The Mental Health and Wellbeing of Siblings of Children with Mental Health Problems: Two Decades of Research

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TABLE OF CONTENTS

Summary

Childhood mental health problems (MHPs) can have a significant long-term impact on the lives of children and on the systems to which the child belongs, including family and school. For example, it is widely accepted that there is a bi-directional, interactional relationship between family functioning and childhood mental health, and extensive research has explored this relationship (Parritz & Troy, 2011). However, the role of siblings has largely been ignored.

Sibling relationships have been identified as making significant contributions to our psychological wellbeing (Dunn, 1983; Fagan & Najman, 2003). For example, sibling relationship problems (e.g. conflict) have been linked to negative attributes such as depressive symptoms and decreased social competence (Milevsky, 2005). Furthermore, existing theoretical frameworks, such as developmental psychopathology (Parritz & Troy, 2011) and impact of illness frameworks (Wallander & Varni, 1992), suggest that siblings of children with MHPs would have an increased risk of MHPs and poorer wellbeing. Yet, little research has been conducted with this population. This dissertation describes an attempt to address this gap and advance our understanding of the mental health and wellbeing of siblings of children with MHPs.

Papers 1 to 4 report on a 20-year systematic review of the existing literature with each study reporting on a different aspect of the mental health and wellbeing of siblings: The prevalence of psychopathology from a categorical and dimensional perspective, the quality of family relationships, and the experiences and coping strategies of siblings. The reviews of the quantitative literature suggest that siblings of children with MHPs are at greater risk of MHPs than control children. Developmental psychopathology risk factors, such as parental psychopathology, were implicated as

predictors of the mental health of siblings. However, the data were not conclusive due to significant methodological limitations in the literature. The reviews of family relationships and qualitative literature described a significant negative impact on all areas of the sibling's life, including relationships and daily routine. These reviews supported impact of illness frameworks as having a role in the mental health of siblings of children with MHPs.

Papers 5 and 6 report on primary research aimed at exploring the mental health and treatment utilisation of siblings of children with MHPs within a clinical population. The key findings were that these siblings were almost four times more likely to have MHPs compared to the general population and had high rates of treatment utilisation. Furthermore, birth order and age difference were related to MHPs in siblings. These findings have important implications for clinical practice and assessment.

The present dissertation argues that although limited by methodological issues, the literature strongly suggests that siblings of children with MHPs are a high-risk group for poorer mental health and wellbeing. This dissertation highlights the role of multiple theories, including developmental psychopathology and impact of illness frameworks, in the mental health of siblings. Methodological guidelines and potential treatment and prevention strategies are outlined. This dissertation has contributed significantly to our understanding of siblings of children with MHPs with important implications for both clinical practice and research.

Thesis Declaration

I, Nylanda Ma, certify that this work contains no material which has been accepted for the award of any other degree or diploma in my name, in any university or other tertiary institution and, to the best of my knowledge and belief, contains no material previously published or written by another person, except where due reference has been made in the text. In addition, I certify that no part of this work will, in the future, be used in a submission in my name, for any other degree or diploma in any university or other tertiary institution without the prior approval of the University of Adelaide and where applicable, any partner institution responsible for the joint-award of this degree.

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Chapter 3: Paper One

Ma, N., Roberts, R., Winefield, H., & Furber, G. (2014). The prevalence of psychopathology in siblings of children with mental health problems: A 20-year systematic review. *Child Psychiatry and Human Development.* Advance online publication. doi: 10.1007/s10578-014-0459-1

Chapter 6: Paper Four

Ma, N., Roberts, R., Winefield, H., & Furber, G. (in press). The utility of qualitative metasynthesis: Advancing knowledge on the wellbeing and needs of siblings of children with mental health problems. *Qualitative Psychology*. (Accepted July 17, 2014).

Nylanda LT Ma

Signed: _____ Date: 4 / 8 / 2014

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me to produce the best research possible in the hopes of making their struggle a little bit easier. I am extremely fortunate that I was able to spend the past few years advocating for a group and cause that I am so passionate about. I hope that this thesis will give siblings a voice and encourage those around them to listen.

Dedication

For Rachael and Nirvana.

Chapter 1. Introduction

1.1 Overview of Chapter

The focus of the present thesis is to explore the wellbeing and needs of siblings of children with mental health problems (MHPs). This chapter situates the topic within the existing literature from related areas, and outlines the rationale for the present thesis. First, definitions, prevalence, and consequences of childhood MHPs are outlined. Second, an argument for the need for sibling-based research is presented. Third, four key areas of related research are discussed. Finally, the rationale and aims of this thesis are outlined. The references for this section, the preamble chapters, and the conclusion are presented separately at the end of the thesis document. The individual papers are presented as isolated pieces of work written in publication format. As such, references for individual papers are contained within each chapter and are in the format required by the journal in which the article is published or submitted. Similarly, tables and figures are numbered consecutively within each paper individually.

1.2 Mental Health Problems in Children

1.2.1 Definitions of mental health problems and terminology.

MHPs in childhood, where a child is defined as an individual 18 years of age or younger (United Nations, 1989), can be broadly defined as “alterations in thought, mood, or behaviour that are associated with distress or impaired functioning” (Sawyer et al., 2000, p.3). While this definition appears reasonably simple and clear, difficulties arise when attempting to distinguish between a child who would be perceived as having MHPs and one who is not (Farrell, 1994). The difficulty is that there are no clear cut-off

points between ‘abnormal’ and ‘normal’ (Farrell, 1994). What is considered ‘normal’ may vary from setting to setting, society to society, and from culture to culture (Farrell, 1994).

Bearing these caveats in mind, there have been several attempts to define MHPs in research and practice. Each of these methods however, has several limitations. One of the most common methods is the application of diagnostic criteria. A child’s difficulties or symptoms can be compared to pre-determined diagnostic criteria for specific MHPs, such as that used in the *Diagnostic and Statistical Manual and Mental Disorders* (5th edition, American Psychiatric Association [APA], 2013). If the diagnostic criteria are met (e.g. the number and nature of symptoms and level of impairment reach a set cut-off), the child is then classified as having this particular MHP. This method has several criticisms including a lack of grounding in aetiological theories, limits in the amount of useful clinical information obtained (e.g. two individuals with the same diagnosis may have very different clinical presentations), and high rates of comorbidity (Helzer et al., 2008). Another common method is the use of standardised measures, such as the Child Behaviour Checklist (Achenbach & Rescorla, 2001), where norms are based on the range typically reported for children attending mental health clinics and for non-referred children (Achenbach, 1991). Thus, if a child scores in the former ‘clinical range’, they are considered to have significant MHPs (Achenbach, 1991). However, not all children experiencing significant MHPs will attend mental health clinics and those that are may not necessarily be attending for MHPs but for other issues, such as family problems (Achenbach, 1991). Thus, scoring within the clinical or normal range, as defined by scores typically seen in clients of mental health services, may not necessarily be indicative of the presence or absence of MHPs in a child. It is beyond the scope of this thesis to discuss diagnostic issues at

length. Suffice to say, there are no clear, universally agreed upon conventions to define the presence or absence of MHPs in children.

In the studies presented in this thesis, I chose to use an inclusive definition wherein multiple methods of defining MHPs were used. Given that there is very little research on siblings of children with MHPs, it was appropriate to use a more inclusive definition. I also chose to use a broad category of MHPs rather than diagnosis specific groupings. That is, I focused on siblings of children with a range of MHPs rather than siblings of children with a particular diagnosis. Children presenting to mental health clinics do not necessarily display symptoms or difficulties that fit neatly within a specific diagnostic criteria (Yeh & Weisz, 2001). They may also present with complex comorbidities or difficulties primarily resulting from adverse life events rather than underlying psychopathology (Bird, Gould, & Staghezza, 1993; Verhulst & van der Ende, 1997). Thus, it is useful to provide clinicians and parents with information about the mental health and wellbeing of siblings of children with a range of MHPs rather than diagnosis specific information. There are also few empirical justifications for exploring the siblings of children with particular diagnoses as separate groups. As can be seen in the papers included in this thesis, the research has been inconsistent as to whether there are differences in the psychological outcomes for siblings based on the diagnosis of the target child or index child. Thus, in this thesis, I have focused on siblings of target children with a range of MHPs.

I formulated several criteria based on past research (e.g. Ravens-Sieberer et al., 2008) according to which target children would be classified as having MHPs: a) presence of a prior established psychiatric diagnosis, b) positive screen on diagnostic measures, c) clinically elevated scores on standardised psychological measures, d) referral to, or engagement in mental health treatment, and e) adjudicated in court for

delinquent behaviour. Adjudication for delinquent behaviour indicates that the Juvenile Court has declared that a minor is delinquent. That is, the court has determined that a minor has violated or has attempted to violate laws of that state or country (Joint Committee on Administrative Rules, 2002). This provides compelling legal evidence of MHPs, such as but not limited to conduct problems and substance abuse problems. The focus of this thesis was siblings of target children with primarily mental health related issues – that is, primarily emotional, social, or behavioural problems. Thus, difficulties classified as primarily cognitive or neurological (e.g. intellectual disability), neurodevelopmental (e.g. autism), or physical (e.g. cancer) were not included.

Using the above criteria and definitions, a problem of circularity arises when attempting to define the child with MHPs and their siblings. If multiple children in the family are displaying MHPs, how do we define which child is the 'target child with MHPs' and which is the 'sibling'? Both could equally be classified as a child with MHPs. It is generally assumed or thought that the target child is the first child in the family to develop MHPs. However, no studies included in this thesis have been able to establish temporal links and determine which child was first to develop MHPs. Without these data, there are no clear logical criteria that could be used to distinguish between the child with MHPs and their siblings. For the same reason, no causal links from the data in this thesis can be drawn. If no temporal links can be determined, by definition, we cannot state that MHPs in one child have caused MHPs in their sibling. It may be that both children in the family develop MHPs at similar times in response to a shared family adversity, such as parental death. Alternatively, it may be that one child develops MHPs first and does indeed contribute causally to the development of MHPs in their siblings. There may also be temporal links but no causal links. That is, one child may develop MHPs before their siblings but this does not then mean that this child

caused their sibling to develop MHPs. There are also several issues related to diagnosis and treatment seeking, and not related to casual mechanisms, that may affect which child is first recognised as having MHPs. For example, boys are more likely to be referred for mental health treatment than girls (Green, Clopton, & Pope, 1996). This could lead to boys being labelled the first child in the family to develop MHPs, yet their siblings, especially girls, may have had an earlier onset of symptoms but did not receive clinical attention and recognition. Thus, there are several issues with temporality and causality in this literature. As yet, the data is unable to shed any light on this issue.

In this thesis, I have decided to retain the definitions used in the existing sibling literature and have defined the 'target child' as the child who is the primary target of research or treatment and the 'target sibling' as the brothers or sisters of this child. I recognise that these are somewhat arbitrary definitions and are an important limitation of all of the work in this field. Thus, while interpreting the findings of this thesis, it is important to remember that no causal links or temporal links between the target child's MHPs and the mental health of the sibling can be inferred.

The emotional, behavioural, and/or social outcomes measured in siblings are described in several different ways. These correspond to the particular aspect or perspective that is being discussed. For example, when discussing psychopathology, I use the term 'psychopathology' or 'MHPs'. When discussing the collective outcomes for siblings, that include the outcomes across a number of these areas, I use the phrase 'mental health and wellbeing'. This is the preferred label used in the context of mental health in Australia as reflected in governmental policies (Department of Health, Government of South Australia, 2010) and national initiatives (e.g. National Survey of Mental Health and Wellbeing; Australian Bureau of Statistics, 2008). It is a phrase that allows greater recognition of different cultural understandings of mental health, reflects

a more holistic view of health, and acknowledges that health is more than the absence of illness (Australian Institute of Health and Welfare, 2009).

1.2.2 Impact of mental health problems.

Definitions and terminology aside, childhood MHPs are a significant health concern worldwide. In children aged 4-17 years, 10-14% of children display clinically significant MHPs (Green, McGinnity, Meltzer, Ford, & Goodman, 2005; Sawyer et al., 2000). Focusing on adolescents aged 13-18 years only, this number increases to 27% (Merikangas et al., 2010). These numbers are particularly alarming given the widespread detrimental effect that MHPs can have on the lives of children. For example, children with MHPs have lower health-related quality of life, lower self-esteem, and greater limitations in school and peer activities compared to children without MHPs (Sawyer et al., 2000). Furthermore, MHPs in children can impact on the many systems to which a child belongs, such as family and school. For example, families of children with MHPs can experience significant financial and employment difficulties, social isolation, family relationship breakdowns (e.g. marital conflict) and tension with school staff due to the child's behaviour (Rosenzweig, Brennan, & Ogilvie, 2002). It is not surprising then that considerable resources have been devoted to research on the aetiology of MHPs in children, on specific child populations at risk for MHPs, and on prevention and treatment strategies. However, the majority of this research has focused on child-parent relations. For example, research has extensively explored the relationship between parenting style and MHPs in children and the effect of parental psychopathology on the mental health of children (Parritz & Troy, 2011). Siblings, on the other hand, tend to be overlooked and neglected.

1.3 The Importance of Siblings

There are several reasons why it is important to consider siblings in research on childhood MHPs. These will be outlined in the following sections.

1.3.1 Siblinghood is unique.

Siblinghood is a unique relationship dynamic that is different from all others (Cicirelli, 1982). Unlike relationships with peers, sibling relationships are lifelong and siblings are likely to spend greater amounts of time together than peers, particularly in childhood. Therefore, sibling relationships are more likely to influence behaviour through ongoing, long-term modelling and reinforcement of behaviours (Dunn, 1983; Fagan & Najman, 2003). Furthermore, unlike parents, siblings are closer in age and have similar levels of control or authority. That is, parents are authority figures for children and hold a greater amount of power in the relationship than vice versa. On the other hand, there is less of a gap in the power differential in sibling relationships and siblings are likely to have a more similar degree of control over one another than that seen in parent-child relationships (Cicirelli, 1982). As such, sibling relationships may have attributes not seen in parent-child relationships. For example, siblings might collude to undermine authority and reinforce unhealthy behaviours, such as substance abuse (Bullock & Dishion, 2002). This is considerably less likely to occur in parent-child relationships. In addition, sibling relationships often endure over a longer period of time, beyond the death of parents. Thus, it is important to explore siblings and siblinghood as it is different from any other dynamic and may provide a unique perspective on mental health in childhood and into adulthood.

1.3.2 Sibling relationships contribute to our mental health and wellbeing.

Sibling relationships, in childhood and adolescence, have been identified as making significant contributions to our psychological wellbeing and mental health. Positive attributes of sibling relationships, such as warmth and closeness, have been found to be associated with positive outcomes in children and adolescents. These include increases in prosocial behaviours, higher self-esteem, and higher life satisfaction (Buist, 2010; Dunn, 1983; Milevsky, 2005). Furthermore, positive sibling relationships have been found to compensate for low parental and peer support and to act as a buffer against risk factors and adverse life events (Kempton, Armistead, Wierson, & Forehand, 1991; Milevsky, 2005; Milevsky & Levitt, 2005). Conversely, sibling relationship problems, such as conflict or aggression, have been linked to negative outcomes in childhood and adolescence. These include depressive symptoms, increased aggression, decreased social competence, and increased loneliness (Buist, 2010; Fagan & Najman, 2003; Milevsky, 2005). Feinberg, Solmeyer, and McHale (2012) provide an excellent summary of the empirical findings in this field relating to mental health in childhood and adolescence.

Sibling relationship problems in childhood and adolescence have also been found to significantly predict adult outcomes. For example, sibling violence in adolescence significantly predicts dating violence (either as a victim or perpetrator) in adulthood (Noland, Liller, McDermott, Coulter, & Seraphine, 2004). In addition, childhood sibling relationships influence adult psychological functioning independent of parent-child relationships. For example, childhood sibling relationships significantly predicted major depression in adulthood (20-50 years of age) even after adjustments for parent-child relationship quality (Waldinger, Vaillant, & Orav, 2007). Parent-child relationship quality did not predict major depression in adulthood when a family history

of depression and sibling relationship quality were taken into account (Waldinger et al., 2007). Therefore, siblings and sibling relationships should be explored as they have short-term and long-term effects on mental health, interpersonal behaviours, social functioning, and psychological wellbeing.

1.3.3 Siblings share risk factors for psychopathology.

Several theoretical frameworks, as discussed in detail in later sections, suggest that siblings of children with MHPs are exposed to several risk factors for psychopathology or MHPs. Developmental psychopathology frameworks, for example, propose that psychopathology is developed in the context of interactions between genetic, family environment, and non-shared or individual risk factors (Parritz & Troy, 2011). Brothers and sisters have similar genetic backgrounds and may be raised in similar family environments. For example, half the genes of full brothers and sisters, sharing the same birth mother and father, will (on average) be identical (Griffiths, Miller, Suzuki, Lewontin, & Gelbart, 2000). Because of this, siblings of children with MHPs likely share genetic and/or family environment risk factors with the child with MHPs and are likely to have an increased risk of psychopathology. Research with adult siblings of individuals with MHPs bares this out. Siblings of individuals with schizophrenia, for example, have been found to have higher rates of substance use disorders and increased vulnerability to developing schizophrenia when compared to siblings of controls (Smith, Barch, Wolf, Mamah, & Csernansky, 2008). Thus, theoretical frameworks suggest that siblings of children with MHPs may be a high-risk group for developing psychopathology and mental health difficulties.

Sibling research may also contribute to the development and continued refinement of such theoretical frameworks. Research with siblings may highlight

sibling-specific aetiological pathways and risk factors, such as sibling conflict, that contribute to the development of MHPs. Such research could significantly contribute to our understanding of psychopathology and inform developmental psychopathology and aetiological theories.

1.4 Contributions from Related Fields

As highlighted in the previous section, it is important and valuable to conduct research with siblings within the context of mental health. Although little research has been conducted with siblings of children with MHPs, findings from four related fields contribute to our understanding of these siblings. The key findings from these areas and how they relate to siblings of children with MHPs are discussed in the following section. Similarly, no known comprehensive theoretical model has been developed that proposes risk factors and pathways specifically relating to the mental health and wellbeing of siblings of children with MHPs. Models developed in related fields however, may highlight pathways that may also play a role in the mental health and wellbeing of siblings and provide a foundation for the future development of a model specifically for siblings of children with MHPs. These are also outlined in the following sections.

