rating of the included studies showed that only two studies had high methodological quality, i.e., Vedel Kessing et al (2020) and Arajärvi et al. (2006). Both these studies reported on psychopathology for the sibling of persons with SMI.

Findings from the present review were that overall, studies of persons with SMI had some positive outcomes (e.g., a more positive self-concept compared to controls), some comparable outcomes as the controls (e.g., QoL), and some negative outcomes. The negative outcomes included lower psychosocial adjustment, lower quality of the sibling relationship, higher rates of psychiatric disorders, having history of previous psychological problems, increased level of emotional distress, and higher rate of family history of psychiatric disorders. The siblings also reported more emotional and practical burden.

Mental Health and Well-Being in Siblings of Persons With SMI

Regarding positive outcomes, the siblings appeared to use more problem-focused coping, be more involved in mental health arenas, and have more positive self-concept compared to controls. The siblings were also shown to have better mental health knowledge than the general population. In the research on coping strategies, problem-focused coping seems to determine level of well-being (Avcioglu et al., 2019). Problem-focused coping strategies aim to solve problems and reduce the effects of stressful events (Orsmond et al., 2009). In line with this, Avcioglu et al. (2019) have suggested that siblings of persons with SMI may use problem-focused coping as a strategy to escape the stressors. Problem-focused coping can be used in settings where a person faces difficult circumstances. This is a coping strategy associated with appraising an illness/problem as a challenge. In contrast, appraising the illness/problem as a threat seems to be more associated with emotion-focused coping, a strategy perhaps more associated with depression and reduced self-esteem (Dysvik et al., 2005). For siblings of persons with SMI this may explain why use of problem-focused coping was associated with mental health and well-being. Perhaps these siblings appraise the SMI illness as a manageable challenge, something they have the resources to cope with, as opposed to a threat they could not manage. Other studies have demonstrated associations between problem-focus in general and high self-esteem (Terry, 1994), and that self-esteem is an important factor for coping (Lazarus & Folkman, 1984). The present review points to several positive outcomes in siblings of persons with SMI.

Siblings of persons with SMI seem to participate more in mental health arenas (e.g., choosing a career in the helping professions). This finding is in line with previous research suggesting that siblings develop alternative relationships and activities than gives them a source of satisfaction and feeling of autonomy (Hoover & Franz, 1972). The concept of constructive escape has been used in the sibling research field as a way of letting the child be temporarily relieved from stress in the environment, either mentally or physically, through activities or relationships outside the family. The activities must be positive in nature, bring the child pleasure, and not carry with them negative repercussions. Literature, art, play, music, school activities and organizational functions may be examples of constructive escape activities (Kinsella et al., 1996). This may be one possible explanation for the findings of participation in mental health arenas in the present study.

One study found that sisters of persons with SMI had a more positive self-concept compared to controls (Verte et al., 2003). This is in line with previous findings where siblings of children with ASD had a significantly better self-concept than a normative sample (Mates, 1990). Dyson (1996) have suggested that better self-concept in siblings may be the result of increased family emphasis upon personal growth in families with disabled children. One can speculate that this may be the case also for families with family members with other disabilities or SMI. The results regarding self-concept for siblings compared to controls in the current review is also supported by Kinsella et al's (1996) findings that siblings of persons with SMI may develop specific strengths from living with a sibling's illness. Thus, having a sister or brother with a SMI may lead to a resourcefulness in the siblings of a person with SMI.

The siblings of persons with SMI were found to have better mental health knowledge than the general population. This result is in line with the more general finding that having a relationship with a person with a mental illness is associated with better mental health knowledge (Henderson & Thornicroft, 2013). In general, knowledge and higher education has been found to be a protective factor for the youths (Centers for Disease Control and Prevention, 2009). It is plausible that despite the negative outcomes, the siblings of persons with SMI develop better mental health knowledge, and this may for some protect against negative outcomes for some siblings' SMI.

Regarding the negative outcomes, studies in the present review showed 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. The siblings also reported poorer academic functioning during adolescence and more emotional and practical burden. Furthermore, they had higher rates of family history of psychiatric disorder. The two large register studies included, with the highest score in the scientific rating of quality, where some of the studies reporting on these psychopathologies regarding the siblings of persons with SMI.

Sibling relationship quality was rated lower for the siblings of persons with SMI. This is in accordance with previous studies reporting experiences of stress and negative outcomes (e.g., despair, fear, guilt, and helplessness) in siblings when their sister or brother was diagnosed with SMI (Sin et al., 2012). Others have found associations between history of violent behavior by a brother or sister with SMI and poorer sibling relations (Bowman et al., 2015). Good sibling relationships have proven to be beneficial for emotional, social, and cognitive development, and an important protective factor for mental health (Brody, 1998; Cicirelli, 1995; Gass et al., 2007; Howe et al., 2005). Thus, the presence of SMI in a sibling may be associated with less resources for siblings to handle difficulties arising in their lives.

