Psychoeducation for siblings of people with severe mental illness

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TABLE OF CONTENTS

[Intervention Review]

Psychoeducation for siblings of people with severe mental illness

Jacqueline Sin[1], Cheryl D Jordan[2], Elizabeth A Barley[2], Claire Henderson[3], Ian Norman[2]

[1]South West London & St. George's Mental Health NHS Trust, London, UK. [2]Florence Nightingale Faculty of Nursing and Midwifery, King's College London, London, UK. [3]Health Service and Population Research Department, Institute of Psychiatry, Psychology and Neuroscience, King's College London, London, UK

Contact address: Jacqueline Sin, South West London & St. George's Mental Health NHS Trust, Springfield Hospital, 61 Glenburnie Road, London, SW17 7DJ, UK. Jacqueline.sin@swlstg-tr.nhs.uk. jacqueline@urbanfuture.org.

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ABSTRACT

Background

Many people with severe mental illness (SMI) have siblings. Siblings are often both natural agents to promote service users' recovery and vulnerable to mental ill health due to the negative impact of psychosis within the family. Despite a wealth of research evidence supporting the effectiveness of psychoeducation for service users with SMI and their family members, in reducing relapse and promoting compliance with treatment, siblings remain relatively invisible in clinical service settings as well as in research studies. If psychoeducational interventions target siblings and improve siblings' knowledge, coping with caring and overall wellbeing, they could potentially provide a cost-effective option for supporting siblings with resulting benefits for service users' outcomes.

Objectives

To assess the effectiveness of psychoeducation compared with usual care or any other intervention in promoting wellbeing and reducing distress of siblings of people affected by SMI.

The secondary objective was, if possible, to determine which type of psychoeducation is most effective.

Search methods

We searched the Cochrane Schizophrenia Group Trials Register and screened the reference lists of relevant reports and reviews (12th November 2013). We contacted trial authors for unpublished and specific data on siblings' outcomes.

Selection criteria

All relevant randomised controlled trials focusing on psychoeducational interventions targeting siblings of all ages (on their own or amongst other family members including service users) of individuals with SMI, using any means and formats of delivery, i.e. individual (family), groups, computer-based.

Data collection and analysis

Two review authors independently screened the abstracts and extracted data and two other authors independently checked the screening and extraction process. We contacted authors of trials to ascertain siblings' participation in the trials and seek sibling-specific data in those studies where siblings' data were grouped together with other participants' (most commonly other family members'/carers') outcomes. We calculated the risk difference (RD), its 95% confidence interval (CI) on an intention-to-treat basis. We presented continuous data using the mean difference statistic (MD) and 95% CIs. We assessed risk of bias for the included study and rated quality of evidence using Grading of Recommendations Assessment, Development and Evaluation (GRADE).

Main results

We found 14 studies that included siblings amongst other family members in receipt of psychoeducational interventions. However, we were only able to include one small trial with relevant and available data (n = 9 siblings out of n = 84 family member/carer-participants) comparing psychoeducational intervention with standard care in a community care setting, over a duration of 21 months. There was insufficient evidence to determine the effects of psychoeducational interventions compared with standard care on 'siblings' quality of life' (n = 9, MD score 3.80 95% CI -0.26 to 7.86, *low quality of evidence*), coping with (family) burden (n = 9, MD -8.80 95% CI -15.22 to -2.34, *low quality of evidence*). No sibling left the study early by one year (n = 9, RD 0.00 CI -0.34 to 0.34, *low quality of evidence*). Low quality and insufficient evidence meant we were unable to determine the effects of psychoeducational interventions compared with standard care on service users' global mental state (n = 9, MD -0.60 CI -3.54 to 2.38, *low quality of evidence*), their frequency of re-hospitalisation (n = 9, MD -0.70 CI -2.46 to 1.06, *low quality of evidence*) or duration of inpatient stay (n = 9, MD -2.60 CI -6.34 to 1.14, *low quality of evidence*), whether their siblings received psychoeducation or not. No study data were available to address the other primary outcomes: 'siblings' psychosocial wellbeing', 'siblings' distress' and adverse effects.

Authors' conclusions

Most studies evaluating psychoeducational interventions recruited siblings along with other family members. However, the proportion of siblings in these studies was low and outcomes for siblings were not reported independently from those of other types of family members. Indeed, only data from one study with nine siblings were available for the review. The limited study data we obtained provides no clear good quality evidence to indicate psychoeducation is beneficial for siblings' wellbeing or for clinical outcomes of people affected by SMI. More randomised studies are justified and needed to understand the role of psychoeducation in addressing siblings' needs for information and support.

PLAIN LANGUAGE SUMMARY

Psychoeducation for brothers and/or sisters of people with severe mental illness (SMI)

Review question.

This review compares psychoeducation for siblings of people with severe mental illness versus standard care or any other intervention as a means of improving their own wellbeing and quality of life as well as coping with the care-giving for their mentally ill siblings.

Background.

Psychoeducation programmes aim to improve knowledge and understanding of mental health. Family members, inlcuding siblings, of people with severe mental illness are often offered psychoeducation. It is supposed that increased knowledge will help the brother or sister to cope more effectively with providing care for their mentally ill sibling and enhance their own wellbeing. Psychoeducational interventions involve an interaction between the information provider and the sibling of the mentally ill person. This can be delivered in different ways, such as face-to-face or via online forums or by a mixture of these methods.

Study characteristics.

A search for randomised trials investigating psychoeducation for the siblings of people with severe mental illness was run in 2013. Results of the search suggest that brothers and sisters form a small proportion of family members participating in studies of this kind. Only one study meeting the review criteria was found. This study included nine siblings and compared a psychoeducational intervention with standard care in a community care setting, over a period of 21 months.

Key results.

Better outcomes in terms of coping were identified for those siblings who received psychoeducation. However, the number of participants was small and the quality of evidence low, and there is no conclusive evidence that psychoeducation is of benefit for brothers/sisters in this and other important areas (such as wellbeing, quality of life) or for the outcomes of people with mental illness (such as mental state, hospital admission or length of hospital stay).

Quality of the evidence.

Further studies are needed to understand the role of psychoeducation in specifically helping brothers and/or sisters to cope with providing care for their mentally ill siblings. The scarcity of good quality studies means that it is not possible to assess which type of psychoeducation is the most effective, although interventions using a group format that brings many family members together to receive education and share their experiences seem well-received by the participants.

This plain language summary has been written by a consumer: Ben Gray, Senior Peer Researcher, McPin Foundation.http://mcpin.org/

SUMMARY OF FINDINGS FOR THE MAIN COMPARISON *[Explanation]*

Psychoeducation compared with Standard Care for siblings of people with severe mental illness

Patient or population: Siblings of people with severe mental illness
Settings: Community
Intervention: Psychoeducation
Comparison: Standard care

Outcomes	Illustrative comparative risks* (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)	Comments
	Assumed risk	Corresponding risk				
	Standard care	Psychoeducation				
Siblings' quality of life - by 12 months (reversed FAD, high = good) Reversed FAD. Scale from: 4 to 28. Follow-up: 12 months		The mean siblings' quality of life - by 12 months (reversed FAD, high = good) in the intervention groups was **3.8 higher** (0.26 lower to 7.86 higher)		9 (1 study)	⊕⊕◯◯ **low**[1,2]	One study provided data on 9 siblings but found no significant differences between psychoeducation or standard care
Siblings' coping (in terms of burden) - by 12 months (FBIS, high = poor) Follow-up: 20 months		The mean siblings' coping (in terms of burden) - by 12 months (FBIS, high = poor) in the intervention groups was **8.8 lower** (15.22 to 2.38 lower)		9 (1 study)	⊕⊕◯◯ **low**[2,3]	One study including 9 siblings looked at (coping with) burden and found significant differences favouring psychoeducation
Leaving the study early for any reason - by 12 months	**Study population**		See comment	9 (1 study)	⊕⊕⊕◯ **low**[2]	Risks were calculated from pooled risk differences. One study including 9 siblings but no

	See comment	See comment[2]			one left the study early from either psychoeducation or standard care
	Moderate				
	0 per 1000	**0 per 1000** (0 to 0)[2]			
Service users' general mental state - by 12 months (BPRS, high = poor)		The mean service users' general mental state - by 12 months (BPRS, high = poor) in the intervention groups was **0.6 lower** (3.54 lower to 2.34 higher)	9 (1 study)	⊕⊕◯◯ **low**[4]	One study including 9 siblings looked at their unwell brother's or sister's mental state but found no significant differences between groups
Service users' number of re-hospitalisation to a psychiatric inpatient unit over six months - by 12 months		The mean service users' number of re-hospitalisation to a psychiatric inpatient unit over six months - by 12 months in the intervention groups was **0.7 lower** (2.46 lower to 1.06 higher)	9 (1 study)	⊕⊕◯◯ **low**[2]	One study including 9 siblings looked at service users' rehospitalisation rate but found no significant differences between groups
Service users' inpatient bed occupancy in terms of average days of hospital stay over 6 months - by 12 months		The mean service users' inpatient bed occupancy in terms of average days of hospital stay over 6 months - by 12 months in the intervention groups was **2.6 lower**	9 (1 study)	⊕⊕◯◯ **low**[4]	One study looked at service users' duration of hospital stay but found no significant differences between groups

		(6.34 lower to 1.14 higher)			
Service users' specific aspect of quality of life, i.e. social functioning - by 12 months (SLOF, high = good) Scale from: 43 to 215.		The mean service users' specific aspect of quality of life, i.e. social functioning - by 12 months (SLOF, high = good) in the intervention groups was **21.4 higher** (2.71 lower to 45.51 higher)	9 (1 study)	⊕⊕◯◯ **low**[2,5]	One study including 9 siblings and their unwell brother or sisters looked at service users' quality of life by means of level of functioning

*The basis for the **assumed risk** (e.g. the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% confidence interval) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).
CI: Confidence interval; **RR:** Risk ratio;

GRADE Working Group grades of evidence
High quality: Further research is very unlikely to change our confidence in the estimate of effect.
Moderate quality: Further research is likely to have an important impact on our confidence in the estimate of effect and may change the estimate.
Low quality: Further research is very likely to have an important impact on our confidence in the estimate of effect and is likely to change the estimate.
Very low quality: We are very uncertain about the estimate.

[1] Quality of life was measured with family functioning, one of many dimensions of QOL.
[2] Downgraded one level due to imprecision (only 1 study with 9 siblings and their unwell brothers/sisters were included).
[3] Coping is measured by FBIS on siblings' burden.
[4] Downgraded one level dues to imprecision (only 1 study with 9 service users' outcome was reported).
[5] Service users' quality of life was measured with SLOF - specific level of functioning scale.
BPRS: Brief Psychiatric Rating Scale
FAD: Family Assessment Device
FBIS: Family Burden Interview Schedule
SLOF: Specific Level of Functioning Scale

BACKGROUND

Description of the condition

Severe mental illness (SMI) may be defined according to three dimensions: 1. A non-organic psychotic disorder; 2. Treatment duration lasting for two years or more; and 3. Disability resulting in difficulties in social and occupational functioning (Ruggeri 2000). The narrowest definition of psychosis is described as a break in reality testing as manifested by delusions or hallucinations into which an individual has no insight (APA 2000; APA 2013) and subsequently causes disturbances in functionality and relationships, despite ongoing treatment and care. Psychosis is characterised by psychotic symptoms, for example: distortion of thinking and perception, delusions, hallucinations, disordered thinking and blunting or incongruity of emotional responses. The cluster of schizophrenia and related disorders (e.g. schizoaffective disorder, schizophreniform disorder and delusional disorder) are considered the most common psychotic disorders (WHO 1992). Individuals with "early onset psychosis" or "first episode psychosis" and those who are receiving treatment and support from early intervention services are also regarded as having SMI due to similarities in presentation and the impact on treatment duration and disability (NICE 2014). Bipolar disorder is characterised by repeated episodes during which the individual's mood and activity are substantially disturbed, alternating between elevated mood and activity which is often correlated with psychotic symptoms, and decreased energy and activity (WHO 1992).

Onset of SMI tends to peak around the late teenage years and early adulthood. A 2004 review of previous surveys estimated the 12-month prevalence rate of Type 1 bipolar disorder to be 0.72% and the lifetime prevalence rate to be 0.8% (Waraich 2004). The prevalence of schizophrenic disorders based on a 2005 review of surveys in 46 countries (Saha 2005) found a median of 0.4% for lifetime prevalence up to the point of assessment and 0.3% in the 12-month period prior to assessment. Moreover, the lifetime morbid risk, that is the number of people estimated to develop schizophrenia at some point in their life, was estimated to be "about seven to eight individuals per 1000" (0.7% to 0.8%). The prevalence of schizophrenia was found to be consistently lower in poorer countries than in richer countries but did not differ between men and women or rural or urban dwellers. In addition to the direct impact of SMI on the health of service users, it is widely recognised that coping with a SMI can be challenging and difficult not just for the individuals but for everyone closely related to them. Their siblings are also vulnerable to mental ill health due to the negative impact of psychosis within the family (Sin 2012a; Smith 2009).