1.4.1 Normative or general population sibling literature.

Sibling relationship research with the general population has shown a significant association between the functioning of the relationship and the mental health of children. Drawing on this literature, three key sibling models have been developed to illuminate the pathways by which siblings and sibling relationships may influence mental health and wellbeing.

1.4.1.1 Brody's (1998) family experience and sibling relationship model.

Brody (1998) developed a heuristic model of mediators between family experiences and sibling relationship quality, as depicted in Figure 1. Past research has shown a strong empirical link between sibling relationships and mental health. Thus, a model that can inform our understanding of factors that influence sibling relationship quality can broaden our understanding of how these relationships might impact mental health and potential prevention or intervention strategies. Brody suggested that sibling relationships influence and are influenced by three key family experiences: Parent-child relationship, differential parental treatment (i.e. parental favouritism), and parental management of sibling conflict. The relationship between these family experiences and sibling relationship quality is mediated by four key factors: Interpersonal behaviour patterns, emotion regulation and coping styles (or a sense of security and safety), attributions for relational events, and internalisation or rejection of norms governing aggression and fairness.

As an example to illustrate these concepts, whether parents intervene or do not intervene into escalating sibling conflict can impact on sibling relationship quality and is mediated by several pathways. If parents intervene, this can help their children learn and develop prosocial behaviours, such as effective conflict resolution, which can improve sibling relationship quality. Further, parental intervention into sibling conflict can build a sense of security and safety for children (i.e. parents are there to help and protect them). A sense of security and safety facilitates healthy attachments and can improve the quality of sibling relationships (Brody, 1998).

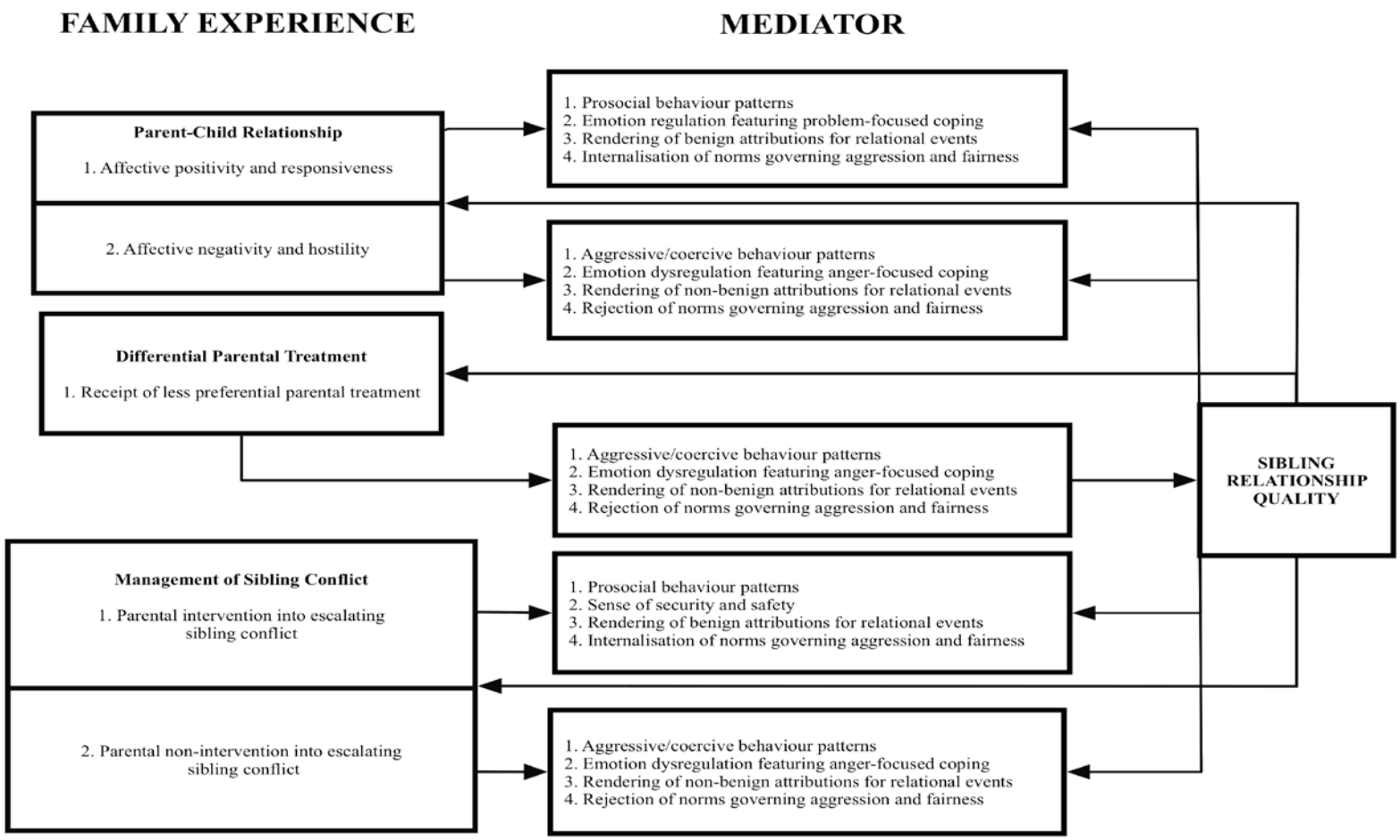

Figure 1. Brody's (1998) family experience and sibling relationship model. Adapted from "Sibling relationship quality: Its causes and consequences," by G.H. Brody, *Annual Review Psychology, 49,* p.11. Copyright 1998 by Annual Reviews Inc. Adapted with permission.

Brody's (1998) model was developed based on the empirical and theoretical literature available at the time. More recent empirical findings also support the individual links in Brody's model. For example, sibling relationship quality is associated with children's emotional and behavioural regulation or dysregulation (Volling, McElwain, & Miller, 2002). However, to the best of my knowledge, no studies have conducted meditational analyses to examine the complete pathways proposed by Brody. It should also be noted that Brody's model focuses on parent-child family processes and their link to sibling relationship quality. There is a range of other family processes, such as marital relationship quality, that significantly influence sibling relationship quality (Volling et al., 2002). These processes may indirectly influence sibling relationship quality through the key family processes already highlighted in Brody's model. For example, marital relationship quality is significantly associated with parent-child relationship quality (Erel & Burman, 1995). They may then function as an extension to Brody's model, rather than a missing element. However, they may also have a direct influence on sibling relationship quality or on the mediators. For example, marital relationship quality is significantly associated with emotional regulation in children (Volling et al., 2002). Thus, while Brody's model contributes to our understanding of the link between family processes and sibling relationship quality, a more comprehensive and inclusive model is needed.

1.4.1.2 Feinberg and colleagues' (2013) model of pathways from sibling relationship to adjustment problems.

The second important model in this area (Figure 2) incorporates empirical findings and pre-existing theories to highlight the pathways from sibling relationships to the development of MHPs (Feinberg, Solmeyer, Hostetler, Sakuma, Jones, &

McHale, 2013). The model consists of four pathways. In the first pathway, sibling relationships provide a developmental context that may lead to the development of a generalised coercive interpersonal style. If a sibling gives into the demands of the other, in response to coercive behaviours or negative behaviours, this interpersonal style is reinforced. The interpersonal style can generalise to other relationships and settings and lead to difficulties with peers, teachers, and school bonding. Difficulties in these areas can lead to depressive symptoms, associating with deviant peers, and externalising or risky behaviours. In the second pathway, siblings can mutually reinforce deviant behaviours, expose each other to risky situations and behaviours (e.g. substance use), encourage deviant peer associations, and can collude to undermine authority figures. These processes, termed sibling deviancy training, and originally put forward by Patterson (1984), can increase externalising or risky behaviour. In the third pathway, sibling relationship problems, such as conflict and low support, contribute directly to depressive symptoms. In the fourth pathway, sibling relationship problems impact on parenting styles. For example, ongoing sibling conflict and violence may increase parental stress. Parental stress may in turn lead to reduced parental capacity and the use of unhelpful parenting behaviours, such as unfair treatment, diminished monitoring, and parent-child conflict. Impaired parenting can lead to depressive symptoms and externalising behaviour problems and may increase sibling deviance training processes (e.g. diminished monitoring may increase opportunities to engage in deviant activities and associate with deviant peers).

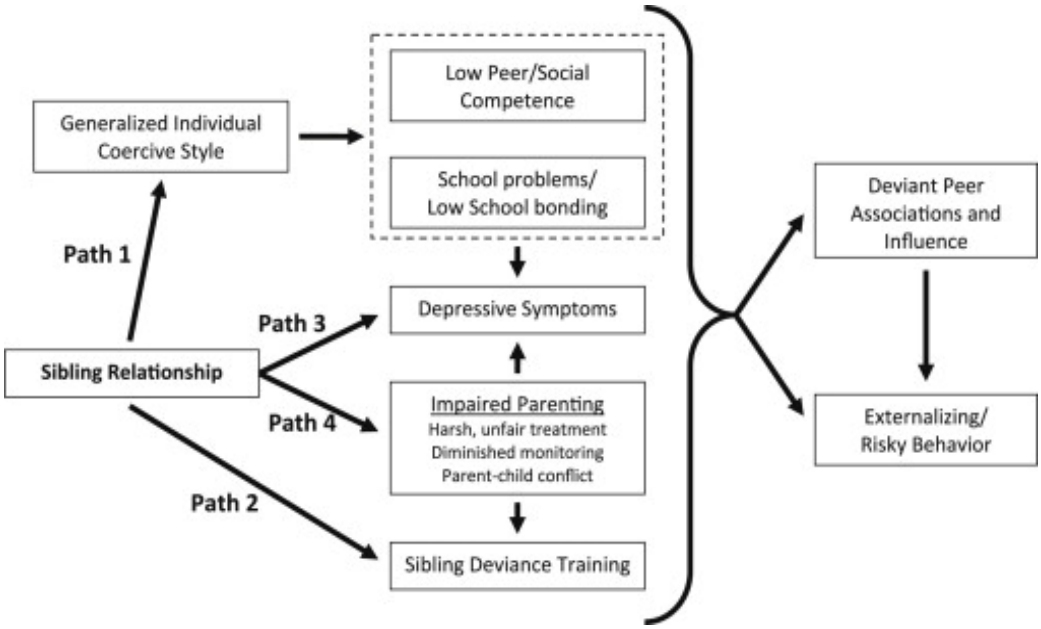

*Figure 2***.** Feinberg and colleagues' (2013) model of pathways from sibling relationship to adjustment problems. Reprinted from "Siblings are special: Initial test of a new approach for preventing youth behaviour problems" by M.E. Feinberg et al., *Journal of Adolescent Health, 53,* p. 167. Copyright 2013 by Society for Adolescent Health and Medicine. Reprinted with permission.

Each of these pathways has been extensively explored in the existing empirical literature (see Feinberg et al., 2012; 2013 for a summary). For example, sibling deviance training processes have been shown to account for 33.8% of variance in child behavioural problems (Bullock & Dishion, 2002). While this model has highlighted several sibling-specific pathways to the development of MHPs, it was designed as a universal model that would apply to the general population. As such, it does not include factors that are likely to influence the mental health of siblings of children with MHPs, such as genetic factors and impact of illness factors (see section 1.4.3). For example, 'illnesses' in children can have a detrimental impact on the mental health and wellbeing of family members and may contribute to the wellbeing of siblings. Furthermore, this model is unidirectional, looking at the pathways from sibling relationships to MHPs. As

a result, this model does not include factors that influence sibling relationship quality and does not include bi-directional relationships and feedback loops. For example, impaired parenting may be influenced by sibling relationship problems but impaired parenting can also lead to sibling relationship problems (see Brody's model in section 1.4.1.1). The model also does not account for anxiety symptoms. Sibling relationship quality has been shown to predict anxiety problems (Dunn, Slomkowski, Beardsall, & Rende, 1994), but this is not included in the model. Last, the model focuses on adjustment problems and does not consider protective effects of sibling relationships. Therefore, while it is arguably the most comprehensive sibling-focused model in the context of mental health, it does not capture all of the factors that are likely to influence the mental health and wellbeing of siblings of children with MHPs.

1.4.1.3 Birth order theory.

The third sibling-focused theoretical framework focuses on the effect of birth order on the mental health and psychological functioning of siblings. The 'sibling position' or birth order theory, originally proposed by Bowen (1978), holds that birth order is linked to fixed personality traits or profiles. As such, based on birth order information alone, an individual's personality profile can be accurately predicted (Bowen, 1978). Although the specific details of Bowen's theory have not been supported by the empirical literature, research has confirmed an association between personality traits and birth order, even after controlling for number of siblings in the family (Eckstein, 2000; Miller, Anderson, & Keala, 2004; Sulloway, 1995). The theory has since been extended to include a range of psychological, cognitive, achievement, and health outcomes. For example, first-born children have been found to have significantly less trait anxiety than third-born children and higher self-esteem than

second-born and youngest children (Gates, Lineberger, Crockett, & Hubbard, 1988). Birth order has also been found to relate to health outcomes. For example, later-born children have significantly lower odds of having allergy-related health problems compared to first-born children, independent of the effect of sibship size (Bernsen, de Jongste, & van der Wouden, 2003). Extensive research has also explored the role of birth order in intelligence and cognitive development, based on observations of enhanced cognitive development in later-born children (Kristensen & Bjerkedal, 2007). Birth order may also play a role in impact of illness pathways in families of children with disabilities (Breslau, 1982), which are discussed in detail in later sections. It has been hypothesised, for example, that siblings born before the 'affected' child may have spent their critical development years in a more 'normal' family environment (Breslau, 1982). This may lead to fewer adjustment problems in older siblings. At the same time, older siblings may be required to engage in more caretaking activities, which may have a significant impact on their mental health and wellbeing.

There are multiple criticisms of this work with most arguing that birth order research fails to account for gender differences, age-spacing, number of siblings, and the effects on the environment (Miller et al., 2004). For example, in the case of cognitive development, it may not be birth order relations per se that predict cognitive development, but that additional children in the family increase the environmental stimulation for learning and provide another source of modelling (Kristensen & Bjerkdal, 2007). Further, age-spacing, or the age difference between siblings, may mediate the effect of birth order (Breslau, 1982). Yet, age difference is rarely accounted for or considered when testing birth order effects (Miller et al., 2004). Findings have also been inconsistent across studies regarding the predictive value of birth order (Miller et al., 2004). Despite the limitations, birth order theory highlights the role of

sibling relations and dynamics and the effect these may have on family environments and psychological outcomes. It points to another factor that may be important to consider in sibling research.

While these three sets of theories and frameworks significantly advance our understanding of sibling-based processes and their relationship to mental health, they are normative models designed for the general population. They do not specifically apply to siblings of children with MHPs and do not account for the range of factors that may influence the mental health of this group of siblings, such as genetic risk factors. However, they provide a useful theoretical foundation that research with siblings of children with MHPs could potentially build upon.

1.4.2 Twin studies.

Arguably, the most significant advances in our understanding of the aetiology of MHPs have come from a special class of sibling studies – twin studies. In full siblings, there are three different classes of siblings with varying degrees of genetic similarity: Monozygotic twins, dizygotic twins, and singleton or non-twin siblings. Because these types of siblings have different degrees of genetic overlap, comparisons across these groups in rates of MHPs can illuminate the relative contributions of genetic, shared family environment, and non-shared factors (Griffiths et al., 2000). In twin studies, it is assumed that family environment factors have an equal impact on siblings and therefore any significant differences in risk of MHPs seen across these groups must be due to differences in genetic similarity (Kendler, 2001). For example, if rates of concordance for depression (i.e. both siblings meet criteria for depression) are higher for monozygotic twins than dizygotic twins, this implies that genetic factors play a significant role in the individual risk of depression, as the former have a greater degree

of genetic overlap than the latter. Similarly, if rates of concordance for depression between twins reared together and those reared apart are similar, this implies that genetic factors are central. That is, growing up in different family environments did not lead to significant differences in risk.

Findings from twin studies, while typically emphasising genetic contributions, have shown that the development of MHPs is multiply determined by genetic, shared family environment, and non-shared factors (Parritz & Troy, 2011). Genetic mechanisms have been strongly implicated for particular types of MHPs or psychopathology. Schizophrenia, for example, is largely influenced by genetic mechanisms with estimates of up to 84% of individual risk of schizophrenia accounted for by genetic factors (Cardino et al., 1999). Several susceptibility genes (e.g. COMT and NRG1) have been identified, and evidence has amassed supporting their role in the development of schizophrenia (Harrison & Weinberger, 2005). Shared family environment factors have also been strongly implicated in the development of MHPs. In a large study including surveys conducted in 21 countries, childhood adversities were found to significantly predict adult psychiatric disorders (Kessler et al., 2010). Parental divorce, parental death, child abuse and/or neglect, and family economic adversities were all significant predictors (Kessler et al., 2010). Collectively, childhood adversities accounted for 29.8% of all first-onset adult psychiatric disorders (Kessler et al., 2010). Non-shared environmental factors also significantly contribute to MHPs. For example, adolescents who perceived greater maternal negativity compared to their sibling were more likely to display depressive symptoms (Pike & Plomin, 1996). Individual experiences or perceptions of maternal negativity had an effect on adolescent depression independent of the effects of genetics and shared family environment factors (Pike & Plomin, 1996).

While each of the types of factors discussed above has been found to significantly predict MHPs, it is widely accepted that it is the interaction between these factors that underpin the development of MHPs. For example, while a strong association has been found for particular high-risk genes and the development of schizophrenia, not all people with the high-risk genes go on to develop schizophrenia (Tienari et al., 2004). This suggests that other factors or triggers need to occur for high-risk genes to be expressed or that other factors or triggers play a key role in the development of schizophrenia (Kendler & Eaves, 1986). Thus, there has been extensive research examining gene x environment interactions in the development of schizophrenia. For example, it is hypothesised that high-risk genes for schizophrenia predispose an individual to be more sensitive to environmental triggers and it is this combination that increases the likelihood of developing schizophrenia (Tienari et al., 2004). Research has identified childhood rearing environments to be a key environmental trigger. In a 12-year follow-up of adoptees with high and low genetic risk for schizophrenia, it was found that adoptees with high genetic risk were significantly more like to develop schizophrenia if raised in an adverse family environment (e.g. conflict, lack of affect, constricted communication, and high enmeshment) than if raised in a healthy family environment (Tienari et al., 2004). The opposite was also found where the risk of developing schizophrenia was significantly reduced if raised in a healthy family environment, despite having high genetic risk (Tienari et al., 2004). Adoptees with low genetic risk did not significantly differ depending on the type of rearing environment (Tienari et al., 2004). Thus, adoptees with high genetic risk were significantly more sensitive to both the effects of adverse and healthy family environments compared to those with low genetic risk (Tienari et

al., 2004). These findings present an excellent example of how genetic risk and environmental risk may interact in the development of MHPs.