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

If adjusted for the sibling relationship, the family ecology model of Pederson and Revenson's (2005) may help explain some of the negative outcomes in siblings of persons with SMI. For instance, the role redistribution mediational hypothesis suggested to occur when parents have a physical illness may also occur for siblings with a sister or brother with SMI. After the onset of SMI, these siblings may be required to take on extra chores, tasks, or responsibility. This may reduce the time and energy available for 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). According to the family ecology model, illness in family members may also impact the well-being of youth by creating daily hassles, such as increased chores (Pedersen & Revenson, 2005).

Even though the family framework can partly explain some of the negative outcomes for siblings of persons with SMI, this model has limitations. First, it does not mention contextual factors beyond the family, e.g., family support systems, socioeconomic factors, quality of the health and social care system. Second, it does not include individual characteristics of the sibling of persons with SMI that may affect their well-being. However, and importantly, the framework does not mention the contribution of genetic vulnerability to the mental health and well-being of siblings. Bauminger and Yirmiya's (2001) diathesis-stress model may contribute to a more comprehensive understanding of the siblings. Orsmond et al. (2009) adopted this model to explain the genetic and environmental mechanisms influencing siblings of individuals with ASD. They hypothesized that the genetic vulnerability of the siblings interacts with environmental stress to influence sibling outcomes. The genetic vulnerabilities take form as broader autism phenotype (BAP) characteristics in the sibling and family history of ASD. Stress was conceptualized as three variables: behavior problems in the brother or sister with an ASD, life events, and maternal depressive symptoms. The model may apply to genetic and environmental influences for siblings of individuals with other SMI as well, especially since many of the severe mental illnesses are disorders with genetic components (Phelan, 2002). Orsmond et al. (2009) found partial support for their diathesis-stress model for siblings of individuals with ASD, primarily through findings that sibling BAP characteristics (i.e., greater risk for ASD as well as a range of related impairments) were associated with depressive and anxiety symptoms in siblings. However, these associations were found only in the presence of high level of stressful life events. The diathesis-stress model should be evaluated, also for sibling outcomes regarding SMI's beyond ASD.

Some studies reported that siblings had comparable level of QoL and perception of self as controls. Furthermore, siblings also were not more susceptible to adaptation problems (i.e., measured by behavior problems, social competence, and self-concept) than siblings of persons without SMI. The fact that both negative and positive outcomes for siblings of persons with SMI have been found, may be related to Powell and Gallagher's (1993) theory of outcomes on a continuum. The functioning of siblings of persons with SMI's functioning may vary from negative outcomes on the one end to positive outcomes on the other end. Also, there are large heterogeneity in symptom presentation of a SMI (Lord et al., 2000). Trubia et al. (2016) emphasize that being a sibling of a child with ASD showing aggressive behavior may influence the functioning of siblings differently compared to if the brother or sister mostly demonstrate repetitive behaviors and scarce eye contact.

The mixed results as well as the findings about negative outcomes demonstrate the need for continued research on this field. It has been suggested that mixed results found in previous studies may be a result of studies applying different measures, methods, informants, and control groups (Meadan et al., 2010). The current review addressed one of these issues by including only studies with siblings of persons with no SMI as comparison group. However, even with this exclusion criterion the results were mixed. To better understand the findings regarding the siblings of persons with SMI future research should, in addition to using the same type of control groups, also evaluate the same outcomes with same measurements and samples across the studies.

Factors Associated With Sibling Mental Health and Well-Being

Both demographic factors, level of maternal care and quality of sibling relationship were associated with sibling outcomes. For instance, sisters reported higher subjective burden (i.e., emotional distress) than brothers. This is in line with previous research showing that females are more likely to provide care for siblings or other relatives with SMI, and that these care responsibilities are associated with more mental health problems, insomnia, somatic symptoms, and lower life satisfaction (Haugland et al., 2020; Shivers & Textoris, 2021). These differences may be explained by gender theories that claim that there are differences in levels of emotional awareness and expression between males and females, with women generally tending to have higher emotional awareness than men (Mintz & O'Neil, 1990). Barak and Solomon (2005) proposed that a tendency for females to disclose emotions may put women in higher risk for emotional distress due to becoming a source of comfort and support for their parents as well as their sibling with SMI.

Adolescent siblings scored higher on depression than siblings below 12 years. This finding is supported by previous research showing that many young adults take care of family members with different illnesses (Haugland et al., 2020). The term 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 illness, mental health issues, or substance misuse problems (Becker & Becker, 2008). This age period, often termed emerging adulthood, may be a particularly difficult life phase, as the young carer may struggle to balance their time between caring and being independent. Haugland et al. (2020) found several risk factors associated with being a carer, for instance financial stress, family structure, and coming from migrant families. Furthermore, emotional reactions due to the caring responsibilities may leave young carers with limited time for social life, relaxation, and leisure activities. Even though siblings in the present review not necessarily had taken on large caring responsibilities for their sibling with SMI, the findings from the carer literature may apply to some of them and partly explain the risk of more mental health difficulties and poorer psychological well-being.