Description of the intervention

Psychoeducation is an intervention which aims to instil information or knowledge on the illness condition and its management (NICE 2014; Xia 2011). Psychoeducational interventions can be delivered as a group or individual programme involving interaction between the information provider and participants, using different delivery modes, including face-to-face (e.g. Smith 1987a), online virtual forum (Rotondi 2010), and a mix of different delivery modes (Szmukler 2003). The purpose of psychoeducation involving families of service users with SMI is to enhance their understanding of the illness and promote their management and caring of service users in their usual environment. Psychoeducational interventions often have multiple components which may consist of, for instance: cognitive and/or behavioural training elements, peer support and/or discussion, with the primary aim of enhancing problem-solving and/or coping with caring-related or illness management issues (Xia 2011).

How the intervention might work

Psychoeducational interventions frequently have education as a cardinal feature and prime aim. The education content often includes information on the illness condition and problem solving and coping strategies for common caring issues, such as managing illness symptoms and related problems encountered by family carers (Birchwood 1992; Szmukler 2003). Such theoretical underpinnings suggest that improved knowledge and understanding of the illness can dispel myths, alleviate anxiety and worries and thus reduce distress in family members of individuals affected by SMI (Birchwood 1992; Smith 1990). Psychoeducation may also enhance family members' optimism and capability in enlisting community resources concerning their roles and their contribution to service users' ongoing recovery and management (Birchwood 1992). Some earlier studies have demonstrated that short-term and simple educational interventions are effective in improving family carers' knowledge about the illness and its management, leading to a reduction in their stress levels, perceived burden and sense of fear, anxiety and isolation (Pakenham 1987; Smith 1987a). These changes have also been found to correlate with increased optimism in the family's role in treatment (Smith 1987a) and an improved home life environment (Cozolino 1988). The stress-appraisal and coping theory (Lazarus 1966), a theoretical framework commonly used in many psychoeducational interventions (e.g. Szmukler 2003), goes further asserting that increased knowledge is one of the best mediating factors in stress-appraisal, enhancing participants' perceived efficacy and coping, if the knowledge also impacts on management strategies. Bandura's self-efficacy theory (Bandura 1977; Bandura 1988) also finds resonance in some psychoeducational interventions, where the intervention itself does not aim to change the caring situation but shape the participants' self-efficacy (i.e. how well they believe they could cope with the caring situation) and mediate the correlated perceived burden of care and anxieties. It is therefore common that psychoeducational

interventions include general problem-solving and coping strategies as a way to enhance coping and self-efficacy. It is well established that high expressed emotion (EE) (Brown 1958; Brown 1962; Brown 1972) is a strong predictor of relapse. Longitudinal studies suggest that EE is strongly correlated with caregiver burden in addition to service users' relapse rate, in that caregivers experience a higher level of burden when they are emotionally overinvolved, critical or hostile to service users (or their behaviour). Through relieving the burden of care, psychoeducation may also reduce EE (Gonzalez-Blanch 2010). Psychoeducation also aims to reduce the frequency at which negative emotion is expressed to the affected family member, in the form of critical comments and hostility. Especially in multi-modal interventions, psychoeducation plays a significant but not exclusive role in the outcomes of interventions in changing family members' attitude, perception and behaviour towards the individual with SMI and/or the illness positively (Birchwood 1992; Leff 1989).

Despite the various benefits of psychoeducation for family carers mentioned above, some studies suggest these benefits are sometimes short-lived (e.g. Pakenham 1987; Smith 1987a). Moreover, improved knowledge, though often achieved through psychoeducation, does not necessarily have a significant impact on other important primary outcomes, such as EE, family beliefs and behaviour towards service users with SMI (Birchwood 1992; Chan 2009a). Some researchers therefore suggest augmenting psychoeducation with more intensive and complex interventions (such as family therapy and cognitive behaviour therapy) conducted over a longer period of time (Chan 2009a; Leff 1989).

Why it is important to do this review

Many people have a sibling; for instance, in the UK over 80% of the general population has at least one sibling (Smith 2009). The sibling relationship often outlives other relationships, including marriages and parenthood (Sin 2012a). The quality of the sibling relationship, especially during adolescence and early adulthood, is a predictive factor in siblings' future involvement in caring for a brother or sister who has SMI (Greenberg 1999), as well as being associated with a higher quality of life (Smith 2007) and a more promising recovery trajectory (Birchwood 2003) for individuals with a diagnosis of SMI. Siblings are both natural agents to promote service users' recovery and vulnerable to mental ill health due to the negative impact of psychosis on the family (Friedrich 2008; Sin 2008; Sin 2012a). Current research into siblings' experiences and needs suggest that they often do not regard themselves as carers and are rarely involved with statutory health or social services, unlike their parents who often act as the primary carers (Sin 2012a; Smith 2009). Nonetheless, siblings' experiences of subjective and objective burden of caring may be similar to that of the primary carers (Magliano 1999). Similarly, siblings' adaptation and grief over the onset of psychosis in their brother or sister may be similar to that experienced by other family members (Patterson 2002). A small body of research in early onset psychosis (Sin 2012a) and schizophrenia (Friedrich 2008) highlights siblings' need for information about the illness, ways to promote recovery in the service user and coping strategies, all of which are key elements in many psychoeducation programmes. Existing systematic reviews on psychoeducation (e.g. NICE 2014; Xia 2011) and family intervention (e.g. Pharoah 2010) have focused on the service user and overall family outcomes, missing the opportunity to evaluate the effectiveness of psychoeducational interventions for siblings directly. This systematic review aims to address this knowledge gap by investigating the effectiveness of psychoeducation in improving the wellbeing of siblings of individuals affected by SMI. We also aim to identify the active essential ingredients in such interventions to inform the development of future psychoeducational interventions targeting siblings directly, which may further enhance benefits for service users.

OBJECTIVES

To assess the effectiveness of psychoeducation compared with usual care or any other intervention in promoting wellbeing and reducing distress of siblings of people affected by SMI.

The secondary objective was, if possible, to determine which type of psychoeducation is most effective.

METHODS

Criteria for considering studies for this review

Types of studies

All relevant randomised controlled trials that compared psychoeducation for siblings of people with SMI with usual care or any other intervention. If a trial was described as 'double blind' but implied randomisation, we planned to include such trials in a sensitivity analysis (see Sensitivity analysis). If their inclusion did not result in a substantive difference, they would have remained in the analyses. If their inclusion did result in important clinically significant, but not necessarily statistically significant differences, we would not have added the data from these lower quality studies to the results of the better trials, but would have presented such data within a subcategory. We excluded quasi-randomised studies, such as those allocating by alternate days of the week.

Types of participants

Brothers and sisters of all ages of adolescents (aged 11 to 17) and adults (aged 18 and over) with severe mental illness as defined in the former section 'Description of the condition', and treated in any setting. We planned to include studies with populations including siblings of people with diagnoses other than SMI as defined by this review, e.g. severe depression/anxiety (as these diagnoses are covered by other Cochrane review groups), but only if ≥ 50% had a psychosis-related disorder or if data specific to siblings of people with SMI were reported independently. Study populations including people other than siblings of individuals with SMI, for instance other family members or carers, would be included if the data specific to siblings were published or obtainable from the study authors.

The definition of siblings is inclusive to incorporate the full range of possible family structures (NICE 2014). Therefore, in addition to biological siblings, half-, adopted-, and step-siblings were included.

Types of interventions

1. Psychoeducation/Psychoeducational intervention

In a previous Cochrane review (Xia 2011), psychoeducational interventions were defined as programmes involving interaction between information providers and service users and/or carers in either an individual or group format. To qualify as a psychoeducational intervention, the education element that instils knowledge or information on the illness condition and its management, must had been significant within the design and be prominent in terms of time duration within the overall content or duration of the multi-modal interventions (comprising at least 50% of the total duration based on the programme's manual content) and be professionally led, although co-facilitation by a lay-person was not excluded. Brief interventions that focused purely on didactic education or health-information using textual or video materials solely, would have been classified as bibliotherapy rather than psychoeducation (NICE 2014). Such bibliotherapies, which did not include interactions between the professional facilitator and the participants, would be excluded. Mutual support groups that from the outset were facilitated solely by lay-persons or family members or siblings would also be excluded. The target participants of psychoeducation interventions might be the person with SMI or their family members or both. This review is concerned with psychoeducational interventions that targeted or included siblings as participants although other family members or relatives and service users might also be included in the interventions.

We considered interventions with a short duration (10 sessions or fewer; or where the number of sessions was not stated but were delivered over a 10-week period, or less) as 'brief' and interventions of longer duration (more than 10 sessions, or where the number of sessions was not stated but were delivered over a period longer than 10 weeks) as 'standard', in line with a previous review on psychoeducation targeting service users with schizophrenia (Xia 2011).

Any intervention that met the criteria as defined above would be included. We also planned to compare psychoeducational interventions that used different modes of delivery or design with each other. For studies in which people were given additional treatments within psychoeducation, data would only be included if the adjunct treatment was evenly distributed between groups and it was only psychoeducation that was randomly assigned.

2. Placebo, no intervention, usual or standard care or any other intervention other than psychoeducation

Any intervention other than psychoeducation whose content, mode of delivery and design were clearly defined, e.g. counselling, cognitive behavioural therapy, family therapy, would be included as a comparison, in addition to placebo, no intervention, usual or standard care. We defined "usual or standard care" as the normal level of psychiatric care or services provided in the geographical area for siblings where the trial was carried out. These psychiatric care and services provided for siblings of service users, in most circumstances, were minimal and most often included sign-posting to information and voluntary services for carers/families (Sin 2012a; Smith 2009).

Types of outcome measures

Since psychoeducation usually aims to impact on outcomes ranging from immediate changes (such as changes in knowledge) to changes in more intricate behavioural and attitudinal outcomes that may take longer to change, we planned to treat all outcomes as either short term (less than one month), medium term (two to five months) or long term (more than six months) following completion of the psychoeducational intervention.

Primary outcomes

1. Siblings' psychosocial wellbeing

1.1 Average change or endpoint scores in wellbeing scores; generic or specific to the siblings' adjustment to psychosis in their brother or sister; physical, psychological, social, cognitive, or functioning.

2. Siblings' quality of life

2.1 Average change or endpoint in quality of life scores; generic or specific to the siblings' adjustment to psychosis in their brother or sister; physical, psychological, social, cognitive, or functioning.

3. Siblings' distress

3.1 Average change or endpoint scores in emotional distress as experienced by siblings specifically depression or anxiety.
3.2 Average change or endpoint scores in worry or fear scales as experienced by siblings.

Secondary outcomes

1. Siblings' knowledge about SMI

1.1 Average change or endpoint scores in siblings' knowledge of SMI.
1.2 Average change or endpoint scores in siblings' understanding of the service user's illness or behaviour.

2. Siblings' coping (attitude, perception and behaviours towards the service user)

2.1 Average change or endpoint scores in siblings' coping.
2.2 Average score or change in siblings' perceived efficacy in coping.
2.3 Average change in siblings' attitudes towards the service user or towards SMI.
2.4 Average change in siblings' behaviour towards the service user or towards psychosis.

3. Siblings' perceived social support or use of social/community support services

3.1 Average change in siblings' perception of or perceived social support scores.
3.2 Average change in siblings' community and/or social service utilisation.

4. Siblings' satisfaction with the intervention

4.1 Leaving the study early
4.2 Siblings' satisfied with the intervention.
4.3 Average change in satisfaction score with care for either siblings or service users.

5. Adverse effects/events affecting siblings

5.1 Any general adverse effects affecting siblings.
5.2 Suicide and all causes of mortality in siblings.

6. Service users' mental state

6.1 Any change in service users' general mental state.
6.2 Clinically important change in specific symptoms.
6.3 Average endpoint general mental state scores.

7. Service users' quality of life

7.1 Average change or endpoint scores in service users' quality of life.
7.2 Average change or endpoint scores in service users' specific aspects of quality of life, i.e. social functioning.
7.3 Average change or endpoint scores in service users' specific aspects of quality of life, i.e. family relationships.

8. 'Summary of findings' table

We used the GRADE approach to interpret findings (Schünemann 2008) and we used the GRADE profiler (GARDEPRO) to import data from RevMan 5.2 (Review Manager) to create 'Summary of findings' tables. These tables provide outcome-specific information concerning the overall quality of evidence from each included study in the comparison, the magnitude of effect of the interventions examined, and the sum of available data on all outcomes we rated as important to patient-care and decision making. We aimed to select the following main outcomes for inclusion in the 'Summary of findings' table.

1. Siblings' wellbeing.
2. Siblings' quality of life.
3. Siblings' distress.
4. Siblings' knowledge about SMI.
5. Siblings' coping.
6. Satisfaction with care for either siblings or the service users.
7. Adverse effects/events affecting siblings.
8. Service users' mental state.

However, data were not available for *Siblings distress*, *Siblings knowledge about SMI, and Adverse effects / events affecting siblings* and we used the available data to create Summary of findings for the main comparison.

Search methods for identification of studies

No language restriction were applied, within the limitations of the search.

Electronic searches

The Trials Search Co-ordinator searched the Cochrane Schizophrenia Group Trials Register, 12 November 2013, using the phrase:
[(*Sibling* or *brother* or *sister* or *family* or *relative* or *relation* or *carer*) AND (*Psychoeducat*) in interventions of STUDY or title of REFERENCE) OR (*family Psychoeducat* or *Psychoeducation family* or *Psychoeducational family* in interventions of STUDY)]
The Cochrane Schizophrenia Group's Trials Register is compiled by systematic searches of major databases, handsearches of relevant journals and conference proceedings (see Group Module). Incoming trials are assigned to relevant existing or new review titles.

Searching other resources

1. Reference searching

We planned to inspect references of all included studies for further relevant studies.