As we can see, twin studies have made significant advancements to our understanding of the aetiology of MHPs. However, there are several limitations of twin studies that are particularly relevant to work with siblings of children with MHPs. Firstly, it has been argued that twin family compositions are not representative of the general population and the findings would therefore not generalise to non-twin, singleton siblings (Lombroso, Pauls, & Leckman, 1994). A priori, we would expect differences in psychological outcomes and mechanisms between these two groups. Twins and non-twin siblings differ in the degree of genetic similarity, in developmental environments (e.g. prenatal environments and stresses), and age-related factors (e.g. sharing classes and peer groups). Furthermore, twins are thought to share a unique interpersonal attachment, different from that of non-twin siblings (Tancredy & Fraley, 2006), such as the formation of a special secret language (Rutter & Redshaw, 1991). While this interpersonal attachment can strengthen the bond between twins, twin siblings may also have greater levels of rivalry (Rutter & Redshaw, 1991). Because twins are the same age, presumably there is greater competition over the same resources. That is, they may compete over the attention of their parents and friends, may compete to achieve the same milestones, and may compete to create a sense of individuality (Rutter & Redshaw, 1991). In addition, because twins are, for all intents and purposes, the same age, birth order effects seen in non-twin sibling dynamics cannot be explored. Rutter & Redshaw (1991) provide an excellent, although now somewhat dated, summary of the differences between twin siblings and non-twin, singleton siblings on a range of outcomes, including psychological and cognitive development. In terms of the recent empirical literature, the findings are inconsistent.

Based on genetic studies that included singleton siblings of twin pairs, the findings suggest there is no significant special twin-sibling environment effect (e.g. Ehringer, Rhee, Young, Corley, & Hewitt, 2006). However, in studies that have examined the differences between twin and non-twin families, there have been several significant differences. For example, mothers of twins had significantly more symptoms of depression than mothers of singleton siblings (Thorpe, Golding, MacGillivray, & Greenwood, 1991). Furthermore, twin siblings have been found to report increased closeness in the sibling relationship and have been found to exhibit greater shyness in social interactions than singleton siblings (DiLalla, Millineaux, & Elam, 2008). Although the findings are not entirely conclusive as to whether there are important differences between twin siblings and non-twin, singleton siblings, there is sufficient evidence to question if the findings of twin studies generalise to non-twin siblings.

Second, twin studies have historically focused on concordant traits and continuous or dimensional traits. Due to a focus on the heritability of particular traits, twin studies have historically only assessed twins for concordant MHPs (i.e. the same type of symptom or MHP). Similarly, twin studies have focused on assessing continuous or dimensional behavioural traits, such as symptom counts, with less research on absent/present categorical diagnostic data. This may be because continuous measures and concordance data are ideally suited to calculating correlational data that form the basis of most heritability estimate calculations (Griffiths et al., 2000). To advance our understanding of mental health, multiple aspects of mental health represented by multiple forms of data are needed (Rutter, 2011). It should be noted though that there has been a recent shift towards including these form of data in twin studies (e.g. Hettema, Prescott, & Kendler, 2001b).

Last, because twin studies are typically population-based, they do not purposively sample families where one twin or child in the family has MHPs. As a result, they are not able to explore the impact that MHPs have on the family system and on the lives of family members, including other siblings. There is a large body of literature, as discussed in the following section, which highlights the impact that types of illnesses can have on family members. In particular, illness or disability in a family member can have a significant detrimental effect on family functioning and the wellbeing and mental health of family members. Thus, while twin studies have contributed immensely to our understanding of MHPs and aetiological mechanisms, there remain gaps in our knowledge that twin studies are not well suited to explore.

1.4.3 Siblings of children with special needs.

Contrary to twin studies that have focused on genetic mechanisms and gene x environment interactions, research with siblings of children with special needs has focused on the impact of illness as a primary aetiological mechanism for MHPs. Instead of developmental psychopathology frameworks, this field's origins are in socioecological and family systems theories, which hold that childhood functioning is influenced by and influences the systems to which children belong, such as family and school (Bronfenbrenner, 1979; Hoffman, 1981). On the basis of these theories, researchers in the field hypothesise that the mental health and wellbeing of family members is impacted by the target child's illness or special needs (Austin, 1993). Extensive research has been conducted to explore this hypothesis. However, support for this hypothesis, in regards to siblings, has not been consistently found.

In a meta-analysis of 50 studies, siblings of children with a chronic illness had significantly poorer psychological, social, and cognitive functioning than control

siblings (Sharpe & Rossiter, 2002). The overall weighted mean effect size however, was small (*d*=-0.20) based on conventions for Cohen's *d* (Cohen, 1988). A more recent meta-analysis with a focus on psychological functioning and with greater methodological sensitivity (e.g. excluding studies with samples ≤ 10 and requiring inclusion of a matched control group), similarly reported small effect sizes (Vermaes, van Susante, & van Bakel, 2012). This was reported for internalising problems (*d*=0.17), externalising problems (*d*=0.08), and positive self-attributes (*d*=-0.09). However, both authors noted significant heterogeneity in effect sizes across the included studies (Sharpe & Rossiter, 2002; Vermaes et al., 2012).

In contrast, other syntheses have reported no significant negative impact on siblings of children with special needs. In a systematic review of 65 studies with siblings of children with cancer, the majority of higher quality studies found that siblings did not have elevated scores on measures of psychological functioning, including anxiety and behavioural problems (Alderfer et al., 2010). Similarly, in a critical analysis of the literature on siblings of children with disabilities, the majority of higher quality studies found no significant emotional and behavioural differences between siblings of children with disabilities and controls (Summers, White, & Summer, 1994). Across all of the literature on siblings of children with special needs, including the studies discussed above, considerable heterogeneity across primary studies has been noted. This has stimulated research and theory development on why some siblings have been found to have significant psychological difficulties but not others. A great deal of research has explored what factors might moderate the relationship between the special needs of the child and the functioning of the sibling.

Several models have been developed in response to this and these models may shed light on processes that may similarly apply to siblings of children with MHPs.

Wallander and Varni developed the primary theoretical framework used to inform research and practice with siblings of children with special needs (Figure 3). This model, based on empirical literature and theoretical developments, outlines the various factors that may play a role in the adaptation or adjustment of children with chronic illnesses (Wallander & Varni, 1992; Wallander, Varni, Babani, Banis, & Wilcox, 1989). The applications of this model, the disability-stress-coping model, have since been extended to research on the functioning of the child's family members (e.g. Manuel, 2001). Outlined in the model are several categories of risk and resistance factors that may contribute to the adaptation of children with chronic illnesses and their families. The risk factors are disease/disability (e.g. diagnosis, handicap severity), functional independence, and psychosocial stressors (e.g. daily hassles, handicap-related problems). The resistance factors are intrapersonal factors (e.g. temperament, problem solving ability), socio-ecological factors (e.g. family environment, social support), and stress processing (i.e. cognitive appraisal and coping strategies). These factors, both in isolation and through interactions or interrelations with one another, influence the adaptation (i.e. mental health, social functioning, and physical health) of children and their families. For example, a diagnosis of paraplegia may exert a direct influence on psychosocial stressors (e.g. daily tasks such as helping with dressing, cooking, and bathing), which in turn influences the adaptation of family members by increasing daily stress, reducing time for one's own goals and activities, and caregiver burden. It may also be that this diagnosis exerts an indirect influence on psychosocial stressors via functional independence. An individual with paraplegia may be highly independent or dependent resulting in fewer or greater daily help requirements for family members, respectively. Thus, the diagnosis alone may not determine the

presence of psychosocial stressors, but it may be the interaction or relationship between diagnosis and functional independence.

Another model, the social model of disability, which focuses on the social inequalities faced by people with disabilities, has similarly been extended to include the impact on their family members (Dowling & Dolan, 2001). This model focuses on the idea that disabilities are not necessarily in and of themselves 'disabling', but it is the way in which society and social organisations (e.g. workplaces) fail to accommodate and organise around difference that is disabling (Dowling & Dolan, 2001). Applying this to family members, an individual with a disability may face prejudice and barriers to obtaining employment and may rely on family members financially, increasing family stress and anxiety. Furthermore, family members may not openly discuss their difficulties with their social network due to fears around stigma and prejudice. This may lead to isolation, reduced social support, and poorer psychological functioning and wellbeing. Very little research has explored these factors in the context of siblings and psychological functioning. However, research does support the role of stigma and prejudice in the challenges faced by siblings of children with special needs. For example, siblings are aware that their brother or sister is a target for prejudice, especially by other children, and at times, they become targets themselves by virtue of having a sibling with special needs (Stalker & Connors, 2004). Furthermore, social inequalities may contribute to the quality of the relationships between the sibling and the child with special needs. For example, siblings may feel anger and frustration towards the child due to their functional limitations and at the same time may also feel more protective towards them (Dauz-Williams, Piamjariyakul, Graff, & Stanton, 2010).

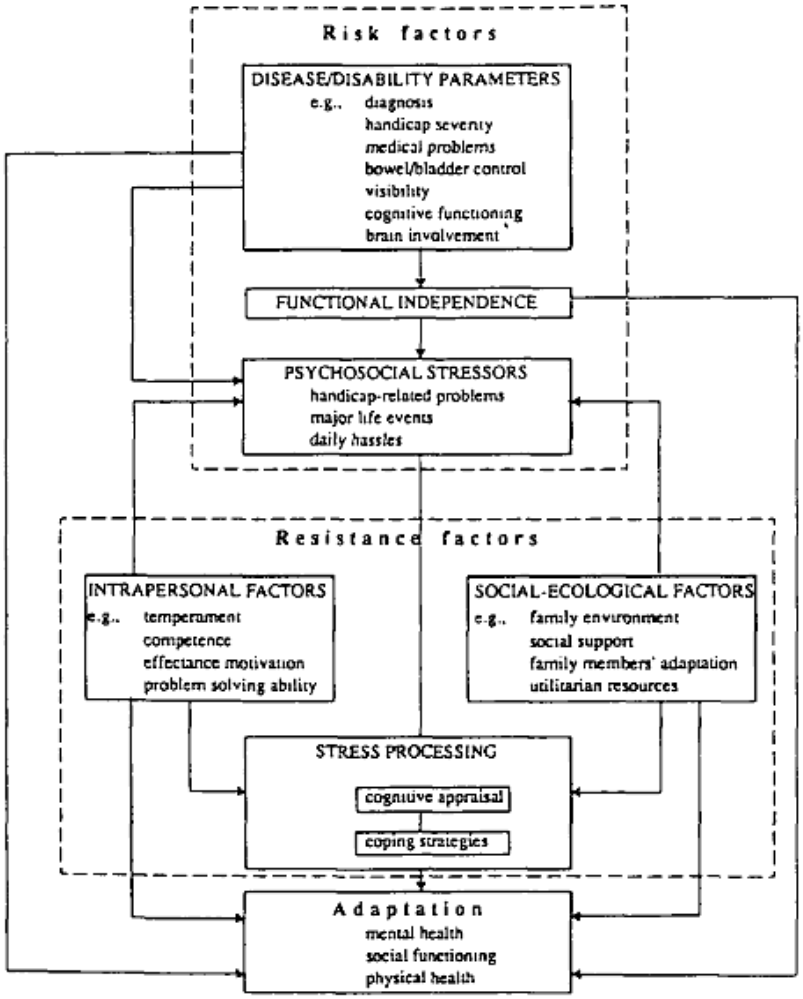

Figure 3. Wallander and colleagues' (1989; 1992) disability-stress-coping model. Reprinted from "Family resources as resistance factors for psychological maladjustment in chronically ill and handicapped children," by J.L. Wallander et al., *Journal of Pediatric Psychology, 14,* p.171. Copyright 1989 by Plenum Publishing Corporation. Reprinted with permission.

Social inequalities may also affect the parent-sibling relationship. In families of children with special needs, most parental resources are typically directed to the child with special needs due to increased caregiving requirements. This may lead siblings to feel angry, resentful, or sad, and negatively impact siblings' self-concept and the parent-sibling relationship (Dauz-Williams et al., 2010).

These models may also be relevant to siblings of children with MHPs. Drawing on the disability-stress-coping model, siblings likely face a number of psychosocial stressors associated with having a brother or sister with MHPs. These psychosocial stressors may differ depending on the type of MHP or diagnosis in the target child. Siblings of children with anxiety disorders may miss out on family outings, such as theme parks, because the child is fearful of crowded social situations. Siblings of children with severe depression or psychosis may have to make several hospital visits that disrupt their daily routines. Drawing on the social model of disability, stigma surrounding mental illness remains prevalent with continuing prejudices and misconceptions (Angermeyer & Dietrich, 2006). This may lead to delays in seeking treatment for children with MHPs and may lengthen the time that siblings must cope with the challenges associated with having a brother or sister with MHPs. It may also lead to delays in seeking treatment for siblings that are also displaying MHPs.

However, as these models are based on research with families of children with physical illnesses and developmental disabilities, they do not give sufficient consideration to risk factors related to familial MHPs. That is, they do not give due consideration of increased genetic and shared family environment risk factors that are likely to impact on the mental health of siblings. Thus, while normative studies, twin studies, and special needs literature have made significant advances that contribute to our understanding of siblings of children with MHPs, each also has important

limitations. Of note, they are unable to fully integrate developmental psychopathology and impact of illness frameworks that likely both apply to siblings of children with MHPs.

1.4.4 Family or familial aggregation studies.

Family studies or familial aggregation studies recruit family members of individuals with MHPs to explore the familial aggregation or transmission of MHPs and psychopathology. That is, if one member of the family has MHPs, what is the likelihood or risk of MHPs in other members of their family? Because family studies recruit families that have a member with MHPs, they are able to explore the role of genetic, shared family environment, and non-shared environment factors in developmental psychopathology aetiological frameworks. Further, they are able to explore the impact that having a family member with MHPs has on the mental health and wellbeing of other family members. In this way, family studies are able to overcome some of the limitations in the fields discussed above.

Family studies have shown a pattern of aggregation of MHPs within families and highlight the increased risk of MHPs and psychopathology in family members of individuals with MHPs. In a meta-analysis of the familial aggregation of anxiety disorders, first-degree relatives had a significantly greater risk of the same type of anxiety disorder as the target individual (Hettema, Neale, & Kendler, 2001a). First-degree relatives were five times more likely to have panic disorder if their family member had panic disorder, than those without a family member with panic disorder (Hettema et al., 2001a). Similarly, first-degree relatives were six times more likely to have generalised anxiety disorder (GAD) if their family member had GAD (Hettema et al., 2001a). Significantly higher rates of MHPs in family members have been found

across a range of psychiatric diagnoses including depression, substance abuse, and conduct or antisocial behaviour problems (Kendler, Davis, & Kessler, 1997). While the focus in family studies has tended towards examination of concordant MHPs, similar findings have also been noted in studies of discordant MHPs. In a large study across 10 countries with a total 51,507 participants, parental psychopathology was significantly associated with an increased risk of a broad range of MHPs in children (McLaughlin et al., 2004). For example, children with one parent with substance abuse problems were two to three times more likely to have substance abuse problems (McLaughlin et al., 2004). The odds ratio (OR) increased to 4.3 if both parents had substance abuse problems (McLaughlin et al., 2004). However, children from these families were also significantly more likely to have a mood disorder (OR=2.3), an anxiety disorder (OR=3.0), or behavioural problems (OR=3.7; McLaughlin et al., 2004).

Family study researchers have tended to focus on developmental psychopathology pathways to explain these findings. For example, combining twin and family study concordance data revealed a significant role for genetic factors in the aetiology of panic disorder (Hettema et al., 2001a). Approximately 30-40% of individual variance in risk was attributed to genetic factors (Hettema et al., 2001a). Using structural equation modelling, the best-fitting model for accounting for variance in individual risk included only genetic and non-shared environment factors, not shared family environment factors (Hettema et al., 2001a). As previously discussed, there is a focus in family studies on concordance and, thus, on examining the aetiology of specific MHPs or clusters of MHPs (e.g. behavioural problems). However, because family members have an increased risk for discordant MHPs also, it has been argued that genetic and gene x environment factors impart a broad vulnerability to MHPs rather than a disorder-specific influence (McLaughlin et al., 2012). Impact of illness

factors have also been explored, though this has been mostly limited to the qualitative literature. For example, children report that having a parent with MHPs leads to family relationship strain, academic and school difficulties, and chaotic home lives (Garley, Gallop, Johnston, & Pipitone, 1997). These impact of illness experiences are likely to significantly influence the mental health and wellbeing of these children.

Family studies have significantly contributed to our understanding of MHPs and have the potential to allow a convergence of developmental psychopathology and impact of illness theories in understanding the development of MHPs. However, the literature has neglected siblings, focusing more on parent-child aggregation and transmission.

1.5 Rationale and Aims of Thesis

In sum, two main points arise from a consideration of the theoretical and empirical literature on families, siblings, and mental health. The literature highlights the influential role of sibling processes on mental health and wellbeing in normative and clinical populations. This literature justifies the continued research of sibling populations. The literature also suggests that siblings of children with MHPs may be a specific group of siblings that are particularly at risk of poorer mental health and wellbeing. That is, they are exposed to risk factors highlighted across all of the theories described above. Yet, very little research has been conducted with this population. Further, to the best of my knowledge, the research has not yet been systematically synthesised. In a preliminary survey of the literature, I noted several issues in the literature that point to the need for systematic synthesis.

First, I found that data on siblings of children with MHPs were spread across several different fields. For example, some data were in sibling-focused family studies,

some in twin studies where non-twin siblings acted as a control group, and some in the impact of illness literature. A systematic review is needed to draw these data together in a systematic and comprehensive fashion to enable conclusions to be formed based on all of the existing data. Second, because sibling data are spread across several fields, each with their own favoured theoretical frameworks, there has been little cross-consideration and integration of these frameworks. A systematic review may serve to draw the data together and enable integration of these frameworks. Third, I found inconsistencies across the literature in terms of statistically significant findings and effect size estimates, similar to that found in the literature on siblings of children with special needs. A systematic review of the literature with a focus on methodological quality may shed light on these inconsistencies. Fourth, I noted several methodological limitations in the existing literature. These methodological limitations persisted over time and the same limitations tended to be continuously replicated. Without a systematic review, there has been little guidance on appropriate methodology when working with siblings of children with MHPs. Similarly, some aspects of the mental health of siblings had been sufficiently explored by past research but later research continued to examine these aspects. Without a systematic review updating the state of knowledge in the field, researchers have little guidance on what questions had been answered and key areas that need additional research. Last, I found that despite decades of research, siblings of children with MHPs continue to be overlooked and effective prevention or intervention programs have not been developed. A systematic review may give more weight to the existing research and allow more conclusions and clinical implications to be formed.