Higher education was associated with less burden among siblings of persons with SMI. This may be explained by increased availability of information among siblings with higher education. According to Barak and Solomon (2005) knowing about the illness helps the siblings to accept the symptoms and to distinguish between the sibling and the illness (Barak & Solomon, 2005).

Even though some factors associated with sibling outcomes were identified, very limited research was found regarding moderating factors. The studies mainly addressed demographic factors whereas important factors such as contextual factors were beyond individual characteristics and family relations, as well as genetic factors were absent. Future research needs to fil this gap by examining a range of variables on different levels, for example cultural differences in how health care and social systems offer care for persons with SMI and how risk and protective factors at societal level may influence the mental health and well-being of siblings of persons with SMI.

Strengths and Limitations

The strengths and limitations of the current study are related to the search process, criteria for the inclusion of literature, the coding of descriptive data and scientific quality, and limitation regarding inference of causality.

Including three databases may be seen as a strength of the current study. Even though there were many duplicates, choosing to look for literature in PsycINFO, CINAHL and Web of Science gave an opportunity to get more relevant hits than choosing one or two databases. Also, the choice of search terms and combinations of these included a mix of different mental conditions and terms for siblings' mental health and well-being which made it possible to get more relevant hits. Even though several terms for SMI were used few studies regarding other diagnosis than schizophrenia, ASD, BD and psychosis came up. Thus, although the impact of other illnesses on family members can be profound and is important to understand, little is known about siblings of individuals with other prevalent diagnosis, e.g., major depression, eating disorders. Even though we included eating disorders in our inclusion criteria, none studies with this illness were included in this review.

Previous research has emphasized the need to focus on defined comparison groups, larger studies and methods that are comparable across different studies (e.g., using the same measures). Meadan et al. (2010) suggested that one reason for these mixed results in sibling outcomes may be the methodological weaknesses in the studies. A strength of the current review is that only studies that compared siblings of individuals with SMI to either siblings of individuals without SMI or a normative sample were included. This made the results easier to interpret indicating that the findings can not be a result of using different control groups in different studies. Another explanation for the mix of positive and negative outcomes among siblings may be the use of different measures in the included studies. This shortcoming in the research studies should be addressed in future studies on siblings of persons with SMI.

After the screening process and selection of studies, the included studies were coded for descriptive date and methodological quality. Even though a systematic evaluation of methodological quality was done by the MMAT (Hong et al., 2018) coding, the studies on relevant variables (e.g., research question, sampling strategy and measurements), there were some limitations in this process. First, not all the included studies were double coded. Second, the coders rated differently on many of the studies, some more conservative than others. The findings regarding methodological quality of the studies showed that there were two studies with high scores, whereas all others had several important weaknesses, especially regarding the sampling strategy. Also, not all variables were reported on in the studies. Even though the process of coding the scientific quality of the studies had several flaws, overall, the assessment of the quality gave a picture of the studies strengths and limitations, also pointing to the need for improvements in future studies on siblings of persons with SMI (e.g., larger samples and the use of same measures across studies).

Most of the included studies were cross-sectional. Therefore, it is not possible to conclude about causality between sibling outcomes and SMI in a brother or sister. The studies show that the mix of positive, negative, and comparable outcomes, and demographic and environmental factors are associated factors, but they do not give answers to which direction this relationship goes or if there are other factors that can explain the relationship. Future research, including longitudinelle studies, may help explain the direction of these associations.

Implications

Because of the methodological limitations in previous research on the sibling field, future research needs to focus on few comparison groups and measures, and larger more, representative samples. This will improve possibilities to generalize from the findings.

The current results show that there are both positive and negative mental health and well-being outcomes for siblings of persons with SMI. These findings may be of relevance for intervention programs aiming to support and help families when a member has a SMI, for instance by enhancing self-esteem and increase the use of problem-focused coping in siblings. Siblings of individuals with SMI need to be acknowledged and to receive appropriate support. Some siblings may experience an objective and/or emotional burden. Also, because the findings show that the risk for negative outcomes may be higher in early adolescence and late adulthood interventions should be targeted here. There also seems to be an unbalance in the sibling sex represented in the studies with more males in the mentally ill samples. Future research should focus on this and work to enhance more balance in the samples regarding the sibling sex.

The ecology family model proposed by Pedersen and Revenson (2005) highlights how different family stressors impact the family members when someone develops an illness. Future research needs to develop models to explain the outcomes of siblings of persons with SMI and factors associated with outcomes. This may increase our understanding of how the sibling's illness may influence other family members' and mediating mechanisms for these associations.

Conclusion

Siblings of persons with SMI have both positive, negative, and comparable outcomes as controls. Increased risk for mental health problems in siblings of persons with SMI needs to be acknowledged by authorities, as well as professionals working within health, and educational systems. There is a need to develop interventions aiming to support and prevent problems in the siblings, and a family perspective needs to be considered when treating persons with SMI. Also, limitations in the research studies need to be addressed and improvement in methodological quality is needed in future research.

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