2. Personal contact

We planned to contact the first and/or corresponding author of each included study for information regarding unpublished trials and data.

Data collection and analysis

We performed the review and meta-analyses following the recommendations of *Cochrane Handbook for Systematic Reviews of Interventions* (Higgins 2011). The analyses were performed using Review Manager (RevMan 5.2).

Selection of studies

Two review authors (JS and CJ) independently examined titles and abstracts from the searches for relevance. A random 20% sample were independently re-inspected by IN and EB to ensure reliability. Where disputes arose, the full report was acquired for more detailed scrutiny. Full reports of any study that appeared relevant were obtained and inspected by JS and CJ who then independently assessed each text for eligibility based on the above inclusion criteria. Again, a random 20% of reports were re-inspected by IN and EB in order to ensure reliable selection. Where it was not possible to resolve disagreement by discussion, we made attempts to contact the authors of the study for clarification. We kept a record of the included study and all excluded studies. If it was not possible to obtain sufficient information to judge whether a study was eligible for inclusion, we recorded the study as 'awaiting classification'.

Data extraction and management

1. Extraction

Review authors JS and CJ extracted data from the one included study. Had we been able to include more studies, to ensure reliability, EB and IN would have independently extracted data from a random sample of these studies, comprising 10% of the total. Again, we would have discussed any disagreement, documented decisions and, if necessary, contacted authors of studies for clarification. With remaining problems, CH helped clarify issues and we documented those final decisions.

We extracted data presented only in graphs and figures whenever possible, but included these only if two review authors independently had the same result. We contacted authors through an open-ended request in order to obtain missing or unpublished information or for clarification whenever necessary. If we had included multi-centre studies, where possible, we planned to extract data relevant to each component centre separately.

2. Management

2.1 Forms

We extracted data onto standard, simple forms.

2.2 Scale-derived data

We included continuous data from rating scales only if:
a) the psychometric properties of the measuring instrument had been described in a peer-reviewed journal (Marshall 2000); and
b) the measuring instrument was not written or modified by one of the trialists for that particular trial. Partial use of a validated instrument would only be included if complete subscale results were available for interpretation.
Ideally. the measuring instrument should either be i. a self-report or ii. completed by an independent rater or relative (not the therapist). We realised that this is not often reported clearly; we planned to note if this was the case or not in Description of studies.

2.3 Endpoint versus change data

There are advantages of both endpoint and change data. Change data can remove a component of between-person variability from the analysis. On the other hand, calculation of change needs two assessments (baseline and endpoint), which can be difficult in unstable and difficult to measure conditions such as schizophrenia. We decided primarily to use endpoint data, and only use change data if the former were not available. We combined endpoint and change data in the analysis as we used mean differences (MD) rather than standardised mean differences (SMD) throughout (Higgins 2011).

2.4 Skewed data

Continuous data on clinical and social outcomes are often not normally distributed. To avoid the pitfall of applying parametric tests to non-parametric data, we aimed to apply the following standards to all data before inclusion:
a) standard deviations (SDs) and means are reported in the paper or obtainable from the authors;
b) when a scale starts from the finite number zero, the SD, when multiplied by two, is less than the mean (as otherwise the mean is unlikely to be an appropriate measure of the centre of the distribution (Altman 1996);
c) if a scale started from a positive value (such as the Positive and Negative Syndrome Scale (PANSS), (Kay 1986)), which can have values from 30 to 210), we planned to modify the calculation

described above to take the scale starting point into account. In these cases skew is present if 2 SD > (S-S min), where S is the mean score and 'S min' is the minimum score.
Endpoint scores on scales often have a finite start and end point and these rules can be applied. Skewed data pose less of a problem when looking at means if the sample size is large (>200) and we planned to enter these data into the syntheses. We planned to present skewed endpoint data from studies of less than 200 participants in 'other tables' within the data analyses section rather than enter such data into statistical analyses.

When continuous data are presented on a scale that includes a possibility of negative values (such as change data), it is difficult to tell whether data are skewed or not. We planned to present and enter skewed change data into analyses.

2.5 Common measure

To facilitate comparison between trials, we intended to convert variables that can be reported in different metrics, such as days in hospital (mean days per year, per week or per month) to a common metric (e.g. mean days per month).

2.6 Conversion of continuous to binary

Where possible, we intended to convert outcome measures to dichotomous data. This can be done by identifying cut-off points on rating scales and dividing participants accordingly into 'clinically improved' or 'not clinically improved'. In general, we assumed that if there had been a 50% reduction in a scale-derived score such as the Brief Psychiatric Rating Scale (BPRS, Overall 1962) or the PANSS (Kay 1986), we could consider this as a clinically significant response (Leucht 2005; Leucht 2005a). If data based on these thresholds were not available, we planned to use the primary cut-off presented by the original authors.

2.7 Direction of graphs

Where possible, we entered data in such a way that the area to the left of the line of no effect indicated a favourable outcome for psychoeducation. Where keeping to this made it impossible to avoid outcome titles with clumsy double-negatives (e.g. 'Not unimproved') we planned to report data where the left of the line indicated an unfavourable outcome and would have noted these in the relevant graphs.

Assessment of risk of bias in included studies

Review authors JS, CJ, EB and IN worked independently to assess risk of bias by using criteria described in the *Cochrane Handbook for Systemic Reviews of Interventions* (Higgins 2011) to assess trial quality. This set of criteria is based on evidence of associations between overestimate of effect and high risk of bias of the article such as sequence generation, allocation concealment, blinding, incomplete outcome data and selective reporting.
If the raters disagreed, we would have made the final rating by consensus, with the involvement of another member (CH) of the review group. Where inadequate details of randomisation and other characteristics of trials were provided, we would have contacted authors of the studies in order to obtain further information. We planned to report non-concurrence in quality assessment, but if disputes arose as to which category a trial was to be allocated, again, we would have resolved these disputes by discussion.
We planned to note the level of risk of bias in both the text of the review and in the 'Summary of findings' table.

Measures of treatment effect

1. Binary data

For binary outcomes, we would have calculated a standard estimation of the risk ratio (RR) and its 95% confidence interval (CI). It has been shown that RR is more intuitive (Boissel 1999) than odds ratios and that odds ratios tend to be interpreted as RR by clinicians (Deeks 2000). For binary data presented in the 'Summary of findings' table, where possible, we planned to calculate illustrative comparative risks as the number needed to treat to benefit/harm (NNTB/NNTH) statistic with its confidence intervals is intuitively attractive to clinicians but is problematic both in its accurate calculation in meta-analyses and interpretation (Hutton 2009).

2. Continuous data

For continuous outcomes, we calculated mean differences (MD) with 95% CIs for comparisons between groups. We preferred not to calculate effect size measures, standardised mean difference (SMD). However, if scales of very considerable similarity had been used, we would have presumed there was a small difference in measurement, and we would have calculated effect size and transformed the effect back to the units of one or more of the specific instruments.

Unit of analysis issues

1. Cluster trials

Studies increasingly employ 'cluster randomisation' (such as randomisation by clinician or practice) but analysis and pooling of clustered data poses problems. Firstly, authors often fail to account for intra-class correlation in clustered studies, leading to a 'unit of analysis' error (Divine 1992) whereby P values are spuriously low, CIs unduly narrow and statistical significance overestimated. This causes type I errors (Bland 1997; Gulliford 1999).

If clustering had not been accounted for in primary studies, we planned to present data in a table, with a (*) symbol to indicate the presence of a probable unit of analysis error. In subsequent versions of this review, if we find such cluster studies, we will seek to contact first authors of studies to obtain intra-class correlation coefficients (ICCs) for their clustered data and to adjust for this by using accepted methods (Gulliford 1999). If clustering had been incorporated into the analysis of primary studies, we planned to present these data as if from a non-cluster randomised study, but we would have adjusted for the clustering effect.

We have sought statistical advice and have been advised that the binary data as presented in a report should be divided by a 'design effect'. This is calculated using the mean number of participants per cluster (m) and the ICC [Design effect = 1+(m-1)*ICC] (Donner 2002). If the ICC was not reported, we would assume it to be 0.1 (Ukoumunne 1999).

If cluster studies have been appropriately analysed taking into account ICCs and relevant data documented in the report, synthesis with other studies would be possible using the generic inverse variance technique.

2. Cross-over trials

A major concern of cross-over trials is the carry-over effect. It occurs if an effect (e.g. pharmacological, physiological or psychological) of the treatment in the first phase is carried over to the second phase. As a consequence, on entry to the second phase the participants can differ systematically from their initial state despite a wash-out phase. For the same reason, cross-over trials are not appropriate if the condition of interest is unstable (Elbourne 2002). As both effects are very likely in severe mental illness, we would only have used data of the first phase of cross-over studies.

3. Studies with multiple treatment groups

Had we included a study that involved more than two intervention groups compared against a control, if relevant, we planned to present the additional intervention groups in additional comparisons. If data were binary, we planned to simply add and combine these within the two-by-two table. If data were continuous, we planned to combine the data following the formula in section 7.7.3.8 (Combining groups) of the *Cochrane Handbook for Systemic reviews of Interventions* (Higgins 2011). Where the additional treatment arms were not relevant, we would not have used these data.

Dealing with missing data

1. Overall loss of credibility

At some degree of loss of follow-up, data must lose credibility (Xia 2009). We chose that, for any particular outcome, should more than 50% of data be unaccounted for, we would not reproduce these data or use them within analyses. If, however, more than 50% of those in one arm of a study were lost, but the total loss was less than 50%, we would address this within the 'Summary of findings' table/s by down-rating quality. Finally, we would have also downgraded quality within the 'Summary of findings' table/s should loss be 25% to 50% in total.

2. Binary

In the case where attrition for a binary outcome was between 0% and 50% and where these data were not clearly described, we planned to present data on a 'once-randomised-always-analyse' basis (an intention-to-treat analysis). We would have assumed those leaving the study early to have the same rates of negative outcome as those who completed, with the exception of the outcome of death and adverse effects. For these outcomes, the rate of those who stayed in the study - in that particular arm of the trial - would be used for those who did not. We planned to undertake a sensitivity analysis to test how prone the primary outcomes were to change when data only from people who completed the study to that point were compared to the intention-to-treat analysis using the above assumptions.

3. Continuous

3.1 Attrition

In the case where attrition for a continuous outcome was between 0% and 50%, and data only from people who completed the study to that point were reported, we would have reproduced these.

3.2 Standard deviations

If SDs were not reported, we planned first try to obtain the missing values from the authors. If not available, where there were missing measures of variance for continuous data, but an exact standard error (SE) and CIs available for group means, and either the 'P' value or 't' value available for differences in mean, we could have calculated them according to the rules described in the *Cochrane Handbook for Systemic reviews of Interventions* (Higgins 2011). When only the SE is reported, SDs are calculated by the formula SD = SE * square root (n). Chapters 7.7.3 and 16.1.3 of the *Cochrane Handbook for Systemic reviews of Interventions* (Higgins 2011) present detailed formulae for estimating SDs from P values, t or F values, CIs, ranges or other statistics. If these formulae did not apply, we would have calculated the SDs according to a validated imputation method which is based on the SDs of the other included studies (Furukawa 2006). Although some of these imputation strategies can introduce error, the alternative would be to exclude a given study's outcome and thus to lose information. We nevertheless planned to examine the validity of the imputations in a sensitivity analysis excluding imputed values.

3.3 Last observation carried forward

We anticipated that in some studies the method of last observation carried forward (LOCF) would be employed within the study report. As with all methods of imputation to deal with missing data, LOCF introduces uncertainty about the reliability of the results (Leucht 2007). Therefore, if LOCF data had been used in the trial, if less than 50% of the data had been assumed, we would reproduce these data and indicate that they are the product of LOCF assumptions.

Assessment of heterogeneity

1. Clinical heterogeneity

If we had included more studies, we planned to consider all included studies initially, without seeing comparison data, to judge clinical heterogeneity. We would simply have inspected all studies for clearly outlying people or situations which we had not predicted would arise. When such situations or participant groups arose, these would have been fully discussed.

2. Methodological heterogeneity

Had we included more trials, we planned to consider all included studies initially, without seeing comparison data, to judge methodological heterogeneity. We would simply have inspected all studies for clearly outlying methods which we had not predicted would arise. If such methodological outliers arose, we would have discussed these fully.

3. Statistical heterogeneity

3.1 Visual inspection

We planned to inspect graphs visually to investigate the possibility of statistical heterogeneity.

3.2 Employing the I^2 statistic

We planned to investigate heterogeneity between studies by considering the I^2 method alongside the Chi^2 'P' value. The I^2 provides an estimate of the percentage of inconsistency thought to be due to chance (Higgins 2003). The importance of the observed value of I^2 depends on i. magnitude and direction of effects and ii. strength of evidence for heterogeneity (e.g. 'P' value from Chi^2 test, or a CI I^2). We would have interpreted an I^2 estimate greater than or equal to around 50% accompanied by a statistically significant Chi^2 statistic as evidence of substantial levels of heterogeneity (Section 9.5.2 - Higgins 2011). When substantial levels of heterogeneity were found in the primary outcome, we would have explored reasons for heterogeneity (Subgroup analysis and investigation of heterogeneity).

Assessment of reporting biases

1. Protocol versus full study

Reporting biases arise when the dissemination of research findings is influenced by the nature and direction of results. These are described in section 10.1 of the *Cochrane Handbook for Systemic reviews of Interventions* (Higgins 2011). We would have tried to locate protocols of included randomised trials. If the protocol was available, outcomes in the protocol and in the published report would be compared. If the protocol was not available, outcomes listed in the methods section of the trial report would be compared with actually reported results.