Broadly, the main aim of this thesis was to explore the mental health and wellbeing of siblings of children with MHPs. To achieve this, I conducted a systematic

review of the literature to a) clarify the current state of evidence, b) assess the methodologies used, c) formulate theoretical and clinical implications beyond that found in primary studies, and d) highlight what is not yet known in this field and form recommendations for future research. In addition, based on the findings of the systematic review, I conducted a primary research study that could address some of the gaps in the literature and begin the process of taking what has been done to inform and advance what comes next.

There are two key points to consider when reading this thesis in regards to aims. While there is considerable discussion of theoretical frameworks and aetiological pathways across the thesis, my intention was not to synthesise research on the aetiology of MHPs in children. This would be better achieved by synthesising findings from twin studies and family-based studies. Second, my focus was on psychosocial outcomes (i.e. emotional, behavioural, and social). I, therefore, did not include data on cognitive, neurological, and physiological measures. A large body of literature on siblings of individuals with MHPs has focused on these measures and meta-analyses of these data have been published (e.g. Chan, Xu, Heinrichs, Yu, & Gong, 2010; Sitskoom, Aleman, Ebisch, Appels, & Kahn, 2004). Thus, I chose to focus on psychosocial outcomes.

This thesis, then, is focused on providing an extensive and comprehensive exploration of the mental health and wellbeing of siblings of children with MHPs, a population and area of functioning that has been neglected across the literature. Additional methodological details and linkage between studies are described in more detail in the following section.

Chapter 2. Preamble for the Systematic Review

2.1 Overview of Chapter

This chapter provides additional details regarding the systematic review research process used in this thesis that could not be outlined in the individual papers. It is important to note that these papers were written in consideration of the journals' space requirements, referencing style preference, and research scope. The papers presented in this thesis have been influenced by the comments, suggestions, and preferences of blind reviewers and editors. As such, the individual papers may shift from the main aims in this thesis. In particular, the paper in Chapter 6 focuses on the methodology used rather than the content of the findings. The methodology used in that paper is a relatively recent development in research methodology (Sandelowki & Barroso, 2007), and the editors believed that this paper could have significant value and could significantly contribute to the advancement of research methodologies. Thus, this paper reflects a more methodologically driven discussion. However, in this body of this thesis, these findings are considered and interpreted within the context of the main aims of this thesis and within the context of the mental health and wellbeing of siblings of children with MHPs.

2.2 Systematic Review Methodology

Chapters 3, 4, 5, and 6 present the individual papers that report on the results of a systematic review of the literature on the mental health and wellbeing of siblings of children with MHPs. The research process used in this thesis began with a cursory examination of the literature to determine what past research had demonstrated, and what gaps remained in the literature. Following a cursory examination of the existing

literature on siblings of children with MHPs, it became apparent that there were inconsistent findings across the literature and that relevant data were spread across several different fields of research. It was decided, at this point, that a more systematic approach was necessary to gain a comprehensive understanding of the current state of evidence on the mental health and wellbeing of siblings of children with MHPs. Having found no existing systematic review in the literature, this became the first step in the research process used in this thesis.

Although the data from the systematic review is presented as four individual papers, each of these resulted from a single systematic search. The methodology of this main or parent systematic review, including the decision to present the data separately, is outlined here, as it was not discussed in detail as a single search strategy in the individual papers. To develop the search and analysis strategy for the systematic review, several established systematic review guidelines were consulted (e.g. Higgins & Green, 2011; Petticrew & Roberts, 2006). A list of search terms was developed, with the guidance of a specialist librarian, to capture the key concepts of the research question, such as *sibling, first-degree relative, child, emotional, psychiatric,* and *social*. A full list of the search terms used is provided in Appendix A. Four electronic databases were searched: Cumulative Index to Nursing and Allied Health Literature (CINAHL), PsycINFO, PubMed, and SciVerse Scopus. To ensure the search results were current yet extensive, the databases were searched for articles published from January 1990 to July 2012.

The selection process used in the systematic review included multiple steps. These are shown in Figure 4. The initial search resulted in 6,182 records. After removing duplicates and records that were not journal articles (e.g. book reviews), 3,853 records remained. These records were screened against the following

inclusion/exclusion criteria. To be included in the systematic review, the study had to a) be an article published in English and in a peer-reviewed journal, b) report primary quantitative or qualitative data, and c) report data on the emotional, behavioural, and/or social wellbeing of siblings of target children with MHPs. The definition of target children with MHPs has been previously described in section 1.2.1. Because the primary interest was in the mental health and wellbeing of siblings of children with MHPs and whether these differed from that of 'normal' or control siblings, an additional criterion was applied to quantitative articles. The articles had to report data that would allow comparisons to a control population, such as data from a matched control group or standardised normative data. This did not apply to qualitative studies. In qualitative synthesis, comparisons to a control population are achieved through comparing the included studies to a separate sample of qualitative studies conducted with an extra-study population (Sandelowski & Barroso, 2007). That is, comparisons are conducted across studies, not within studies. Therefore, qualitative studies did not need to include data from a control group or normative data (see Chapter 6 for more details). Studies were excluded if a) the target child's difficulty was not primarily a MHP (e.g. chronic illness), b) purposive sampling of siblings of target children according to the presence or absence of psychopathology was used, and c) the study focused exclusively on twin sibling pairs. Additional inclusion and exclusion criteria that pertain to the types of data analysed in the individual papers is described in the relevant paper.

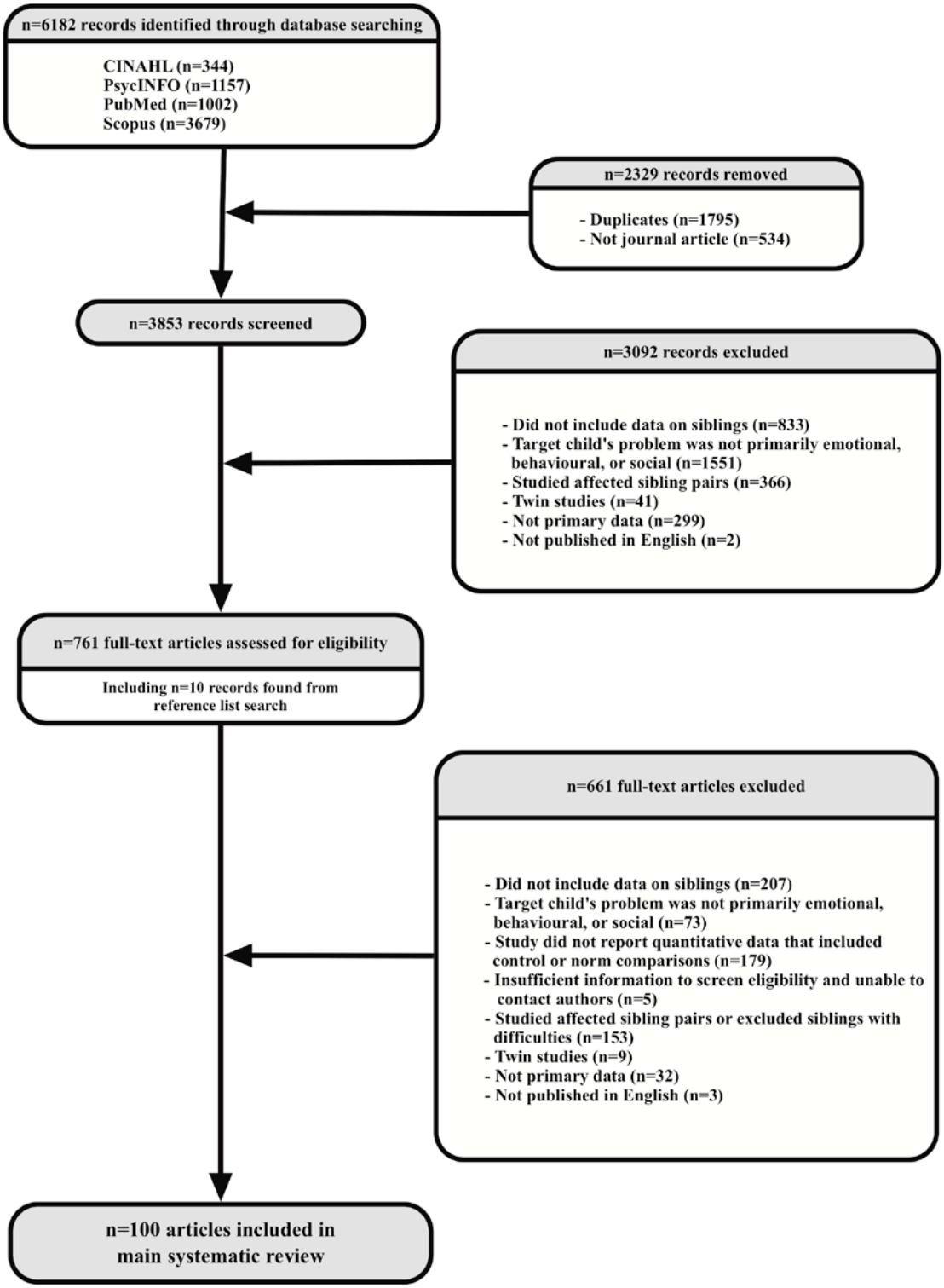

Figure 4. Flow of studies included in the systematic review.

Titles and abstracts were screened according to these inclusion/exclusion criteria. If this did not provide sufficient information, full-text versions of the articles were accessed and screened for eligibility. In cases where the full-text articles did not provide sufficient information, attempts were made to contact the original authors. If they did not reply, the study was excluded. To limit data-selection bias, an independent researcher reviewed a randomly selected sample of articles (10% of full-text articles) for eligibility and 100% inter-rater agreement was reached. The reference lists of the eligible articles were searched for records not captured by the initial search strategy. This lead to an additional 10 records, none of which were eligible. From this selection process, a total of 100 eligible articles were identified.

Data on the mental health and wellbeing of siblings of children with MHPs were extracted, including data on moderators of mental health and wellbeing, from each article. If studies recruited participants from the same population pool, several extraction methods were used to ensure the participants were not counted twice. First, only unique data points were extracted from the studies. For example, one study may have reported the prevalence of social anxiety and another may have reported the prevalence of depression. In this case, both of these data points were extracted. If studies reported the same data and drew upon the same participant pool, the data from the highest quality study were analysed (see papers for quality criteria). For studies that segregated data by categories not relevant to the systematic review (e.g. sibling sex or age), the data were pooled into a total sibling score. Sibling sex or age data were retained and included in the discussion of moderators of the mental health and wellbeing of siblings of children with MHPs.

Data extraction resulted in a large dataset and it became apparent that it was necessary to segregate these data in some way. Segregation of the data allowed the

analysis of a large dataset to be more manageable, allowed due consideration and in-depth examination of the findings from the primary studies, and was more suitable for the length of journal publications. From a cursory examination, I found that all of the data could be characterised as describing one of four different perspectives on the mental health and wellbeing of siblings of children with MHPs. These were prevalence rates of psychopathology, continuous/dimensional data on mental health, family relationship quality, and qualitative research on the experiences of siblings. These were clearly defined and distinct categories and were the most appropriate way of segregating the large dataset. Chapters 3, 4, 5, and 6 present the papers on each of these categories, respectively. Appendices B, C, D, and E contain the main data extraction tables that were not included in the published or submitted papers.

There were two overarching analytic choices that applied to the quantitative papers (Chapters 3, 4, and 5). Because these choices impacted on the majority of the analyses conducted in the systematic review, the process behind them will be briefly outlined here. The individual papers discuss in greater detail the specific analytic strategies used for that particular body of work. First, it was decided following data extraction that a meta-analysis was inappropriate. Significant heterogeneity across studies was noted in key study characteristics, such as sample size, type of MHP in the target child, measures used, and outcomes measure. While study characteristics can be controlled for or analysed using sub-group meta-analytic techniques (Davey Smith & Egger, 2001), there were insufficient data to warrant the use of these strategies. For example, in most cases, only two or three studies with a control group assessed siblings for the same type of psychopathology and these typically differed in the type of MHP in the target child and the measures used. As noted by several researchers (e.g. Valentine, Pigott, & Rothstein, 2010), while a meta-analysis of only two or three studies can be

conducted, it is likely to only be informative when the studies are close replications of one another. This was not the case in this review. Second, it was decided that sibling data would not be statistically compared to normative data in studies that did not recruit a matched control group. Doing so may have introduced additional sources of error through imperfect matching of comparison groups (e.g. on age and gender) and due to considerable differences in study characteristics, such as sample size. Given a substantial proportion of the included studies had significant methodological limitations, it was deemed inappropriate to further compound these issues by conducting statistical comparisons to normative data. Thus, the papers present narrative syntheses of the existing literature with a focus on methodological quality and on consistent findings across studies.

In the narrative synthesis for the quantitative papers, four different methods of understanding the relationship between target sibling scores and control sibling or general population scores are discussed. First, absolute effect size or raw differences and relationships between scores are discussed (Sullivan & Fein, 2012). This was appropriate given that many studies did not recruit a control group and thus, statistical comparisons could not be discussed. Although this type of data is not conclusive, it can be informative and can suggest a trend in the data when considered within the context of findings from multiple studies and with attention to consistency across studies. Second, statistical significance of the difference between the target and control sibling scores is discussed. Third, effect sizes (i.e. odds ratios or Cohen's *d;* Cohen, 1988) are discussed with reference to the associated interpretive guidelines. Last, effect size confidence intervals are discussed with reference to the width of the confidence interval. As recommended by systematic review guidelines (Higgins & Green, 2011) and as noted by several researchers (e.g. Valentine et al., 2010), it is essential to

consider multiple statistical comparison measures when interpreting research findings. Each of these statistics is impacted by several variables and do not necessarily correspond to 'true' effects. For example, *p*-values and statistical significance is impacted by sample size and power and a non-significant result does not necessarily mean that there is no difference between these groups (Goodman, 2008). Null effect is a possibility but the range of plausible effects, as shown in confidence intervals, are equally likely (Goodman, 2008). Given that the majority of the studies in this review were considered to be lower in quality and did not recruit sufficient sample sizes, based on Cohen's (1992) power analyses (see individual papers), it was especially important in this review to consider multiple comparison statistics.

2.3 Key Findings From Systematic Review Papers

The paper presented in Chapter 3 describes the findings on the prevalence of psychopathology in siblings of children with MHPs. Siblings of children with MHPs were found to have higher prevalence rates of psychopathology compared to control children and general population prevalence rates. Although there was a trend for concordant disorders in the target child and target sibling, siblings were also found to have an increased risk of other types of psychopathology. These findings suggest that siblings of children with MHPs are at risk of developing a broad range of psychopathologies.

The paper presented in Chapter 4 describes continuous data on the psychosocial functioning of siblings of children with MHPs. It takes a dimensional approach to understanding the mental health of siblings as opposed to the categorical approach of the previous paper. Siblings of children with MHPs were found to have poorer psychosocial functioning across a range of domains compared to control children (e.g.

social problems). There was a trend for siblings to have greater impairment in particular areas of functioning including social problems, delinquent behaviour problems, anxious/depressed behaviour, and somatic complaints. This trend was found regardless of the type of MHP in the target child.

The paper presented in Chapter 5 describes the quality of family relationships for siblings of children with MHPs. Siblings of children with MHPs had more negative (e.g. hostile) and less positive (e.g. supportive) relationships with the target child and with their parents. This was reported across several different measures and across reports from the perspective of parents, target children, and siblings. There were two possible exceptions. First, siblings of children with conduct disorder (CD) had more positive sibling relationships than control children. Second, siblings of children with anxiety disorders did not significantly differ from controls on sibling relationship quality.

The paper presented in Chapter 6 describes the experiences and challenges faced by siblings of children with MHPs and the coping strategies they use. Siblings of children with MHPs experience a range of challenges due to having a brother or sister with MHPs. These include conflict and violence, family relationships problems (e.g. parental favouritism), and an increase in caregiving responsibilities. These challenges have a significant emotional impact on the wellbeing of siblings of children with MHPs. While the majority of these were described as operating mostly within the home setting, siblings also described how these challenges extended to all other areas of their life. For example, siblings described also needing to care for, protect, and monitor the target child at school. Siblings used a number of strategies to cope with these challenges. These included accommodating around the target child's behaviour and moods, avoiding the target child and avoiding being at home, and normalising their

experiences within the context of ‘normal’ family dynamics and dysfunctions. Some trends were noted in regards to the type of MHP in the target child. For example, siblings of children with anxiety disorders did not describe experiencing violence in the home. However, experiences and coping strategies were, for the most part, shared across siblings regardless of the type of MHP in the target child.

Across all of these papers, two consistent findings emerged. First, siblings of children with MHPs appear to have a broad risk of psychosocial impairment. That is, siblings of children with MHPs experience difficulties in all areas of mental health and wellbeing and across a range of settings. This was found regardless of the type of MHP in the target child, with the possible exception of target children with anxiety disorders. Second, with the exception of the qualitative literature, the majority of studies had significant methodological limitations. Poor methodological quality in the existing literature has limited the conclusions that can be formed and has limited advancements in the scientific knowledge base in this field.

2.4 Contribution to Knowledge

Each of the papers had specific limitations and implications, which are discussed within each chapter. Overall, this systematic review significantly contributed to our understanding of siblings of children with MHPs. It was the first known attempt to synthesise the literature on the mental health and wellbeing of siblings of children with MHPs. It clarified the current state of evidence highlighting what is known and what areas require further exploration. Importantly, the systematic review included findings on multiple aspects or perspectives on mental health and wellbeing. In doing so, it has provided a holistic, comprehensive summary of the mental health and wellbeing of siblings. Several key research gaps that remain in the literature were

highlighted. These provide guidance for future researchers and encourage future research that builds on existing evidence in a meaningful way.

This systematic review also highlighted common methodological problems that occur in sibling research and several methodological recommendations were outlined. In this way, it has contributed to improvements in methodological quality, will enable more robust conclusions to be formed, and will significantly advance research in this field. Theoretical and clinical implications resulting from this review, as discussed in the individual papers, may advance the development of sibling theories in mental health and may advance prevention, treatment, and support efforts for target siblings. Last, this systematic review may draw much needed attention to siblings of children with MHPs. It provides a higher level of evidence than primary studies and may attract greater recognition of this high-risk population from clinicians, researchers, and funding bodies.

Chapter 3. Paper One

The Prevalence of Psychopathology in Siblings of Children with Mental Health Problems: A 20-Year Systematic Review

(ACCEPTED FOR PUBLICATION)

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Statement of Authorship

Nylanda Ma (Candidate)

I was responsible for the design and implementation of the systematic review, including development of the search strategy, collection of the included studies, data extraction, data analysis, and writing the manuscript. I served as corresponding author and was responsible for manuscript submission, revisions, and responses to journal reviews.

Signed: Nylanda LT Ma _____ Date: 4/8/2014

Dr. Rachel Roberts, Prof. Helen Winefield, & Dr. Gareth Furber (Co-authors)

We provided ongoing supervision through the systematic review and there was ongoing collaboration between Nylanda Ma and ourselves in refining the direction of research and methodology. We commented on manuscript drafts, made suggestions on the presentation of the material in the manuscript, and provided guidance on journal revisions and responses. We hereby give our permission for the inclusion of this paper in Nylanda Ma's submission for the degree of Doctor of Philosophy from the University of Adelaide.

Signed: Dr. Rachel Roberts _____Date: 4/8/2014

Signed: Prof. Helen Winefield _____Date: 4/8/2014

Signed: Dr. Gareth Furber _____ Date: 4/8/2014

Abstract

While the importance of looking at the entire family system in the context of child and adolescent mental health is well recognised, siblings of children with mental health problems (MHPs) are often overlooked. The existing literature on the mental health of these siblings needs to be reviewed. A systematic search located publications from 1990 to 2011 in four electronic databases. Thirty-nine relevant studies reported data on the prevalence of psychopathology in siblings of target children with MHPs. Siblings of target children had higher rates of at least one type of psychopathology than comparison children. Risk of psychopathology varied across the type of MHP in the target child. Other covariates included sibling age and gender and parental psychopathology. Significant variations and limitations in methodology were found in the existing literature. Methodological guidelines for future studies are outlined. Implications for clinicians, parents, and for future research are discussed.

Keywords: Sibling; prevalence; mental health problems; child; systematic review

Mental health problems (MHPs) in children have a significant impact, not only on the lives of these children, but also on the multiple systems in which the child is involved (e.g. family and school systems) [1]. In line with this, there is a large body of research on children with MHPs and their families. This research however has largely ignored siblings, focusing mainly on parents of children with MHPs and children of parents with MHPs. There are three related fields that inform our understanding of the mental health of siblings. These are discussed briefly below with attention paid to their limitations in terms of contributions to our understanding of siblings and why research focused on siblings of children with MHPs is needed.

Twin studies

Much of our understanding of childhood MHPs has come from findings of twin studies. Findings from twin studies have consistently shown high rates of concordant MHPs in twin siblings. For example, a concordance rate of 48% for schizophrenia has been reported for monozygotic twins [2]. The predominant theory in developmental psychopathology holds that MHPs are multiply determined by the interactions between genetic, shared environment, nonshared or individual risk factors, and protective factors [3]. Within each of these factorial categories, several different pathways have been hypothesised and implicated in the development of psychopathology. Both genetic factors, such as individual genotypes, and shared environment factors, such as impaired parenting have been associated with the development of several types of MHPs in children including conduct problems and anxiety problems [3]. Interplays between genetic and shared environments have also been linked to psychopathology (see [4] for a comprehensive review). In the case of ADHD, for example, a passive gene x environment interaction between adverse childhood environments (e.g. family conflict) and 5-HTTLPR, a serotonin promoter transporter gene polymorphism, has been shown

to influence the risk of ADHD. Variations in this polymorphism (i.e. SS, SL, or LL) have been found to result in variations in the rate of uptake of serotonin with the LL genotype being linked to the higher maximal uptake of serotonin compared to the SS or SL genotype [5]. A high childhood adversity index significantly increased the risk of ADHD for children with the SS and SL genotypes, whereas childhood adversity did not significantly increase risk for children with the LL genotype [6]. Lastly, nonshared environmental or individual factors have been shown to influence the development of psychopathology in children. Adaptive coping strategies, for example, have been found to be a protective factor for children who are exposed to risk factors for psychopathology [7]. Although genetic, shared environment, and nonshared environment factors and the interplay between them have been linked to a broad range of MHPs, the specific risk factors, pathways, interactions, and relative contributions of these differ depending on the type of MHP. For example, as described above, while genetic and gene x environment interactions explain a considerable amount of individual variation in risk of ADHD, reported heritability estimates are lower for depression and anxiety and may point to a more prominent role for non-shared environment factors, such as stressful life events, than is found for ADHD [8]. Given the wide range of variables that can influence child development and the numerous ways in which these can interact, despite decades of research, there remain significant gaps in our understanding of how psychopathology develops in children.

Due to the value of twin studies in examining the relative contributions of genetic and shared and non-shared environmental factors, the majority of research has focused on this population. There have been several criticisms of twin studies however, several of which have particular relevance to this review. Firstly, it has been argued that twin family compositions are not representative of the general population [9]. More

specifically, findings from twin siblings may not apply to non-twin or singleton siblings. Theoretically, we would expect differences across these groups due to differences in the degree of similarity in genetic backgrounds, developmental environments (e.g. prenatal), and age-related factors (e.g. sharing classes, having the same peer group). The research in this area has been inconsistent. While some studies have demonstrated no significant special twin-sibling environment [8], other studies have reported significant differences between twin and non-twin families, for example, in the presence of psychopathology in parents [10], perceptions of family environments [11], in the quality of sibling relationships and on psychological measures, such as shyness [12]. Second, due to a focus on the heritability of particular traits, twin studies typically only assess concordant psychopathologies although studies examining twins for discordant psychopathologies are increasing. Third, although there has been a recent shift towards including absent/present diagnostic data, twin studies have traditionally been limited to continuous behavioural trait measures, such as symptom counts. Last, because twin studies are population-based and do not purposively sample the population for families where one twin or one child in the family has MHPs, they do not explore the impact that psychopathology has on the family system and on the lives and mental health of family members. Qualitative research has highlighted the difficulties associated with having a family member with MHPs, including a chaotic, unpredictable home life and strained family relationships, which impact on the mental health of family members [13]. Thus, while twin studies have contributed immensely to our understanding of psychopathology, there are still several gaps in our knowledge that twin studies are not apt to address.

Siblings of children with disabilities or special needs

Research with siblings of other types of 'affected' children, such as those with neurodevelopmental disabilities or chronic health conditions, has extensively explored the impact of health problems on family members. This research is based on socio-ecological and family systems theories that hold that individuals are influenced by the systems to which they belong (e.g. family, school) and vice versa [14,15]. Therefore, it would be expected that family members would be impacted by the child's illness. Findings thus far have suggested a negative impact on the functioning of family members. For example, research has demonstrated a significant association between having a brother or sister with a disability and poorer psychosocial functioning in siblings [16]. However, considerable variation has also been noted across studies [16]. In line with socio-ecological and family systems theories, the primary focus in this field has been on exploring factors relating to the impact of the illness on the family system as possible explanations for individual variations in psychosocial functioning among these siblings. Researchers have suggested several pathways by which the child's disability may impact on the siblings' mental health. The most influential model in this field was a conceptual model developed by Wallander and Varni [17] that highlighted risk and protective factors that contribute to the psychosocial adjustment of siblings. They outlined seven interrelated factor categories, each with numerous individual factors: Disease and disability (e.g. diagnosis, severity), functional independence (e.g. ambulation, communication), psychosocial stress (e.g. daily hassles), adjustment/adaptation (e.g. social, physical), personal factors (e.g. temperament, problem solving skills), socio-ecological (e.g. family members' adjustment, social support) and stress processing (e.g. coping strategies, cognitive appraisal). Research in this area has supported some of the proposed pathways. For example, the psychosocial

adjustment of siblings of children with chronic health conditions has been shown to significantly relate to the impact of the child's illness on day-to-day functioning [18]. It was hypothesised that this was due to increased caretaking responsibilities and related psychosocial stress and decreased parental attention and family relationship problems often reported by siblings [18]. Qualitative studies with siblings of children with MHPs have found that these siblings and their parents describe similar experiences of how MHPs impact on their lives [13]. Thus, we would expect that siblings of children with MHPs would similarly be at risk of poorer mental health compared to siblings of typically developing children and that similar impact of illness pathways may be involved. While findings from research with siblings of children with special needs have highlighted the impact of illness on family members, this field has largely neglected siblings of children with MHPs. Furthermore, as the focus in this field is on illness impact, the role of genetic and shared environment risk factors is typically not considered.

In sum, twin studies highlight the role of genetic, shared environment, and non-shared environment factors in the development of psychopathology and research with siblings of children with special needs highlights the role of the child's illness and the impact on the family system. However, both fields have limitations and have not been able to integrate findings and theories from other fields.

Family studies

Family studies or familial aggregation studies, on the other hand, are able to overcome some of these limitations. Because these studies recruit families that have a member with MHPs, they are able to explore genetic and shared and non-shared environmental risk factors as well as how MHPs impact the family system. Further, these studies are not limited to recruiting twin families and are able to explore the risk

of psychopathology for siblings in non-twin families. Findings from these studies, consistent with twin studies, show a pattern of aggregation of MHPs within families. This has been demonstrated across a broad range of psychopathologies [19]. While the majority of these studies maintain a focus on heritability and assess family members for concordant psychopathologies, a large number of studies have assessed a broad range of psychopathologies in family members. Parental depression, for example, is associated with increased risk of affective disorders, substance use disorders (SUDs), and behavioural problems in their offspring [20]. A similar pattern of a broad risk for psychopathology has been found for children of parents with a range of MHPs [20]. Because family members have an increased risk of discordant psychopathology also, it has been argued that genetic and gene x environment factors impart a broad vulnerability to psychopathology rather than a disorder-specific vulnerability [20]. Family study methodology can also be used to explore the impact of MHPs on other family members and family functioning overall because families are purposively sampled for the presence of MHPs in a family member. Consistent with findings from research with siblings of children with special needs, family members of individuals with MHPs report that these problems have a significant impact on their life. For example, children of parents with MHPs report family relationship strain, academic and school difficulties, fears about developing MHPs, chaotic home lives, and having to take on caretaking roles [21].

While family studies significantly contribute to our understanding of psychopathology, the vast majority of the research examines parent-child transmission. Despite the significant contribution of siblings to the family system and to our mental health, siblings of children with MHPs have been neglected in the family study literature. Unlike many non-familial relationships, sibling relationships are lifelong and

are more likely to influence behaviour through ongoing long-term modelling and reinforcement [22,23]. Sibling relationship problems such as conflict or aggression, for example, have been linked to increased aggression, decreased social competence, and increased loneliness [23-25]. Furthermore, because siblings similarly share genetic predispositions and risk factors related to shared family environments, it would be expected that they would be a high risk of developing psychopathology compared to siblings of typically developing children [3]. Yet, very little research has explored the risk of psychopathology in siblings of children with MHPs.

Universal models have been developed to describe sibling pathways in the development of psychopathology. However, these are generalised models that do not take into account the impact of living with a brother or sister with MHPs and, therefore are insufficiently comprehensive to account for the numerous mechanisms relevant to siblings of children with MHPs (e.g. genetic influences). Feinberg, Sakuma, Hostetler, and McHale [26] for example, outlined a conceptual model of pathways from sibling conflict to sibling adjustment problems. The model includes two primary pathways: An extra-famililal peer/school pathway and a family relationship pathway. The extra-familial pathway is based on the idea that sibling conflict provides a negative socialisation environment that generalises to other social relationships leading to deviant peer associations. These associations reinforce positive attitudes towards risky behaviour, increase exposure to substances, and increase opportunities for learning and expressing maladaptive behaviours. The familial pathway includes sibling deviance training, which increases the risk of developing MHPs through modelling, reinforcement, and collusion to undermine authority mechanisms. This pathway also includes broader parenting risk factors, such as decreased monitoring and harsh and inconsistent parenting styles, which are positively correlated with sibling conflict and

with risk of MHPs in children. While this model contributes to our understanding of how psychopathology might develop across siblings, it does not include genetic influences or factors relating to the impact of MHPs on the family system.

The Present Study

Even though, according to the theoretical frameworks and empirical findings from related research fields, siblings of children with MHPs would be at high risk of developing psychopathology compared to siblings of typically developing children, very little research has explored this population. Research with siblings of children with MHPs has slowly amassed over the last 20 years. However, this research has yet to be systematically synthesised. Given that the literature in this area is spread across several fields, as described above (e.g. genetic studies and impact of illness studies), a systematic review is needed to aggregate the research. Furthermore, existing research has reported inconsistent findings. For example, reported rates of obsessive compulsive disorder (OCD) in siblings of children with anxiety disorders vary considerably across studies, ranging from 0% to 35% [27-32]. A systematic review of the literature with a focus on methodological quality would identify the more robust and reliable findings and address the inconsistencies across studies. Lastly, without a systematic review of the literature, researchers in the field have little guidance on what questions need to be answered and on common methodological issues that occur in sibling studies. As a result, future research may continue to explore topics that have been adequately addressed or may continue to perpetuate methodological limitations. Accordingly, the present study aims to synthesise the existing research to: a) clarify the current state of evidence on the prevalence rates and covariates of psychopathology in siblings of children with MHPs, b) assess the methodologies used across studies in this field and

formulate recommendations for future research, and c) highlight what is not yet known regarding psychopathology in siblings to guide future research.

Methods

Identification of Studies

Four electronic databases were searched: Cumulative Index to Nursing and Allied Health Literature (CINAHL), PsycINFO, PubMed, and SciVerse Scopus using terms such as *sibling; first degree relative; child; emotional; behavioural; psychiatric* (see Appendix A for the full list). Studies were included if they reported prevalence rates of or variables associated with psychopathology in siblings of target children with MHPs, reported primary data, and were published in English in a peer-reviewed journal between January 1990 and July 2011. Criteria for the presence of MHPs in target children (≤18 years as set by legal definitions of children [33], no age cut-offs were set for siblings) were defined broadly based on previous research [34], and were as follows: a) presence of an established psychiatric diagnosis, b) positive screen on diagnostic measures, c) clinically elevated scores on psychological measures, d) referred to or engaging in mental health treatment, or e) adjudicated in court for delinquent behaviour suggesting the presence of emotional and/or behavioural difficulties.

Using these criteria and definitions, a problem of circularity arises. If the sibling also has MHPs or meets criteria for a psychiatric diagnosis, both children could equally be defined as the target child. In this study, we have retained the definitions used in the included studies and have defined the ‘target child’ as the one who is the primary target of treatment or research and the ‘target sibling’ as the brothers and/or sisters of this child. Control children are similarly defined and the term ‘comparison children’ is used to collectively refer to both children in the general population and control siblings. We

recognise that these are arbitrary definitions and this should be considered alongside the results. Further, it should be noted that based on this definition and the data available, no causal or temporal links between the target child's MHPs and the mental health of the sibling can be inferred.

Articles identified by the search strategy were also screened according to the following exclusion criteria: a) the target child's difficulty is not primarily a MHP, b) purposive sampling of siblings of target children according to the presence or absence of psychopathology was used, and c) the study focused exclusively on twin sibling pairs. The reasons for focusing on non-twin sibling pairs have been outlined above.

The initial search identified 4,387 records after removing duplicates. After screening titles and abstracts, 761 full-text articles were reviewed. The majority were ineligible because the target child did not have a primary MHP and because data on the target sibling was not reported. The final pool of studies consisted of 39 articles based on 24 independent participant samples. Figure 1 summarises the article selection process.

Data Analysis

The main outcomes in this review are prevalence rates of psychopathology in target siblings and variables associated with psychopathology in these siblings (covariates). For studies that segregated data by categories not relevant to this systematic review (e.g. sibling sex or age), the data were pooled into a total sibling prevalence rate. Difference of proportions tests and odds ratios (OR) were calculated or directly extracted from studies.

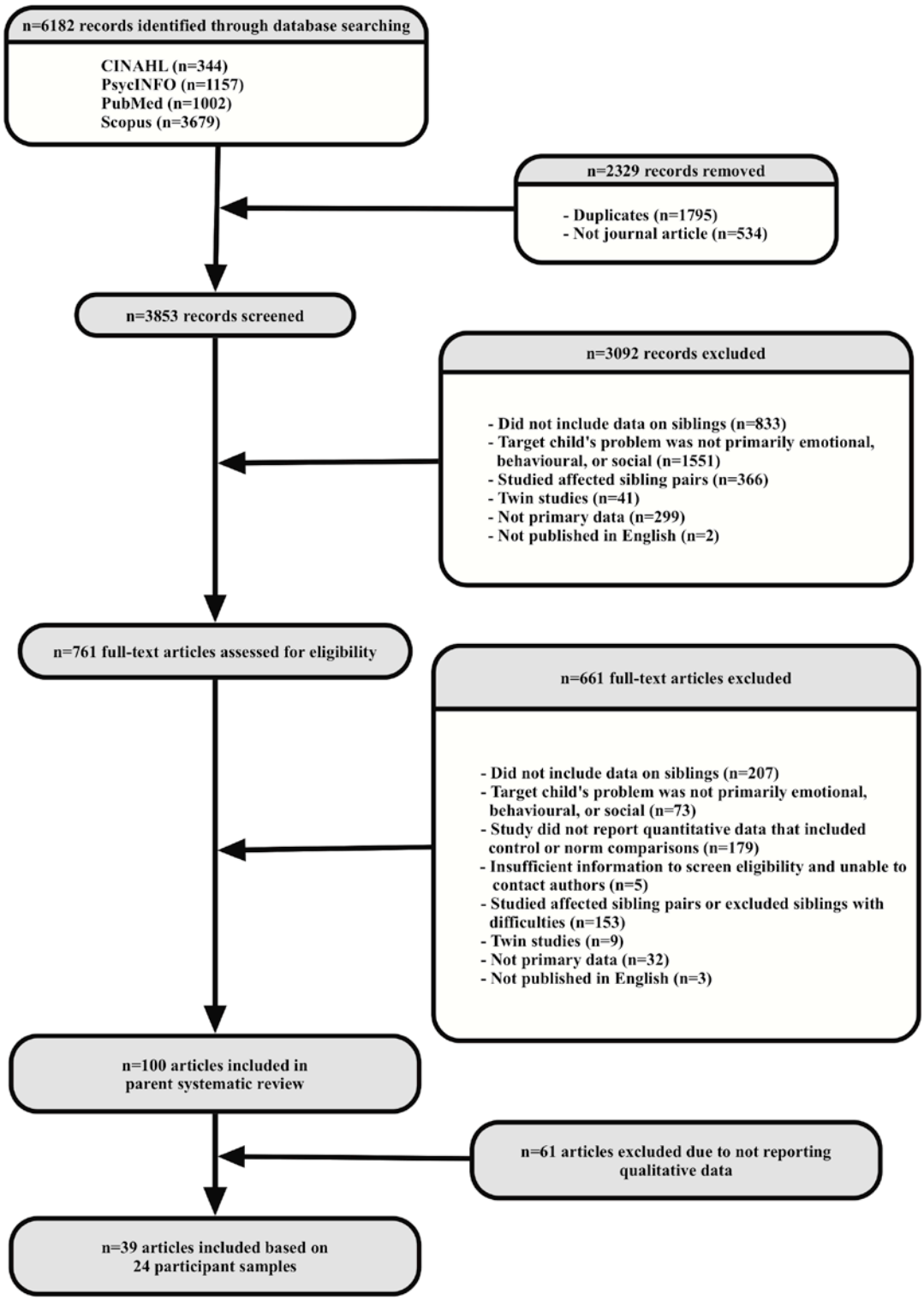

Figure 1. Flow of included studies.