2. Funnel plot

Reporting biases arise when the dissemination of research findings is influenced by the nature and direction of results (Egger 1997). These are again described in Section 10 of the *Cochrane Handbook for Systemic Reviews of Interventions* (Higgins 2011). We are aware that funnel plots may be useful in investigating reporting biases but are of limited power to detect small-study effects. As we only included one study we did not use funnel plots. In future versions of this review, where funnel plots are possible, we plan to seek statistical advice in their interpretation.

Data synthesis

We understand that there is no closed argument for preference for use of fixed-effect or random-effects models. The random-effects method incorporates an assumption that the different studies are estimating different, yet related, intervention effects. This often seems to be true to us and the random-effects model takes into account differences between studies even if there is no statistically significant heterogeneity. There is, however, a disadvantage to the random-effects model. It puts added weight onto small studies which often are the most biased ones. Depending on the direction of effect, these studies can either inflate or deflate the effect size. We chose to use the random-effects model for all analyses.

Subgroup analysis and investigation of heterogeneity

1. Subgroup analyses - only primary outcomes

We planned to conduct three syntheses which compare psychoeducation with i) all comparators (treatment as usual (TAU), standard care, placebo, any other active treatment), ii) all comparators, excluding any active treatment (TAU, standard care, placebo only); and iii) any active treatment only.

1.1 Siblings of service users with different diagnoses

We are interested in whether siblings of service users with different diagnoses (e.g. schizophrenia, Type I bipolar disorder, early onset

psychosis) would have similar benefits or effects from the interventions. We proposed to undertake comparisons only for primary outcomes to minimise the risk of multiple comparisons.

1.2 Intervention types

We anticipated undertaking subgroup analyses investigating the different lengths of intervention durations: interventions with brief duration (10 sessions or less or, where the number of sessions was not stated but which was delivered within 10 weeks or less) and interventions with longer duration (more than 10 sessions or where the number of sessions was not stated but which were delivered in more than 10 weeks). We also planned to present data on intervention programmes using an individual (i.e. one information provider seeing one participant or participants from one family) and using a group format (i.e. more than two participants or participants from one family/service users involved in the sessions). These data, although synthesised overall, would, if possible, be presented in subgroups. We proposed to undertake comparisons only for primary outcomes to minimise the risk of multiple comparisons.

2. Investigation of heterogeneity

If inconsistency was high, this would have been reported. First, we planned to investigate whether data had been entered correctly. Second, if data were correct, we would have visually inspected the graph and successively removed outlying studies to see if homogeneity was restored. For this review, we decided that should this occur with data contributing to the summary finding of no more than around 10% of the total weighting, we would present data. If not, we would not pool data and would discuss relevant issues. We know of no supporting research for this 10% cut off but are investigating use of prediction intervals as an alternative to this unsatisfactory state.

When unanticipated clinical or methodological heterogeneity was obvious, we would simply state hypotheses regarding these for future reviews or versions of this review. We did not anticipate undertaking analyses relating to these.

Sensitivity analysis

1. Implication of randomisation

We aimed to include trials in a sensitivity analysis if they were described in some way as to imply randomisation. For the primary outcomes, we planned to include these studies and if there was no substantive difference when the implied randomised studies were added to those with better description of randomisation, then we would have used all data from these studies.

2. Assumptions for lost binary data

Where assumptions have to be made regarding people lost to follow-up (see Dealing with missing data), we planned to compare the findings of the primary outcomes when we used our assumption/s and when we used data only from people who complete the study to that point. If there was a substantial difference, we would have reported results and discussed them, but continued to employ our assumption.

Where assumptions have to be made regarding missing SDs data (see Dealing with missing data), we planned to compare the findings of the primary outcomes when we used our assumption/s and when we used data only from people who complete the study to that point. We planned to undertake a sensitivity analysis to test how prone results were to change when completer-only data only were compared with the imputed data using the above assumption. If there had been a substantial difference, we would have reported results and discussed them, but continued to employ our assumption.

3. Risk of bias

We planned to analyse the effects of excluding trials judged to be at high risk of bias across one or more of the domains of randomisation (implied as randomised with no further details available): allocation concealment, blinding and outcome reporting for the meta-analysis of the primary outcome. If the exclusion of trials at high risk of bias did not substantially alter the direction of effect or the precision of the effect estimates, then we would have included data from these trials in the analyses.

4. Imputed values

We planned also to undertake a sensitivity analysis to assess the effects of including data from trials where we used imputed values for ICC in calculating the design effect in cluster randomised trials. If substantial differences were noted in the direction or precision of effect estimates in any of the sensitivity analyses listed above, we would not have pooled data from the excluded trials with the other trials contributing to the outcome, but would have presented them separately.

5. Fixed and random effects

We planned to synthesise data using a random-effects model, however, we would also have synthesised data for the primary outcome using a fixed-effect model to evaluate whether this altered the significance of the results.

RESULTS

Description of studies

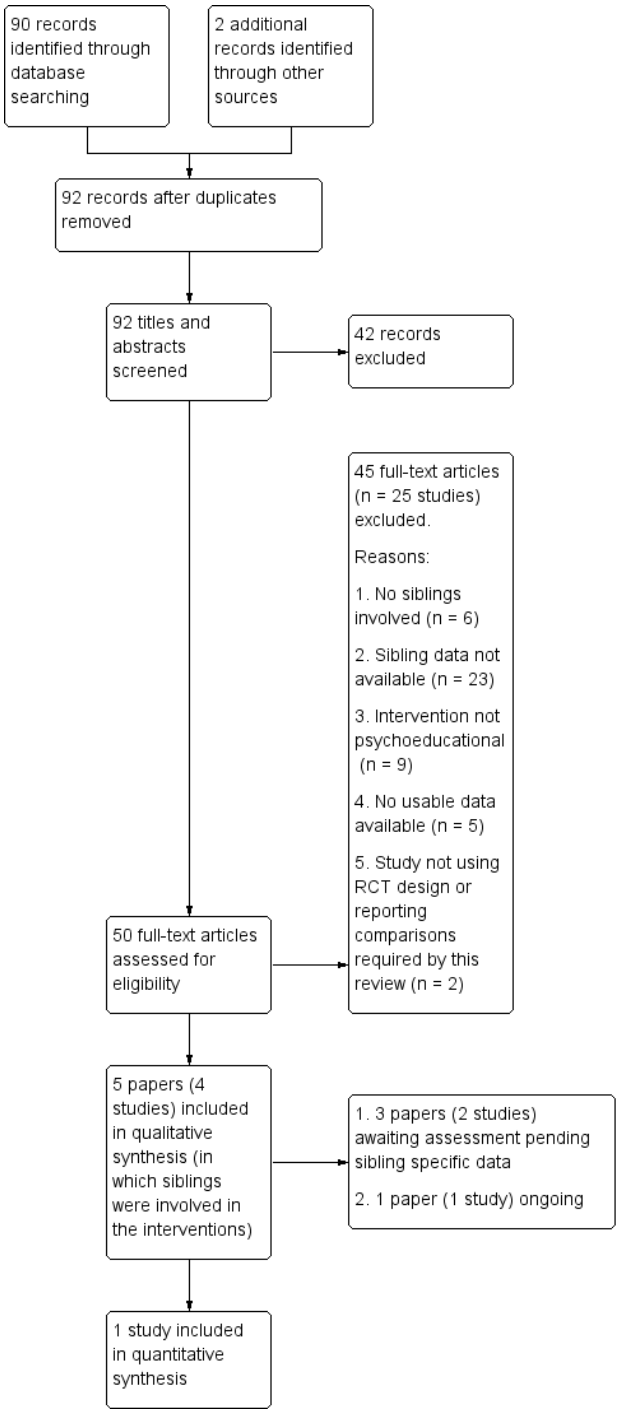
See Characteristics of included studies; Characteristics of excluded studies; Characteristics of studies awaiting classification; Characteristics of ongoing studies.

For more detailed description of each study, please refer to the Characteristics of included studies and Characteristics of excluded studies tables.

Results of the search

The search results from the Cochrane Schizophrenia Group Trials Register yielded 90 unique abstracts. Two further studies were identified by checking through the reference lists of these papers and updated publication of registered trials (i.e. 12th November 2013). All 92 abstracts and titles were independently screened by two authors (JS and CJ) whilst 20% of a random sample of these were screened by two additional authors (EB and IN). Full papers were pulled for abstract-examination that either author (JS, CJ, EB and IN) felt they could not confidently exclude: 50 full-text articles were assessed for eligibility and assessed independently by two authors (JS and CJ), with 20% random sample being screened by two additional authors (EB and IN) again. For trials that implied that siblings were involved and/or siblings' involvement was unclear, we contacted the trial authors for further data by email correspondence; trial authors who did not respond within three weeks were contacted again. In trials in which sibling data were collected, we asked the authors for either the data specific to siblings or a subgroup analysis specific to siblings' data for further analysis to be conducted in this review. Authors' responses are summarised in the Characteristics of included studies; Characteristics of excluded studies; and Characteristics of studies awaiting classification sections. Any papers where there was disagreement between the two authors (JS and CJ) were discussed in the review team for elaboration and operationalisation of the eligibility criteria. The results of the search is summarised in Figure 1.

Figure 1. Study flow diagram.



Initially, 14 studies (reported by 28 papers) met the inclusion criteria in terms of siblings being amongst the family members targeted in a trial of a psychoeducational intervention (Bauml 1997; Chan 2009; Cheng 2005; Chien 2007; Dyck 2002; Fiorillo 2011; Gutierrez-Maldonado 2009; Kane 1990; Lobban 2013; Nasr 2009; Posner 1992; Ran 2002; Smith 1987; Solomon 1996). In these studies siblings tended to comprise a small proportion of the total sample of family member/carer-participants, who are most commonly parents, especially mothers of service users with schizophrenia. In 10 studies that clearly identified kinship of the participants with the service users, the proportion of siblings in the study samples ranged from 3.9% (nine siblings out of 230 family member-participants in Fiorillo 2011) to 29% (21 siblings out of 73 family member-participants in Chan 2009), mean = 13.7%, median = 14%. However, many of these trials no longer had access to their dataset and so it was not possible to extract data specific to siblings for subgroup analysis (e.g. Kane 1990; Solomon 1996). Other trials had included siblings' data within data for a generic group of relatives or friends, which meant that extracting data specific to siblings was, again, impossible (Bauml 1997; Lobban 2013; Smith 1987).

Included studies

See also Characteristics of included studies for descriptions of the study. With additional unpublished data obtained from the trial authors, one study was included that met all the inclusion criteria and provided data specific to siblings for this review (Chien 2007). The included study was based in Hong Kong, China (Chien 2007).

1. Length of trial

The duration of the intervention in Chien 2007 lasted for 36 weeks, including 18 biweekly psychoeducation sessions, each lasting two hours. Chien 2007 followed up the participants at one week post intervention and at 12 months.

2. Design

The only study included in the review was a randomised controlled trial. Blinding was described in the study as using blinded or independent assessors who were not aware of participants' allocation to undertake all assessments (Chien 2007), whilst the therapists who facilitated the interventions and the participants were not blind due to the nature of psychosocial interventions and the lack of placebo condition. The participants' outcomes were taken at three time points: before the intervention, immediately following the intervention which ran for 36 weeks, and at 12-month follow-up (Chien 2007).

2. Participants

The included study provided results on nine siblings who comprised 10.7% of the total participants of 84 relatives in the study (Chien 2007). The trial provided psychoeducational intervention to family members/carers of adults with schizophrenia living in the community, and the siblings included were key caregivers who provided substantial day-to-day care for the service users. To be eligible to join the study, the siblings had to also be living with the service users (Chien 2007). No demographic characteristics specific to the siblings were available although the overall demographic characteristics of all the family members indicate that two-thirds of the family member-participants were female. The mean age of family member-participants in the study was 40.6 (SD = 7.2) years (Chien 2007).

3. Interventions

3.1 Psychoeducation

The study delivered the psychoeducational interventions using a group format facilitated by experienced mental health nurses (Chien 2007). The psychoeducational intervention also involved the service users in part of the group programme in that they attended six out of 18 sessions, which were focused on education about the illness and its management (Chien 2007). The psychoeducation programme evaluated by the study provided education, which was professionally-led, followed by peer support and discussion amongst group participants. Chien 2007 reported adapting the intervention content to address specific ethnic-cultural needs of family carers whilst building upon well-established theoretical modelor frameworks, such as the McFarlane model (McFarlane 2002). Furthermore, engagement strategies that aimed to enhance attendance by participants and retention in the trial were used in the programme, including running sessions at a convenient community location.

3.2. Standard care

The comparison group received usual or standard care, which involved routine psychiatric outpatient and family services only (Chien 2007). These services consisted of monthly medical consultation and advice; individual nursing advice on community healthcare services; brief family education (up to two or three one-hour group sessions) on the service user's illness, medication and treatment plan and counselling provided by clinical psychologists if necessary. The same standard care was also provided to the psychoeducation group participants.

4. Setting

The study was conducted with family members of service users with a diagnosis of schizophrenia, living in the community and receiving ongoing out-patient care (Chien 2007).