If odds ratios were unavailable (no control group or insufficient data), matched population prevalence rates (on country and sibling age and gender) were used as narrative comparison points. Statistical testing on these comparisons was not conducted due to significant heterogeneity between studies (e.g. large discrepancies in sample size).

A meta-analysis was precluded due to significant heterogeneity across studies and few studies including controls and assessing the same psychiatric disorder in target siblings. This article therefore presents a narrative synthesis with particular emphasis on consistent findings across studies and methodological quality. Quality is assessed on the following factors: Sample size, method of assessing sibling prevalence rates, number and type of measures used, data obtained from one or multiple informants, and generalizability of findings. For brevity, types of MHPs or psychopathology that were assessed by only one study were not included in this paper (e.g. siblings of children with schizophrenia or borderline personality disorder). These data can be obtained from the first author upon request.

Results

Characteristics of Included Studies

This review is based on 39 studies, summarised in Table 1, which assessed approximately 7,278 participants across seven geographic locations. The majority of these studies were conducted in Western countries (33 studies) and recruited target children from mental health treatment programs (34 studies). The size of target sibling samples tended to be small (19 studies; $n<100$) with very few recruiting large samples (3 studies; $n>500$). Studies with larger sample sizes are considered to be of higher quality due to increased generalizability and reliability. In this field, samples sizes of 100 or greater are reasonably large relative to other studies. Approximately half of all

included studies employed a control group of siblings of control children who were matched to the target child's demographics (21 studies with two exceptions as described in Table 1). The age range of target children ranged from 4-18 years. The age range for target siblings was 6-23 years and the mean age of each sample closely matched the mean age of target children. Gender proportions in target children samples were often asymmetrical reflecting gender differences in population prevalence. Gender proportions in sibling samples were, in most cases, balanced.

The majority of studies assessed psychopathology in siblings of target children using only one measure (33 studies) and obtained data from only one informant (25 studies). Prevalence rates have been shown to vary across different types of measures and different informants [35,36]. Studies obtaining data from multiple sources are considered to be of higher quality due to increased accuracy and reliability of diagnoses [37]. Further, studies employing structured diagnostic interviews are considered of higher quality than those using family history reports. The use of structured measures has been found to increase diagnostic reliability whereas family history reports often result in underreporting of psychopathology [38,39].

Prevalence rates or risk of psychopathology for siblings of target children were measured using three different methods: a) assessed all available siblings, b) assessed only one sibling, and c) assessed the number of target children with at least one sibling with psychopathology. The first method examines the entire population of siblings for a given sample of target children. Though still subject to sampling error, this method has the least bias and greatest accuracy of the three. The second method estimates the prevalence of psychopathology by examining one of the target child's siblings, typically the closest in age. This method increases sampling error in that siblings were not randomly chosen. The third method assessed the number of target children with at

least one sibling that meets criteria for a psychiatric diagnosis. It is likely to lead to overestimations as it effectively selects any sibling with a psychiatric diagnosis. By treating the target child's siblings as one unit, it may also lead to underestimations when the target child has multiple siblings with psychiatric diagnoses. Therefore, studies assessing all available siblings are considered higher quality than those using the other two methods. Table 1 highlights the methodological strengths and limitations of the included studies.

Siblings of Children with Attention Deficit Hyperactivity Disorder (ADHD)

Eight studies reported on the prevalence rates of ADHD in siblings of children with ADHD. Prevalence rates ranged from 11.3-44.1%. Higher quality studies based on large sample sizes, assessments of all available siblings, multiple informants and/or structured diagnostic interviews reported ADHD rates of 18.0-44.1% (*n*=143-1,647) [40-43]. All of these studies showed that target siblings had significantly higher odds of ADHD than control siblings (OR=1.9-4.6). All participants were Caucasian and all siblings were full biological siblings of target children. The results may not generalise to other ethnicities and to half- or step- siblings.

Poorer quality studies due to small sample sizes, using the least accurate measure of prevalence, one informant, and/or family history reports [44-47] reported rates ranging from 11.3-29.5% and all were higher than population prevalence rates (0.4-5.3%) [48-50]. Based on the most consistent findings and on the quality of the studies, the rate of ADHD in siblings of children with ADHD is likely between 18.0% and 29.4%.

Table 1

Methodological Characteristics of Included Studies

Reference #	Target child MHP	Sibling		Key methodological strengths (S) and weaknesses (W)
		n	Age (y)	
75	ADHD	--	5-18	(S): Diagnostic interview (PACS);
81		1135		Assessed all siblings; Large *n*
44		1647		(W): All Caucasian sample; Recruited
52		1531		target children with ADHD combined type only; No control group; Parent-report only
40	ADHD	--	12.9	(S): Diagnostic interview (DICA; DISC;
51		31		SADS); Assessed all siblings; Control
54		113	12.6	group (SC; *n*=33-143); Multiple
79		174		informants (P/S)
77		106		(W): All Caucasian sample; All male
41		169-174	15.6	target children (except [26] which
80		174		recruited all female target children)
76		152	17.1	
82		169-174		
83		169-174		
42		143	14.0	
55	ADHD	51	15.5	(S): Diagnostic interview (SADS);
	Bipolar	95	15.7	Multiple informants (P/S); Assessed all siblings; Control group (SC; *n*=109)
				(W): Excluded target with ADHD inattentive or SUDs;
45	ADHD	409	-	(S): Multiple informants (P/S); Large *n*; Assessed all siblings
				(W): FH only; All Caucasian sample
46	ADHD	-	-	(S): Multiple measures (SADS; Wender Utah Rating Scale); Multiple informants (P/S)
				(W): No control group; Assessed % of targets with sibling

Table 1 (cont.)

Methodological Characteristics of Included Studies

Reference #	Target child MHP	Sibling		Key methodological strengths (S) and weaknesses (W)
		n	Age (years)	
43	ADHD	93	-	(S): Multiple measures (DISC; CRS; BASC); Multiple informants (P/T); Assessed all siblings (W): No control group; Only prepubertal target children
56	ADHD ANX	36 86	5+	(S): Diagnostic interview (SCID; SADS); Multiple informants (P/S); Assessed all siblings; Control group (SC; *n*=67) (W): FH from parents for siblings not interviewed
47	ADHD	77	4-18	(S): Control group (C; *n*=14) (W): Symptom scale (CSI); Parent report only; Prepubertal target children only; Assessed sibling closest in age
57	CD	9	-	(S): Multiple measures (DIS; FH); Multiple informants (S/C); Assessed all siblings; (W): Required target have SUD; Small *n*; No control group
59	CD/ODD SUD	14 41	-	(S): Multiple informants (P/C) (W): FH only; Assessed % of targets with sibling; Small *n*; Adolescent targets only; No control group
63 62	Delinquent	126 69	6-10	(S): Diagnostic interview (DISC) (W): Parent report only; Assessed random sibling; Adolescent targets and younger prepubertal siblings only; All male target and sibling sample; No control group
66	Affective	86 45	13.2 12.5	(S): Diagnostic interview (SADS); Multiple informants (P/S); Assessed all siblings; Control group (SC; *n*=77) (W): Prepubertal target children only

Table 1 (cont.)

Methodological Characteristics of Included Studies

Reference #	Target child MHP	Sibling		Key methodological strengths (S) and weaknesses (W)
		n	Age (years)	
64	DEP	10	18.17	(S): Diagnostic interview (SADS; SCID); Multiple informants (P/S); Control group (SC; *n*=6) (W): Adolescent targets only; Small *n*
29	ANX	65	6-18	(W): FH only; Parent report only; Assessed sibling closest in age; Assessed % targets with sibling; No control group
30	OCD	123	-	(S): Multiple measures (SADS; SCID; STOBS; FH; Clinical records); Multiple informants (P/C/S); Large *n*; Assessed all siblings; Control group (SC; *n*=62) (W): Control target group not free of psychopathology
32	OCD	-	-	(S): Multiple informants (P/C) (W): FH; Assessed % targets with sibling; No control group
28	OCD	58	6-23	(S): Multiple measures (SADS; DICA; FH; Clinical observations); Multiple informants (P/S); Assessed all siblings (W): Small *n*; Raters not blind to OCD status of target for sibling OCD rates; No control group
68	OCD	53	12.3	(S): Multiple measures (DICA; Y-BOCS; LOI); Multiple informants (P/S); Assessed all siblings; Control group (SC; *n*=61) (W): Small *n*; Raters not blind
31	OCD	-	-	(W): Review of clinical records only; Assessed % targets with sibling; No control group

Table 1 (cont.)

Methodological Characteristics of Included Studies

Reference #	Target child MHP	Sibling		Key methodological strengths (S) and weaknesses (W)
		n	Age (years)	
72	CD/SUD	173	17.5	(S): Diagnostic interviews (DISC; CIDI-SAM); Large *n* (W): Sibling report only; Adolescent targets only; No control group
71	CD/SUD	315	-	(S): Diagnostic interviews (DISC; DIS;
73		245	16.9	CIDI-SAM); Large sample; Control group (SC; *n*=163-307); (W): Assessed sibling closest in age; Adolescent targets only
65	DEP/ANX	31	8-17	(S): Diagnostic interviews (SADS); Multiple informants (P/S); Assessed all siblings; Control group (C; *n*=40) (W): Small *n*
78	ADHD/CD/ Affective	-	-	(S): Multiple informants (P/S/T/C); Assessed all siblings (W): Behavior checklist based on DSM-III criteria, unknown validity and reliability; No control group; Examined concordance only
74	ADHD/CD/ ODD	41	9.51	(W): Emory Diagnostic Rating Scale, unknown validity and reliability; Parent-report only; Local twin sample as comparison; Small *n*

Note. Type of mental health problems (MHP): ADHD: Attention Deficit Hyperactivity Disorder; ANX: Anxiety disorders; CD: Conduct Disorder; DEP: Depressive disorders; OCD: Obsessive-Compulsive Disorder; ODD: Oppositional Defiant Disorder; SUD: Substance Use Disorder; Control group: SC: Siblings of control children matched to target child demographics; C: Control children matched to target child demographics; Measure: BASC: Behavioral Assessment Scale for Children; CIDI-SAM: Composite International Diagnostic Interview-Substance Abuse Module for DSM-IV; CSI: Child Symptom Inventory; DICA: Diagnostic Interview for Children and Adolescents; DIS: Diagnostic Interview Schedule; DISC: Diagnostic Interview Schedule for Children; FH: Family History report; LOI: Leyton's Obsessional Inventory; PACS: Parental Account of Childhood Symptoms; SADS: Schedule for Affective Disorders and Schizophrenia; SCID: Structured Clinical Interview for DSM-III-R; Y-BOCS: Yale-Brown Obsessive Compulsive Scale; Informant: P: Parent; T: Teacher; C: Target child; S: Sibling

Five studies examined the prevalence of CD/oppositional defiant disorder (ODD) in siblings of children with ADHD. All of the studies were of good quality employing large sample sizes, assessments of all available siblings, multiple informants, and/or structured diagnostic interviews [44,51,52]. Ranges of prevalence rates were as follows: CD (7.0-11.0%), ODD (15.5-30.0%), and CD/ODD (17.1-25.8%). In comparison to population prevalence rates, the rates of ODD and CD/ODD in target siblings were considerably higher (v 2.7% and 5.8%, respectively) [48]. In comparison to control siblings, siblings of male target children with ADHD had significantly higher rates of CD and ODD (OR=2.7 and 2.4-3.1, respectively). There was one exception: Target siblings with a mean age of 11-12 years did not significantly differ from controls on rates of CD. However, this sample had not yet passed the median age of onset for CD (11.6 years) [53]. Four years later, when the mean age of the sample had increased, the difference was statistically significant. Siblings of female target children had similar prevalence rates to siblings of male targets and were almost twice as likely to have CD and ODD than control siblings. However, the difference was not statistically significant [42].

Only two studies examined the prevalence of substance use disorders (SUDs) in siblings of children with ADHD and both studies drew upon the same participant pool (*n*=152-174) [41,44]. Both studies were of good quality employing large sample sizes, assessments of all available siblings, multiple informants, and structured diagnostic interviews. Prevalence rates were as follows: Alcohol use disorders (3.0-11.0%), drug use disorders (1.0-17.0%), and any alcohol or drug use disorder (9.0-17.0%). Similar rates were reported for control siblings and no significant difference was found between groups.

Four studies examined the prevalence and risk of affective disorders in siblings of children with ADHD. Three of these were of high quality using structured diagnostic interviews, multiple informants, assessments of all siblings, and large sample sizes [41,54,55]. The reported rates were as follows: Depression (6.2-10.0%), recurrent depression and/or bipolar disorder (0.9-19.6%), and dysthymia (0.9-4.0%). All of these studies reported no significant difference between target siblings and control siblings. There was one exception: Target siblings of mean age 15-16 years had significantly higher rates of depression than controls (OR=2.26) [41]. The remaining study [45] found that target siblings did not have higher rates of depression (0.5%) or bipolar disorder (0%) compared to population prevalence rates (0.9%) [48]. These findings were based on family history reports from parents and siblings and may have underestimated the rate of affective disorders.

Only two studies examined the prevalence of anxiety disorders in siblings of children with ADHD. Both studies used similar methodologies but varied in the anxiety disorders assessed. In a small study, target siblings (*n*=36) did not significantly differ from control siblings in the rate of anxiety disorders (8.3% v 10.8%, respectively) [56]. In a larger study, target siblings (*n*=169-174) significantly differed from control siblings on rates of social phobia, separation anxiety disorder, and on having two or more anxiety disorders (OR=2.4-3.6, 3.4-3.5, and 2.6, respectively) [41]. For all other types of anxiety disorders, siblings of children with ADHD did not significantly differ from control siblings.

Siblings of Children with CD/ODD or SUDs

Two studies examined the prevalence of psychopathology in siblings of children with CD/ODD or SUDs. Both had methodological limitations. The first study reported a rate of 77.8% for alcohol dependence for siblings of adults with childhood CD (*n*=9)

[57]. It should be noted that the target adults with childhood CD were also required to have a history of early onset alcoholism (<25 years). Given that there is a high degree of familial aggregation for SUDs, it is likely that the rate of alcohol dependence in this sample was elevated [58]. The second study assessed the proportion of target children with a sibling with SUDs [59]. Of target children with CD/ODD and with SUDs, 28.0% and 24.0% respectively, had siblings with SUDs. Across these studies, the prevalence rates were substantially higher than population prevalence rates (11.4-12.5%) [60,61]. The second study also assessed ADHD, CD/ODD, and affective disorders. Of target children with CD/ODD, 7.0%, 50.0%, and 7.0-21.0% had siblings with ADHD, CD/ODD, and affective disorders, respectively. The proportions for target children with SUDs were 10.0%, 36.0%, and 6.0-10.0%, respectively. Only percentages of CD/ODD across these groups were higher than population prevalence rates (19.6%) [61]. However, it is difficult to compare these figures to population prevalence rates, as the outcomes are not directly comparable. Population prevalence estimates the number of children in the population with the particular problem while this study examined the number of targets with a sibling with the particular problem.

Siblings of Children Adjudicated for Delinquency

Two studies, drawing on the same participant pool, examined the prevalence of psychopathology in siblings of children adjudicated for delinquency [62,63]. Both had methodological limitations and were limited in generalizability to younger brothers currently living with male target children. The prevalence rates in target siblings (*n*=69-129) were as follows: ADHD (11.6-21.4%), CD (2.9-3.2%), ODD (8.7-11.9%), affective disorders (1.4-2.0%), and anxiety disorders (2.9-31.0%). Lower estimates were reported by Pine and colleagues [62] whose sample had significantly lower ratings on mental health symptoms due to additional medical exclusion criteria. Therefore, the

findings from Wasserman and colleagues [63] are more reliable. Across both studies, target siblings had higher rates of psychopathology compared to population prevalence rates [49]. However, rates for affective disorders were equivalent to population prevalence rates (2.0%) [49].

Siblings of Children with Affective Disorders

Four studies examined the prevalence of psychopathology in siblings of children with affective disorders. The prevalence of affective disorders ranged from 6.2-60.0% showing considerable variability across studies. The highest estimate was based on only 10 siblings and may not be reliable [64]. Another study based on a small sample size (*n*=31), reported the lower estimates in the prevalence range[1] [65]. Higher quality studies that recruited larger sample sizes and matched control groups reported less variable rates [55,66]. Rates for affective disorders were consistently within 18.6-24.7% (except recurrent depression: 6.2%). These studies found significantly higher rates of affective disorders (except recurrent depression only) in target siblings than control siblings. Hazard ratios ranged from 3.5-7.2.

Rates of other psychopathology were assessed by the two small studies described above [64,65]. The prevalence rates reported were as follows: ADHD (13.0%), CD (16.0%), SUDs (30.0%), and anxiety disorders (6.5-20.0%). All reported rates for target siblings were higher than control siblings and population prevalence rates [48,49]. Due to limited data, no firm conclusions can be made.

Siblings of Children with Anxiety Disorders

Seven studies examined the prevalence of anxiety disorders in siblings of children with anxiety disorders. Prevalence rates for all anxiety disorders ranged from

1 This study included target children with anxiety disorders. However, the sample was predominantly children with depression (21 v 8 children) and was therefore included here.