5. Outcomes

We identified no studies reporting siblings' outcomes independently which could be included in the review at the current time; however, there is an ongoing trial (Sin 2012a). For all the full texts screened that implied siblings were included in the reported trials, we contacted trial authors to seek data specific to siblings for analysis in the review. Only one trial provided data specific to siblings (Chien 2007), within the review timeframe (four months from the initial search which was conducted in early September 2013). This study used a variety of scales to assess family members' outcomes and service users' clinical responses. The outcome measures reported in the included study are described below.

5.1 Siblings' quality of life

Chien 2007 reported on siblings' quality of life, using the Family Assessment Device (FAD). Chien 2007 used a validated FAD (Epstein 1983) translated into Chinese with reversed scoring in which higher scores indicate healthy family functioning.

5.2 Siblings' coping (with burden)

Chien 2007 measured coping with burden using the Family Burden Interview Schedule (FBIS) (Pai 1981). FBIS is a 24-item scale used to assess the burden of care placed on families in caring for a mentally ill family member. It assesses six domains of the carer's burden: the effects on family finances, routines, leisure time, interaction, and physical and mental health. Higher scores indicate a greater burden of care (Pai 1981).

5.3 Siblings' satisfaction with the intervention

5.3.1 Leaving the study early**:** Chien 2007 reported participant attrition rates.

5.4 Service users' mental state

Chien 2007 reported on service users' general mental state, using the Brief Psychiatric Rating Scale (BPRS) (Overall 1962). The BPRS is an 18-item scale measuring positive symptoms, general psychopathology and affective symptoms. Higher scores indicate higher severity of symptoms. Chien 2007 also reported service users' average number of re-hospitalisation and inpatient bed occupancy in terms of average days of hospital stays in the previous six months, at pre-test and post-test time points and at 12-month follow-up.

5.5 Service users' quality of life

Service users' specific aspects of quality of life, i.e. social functioning, was reported by Chien 2007 using the Specific Level of Functioning Scale (SLOF) (Schneider 1983). SLOF is a Likert-type scale with possible scores ranging from 43 to 215, with higher scores indicating better outcomes.

5.6 Missing outcomes

Chien 2007 did not report data on siblings' wellbeing or distress, the primary outcomes we were interested in. There was also no data on siblings' knowledge, perceived social support, or adverse effects or events affecting siblings, from the single study we included.

Excluded studies

Studies excluded from the review are described in Characteristics of excluded studies. Fourty-two papers were excluded at the title and abstract-screening stage because they did not meet the inclusion criteria for the review. In the full-text screening stage covering 50 papers, we excluded 45 papers reporting on 25 studies: four studies were excluded because siblings were not included in the trials (Lacruz 1999; So 2006; Rotondi 2011; Sharif 2012); 10 studies collected siblings' data as they were participants in the trials but such data were grouped into the overall family members/carers dataset that was not made available or was no longer accessible for the review (Bauml 1997; Cheng 2005; Dyck 2002; Kane 1990; Lobban 2013; Nasr 2009; Smith 1987; Solomon 1996; Fiorillo 2011; Posner 1992); five studies investigated multi-modal interventions that did not meet our intervention definition (Chien 2010; Hogarty 1986; Magliano 2006; Roncone 2000; Schepp 2009); five studies provided no usable data and/or authors were not contactable for further data or clarification (Gutierrez-Maldonado 2009; O'Callaghan 2009; Shinde 2005; Weng 1994; Ying 2006); and one study compared the same psychoeducation intervention for two different durations, and so did not meet the inclusion criteria for this review (Motlova 2002). Of note, data in Figure 1 - Study flow diagram relate to exclusion of full-text articles.

Studies awaiting assessment

Two studies await assessment pending the availability of the sibling-specific data (Chan 2009; Ran 2002). In both cases, the trial authors were not able to provide the sibling-specific data for our use within the review timeframe. The Ran 2002 study, which was conducted in mainland China had 55 siblings among 326 key relative-participants (17%). In Chan 2009, which was conducted in Hong Kong, China, 29% of the 73 family carer-participants were siblings (n = 21).

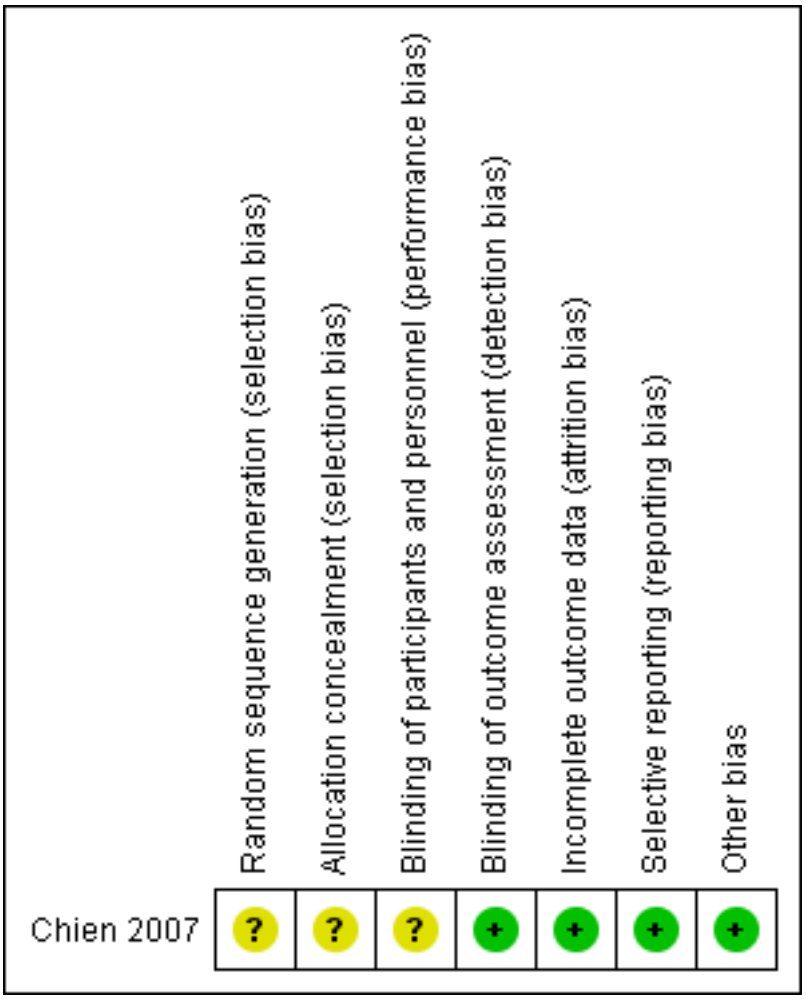
Ongoing studies

One study is an ongoing trial testing an online psychoeducational intervention with siblings of individuals with first episode psychosis and is expected to complete by end of 2015 (Sin 2012).

Risk of bias in included studies

Our overall impression of risk of bias in the included study is represented in Figure 2 whilst assessment of risk of bias of the included study can be found in Characteristics of included studies. The included study was a randomised controlled trial but the randomisation method used was not reported clearly.

Figure 2. 'Risk of bias' summary: review authors' judgements about each risk of bias item for each included study.



Allocation

The included study did not describe the method used to generate the randomisation sequence (Chien 2007). Allocation concealment was also not reported and so it was assigned an unclear risk of selection bias.

Blinding

The only included study used standard care as the comparison, making it impossible to blind the participants (Chien 2007). A researcher blind to group allocation was used in the study (Chien 2007) for collecting both service users' and family members' outcomes. In addition, some participants-rated/self-reported outcome measures were used, however, and as participants were not blind to their allocation, the potential for performance and detection bias remain. We therefore had to rate the risk of bias as (at best) 'unclear'.

Incomplete outcome data

The only included study performed intention-to-treat analysis on the results and reported an attrition rate of 7% in the intervention group and 10% in the control group (Chien 2007).

Selective reporting

We were not able to find any published protocol for the included study. We found no evidence of selective reporting of outcomes as

all the outcomes measured by Chien 2007 were reported within the study article.

Other potential sources of bias

There were no other obvious potential sources of bias.

Effects of interventions

See: **Summary of findings for the main comparison** Psychoeducation compared with standard care for siblings of people with severe mental illness
See also Summary of findings for the main comparison.

Comparison 1 - Any form of psychoeducation versus standard care

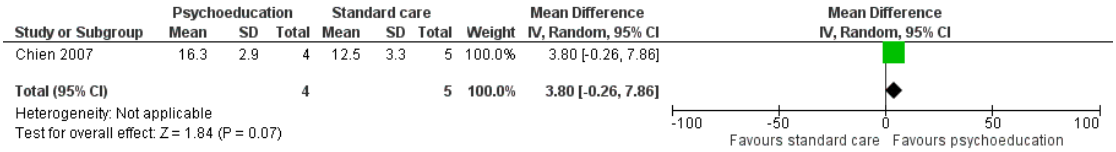
1. Siblings' psychosocial wellbeing

The included study provided no measures for this outcome.

2. Siblings' quality of life

Chien 2007 reported siblings' quality of life in terms of family functioning using the Family Assessment Device (FAD). We found no statistically significant difference in those siblings receiving the intervention, compared to those receiving standard care, at 12 months (1 RCT, n = 9, mean difference (MD) 3.80 95% CI -0.26 to 7.86, Analysis 1.1). Also see Figure 3.

Figure 3. Forest plot of comparison: 1 Psychoeducation versus Standard Care, outcome: 1.1 Siblings' quality of life - at 12 months (reversed FAD, high = good).



3. Siblings' distress

No study reported this outcome.

4. Siblings' knowledge about severe mental illness (SMI)

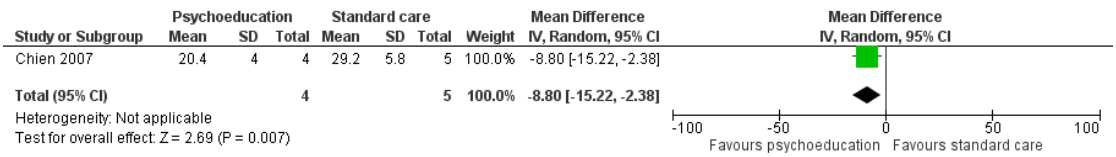
No study reported this outcome.

5. Siblings' coping (attitude, perception and behaviour towards the service users)

5.1 Siblings' coping

Continuous data on siblings' coping in terms of burden of care, measured by the Family Burden Interview Schedule (FBIS) in Chien 2007 showed a significant difference between the intervention and control group; those receiving the intervention had better outcomes on the FBIS scale in the long term (1 RCT, n = 9, MD -8.80 95% CI -15.22 to -2.38, Analysis 1.2). Also see Figure 4.

Figure 4. Forest plot of comparison: 1 Psychoeducation versus Standard Care, outcome: 1.2 Siblings' coping (in terms of burden) - at 12 months (FBIS, high = poor).



5.2 Siblings' perceived efficacy in coping

The included study provided no measures for this outcome.

5.3 Siblings' attitude towards the service user or towards SMI

The included study provided no measures for this outcome.

6. Siblings' perceived social support or use of social/community support services

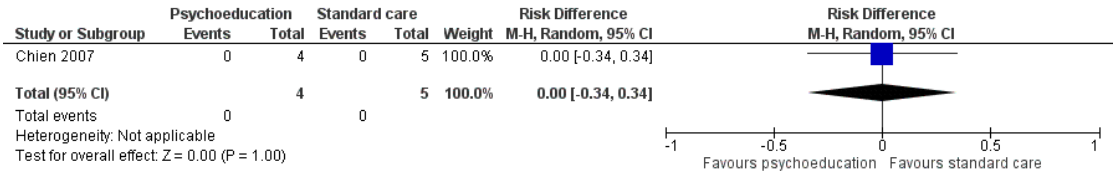
The included study provided no measures for this outcome.

7. Siblings' satisfaction with the intervention

7.1 Leaving the study early

Chien 2007 reported participants' attrition rates: 7% participants (n = 3 out of 42 in total) in the intervention group who did not complete the psychoeducation programme, and 10% participants (n = 4 out of 42 in total) in the control group did not complete post-test, but none of these were siblings (Chien 2007), hence no significant differences were found between groups in terms of siblings leaving the study early (1 RCT, n = 9, risk difference (RD) 0.00 95% CI -0.34 to 0.34, Analysis 1.3). Also see Figure 5.

Figure 5. Forest plot of comparison: 1 Psychoeducation versus Standard Care, outcome: 1.3 Leaving the study early for any reason - at 12 months.



7.2 Siblings' satisfaction with the intervention

The included study provided no measures for this outcome.

8. Adverse effects/events affecting siblings

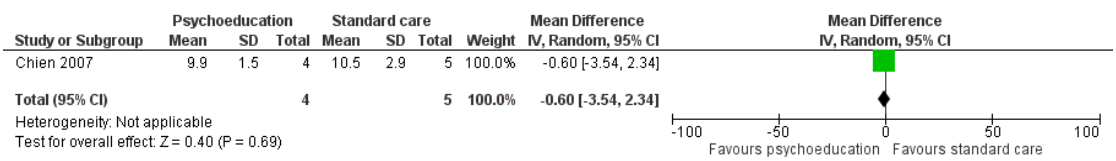
The included study provided no measures for this outcome.

9. Service users' mental state

9.1 Service users' general mental state

Chien 2007 reported on service users' general mental state but no significant differences in terms of average Brief Psychiatric Rating Scale (BPRS) scores at 12 months were found (n = 9, MD -0.60 95% CI -3.54 to 2.34, Analysis 1.4). See Figure 6.