0.0-21.7%. Higher quality studies employing multiple measures and recruiting multiple informants and all available siblings reported the highest prevalence rates for OCD (14.6%) [30] and for any anxiety disorder (21.7%) [56]. The rate of OCD was five times greater than population prevalence rates (2.5%) [67] and the rate of anxiety disorders was double that of control siblings (10.8%) [56]. However, for the latter, the difference was not statistically significant. This study had a small sample size (n=36) and power may not have been sufficient. Two other good quality studies based on the use of multiple measures and data from multiple informants, though also limited in sample size, reported considerably lower rates. The first, conducted by Lenane and colleagues [28] reported rates for OCD (5.0%) and other anxiety disorders, such as separation anxiety disorder (0.0-2.0%). The rate of OCD was double that of the general population [67] but all other anxiety disorder rates were less than population prevalence rates [61]. The second study, conducted in India, found that in both target sibling and control sibling samples, none met criteria for OCD or subthreshold OCD [27]. However, research suggests that Indian populations have lower rates of OCD than Western countries and thus prevalence differences across countries may account for this divergent finding [68].

The remaining studies relied on family history reports or clinical records [29,31,32]. Three studies reported rates of OCD (4.6-6.0%) that were consistent with Lenane and colleagues' [28] study. Across four studies then, there was significant consistency and little variance in the rate of OCD. For other anxiety disorders, rates ranged from 0.0-14.3%. The highest rates were for any anxiety disorder (12.3-14.3%) with rates for other disorders falling between 0.0-3.1%. All of these reported rates were lower or equal to population prevalence rates [49,61,69].

In sum, based on methodological rigour, the rate of OCD in target siblings is 14.6%. However, based on consistent findings, the rate of OCD in target siblings is 4.6-6.0%. Although the studies reporting the latter prevalence rates are poorer in quality, the consistency across four studies cannot be ignored. Consistently across all studies, target siblings did not have significantly higher rates of other anxiety disorders than comparison children.

Few studies examined the prevalence of other forms of psychopathology in siblings of children with anxiety disorders and few types of psychopathology were examined. Three studies examined the rate of affective disorders in target siblings. Two poorer quality studies, as previously described, reported lower rates of depression than the general population (3.6-4.6%) [29,32]. In contrast, Lenane and colleauges' [28] higher quality study, though limited by sample size, reported a higher rate of affective disorder in target siblings than population prevalence rates (18.0% v 14.3%) [61]. However, this figure may be an artefact of the inclusion of adult siblings in this study. Half of the siblings with depression were over 18 years of age. Using data from only the child and adolescent siblings, the prevalence rate drops to 11.1% and is no longer higher than the population prevalence rate. This study also assessed the rates of ADHD, CD, and ODD in target siblings and the rates were 9.0%, 7.0%, and 13.0%, respectively. All were approximately equal to population prevalence rates [70]. Findings on other types of psychopathology are too limited to form any conclusions.

Siblings of Children with Comorbid MHPs

While other studies did not necessarily preclude comorbidity, the following studies are reported separately as they purposively examined these comorbidities. Three studies examined the prevalence of psychopathology in siblings of children with comorbid CD+SUDs. All three studies were similar in quality and methodology, though

the findings are limited to siblings of adolescent targets [71-73]. Rates of abuse disorders ranged from 20.0-40.0% and rates of dependence ranged from 9.5-20.0%. Reported rates were higher than population prevalence rates [49]. Compared to control siblings, target siblings had significantly higher rates of marijuana abuse and dependence and alcohol use disorder (OR=2.3-2.7), but not alcohol dependence. Rates of CD ranged from 33.3-38.0% across studies. These rates were higher than population prevalence rates [49] and were significantly higher than control siblings (OR=3.9).

One study, conducted by Waldman and colleagues [74], examined the prevalence of psychopathology in siblings of children with mixed behavioural problems (*n*=41). The prevalence rates were as follows[2]: ADHD (22.0%), CD (7.0%), and ODD (17.0%). Compared to a local twin sample representative of the geographic area, target siblings had significantly higher rates of ADHD, CD, and ODD (OR=2.9, 7.4, and 3.9, respectively). However, there were a greater proportion of males in the target sibling group than the control group. Given these forms of psychopathology are more prevalent in males [61], the findings may be an artefact of gender differences across the target and control group.

Findings on Covariates

Genetic variables. Three studies examined associations between genes and the prevalence of psychopathology in siblings. First, Waldman and colleagues [74] reported a statistically significant association between the DAT1 allele, a dopamine transporter gene, and ADHD diagnoses within families of children with behavioural problems. This is particularly relevant to ADHD diagnoses as pharmacological treatments for ADHD, in part, take their effect by inhibiting the dopamine transporter [74]. Second, Sakai and colleagues [73] found no significant association between the GABRA2 gene and

[2] Prevalence rates were reported separately according to the severity of the MHP assessed. Only high severity rates are reported here.

alcohol dependence and CD within families of children with CD+SUDs. Both of these studies employed within-family association tests and aggregated all 'affected' children. Altink and colleagues [75] was the only study to test the association between genes and the mental health of the sibling separately. They found that the *DRD4* gene, which has been linked to impulsivity traits such as novelty seeking, prenatal smoking, or the interaction between these did not significantly predict ADHD in siblings of children with ADHD after controlling for the age and gender of the sibling.

Target child and sibling characteristics. Four studies examined the relationship between the target child's age and gender and the prevalence of psychopathology in siblings of children with ADHD [46,56] and siblings of children with OCD [31,32]. All studies reported no significant relationship.

Two studies examined the role of the sibling's age. Both found that younger aged target siblings had significantly greater risk of psychopathology [65,75]. Eight studies investigated the role of the target sibling's gender. For siblings of children with ADHD, male siblings were significantly more likely to have ADHD and SUDs compared to female siblings (OR=2.8-3.6) [40,56,75-77]. In contrast, no gender effect on the prevalence of psychopathology was found for siblings of children with affective disorders [65,66]. Two studies found differential gender effects depending on the type of psychopathology assessed. Lenane and colleagues [28] found that male siblings of children with OCD had higher rates of CD, ODD, and OCD. No gender differences were observed for ADHD and affective disorders. Hopfer and colleagues [71] found for marijuana abuse, both male and female siblings of children with CD+SUDs had significantly greater risk of marijuana abuse compared to control siblings. However, female siblings had higher risk ratios than male siblings (3.1 compared to 2.2, respectively). For marijuana dependence, risk ratios across male and female siblings

were similar (2.2 and 2.3, respectively). However, only male siblings had significantly greater risk than control siblings. Therefore, the effect of gender on the prevalence of psychopathology in target siblings varies across the type of MHP in the target child and across the type of psychopathology assessed in the sibling.

Szatmari, Boyle, and Offord [78] investigated the association between the gender and age composition of sibling pairs (*n*=1,030) and the concordance of MHPs or psychiatric diagnoses. They found that the age composition (older or younger than 12 years) of sibling pairs was not associated with the concordance of ADHD, CD, or affective disorders. In contrast, the gender composition was associated with the concordance of ADHD and affective disorders, but not CD. Male-male sibling pairs had the lowest aggregation of ADHD and affective disorders. Mixed sibling pairs and female-female sibling pairs had the greatest aggregation of ADHD and affective disorders, respectively. The size of the sibship was also associated with the concordance of psychopathology. Sibships of four or more children, compared to two or three, was associated with a slight increase in the concordance of ADHD and affective disorders, but not CD.

Last, Lenane and colleagues [28] found that for siblings of children with OCD, increased emotionality increased the likelihood that the sibling had a MHP. No relationship was found between sibling psychopathology and other temperament scales (activity, sociability, or shyness).

Type of MHP in target child. Several studies found that siblings of target children had higher rates of the same type of MHP or psychopathology as the target child compared to siblings of children with other MHPs. This pattern of concordance was found for siblings of children with ADHD (v those with anxiety disorders) [56], siblings of children with anxiety disorders (v those with ADHD) [56], siblings of

children with affective disorders (v those of children with non-affective disorders, with CD, or with ADHD) and those of children with CD (v those of children with ADHD or with affective disorders) [66,78]. However, other studies report no evidence of concordance. This was found for siblings of children with ADHD compared to siblings of children with a range of other MHPs [40] and compared to siblings of children with CD and affective disorders [78]. Further, Hovens and colleagues [59] found that siblings of children with SUDs and those of children with CD/ODD did not significantly differ in the rates of SUDs or CD/ODD. These discrepancies may be in part due to variations in the type of MHP of the target child, the type of psychopathology assessed in the sibling, and the nature of the comparison group. Asides from specifically examining concordance, Hovens and colleagues [59] also found that siblings of children with CD/ODD had significantly higher rates of affective disorders (other than major depression) compared to siblings of children with SUDs.

Several studies also examined the relationship between comorbid problems in the target child and sibling psychopathology. However, the findings are inconsistent across studies. Two studies found that ADHD with comorbid CD/ODD or affective disorders in target child was not associated with increased rates of ADHD, CD, or ODD in their siblings compared to ADHD only [42,79]. In contrast, Christiansen and colleagues [44] found significantly higher rates of ADHD and CD/ODD in siblings of children with ADHD+CD/ODD compared to ADHD only. Further, Faraone and colleagues [42,80] found that siblings of children with ADHD+CD had significantly higher rates of CD and ODD compared to siblings of children with ADHD only and ADHD+ODD.

Last, three studies examined the relationship between the target child's symptoms and the prevalence of psychopathology in their siblings. Lenane and

colleagues [28] found no consistent pattern across the specific OCD behaviours in parents, target children, and siblings of children with OCD. This suggests that OCD behaviours are not directly modelled or learned across family members. The two remaining studies found no significant difference in the prevalence of ADHD in target siblings according to the severity of the target child's ADHD symptomatology [52,81].

Family History of Psychopathology. Five studies examined the association between psychopathology in parents and in the sibling. Biederman and colleagues [54] found no significant association between the prevalence of affective disorders in siblings of children with ADHD and the presence of affective disorders in their parents. In contrast, other studies reported that parental psychopathology significantly increased the risk of psychopathology in siblings. This was found for parental depression and sibling anxiety, depression, and SUDs [64] and for maternal ADHD and sibling ADHD (OR=9.7) [77]. No effect was found for paternal ADHD. In siblings of children with bipolar disorder, having two parents with bipolar disorder versus having one parent did not significantly increase the risk of bipolar disorder in siblings [55]. Last, Hopfer and colleagues [71] analysed familial transmission models. They found that 25-42% of variance in marijuana abuse and dependence in the target child was attributable to factors transmitted from parent to child. However, they also found significant residual sibling correlations suggesting sibling specific environmental factors contribute to the development of SUDs.

Biederman, Faraone, and colleagues also examined the role of a family history (in either the target child or parent) of antisocial disorders or bipolar disorder. Siblings of children with ADHD with a family history of antisocial disorders or bipolar disorder had significantly higher rates of ADHD, CD, SUDs, major depression, and of having two or more anxiety disorders compared those without this family history (*n*=169-174)

[82,83]. In addition, siblings with a family history of bipolar disorder had significantly higher rates of major depression, bipolar disorder, and separation anxiety disorder compared to siblings with a family history of antisocial disorders [82]. In contrast, a family history of antisocial disorders was not associated with the prevalence of ODD in siblings of children with ADHD [83].

Discussion

This systematic review allows several conclusions to be made about the prevalence of psychopathology in siblings of children with MHPs compared to those of control children or the general population. First, in siblings of children with ADHD, there is consistent evidence of significantly higher rates of ADHD and CD/ODD but no significant difference in rates of SUDs and affective disorders compared to control siblings. Second, siblings of children with affective disorders had significantly higher rates of affective disorders than control siblings. Third, siblings of children with anxiety disorders had consistently higher rates of OCD compared to the general population. Last, siblings of children with CD+SUDs had consistently higher rates of SUDs and CD than comparison children. Therefore, there was a trend for siblings of children with externalising problems and those of children with internalising problems to have concordant difficulties. It should not be concluded however that these siblings are not also at increased risk for other psychopathology. The limited data presented in this review suggests that they are at risk for a range of psychopathology. For example, siblings of children with ADHD had similarly greater odds of having ADHD, CD/ODD, and anxiety disorders compared to control siblings. This is consistent with developmental theorists' concept of multifinality which highlights that similar genetic and family environment backgrounds can lead to multiple different outcomes [3]. No conclusions on the rates of other types of psychopathology could be formed due to

limited data, poor quality studies, and the focus in sibling research on rates of concordance. However, the existing literature suggests that siblings of children with CD/ODD, SUDs, delinquent behaviour problems, and mixed behavioural problems have higher rates of psychopathology than comparison children.

These findings have several important implications. First, the findings of this review highlight an alarmingly high prevalence of psychopathology and high risk compared to control children in siblings of children with MHPs. Odds ratios from higher quality studies indicate that target siblings are two to four times more likely to have a psychiatric diagnosis compared to control children. Twin studies have consistently shown that non-twin siblings have significantly lower concordance rates for MHPs [8]. Within this context, non-twin siblings may be viewed as 'low risk'. However, this can underestimate the real-world impact of increased risk for non-twin siblings. Monozygotic twin births occur in 32 per 1,000 births (0.03% of families) [84] whereas, even by the most conservative definition of non-twin siblings as two or more singleton biological children of parents, approximately 28% of families have two or more children [85]. Therefore, while twin siblings have significantly higher risk of shared psychopathology, given their low base rate, they are less likely to have a significant impact on population prevalence rates. On the other hand, non-twin siblings of children with MHPs are considerably more common and thus, a two-fold or four-fold risk compared to control children can have a significant population-level impact. This highlights the important need for research with this population, particularly regarding etiological pathways to inform prevention and treatment programs. As described in this review, the existing literature base is small and methodologically flawed, which is a serious concern given the high risk of psychopathology and the impact of this on

families, health care systems, and other broader socio-ecological systems (e.g. schools and communities) [1].

The findings of this review also highlight a need for increased clinician and parent awareness of the elevated risk to siblings to allow for early intervention. More specifically, there was a trend for concordant MHPs across target children and siblings, particularly across broad categories of externalising and internalising problems. This suggests parents and clinicians should pay particular attention to behaviours in siblings that are concordant with the target child's MHP. Further, efforts should be made to increase clinician and parent awareness of existing support structures for siblings, such as the Siblings Network, and treatment modalities that are inclusive of siblings with mental health issues, such as family therapy [86].

With regard to variables associated with the prevalence of psychopathology in target siblings, several conclusions can be drawn. First, there is consistent evidence that the age and gender of the target child is not associated with the prevalence of psychopathology in the siblings. Given the problem of circularity (see Methods), we would not expect the target child's age or gender to be associated with the prevalence of psychopathology in their siblings. Because the designation of target child and sibling are somewhat arbitrarily assigned and do not imply a causal or temporal link, we would not necessarily expect to find an association between the gender and age of one of these children to the mental health of the other.

Second, there is consistent evidence that the prevalence of psychopathology in siblings is associated with their age and gender. Younger aged siblings were consistently more likely to have a psychiatric diagnosis than older siblings. This provides support for sibling deviance training theory, as previously described, which holds that older siblings increase younger siblings' exposure to maladaptive behaviours,

deviant peers, and substances and increase peer pressure to engage in similar behaviours resulting in increased rates of psychopathology [26,87]. However, sibling deviance training has historically only been applied to CD, SUDs, and other externalising problems [87]. It could be argued that similar pathways also play a role in the development of internalising problems and other forms of psychopathology. For example, older siblings may model maladaptive cognitive biases, such as negative interpretations of ambiguous social cues, and may increase associations with a peer group that provides socialisation to maladaptive attitudes and behaviours, such as self-harming behaviours, which increases the risk of internalising problems in younger siblings. Research on parent-child relations and peer group relations provide some support for these developmental psychopathology pathways. For example, depressive symptoms in peers predict changes in self-harming behaviours in adolescents [88]. Sibling relationship quality, such as low support and increased conflict, has also been implicated in the development of psychopathology, particularly depressive symptoms [26]. Although this pathway does not imply an increased risk to younger siblings, it could be argued that younger siblings are more developmentally vulnerable to developing MHPs in response to interpersonal stress, such as poor sibling relations. For example, younger siblings may not have developed sufficient coping skills, may not have formed their own peer group and support network, developed a secure sense of self or self-worth, or developed secure attachment styles and therefore, may be more vulnerable to the negative effects of poor sibling relations than older siblings who are likely more developmentally advanced. It may also be that younger siblings are more likely to be victimised than older siblings [13]. However, it should be noted that the relationship between younger and older siblings has also been observed in general population prevalence rates where younger children have higher rates of

psychopathology than older children [1]. This suggests that factors other than those relating to sibling pathways may explain why younger children are at greater risk of psychopathology.

Gender effects varied across the type of psychopathology assessed. These associations between psychopathology and sibling gender match those found in the general population. For example, male siblings were more likely to have ADHD, CD, and ODD compared to female siblings, which mirrors general population gender differences [61,89]. Further, there was a trend for male-male sibling pairs to be more likely to have concordant psychopathology compared to male-female sibling pairs, particularly in the case of ADHD. This may, in part, be explained by variations in what genes influence risk across genders [90]. It may be that high-risk genes for ADHD in males are different from high-risk genes in females. If a male target child has high-risk genes for ADHD, it is likely that any male or female siblings would similarly have these genes. However, the high-risk genes may only increase the risk of ADHD in males. Consistent with this, Szatmari and colleagues [78] found significant differences in concordance according to the gender composition of the target child-sibling pair. Genetic risk likely also interacts with environment factors. For example, females who have a genetic predisposition for ADHD may evoke responses from other people that are particularly restrictive in regards to impulsive or restless behaviour. This may inhibit the expression of high-risk genes. Further, there are diagnostic issues surrounding ADHD including biases towards diagnosing ADHD in males and increasing concerns around overdiagnosis of ADHD that should be considered [89]. These findings suggest that sibling gender but also the gender composition of the sibling pair should be obtained, as is similarly done in twin studies.