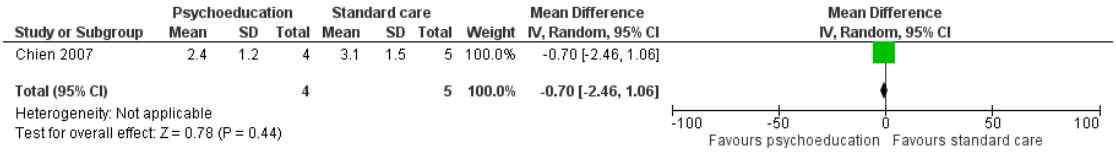
Figure 6. Forest plot of comparison: 1 Psychoeducation versus Standard Care, outcome: 1.4 Service users' general mental state - at 12 months (BPRS, high = poor).



9.2 Service users' number of re-hospitalisation to a psychiatric inpatient unit over six months

The subgroup analysis on siblings' data obtained from Chien 2007 showed no statistical significant differences in service users' average number of readmissions to a psychiatric inpatient unit over the six to 12 month period following completion of the intervention, regardless of whether the siblings received psychoeducation or standard care (n = 9, MD -0.70 95% CI -2.46 to 1.06, Analysis 1.5). Also see Figure 7.

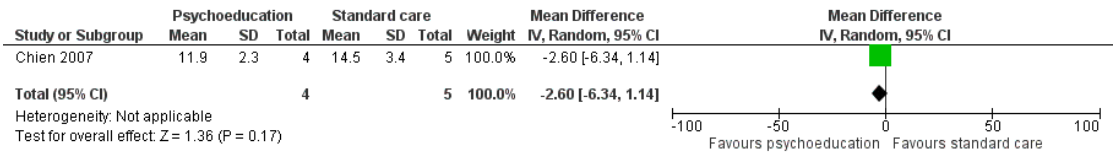
Figure 7. Forest plot of comparison: 1 Psychoeducation versus Standard Care, outcome: 1.5 Service users' number of re-hospitalisation to a psychiatric inpatient unit over six months - at 12 months.



9.3 Service users' inpatient bed occupancy in terms of average days of hospital stays over six months

No significant difference was found between groups in service users' duration of hospital stay over the six to 12 month period, following the completion of the intervention in Chien 2007's trial (n = 9, MD -2.60 95% CI -6.34 to 1.14, Analysis 1.6). See Figure 8.

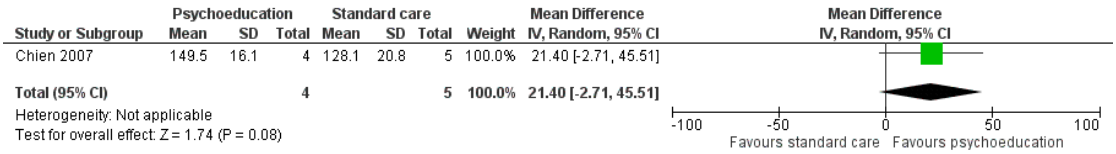
Figure 8. Forest plot of comparison: 1 Psychoeducation versus Standard Care, outcome: 1.6 Service users' inpatient bed occupancy in terms of average days of hospital stay over 6 months - at 12 months.



10. Service users' specific aspect of quality of life

Chien 2007 reported service users' specific aspects of quality of life, i.e. social functioning, using the Specific Level of Functioning Scale (SLOF). No significant differences were found between psychoeducation and standard care in terms of service users' functioning (n = 9, MD 21.40 95% CI -2.71 to 45.51, Analysis 1.7). Also see Figure 9.

Figure 9. Forest plot of comparison: 1 Psychoeducation versus Standard Care, outcome: 1.7 Service users' specific aspect of quality of life, i.e. social functioning - at 12 months (SLOF, high = good).



Subgroup analyses

No subgroup analyses were conducted due to paucity of data. The included study included only service users diagnosed with schizophrenia and their key family carers. The intervention used a group format and ran for eight months (Chien 2007).

Sensitivity analyses

Sensitivity analyses were not possible given the paucity of data.

DISCUSSION

The greatest problem we found in examining the effectiveness of psychoeducational intervention for siblings of people with SMI was the lack of data specific to siblings. Given that the only eligible study we included for this review involved only nine siblings, this renders the review results derived solely from one study with nine siblings (Chien 2007).

Summary of main results

Considering the paucity of data from a single study involving nine siblings amongst 75 other relatives as family carers of people with schizophrenia, the effectiveness of psychoeducation is at best, inconclusive and the results reported in this review should be considered with caution. See also Summary of findings for the main comparison.

The only study (Chien 2007) we included reported a number of siblings' and service users' outcomes we were interested in, such as siblings' quality of life, coping, satisfaction with the intervention and service users' mental state. However, there was a distinct lack of data on our primary outcomes, concerned with siblings' wellbeing, both in terms of positive measures and proxy measures, such as various psychological morbidities.

Amongst the outcomes that were reported, there is some weak evidence to show that psychoeducation has positive effects on siblings' coping with their perceived burden of care. Psychoeducational intervention (at least when delivered as a face-to-face group) seemed well-tolerated, when compared with standard care. However, there is no evidence from Chien 2007 that psychoeducation is associated with better clinical outcomes in service users when their siblings received the intervention. Having said that, there is too little data for us to be able to draw any conclusion regarding the outcomes for both siblings' and their brother's or sister's with severe mental illness.

Definitions of psychoeducational interventions in the literature

Another problem we encountered in the review process is the varying definitions of psychoeducation and/or psychoeducational interventions in the literature. Whilst the review identified psychoeducation as popular and frequently implemented across different clinical settings around the world, this also often meant that the psychoeducational interventions had been adapted and modified to suit local ethnic-cultural needs and local service infrastructure. We came across a number of studies that referred to their interventions as "psychoeducative / psychoeducational (family) therapy / intervention / work", in which psychoeducation was blended with a number of other approaches and components (e.g. Hogarty 1986; Magliano 2006). The components most commonly embedded in these multi-modal family-based interventions include: communication skills and problem-solving practice and role-plays, cognitive, behavioural and/or cognitive-behavioural work with the family members. Many interventions involved the majority of the programme content or duration being spent on practising and adopting such skills or behaviour by family members and carers whilst psychoeducation was used as a pre-requisite for later therapeutic work (e.g. Hogarty 1986; Magliano 2006). Other programmes were referred to using user or carer-friendly terms, such as "supported self-management intervention for relatives" (Lobban 2013), which further complicated the process of identifying psychoeducational interventions. To ensure our selection of included studies was accurate with reference to the intervention definition, whenever we were uncertain about the content of the intervention

from the abstract and title of the article, we obtained the full text for examination and if necessary, contacted the authors for further clarification.

Overall completeness and applicability of evidence

The search identified 14 studies that investigated the effectiveness of psychoeducational interventions targeting family members of people with SMI, inclusive of siblings. Nonetheless, as this review relied heavily on obtaining data specific to siblings from trials and many of the excluded studies had grouped siblings' data into overall family carers/members' outcomes at the outset, the availability of data was compromised despite assistance provided by the trial authors. Four studies had grouped siblings' data with that of other relatives, making extracting siblings' data impossible (Bauml 1997; Dyck 2002; Lobban 2013; Smith 1987). The remaining 10 studies had collected data on 163 siblings (amongst 1259 relatives-participants in total). Four of these studies originated from China (Chien 2007 (included study); Chan 2009; Cheng 2005; Ran 2002), one from Pakistan (Nasr 2009); one from Chile (Gutierrez-Maldonado 2009); one from Italy (Fiorillo 2011) and three from North America (Kane 1990; Posner 1992; Solomon 1996). All targeted family members as key carers for people with schizophrenia living in the community. Eight out of the 10 studies used a multi-family group format for their intervention (e.g. Chan 2009; Fiorillo 2011; Gutierrez-Maldonado 2009); one used an individual family-based approach (Nasr 2009); and another used mixed individual family sessions supplemented with groups for family members (Ran 2002).

However, accessing such data to extract specific siblings' data, in spite of assistance from trial authors, proved challenging. The earlier trials screened for this review were conducted in the late 1980s and early 1990s (e.g. Kane 1990; Smith 1987; Solomon 1996) and the authors no longer had access to the dataset. Authors of some recent trials had similar problems in accessing their data due to changes in work affiliation. Only one study with data specific to nine siblings was included in the review (Chien 2007). The included study, nevertheless, was representative in terms of the group format used in delivering the psychoeducation undertaken in the community out-patient settings for family carers of people with schizophrenia.

Whilst some of the primary and secondary outcomes we had chosen for the review were reported by the included study, we are disappointed with the under-use of participant-oriented outcomes, such as: wellbeing, quality of life, satisfaction, family relationship and communication. In contrast, physician-oriented or psychosocial morbidity outcomes were more frequently used, such as: burden, service users' mental state, re-hospitalisation and inpatient stay. Economic data were largely unreported.

Quality of the evidence

All but one full-text article we screened in the review process were randomised controlled trials (RCTs) but the quality of the trials was variable. Most studies did not provide details about the randomisation process and assessor blinding making judgements of bias difficult. Some studies did not report follow-up outcome measures beyond the end of the intervention, rendering no usable data in the short, medium or long term. Such data reporting problems did not apply to the only study we included, as in contrast, our included study data were clearly reported. Please refer to Figure 2 for a graphic representation of the methodological quality of Chien 2007.

Potential biases in the review process

We believe the process of searching for studies was thorough. We followed the review protocol strictly in the processes of selecting studies for inclusion, data extraction and analysis. Due to the nature of the review, we contacted a large number of authors to try to clarify if siblings' data were collected and to seek subgroup or unpublished data specific to siblings. We were pleased to have received assistance from many authors although a limited amount of data were obtained due to the various reasons outlined above. Although we sought studies investigating effectiveness of psychoeducation for siblings, no (completed) study focused on siblings was identified. Siblings were included as family members- or carer-participants in studies and many studies reported both service users' and family members' outcomes; but more often than not the latter were not the primary outcomes.

Agreements and disagreements with other studies or reviews

Given the paucity of data included in this review, we are unable to draw any firm conclusions from our results, or compare our findings with those of other studies or reviews. An earlier Cochrane review on psychoeducation for people with schizophrenia (Xia 2011) identified psychoeducation as beneficial in improving compliance and reducing readmissions in service users with schizophrenia and participation by family members or carers in the interventions was regarded as pivotal. However, limited results on family carers' outcomes were reported by Xia 2011, in which service users' outcomes took priority. Similar conclusions were drawn by a systematic review conducted by the National Insititute of Clinical Excellence in the United Kingdom (NICE 2014). There are two recent systematic reviews focusing on psychoeducational interventions for family carers/members (Lobban 2013a; Sin 2013). Lobban 2013a reviewed 50 RCTs investigating family-based intervention targeting relatives of people with psychosis to investigate effectiveness of such interventions and to identify the key components of effective intervention packages. Psychoeducation was identified as a

common component in most interventions and 30 (60%) studies showed a statistically significant positive impact of the intervention on at least one relatives' outcome category. However, the authors came to the conclusion that the heterogeneity of study design and intervention content rendered meta-analysis inappropriate and there was no evidence to clearly define the key components of effective intervention (Lobban 2013a). Another review included 44 studies of psychoeducational interventions targeting family members directly (including siblings) (Sin 2013); the authors of this review also concluded that the data from these studies were too heterogeneous for a meta-analysis and provided a narrative report. Whilst psychoeducational interventions appeared to improve family members' knowledge, coping and perceived support, the collective data did not shed much light in its effectiveness on family members' wellbeing or quality of life. Siblings comprised around 3% to 30% of the family member-participants in about one-third of the included studies. Both these reviews hence urged for more work on siblings' outcomes (Lobban 2013a; Sin 2013). In terms of intervention design and modes of delivery, both reviews identified that psychoeducational intervention delivered as a multi-family group programme was more popular with family members than other delivery formats; this might also have an added benefit in enhancing peer support.

AUTHORS' CONCLUSIONS

Implications for practice

1. For siblings and people with severe mental illness

This review shows that siblings are under-represented in psychoeducational interventions provided for family members/carers. This is in spite of many service users having siblings and the key role siblings often play in supporting service users' recovery. Availability of interventions such as psychoeducation should be made known to the service users and their siblings, who should be encouraged to participate in such interventions.

2. For clinicians

Siblings' needs for information should be highlighted to clinicians, to promote provision of psychoeducation for siblings. Existing carers' services including psychoeducational interventions may have been focusing on a named family carer who takes up the key caregiving role, clinicians should endeavour to encourage siblings as well as other family members who may also be significant in supporting service users' recovery, to engage with psychoeducation. Whilst siblings could be identified by clinicians working in mental health services, a range of multi-disciplinary professionals working across primary care, social care and non-governmental organisations may also be well-placed to address siblings' needs and signpost them for interventions. Currently, there is limited evidence available to specify the best design and content of psychoeducational interventions for siblings; clinicians will have to draw on their clinical judgement and experience to consider the intervention content and design, as well as implementation strategies that enhance engagement of siblings. Evidence from the literature suggests that interventions with a group discussion and support feature seem particularly well-received.

3. For policy and decision makers

Considering the under-representation of siblings in family-based psychoeducation as identified in this review, policy and decision makers may wish to review the comprehensiveness of "family based" approaches to ensure intervention-provision extend beyond a single named carer of the family. Currently, most evidence from randomised controlled trials on psychoeducation is focused on key carers (e.g. parents) of people with long term SMI, policy makers lack specific evidence upon which to base decisions or guidelines for adapting psychoeducation for siblings. Funding bodies may wish to make this a priority for future research and intervention-provision in order to increase the uptake of psychoeducation by siblings.

Implications for research

In general, this review shows that there is a lack of studies targeting siblings. Despite a wealth of research evidence showing the effectiveness of psychoeducation in reducing relapse rates and improving compliance in service users, little is known about how these interventions impact on their family members' and siblings' outcomes. Such a gap of understanding is especially poignant when considering that family members' involvement in psychoeducation is common and regarded as essential in influencing service users' outcomes (NICE 2014; Sin 2013). Across the many studies, we found that included family members receiving psychoeducational interventions, siblings' data were collected but not available for this review. Given the changes in family structures and relationships in modern-day societies, future studies targeting family members of people with severe mental illness (SMI) should expand their reach beyond parents. More specifically, the optimum design and delivery of such interventions for siblings also needs further investigation in future studies. Some recent studies have started exploring innovative design and delivery modes using the internet and/or phone support to optimise the flexibility of psychoeducation for family members/carers (e.g. Lobban 2013; Rotondi 2011). These initiatives have the potential to engage siblings in future studies since they fit with the busy life-style of many people. We suggest an outline design for future trials focusing on interventions using online facilitation and delivery (Table 1).

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The Cochrane Schizophrenia Group Editorial Base in Nottingham produces and maintains standard text for use in the Methods section of their reviews. We have used this text as the basis of what appears here and adapted it as required.

The search terms were developed by the Trial Search Co-ordinator of the Cochrane Schizophrenia Group, Samantha Roberts and the contact author of this protocol.

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We are grateful for the assistance provided by the many authors of the RCTs screened for this review in providing us with further information and data.

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* *Indicates the major publication for the study*

CHARACTERISTICS OF STUDIES

Characteristics of included studies *[ordered by study ID]*

Chien 2007

Methods	Allocation: randomised, no further description Blindness: single blind (assessor blind) Duration: approximately 36 weeks within which 18 bi-weekly sessions of 2 hours each were delivered + 12 months follow-up Setting: community, Hong Kong, China
Participants	Diagnosis: Schizophrenia (diagnostic standard not stated) N = 9* 1. Siblings (9) amongst family members (84)**: 22 - 60 years, mean = 40.6 years, SD = 7.2 years** Sex: male (n = 28) and female (n = 56) 2. Service Users Age: 20 - 49, mean = 28.8 years, SD = 4.8 years Sex: male (n = 51) and female (n = 34) History: range of duration of the service users' illness: 1 - 7 years, mean = 3.6 years, SD = 1.8 years Excluded: if family member cared for more than one relative with a chronic mental or physical illness
Interventions	1. Psychoeducation group programme + standard care to family members and patients (attended the educational workshops only): 18 biweekly sessions over 4 stages - orientation and engagement, educational workshop, therapeutic family role and strength rebuilding and termination. Programme designed based on the work of McFarlane 2002 & Chien 2003. N = 4 siblings (out of 42 family members) 2. Standard care group (= routine psychiatric outpatient and family services only, consisted of monthly medical consultation, nursing advice, brief family education and counselling) N = 5 siblings (out of 42 family members)
Outcomes	1. Siblings'* (Family members') quality of life as measured by modified Family Assessment Device (FAD) with a reversed scoring scale*** 2. Siblings'* (Family members') coping: Family Burden Interview Schedule (FBIS) 3. Participants leaving the study early for any reason 4. Service users' general mental state: Brief Psychiatric Rating Scale (BPRS) 5. Service Users' specific aspect of quality of life i.e. social functioning: Specific Level of Functioning Scale (SLOF) 6. Service users' number of re-hospitalisation to a psychiatric inpatients unit over six months 7. Service users' inpatient bed occupancy in terms of hospital stay over 6 months
Notes	* Siblings out of 84 family member/carer-participants**, 4 in the active treatment group and 5 in the control group ** Family members were eligible to participate in this study if they were living with and

	caring for a relative with a primary diagnosis of schizophrenia *** Author contacted: advised that a validated translated FAD with a reversed scoring scale was used in the study

Risk of bias

Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	Unclear risk	Randomisation method not stated.
Allocation concealment (selection bias)	Unclear risk	Not stated.
Blinding of participants and personnel (performance bias) All outcomes	Unclear risk	The therapist could not be blinded in order to facilitate the group. Participants were not blinded
Blinding of outcome assessment (detection bias) All outcomes	Low risk	Single blind - One researcher who administered the pre and post-tests assessments was blinded to the allocation
Incomplete outcome data (attrition bias) All outcomes	Low risk	No incomplete data.
Selective reporting (reporting bias)	Low risk	All measured outcomes were reported.
Other bias	Low risk	None obvious.

Characteristics of excluded studies *[ordered by study ID]*

Study	Reason for exclusion
Bauml 1997	Allocation: randomised Participants: individuals with schizophrenia and their family carers Intervention: psychoeducation group vs standard care Outcomes: advice sought from corresponding author confirmed that sibling data were collected but were collapsed with other relatives who also included friends, children and others, no usable sibling-specific data
Cheng 2005	Allocation: randomised Participants: family carers of individuals with schizophrenia Intervention: psychoeducation group vs standard care Outcomes: advice sought from corresponding author confirmed that sibling-specific data were collected but no longer accessible

(*Continued*)

Chien 2010	Allocation: randomised Participants: patients with schizophrenia and their family carers Intervention: multi-modal psychoeducation with emphasis on care management
Dyck 2002	Allocation: randomised Participants: patients with schizophrenia and their family members Intervention: multi-family psychoeducation group vs standard care Outcomes: advice sought from lead author confirmed that sibling-specific data were collected but no longer accessible
Fiorillo 2011	Allocation: randomised Participants: individuals with schizophrenia and their family members Intervention: multi-family psychoeducation group vs standard care Outcomes: advice sought from lead author confirmed that sibling-specific data were collected but no longer accessible
Gutierrez-Maldonado 2009	Allocation: randomised Participants: family carers of individuals with schizophrenia Intervention: psychoeducation group vs standard care Outcomes: no usable data
Hogarty 1986	Allocation: randomised Participants: individuals with schizophrenia and their family members Intervention: family intervention including psychoeducation as an element vs standard care
Kane 1990	Allocation: randomised Participants: family members of hospitalised patients with long term schizophrenia Intervention: multi-family psychoeducation group vs standard care: Outcomes: advice sought from corresponding author confirmed that sibling-specific data were collected but no longer accessible
Lacruz 1999	Allocation: randomised Participants: individuals with schizophrenia and their key family carers Intervention: psychoeducation vs psychoeducation (in two different programme content) Outcomes: reported no family carers outcomes or data.
Lobban 2013	Allocation: randomised Participants: relatives of people with recent onset psychosis Intervention: psychoeducational supported self-management package (textual or online) vs standard care Outcomes: advice sought from the lead author that 3 siblings were involved in the study but data specific to siblings were not available
Magliano 2006	Allocation: randomised Participants: individuals with schizophrenia and their relatives Intervention: family intervention including psychoeducation as an element vs standard care

(*Continued*)

Motlova 2002	Allocation: randomised Participants: individuals with schizophrenia and their relatives Intervention: psychoeducation groups (weekly a hour programme run over 8 weeks) vs a one-day (8 hours) psychoeducation groups
Nasr 2009	Allocation: randomised Participants: individuals with schizophrenia and their key relatives Intervention: psychoeducation group vs. standard care Outcomes: data specific to siblings not available, authors not contactable
O'Callaghan 2009	Lead-author is deceased and no further detail on study participants (including siblings or not) could be found
Posner 1992	Allocation: randomised Participants: family members of individuals with schizophrenia Intervention: psychoeducational support group vs. waiting list Outcomes: no data specific to siblings could be sought, authors not contactable
Roncone 2000	Allocation: randomised Participants: individuals with schizophrenia and their relatives Intervention: individual family intervention versus multi-family intervention
Rotondi 2011	Allocation: randomised Participants: individuals with schizophrenia and their support person Intervention: online multi-family psychoeducation vs standard care Outcomes: no siblings were involved in the study
Schepp 2009	Allocation: randomised Participants: individuals with schizophrenia and their families Intervention: self-management intervention which included psychoeducation as an element vs standard care
Sharif 2012	Allocation: randomised Participants: individuals with schizophrenia and their key family carers, mostly mothers Intervention: psychoeducation group for family carers vs standard care Outcomes: no sibling-specific data were available.
Shinde 2005	Allocation: not stated Participants: individuals with schizophrenia and their families Intervention: psychoeducation Outcomes: no usable data
Smith 1987	Allocation: randomised Participants: family members of people with schizophrenia Intervention: psychoeducation group vs. postal information booklet Outcomes: advice was sought from the lead author to confirm that a small but unconfirmed number of siblings (<5 out of the total 40 carer-participants) were involved in the trial and their data were collected. However, the data were no longer accessible

(*Continued*)

So 2006	Allocation: randomised Participants: parents of people with schizophrenia, no siblings were involved in the trial
Solomon 1996	Allocation: randomised Participants: relatives of people with SMI Intervention: individualised consultation vs. psychoeducation group vs. standard care Outcomes: advice was sought from the lead author to confirm that 25 siblings participated in the trial but the data were no longer accessible
Weng 1994	Allocation: randomised Participants: individuals with schizophrenia and their families Intervention: psychoeducation group vs standard care Outcomes: no family members' outcomes or data were reported.
Ying 2006	Allocation: randomised Participants: individuals with schizophrenia and their family carers Intervention: educational family intervention vs. standard drug therapy Outcomes: published data do not specify if siblings were involved as participants. The authors were contacted and we were advised that they were unable to confirm whether any siblings were involved and that no unpublished data could be released

SMI: severe mental illness

Characteristics of studies awaiting assessment *[ordered by study ID]*

Chan 2009

Methods	Allocation: randomised
Participants	Adult patients aged between 18 to 62 diagnosed with schizophrenia DSM-IV within the previous 24 months and each with a family member who provided at least 4 hours caring per day (n = 73 patients and 73 family carers)
Interventions	1. Psychoeducation programme that included 10 weekly group sessions (both patients and their family carers attended together) (n = 36 dyad) 2. Standard care (n = 37 dyad)
Outcomes	1. Patients' attitude towards medication treatment as measured by the Rating of Medication Influences 2. Patients' mental state as measured by the Brief Psychiatric Rating Scale 3. Patients' insight into illness as measured by the Insight and Treatment Attitude Questionnaire 4. Family carers' burden of care as measured by the Family Burden Interview Schedule 5. Family carers' self-efficacy in coping with caring as measured by the General Perceived Self-Efficacy Scale 6. Family carers' perceived social support as measured by the Six-item Social Support Questionnaire
Notes	Out of the 73 family carer-participants, 29 were siblings of the patients. Sibling-specific data needed

Ran 2002

Methods	Allocation: cluster (by township) randomised
Participants	Adult patients diagnosed with schizophrenia ICD-10 and Chinese Classification and Diagnostic Criteria of Mental Disorder (CCMD-2-R) and their family carers
Interventions	1. Psychoeducational family intervention - 9 monthly sessions for the patient and his/her family members together, and multiple family workshops that were held once every 3 months (n = 132 dyad) 2. Drug treatment group that consisted of long-term injection of haloperidol decanoate (50-125 mg/month) and/or an oral medication (n = 110 dyad) 3. Standard care (n = 115 dyad)
Outcomes	1. Patients' medication compliance, as measured by therapists' dichotomous rating based on all available information 2. Patients' recognition of mental disease as measured by the General Psychiatric Interview Schedule and Summary Form 3. Relapse rate defined as either a change from a normal or no schizophrenic state to a state of schizophrenia defined by Present State Examination (PSE-9, Chinese translation)-derived criteria, or a marked worsening of schizophrenic symptoms 4. Patients' working ability and rate of mental disability as measured by the Social Disability Screening Schedule 5. Family carers' attitude towards the patient as measured by the Relatives' Beliefs Scale (Ran et al. 2001)
Notes	SIbling specific data needed as 17% of the family carer-participants were siblings

DSM IV: Diagnostic and Statistical Manual, fourth edition
ICD-10: International Classification of Diseases

Characteristics of ongoing studies *[ordered by study ID]*

Sin 2012

Trial name or title	The E Sibling Project - an online information and peer support resource
Methods	Exploratory randomised controlled trial
Participants	Siblings of individuals with first episode psychosis
Interventions	1. Online multi-component psychoeducational intervention 2. Online psychoeducational intervention 3. Online peer-support intervention 4. Control - online information-giving website
Outcomes	1. Siblings' mental wellbeing - Warwick-Edinburgh Mental Wellbeing Scale 2. Siblings' mental health knowledge - Mental Health Knowledge Schedule 3. Siblings' self-efficacy in coping as measured by the Assessment of Perceived General Self-Efficacy 4. Siblings' experiences of care-giving as measured by the Experience of Care-giving Inventory
Starting date	May 2013

Sin 2012 (*Continued*)

Contact information	http://www.controlled-trials.com/ISRCTN01416694/
Notes	Ongoing trial led by members of the review team, expected to complete by end of year 2015

DATA AND ANALYSES

Comparison 1. Psychoeducation versus standard Care

Outcome or subgroup title	No. of studies	No. of participants	Statistical method	Effect size
1 Siblings' quality of life - at 12 months (reversed FAD, high = good)	1	9	Mean Difference (IV, Random, 95% CI)	3.80 [-0.26, 7.86]
2 Siblings' coping (in terms of burden) - at 12 months (FBIS, high = poor)	1	9	Mean Difference (IV, Random, 95% CI)	-8.8 [-15.22, -2.38]
3 Leaving the study early for any reason - at 12 months	1	9	Risk Difference (M-H, Random, 95% CI)	0.0 [-0.34, 0.34]
4 Service users' general mental state - at 12 months (BPRS, high = poor)	1	9	Mean Difference (IV, Random, 95% CI)	-0.60 [-3.54, 2.34]
5 Service users' number of re-hospitalisation to a psychiatric inpatient unit over six months - at 12 months	1	9	Mean Difference (IV, Random, 95% CI)	-0.70 [-2.46, 1.06]
6 Service users' inpatient bed occupancy in terms of average days of hospital stay over 6 months - at 12 months	1	9	Mean Difference (IV, Random, 95% CI)	-2.60 [-6.34, 1.14]
7 Service users' specific aspect of quality of life, i.e. social functioning - at 12 months (SLOF, high = good)	1	9	Mean Difference (IV, Random, 95% CI)	21.40 [-2.71, 45.51]

Analysis 1.1. Comparison 1 Psychoeducation versus standard Care, Outcome 1 Siblings' quality of life - at 12 months (reversed FAD, high = good).

Review: Psychoeducation for siblings of people with severe mental illness

Comparison: 1 Psychoeducation versus standard Care

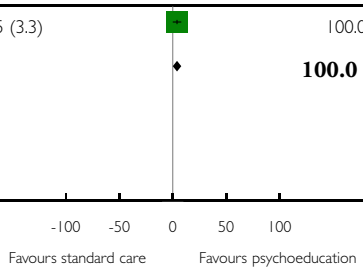
Outcome: 1 Siblings' quality of life - at 12 months (reversed FAD, high = good)

Study or subgroup	Psychoeducation		Standard care		Mean Difference	Weight	Mean Difference
	N	Mean(SD)	N	Mean(SD)	IV,Random,95% CI		IV,Random,95% CI
Chien 2007	4	16.3 (2.9)	5	12.5 (3.3)		100.0 %	3.80 [-0.26, 7.86]
Total (95% CI)	**4**		**5**			**100.0 %**	**3.80 [-0.26, 7.86]**

Heterogeneity: not applicable

Test for overall effect: Z = 1.84 (P = 0.066)

Test for subgroup differences: Not applicable



Analysis 1.2. Comparison 1 Psychoeducation versus standard Care, Outcome 2 Siblings' coping (in terms of burden) - at 12 months (FBIS, high = poor).

Review: Psychoeducation for siblings of people with severe mental illness

Comparison: 1 Psychoeducation versus standard Care

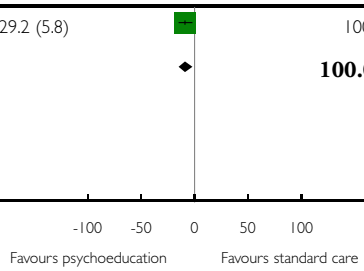
Outcome: 2 Siblings' coping (in terms of burden) - at 12 months (FBIS, high = poor)

Study or subgroup	Psychoeducation		Standard care		Mean Difference	Weight	Mean Difference
	N	Mean(SD)	N	Mean(SD)	IV,Random,95% CI		IV,Random,95% CI
Chien 2007	4	20.4 (4)	5	29.2 (5.8)		100.0 %	-8.80 [-15.22, -2.38]
Total (95% CI)	**4**		**5**			**100.0 %**	**-8.80 [-15.22, -2.38]**

Heterogeneity: not applicable

Test for overall effect: Z = 2.69 (P = 0.0072)

Test for subgroup differences: Not applicable

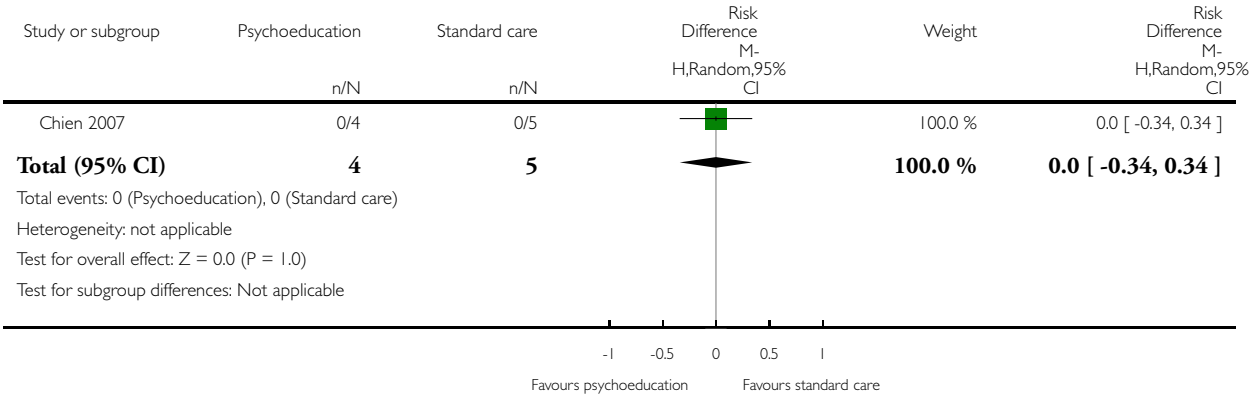


Analysis 1.3. Comparison 1 Psychoeducation versus standard Care, Outcome 3 Leaving the study early for any reason - at 12 months.

Review: Psychoeducation for siblings of people with severe mental illness

Comparison: 1 Psychoeducation versus standard Care

Outcome: 3 Leaving the study early for any reason - at 12 months

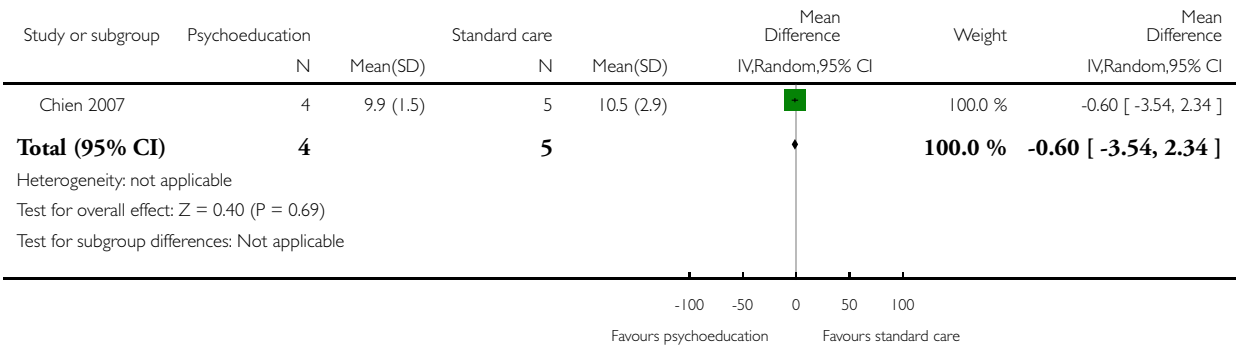


Analysis 1.4. Comparison 1 Psychoeducation versus standard Care, Outcome 4 Service users' general mental state - at 12 months (BPRS, high = poor).

Review: Psychoeducation for siblings of people with severe mental illness

Comparison: 1 Psychoeducation versus standard Care

Outcome: 4 Service users' general mental state - at 12 months (BPRS, high = poor)



Analysis 1.5. Comparison 1 Psychoeducation versus standard Care, Outcome 5 Service users' number of re-hospitalisation to a psychiatric inpatient unit over six months - at 12 months.

Review: Psychoeducation for siblings of people with severe mental illness

Comparison: 1 Psychoeducation versus standard Care

Outcome: 5 Service users' number of re-hospitalisation to a psychiatric inpatient unit over six months - at 12 months

Study or subgroup	Psychoeducation N	Mean(SD)	Standard care N	Mean(SD)	Mean Difference IV,Random,95% CI	Weight	Mean Difference IV,Random,95% CI
Chien 2007	4	2.4 (1.2)	5	3.1 (1.5)		100.0 %	-0.70 [-2.46, 1.06]
Total (95% CI)	**4**		**5**			**100.0 %**	**-0.70 [-2.46, 1.06]**
Heterogeneity: not applicable							
Test for overall effect: Z = 0.78 (P = 0.44)							
Test for subgroup differences: Not applicable							

Analysis 1.6. Comparison 1 Psychoeducation versus standard Care, Outcome 6 Service users' inpatient bed occupancy in terms of average days of hospital stay over 6 months - at 12 months.

Review: Psychoeducation for siblings of people with severe mental illness

Comparison: 1 Psychoeducation versus standard Care

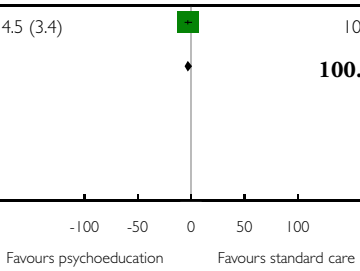
Outcome: 6 Service users' inpatient bed occupancy in terms of average days of hospital stay over 6 months - at 12 months

Study or subgroup	Psychoeducation N	Mean(SD)	Standard care N	Mean(SD)	Mean Difference IV,Random,95% CI	Weight	Mean Difference IV,Random,95% CI
Chien 2007	4	11.9 (2.3)	5	14.5 (3.4)		100.0 %	-2.60 [-6.34, 1.14]
Total (95% CI)	**4**		**5**			**100.0 %**	**-2.60 [-6.34, 1.14]**

Heterogeneity: not applicable

Test for overall effect: Z = 1.36 (P = 0.17)

Test for subgroup differences: Not applicable



Analysis 1.7. Comparison 1 Psychoeducation versus standard Care, Outcome 7 Service users' specific aspect of quality of life, i.e. social functioning - at 12 months (SLOF, high = good).

Review: Psychoeducation for siblings of people with severe mental illness

Comparison: 1 Psychoeducation versus standard Care

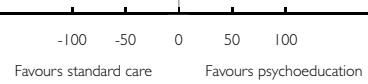
Outcome: 7 Service users' specific aspect of quality of life, i.e. social functioning - at 12 months (SLOF, high = good)

Study or subgroup	Psychoeducation N	Mean(SD)	Standard care N	Mean(SD)	Mean Difference IV,Random,95% CI	Weight	Mean Difference IV,Random,95% CI
Chien 2007	4	149.5 (16.1)	5	128.1 (20.8)		100.0 %	21.40 [-2.71, 45.51]
Total (95% CI)	**4**		**5**			**100.0 %**	**21.40 [-2.71, 45.51]**

Heterogeneity: not applicable

Test for overall effect: Z = 1.74 (P = 0.082)

Test for subgroup differences: Not applicable



ADDITIONAL TABLES

Table 1. Suggested design of future studies

Methods	Allocation: randomised, full explicit description of methods of randomisation and allocation concealment Blinding: single, tested. Setting: community rather than hospital. Duration: 10-18 weeks intervention, and then follow-up to at least one year
Participants	Diagnosis: Siblings (full or otherwise) of individuals with schizophrenia or psychosis (ICD) N = 300* Age: adolescents and adults Sex: both
Interventions	1. Psychoeducation intervention (using online delivery and incorporating a virtual group element), n =150 2. Standard care, n = 150
Outcomes	Siblings' psychosocial wellbeing and quality of life. Siblings' knowledge, coping (with caregiving), perceived social support and use of services Service users' mental state using standardised measures such as BPRS and PANSS, global state, social functioning and quality of life Adverse events: any adverse event recorded Economic outcomes
Notes	*Powered to be able to identify a difference of 20% between groups for primary outcome with adequate degree of certainty

ICD: International Classification of Diseases
BPRS: Brief Psychiatric Rating Scale
PANSS: Positive and Negative Syndrome Scale

CONTRIBUTIONS OF AUTHORS

Jacqueline Sin (JS): protocol development, abstract screening, paper screening, data extraction, analysis and interpretation of data, and writing and reviewing the final report.

Cheryl D. Jordan (CJ): protocol development, abstract screening, paper screening, data extraction, and reviewing the final report.

Elizabeth A. Barley (EB): protocol development, commenting on drafts, checking abstract and paper screening, checking data extraction and analysis, and reviewing the final report.

Claire Henderson (CH): protocol development, commenting on drafts, and reviewing the final report.

Ian Norman (IN): protocol development, commenting on drafts, checking abstract and paper screening, checking data extraction and analysis, and reviewing the final report.

DECLARATIONS OF INTEREST

Authors JS, CH and IN are working on a randomised controlled trial on developing and evaluating the preliminary efficacy of online psychoeducational intervention for siblings of individuals with first episode psychosis, which is expected to complete by end of 2015.

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Internal sources

- King's College London, UK.

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- National Institute for Health Research, UK.

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DIFFERENCES BETWEEN PROTOCOL AND REVIEW

Not applicable.

INDEX TERMS

Medical Subject Headings (MeSH)

*Mental Disorders; Adaptation, Psychological; Caregivers [*education; psychology]; Mental Health [*education]; Quality of Life; Randomized Controlled Trials as Topic; Siblings [*psychology]

MeSH check words

Adolescent; Adult; Humans; Young Adult