Last, the type of MHP in the target child and parental psychopathology were related to sibling psychopathology. However, the association varied according to the type of MHP in the target child, sibling, parent, and the comparison group. There is consistent evidence of familial aggregation and concordance of MHPs or psychiatric diagnoses between target children and siblings and between parents and siblings when compared to controls. This has been similarly found in adult populations and parent-child dyad studies [19,91]. However, when compared to families with other types of psychopathology, a significant association was not always found. Similarly in adult populations, controlling for other psychopathology reduced the effect of familial aggregation [19]. On the basis of such findings and from twin studies, it has been argued that familial aggregation may be explained by a broad genetic vulnerability to psychopathology or shared underlying genetic mechanisms across internalising and externalising forms of psychopathology rather than by the influence of individual genes [19]. The included studies in this review similarly found no conclusive evidence of the role of individual genes in the development of psychopathology.

Beyond this, there were insufficient data to formulate any hypotheses regarding specific pathways for the development of psychopathology in siblings or mechanisms to explain to patterns of risk found in the included studies. Only one study explicitly examined gene-by-environment interactions [75]. Family environment and illness related factors highlighted in research with siblings of children with special needs were also neglected. Only three studies in this review directly examined symptom severity as a possible covariate and all found no significant relationship between the severity of the target child's MHP and psychopathology in siblings. Research with siblings of children with special needs has reported inconsistencies regarding illness severity [18]. It has been hypothesised that illness severity, in and of itself, does not significantly contribute

to MHPs but has indirect effects through its relationship to interference in daily activities [18]. That is, greater severity illnesses, such as cancer, are linked with increased daily interferences due to increased treatment requirements and increased need for supportive care. Research with siblings of children with special needs has confirmed this and has demonstrated a significant relationship between interference in daily activities as a result of the child's disability or treatment and the mental health of siblings [16]. No studies in this review examined this variable.

Research on variables associated with psychopathology in siblings of children with MHPs is lacking, which may be due to the lack of conceptual frameworks that hypothesise causal mechanisms for psychopathology in siblings of children with MHPs. As highlighted in previous sections, several models have been developed in related fields, and although these are not entirely applicable to siblings of children with MHPs, future research on variables associated with the risk of psychopathology in this population should draw upon these models to explore if similar mechanisms are applicable to this population. Each of these aforementioned models (i.e. developmental psychopathology frameworks and impact of illness frameworks) has been linked with psychopathology in children in general (see [3] for a summary of recent findings). Furthermore, the interplay between risk factors described in these models has been linked with psychopathology in children. For example, interpersonal stress and individual physiology (e.g. cortisol reactivity) interact in the development and recurrence of depressive symptoms in adolescents [92]. While conclusions regarding causal mechanisms cannot be formed in this review, it is likely that these factors would play a role in the development of psychopathology in siblings of children with MHPs, as they do in children in general. Therefore, future research should draw upon these models to explore the mechanisms underpinning psychopathology in siblings. What is

gained from focusing on siblings, however, is the discovery of sibling-specific pathways, such as the quality of the sibling relationship and sibling deviance training, in the development of psychopathology. In addition, focusing on siblings of children with MHPs may contribute to our understanding of how similar genetic and shared environment factors, as is shared between siblings, may lead to different outcomes. That is, what factors determine whether or not a child develops MHPs despite similar genetic backgrounds and similar family environments? Therefore, we recommend that future work with siblings focuses on exploring these areas and pathways to the development of psychopathology.

Methodological Limitations of Sibling Studies

This systematic review also highlights several methodological issues in the existing literature. First, no consistent methodology was applied across studies limiting the comparisons and conclusions that could be derived from the data. This was particularly problematic with regard to methods used to assess the prevalence of or risk of psychopathology in siblings of target children. Many studies used the two least accurate and reliable methods of determining the prevalence of psychopathology in target siblings. Second, most studies used only one measure of psychopathology and relied on data from only one informant. Further, several studies did not directly assess psychopathology in siblings but relied on family history reports of their psychiatric history. These methods can lead to significant underreporting, variations in prevalence rates across informants, and variations in prevalence rates across studies [39,93]. Third, less than half of the included studies reported data from a matched control group of siblings. As a result, this review is limited in the conclusions it can make regarding significant differences between siblings of children with MHPs and those of children without MHPs. Fourth, the majority of studies recruited target children from mental

health treatment programs. Given that a substantial proportion of individuals with MHPs do not receive treatment [94], prevalence rates reported in the included studies likely underestimate population prevalence rates. Fifth, most studies focused on examining the rate of concordant MHPs. While there is evidence of concordance between the MHP of the target child and that of the sibling, the findings of this review show that siblings are at risk of other types of psychopathology also. Therefore, ideally the rates of most psychopathology should be assessed. Last, when assessing SUDs, anxiety disorders, and affective disorders, studies varied in the diagnostic levels assessed which limited comparisons across studies. For example, some studies assessed most types of affective disorders, some assessed only one or two, and some assessed rates of affective disorders as a broad category. Studies in this review have found that siblings of children with MHPs do not significantly differ from controls on the rate of having any anxiety disorder, for example, but do significantly differ on rates of specific anxiety disorders [29,41,56]. This highlights the importance of measuring both the broad category disorder as well as the individual disorders.

To address these methodological issues and variations in methodology, we suggest a list of guidelines to be followed in future research within the context of resource limitations. First, ideally data on all available siblings should be obtained. Failing this, data should be obtained on a *randomly* selected sibling. Second, at a minimum, data should be obtained from multiple informants and, ideally, using multiple measures and combining the data. Third, structured diagnostic interviews or other standardised measures should be used. Family history reports should only be used to provide additional data. Last, prevalence rates should be assessed for a broad range of psychopathology. When assessing those with numerous sub-category specific

disorders (e.g. anxiety disorders), both the rate of the specific disorders and the overarching disorder should be assessed.

Limitations

There are several limitations of this review that should be considered when interpreting the results. First, where control groups were not employed, prevalence rates in siblings of children with MHPs were compared to general population prevalence rates. These were not directly matched on age, gender proportions, and SES as would occur with a control group. It was considered inappropriate to statistically compare the prevalence rates across these groups due to significant sample size differences as well as other methodological variations. Therefore, for these studies, only narrative comparisons between siblings of children with MHPs and the general population could be made. Further, research has shown that comparisons to control groups may reduce effect sizes thus the findings in this review may underestimate the odds or risk of psychopathology in siblings [16]. Second, as previously discussed, labelling one child as the target child and labelling the other as the sibling is problematic given the problem of circularity. Further, this labelling may be seen as implying a causal or temporal link between the target child's MHPs and the siblings' mental health. However, this cannot be inferred from the existing literature as no prospective, longitudinal studies or retrospective studies taking into account age or order of onset have been conducted. Third, the heterogeneity of included studies and limited number of studies in this area precluded conducting a formal meta-analysis. Fourth, as this review focused on psychopathology rates, it relied on a dichotomous approach to assessing the mental health of siblings. Although it should be noted that a strength of this review is that the criteria set for the target child having MHPs included a broad range of assessment approaches, including clinically elevated scores on continuous or dimensional

psychological measures. Therefore, this limitation applies only to the rates of psychopathology in siblings reported in this review. This approach has several limitations including the use of artificial cut-offs (such as the number of symptoms that need to be present to receive a psychiatric diagnosis) and restrictions in the amount of clinically relevant data that can be obtained [95]. However, the findings of this review are consistent with a review conducted on published data using a continuous or dimensional approach to mental health assessment [96]. Siblings of children with MHPs were found to have significantly greater impairment on continuous measures of externalising and internalising problems, psychiatric symptom scales, and social scales than control siblings of children without MHPs [96]. Although it should be noted that the studies included in that review were similarly limited by methodological issues and tended to focus on siblings of children with ADHD and externalising problems [96]. Last, the search strategy used in this review could not identify potentially relevant unpublished findings.

Future Directions

This review highlights a lack of comprehensive and methodologically robust research with siblings of children with MHPs. Such research is clustered within research on ADHD with very little research with siblings of children with other types of MHPs. Addressing this is considered the main priority for research in this area. Further research is also needed on factors associated with the prevalence of psychopathology in target siblings. Of particular interest are genetic studies that explicitly examine gene-by-environment interactions and studies designed to test theorised developmental psychopathology pathways, in particular sibling-specific pathways. We also suggest appealing to studies with siblings of children with disabilities or special needs for guidance on variables that may be worthy of investigation in studies with siblings of

children with MHPs. Further, prospective, longitudinal, or retrospective research that is able establish temporal relationships between MHPs in siblings – that is, identify the first child in the family to have MHPs is needed. Establishing a temporal link is essential before causal links can be explored. It may be that one child develops MHPs and their sibling later develops MHPs. This research may reveal causal factors specifically related to having a brother or sister with MHPs that contribute to the development of psychopathology in siblings. For example, living with a child prone to violent outbursts may lead to hypervigiliance and the development of an anxiety disorder in the sibling. Alternatively, there may be no temporal link (i.e. onset occurs at the same time in the child and sibling) suggesting that shared environmental factors that occur at a certain time point, such as marital separation, play a prominent role in the development of psychopathology rather than sibling-specific pathways. Lastly, no research to date has explored if siblings meeting criteria for psychiatric diagnoses receive support or treatment. This is a particularly important issue considering the elevated risk for these siblings.

Summary

Based on the data reported in 39 studies over the past 20 years, this systematic review has highlighted the elevated risk of psychopathology for siblings of children with MHPs. However, this elevated risk compared to control siblings was more conclusively shown for siblings of children with ADHD where the vast majority of the research has been clustered. Some conclusions could be formed for siblings of children with affective disorders, anxiety disorders, and comorbid CD+SUDs. However, these were limited to the rate of concordant MHPs in the siblings. More comprehensive research is needed on siblings of children with mental health problems other than ADHD.

Additional research on variables associated with the prevalence of MHPs in target siblings is also needed. What could be concluded on the basis of the existing literature is that the target child's age and gender was not associated with the prevalence of MHPs in their siblings. In contrast, the age and gender of the sibling and a family history of MHPs were associated with prevalence rates, though the association varied depending on the MHPs in the target child, in the sibling, and in the comparison group. The variables explored were very limited and more research is needed, particularly research exploring etiological pathways such as gene-by-environment interactions.

In addition to a pervasive scarcity of research, there was considerable variation in methodology used across the existing studies with siblings of children with MHPs. Further, a large proportion of these studies had important methodological limitations. This limited the conclusions that could be formed from the existing literature. In line with this, a list of recommended guidelines has been outlined for future research with siblings to encourage methodologically robust studies and consistent methodologies across studies. Other suggestions for future research were outlined. While research on siblings of children with MHPs has begun to emerge, pointing to an elevated risk of psychopathology, there is far more that is yet to be explored and more to be known in order to effectively support these siblings.

References

1. Sawyer MG, Arney FM, Baghurst PS, Clark JJ, Graetz BW, Kosky RJ,…Zubrick SR (2000) The mental health of young people in Australia. Mental Health and Special Programs Branch, Commonwealth Department of Health and Aged Care, Australia
2. Onstead S, Skre, I, Torgersen S, Kringlen E (1991) Twin concordance for DSM-II-R schizophrenia. Acta Psychiatr Scand 83:395-401
3. Parritz RH, Troy MF (2011) Disorders of childhood: Development and psychopathology (2nd edition). Wadsworth, Cengage learning, USA
4. Wermter AK, Laucht M, Schimmelmann BG, Banaschweski T, Sonuga-Barke EJS, Rietschel M, Becker K (2010) From nature versus nuture, via nature and nuture, to gene x environment interaction in mental disorders. Eur Child Adolesc Psychiatry 19:199-210
5. Greenberg BD, Tolliver TJ, Huang SJ, Li Q, Bengel D, Murphy DL (1999) Genetic variation in the serotonin transporter promoter region affects serotonin uptake in human blood platelets. Am J Med Genet 88:83-87
6. Retz W, Freitag CM, Retz-Junginger P, Wenzler D, Schneider M, Kissling C, Thome, J, Rösler M (2008) A functional serotonin transporter promoter gene polymorphism increases ADHD symptoms in delinquents: Interaction with adverse childhood environment. Psychiatry Res 158:123-131
7. Dumont M, Provost MA (1999) Resilience in adolescents: Protective role of social support, coping strategies, self-esteem, and social activities on experience of stress and depression. J Youth Adolesc 28:343-363
8. Ehringer MA, Rhee SH, Young S, Corley R, Hewitt JK (2006) Genetic and environmental contributions to common psychopathologies of childhood and adolescence: A study of twins and their siblings. J Abnormal Child Psychology 34:1-17

9. Lombroso PJ, Pauls DL, Leckman JF (1994) Genetic mechanisms in childhood psychiatric disorders. J Am Acad Child Adolesc Psychiatry 33:921-938
10. Thorpe K, Golding J, MacGillivray I, Greenwood R (1991) Comparison of prevalence of depression in mothers of twins and mothers of singletons. BMJ, 302:875-878
11. Rowe DC (1983) A biometric analysis of perceptions of family environment: A study of twin and singleton sibling kinships. Child Dev 54:416-423
12. DiLalla LF, Millineaux PY, Elam KK (2008) Twins. In: Haith MM, Benson JB, editors. Encyclopedia of infant and early childhood development. Academic Press, San Diego, USA
13. Kendall J (1999) Sibling accounts of attention deficit hyperactivity disorder. Fam Process 38:117-136
14. Bronfenbrenner U (1979) The ecology of human development. Harvard University Press, Massachusetts, USA
15. Hoffman L (1981) Foundations of family therapy: A conceptual framework for systems change. Basic Books, New York, USA
16. Vermaes IP, van Susante AM, van Bakel HJ (2012) Psychological functioning of siblings in families of children with chronic health conditions: A meta-analysis. J Pediatr Psychol 37:166-184
17. Wallander JL, Varni JW (1992) Adjustment in children with chronic physical disorders: Programmatic research on a disability-stress-coping model. In: LaGreca AM, Siegal L, Wallander JL, Walker CE, editors. Stress and coping in child health. Guildford Press, New York
18. Sharpe D, Rossiter L (2002) Siblings of children with a chronic illness: A meta-analysis. J Pediatr Psychol 27:699-710

19. Kendler KS, Davis CGD, Kessler RC (1997) The familial aggregation of common psychiatric and substance use disorders in the National Comorbidity Survey: A family history study. Br J Psychiatry 170:541-548
20. McLaughlin KA, Gadermann AM, Hwang I, Sampson NA, Al-Hamzawi A, Andrade LH, et al. (2012) Parent psychopathology and offspring mental disorders: Results from the WHO World Mental Health Surveys. Br J Psychiatry 200:290-299
21. Garley D, Gallop R, Johnston N, Pipitone J (1997) Children of the mentally ill: A qualitative focus group approach. J Psychiatr Ment Health Nurs 4:97-103
22. Dunn J (1983) Sibling relationships in early childhood. Child Dev 54:787-811
23. Fagan AA, Najman JM (2003) Association between early childhood aggression and internalizing behavior for sibling pairs. J Am Acad Child Adolesc Psychiatry 42:1093-1100
24. Buist KL (2010) Sibling relationship quality and adolescent delinquency: A latent growth curve approach. J Fam Psychol 24:400-410
25. Milevsky A (2005) Compensatory patterns of sibling support in emerging adulthood: Variations in loneliness, self-esteem, depression, and life satisfaction. J of Soc and Pers Relationships 22:743-755
26. Feinberg ME, Solmeyer AR, Hostetler ML, Sakuma KL, Jones D, McHale SM (2013) Siblings are special: Initial test of a new approach for preventing youth behavior problems. J Adolesc Health 53:166-173
27. Reddy PS, Reddy YUJ, Srinath S, Khanna S, Sheshadri SP, Girimaji SR (2001) A family study of juvenile obsessive-compulsive disorder. Can J Psychiatry 46:346-351
28. Lenane MC, Swedo SE, Leonard H, Pauls DL, Sceery W, Rapoport JL (1990) Psychiatric disorders in first-degree relatives of children and adolescents with obsessive compulsive disorder. J Am Acad Child Adolesc Psychiatry 29:407-412

29. Dia DA, Harrington D (2006) Research note: What about me? Siblings of children with an anxiety disorder. Soc Work Res 30:183-188
30. Do Rosario-Campos MC, Leckman JF, Curi M, Quatrano S, Katsovitch L, Miguel EC, Pauls DL (2005) A family study of early-onset obsessive-compulsive disorder. Am J Med Genet Part B 136:92-97
31. Toro J, Cervera M, Osejo E, Salamero M (1992) Obsessive-compulsive disorder in childhood and adolescence: A clinical study. J Child Psychol Psychiatry 33:1025-1037
32. Farrell L, Barrett P, Piacentini J (2006) Obsessive-compulsive disorder across the developmental trajectory: Clinical correlates in children, adolescents, and adults. Behan Change 23:103-120
33. United Nations (1989) Convention on the rights of the child. Retrieved 8 Nov 2013 from http://treaties.un.org
34. Ravens-Sieberer U, Wille N, Erhart M, Bettge S, Wittchen J, Rothenberger A,…Dopfner M (2008) Prevalence of mental health problems among children and adolescents in Germany: Results of the BELLA study within the National Health Interview and Examination Survey. Eur Child Adolesc Psychiatry 17:22-23
35. Jensen PS, Rubio-Stipec M, Canino G, Bird HR, Dulcan MK, Schwab-Stone ME, Lahey BB (1999) Parent and child contributions to diagnosis of mental disorder: Are both informants always necessary? J Am Acad Child Adolesc Psychiatry 38:1569-1579
36. Valo S, Tannock R (2010) Diagnostic instability of DSM-IV ADHD subtypes: Effects of informant source, instrumentation, and methods for combining symptom reports. J Clin Child Adolesc 39:749-760
37. Ramirez Basco M, Bostic JQ, Davies D, Rush AJ, Witte B, Hendrickse W, Barnett V (2000) Methods to improve diagnostic accuracy in a community mental health setting. Am J Psych 157:1599-1605

38. Aboraya A, Rankin E, France C, El-Missiry, A, John C (2006) The reliability of psychiatric diagnosis revisited: The clinician's guide to improve the reliability of psychiatric diagnosis. Psychiatry Edgmont 3:41-50
39. Andreasen NC, Endicott J, Spitzer RL, Winokur G (1977) The family history method using diagnostic criteria: Reliability and validity. Arch Gen Psychiatry 34:1229-1235
40. Biederman J, Faraone SV, Keenan K, Knee D, Tsuang MT (1990) Family-genetic and psychosocial risk factors in DSM-III attention deficit disorder. J Am Acad Child Adolesc Psychiatry 29:526-533
41. Faraone SV, Biederman J, Mennin D, Gershon J, Tsuang MT (1996) A prospective four-year follow-up study of children at risk for ADHD: Psychiatric, neuropsychological, and psychosocial outcome. J Am Acad Child Adolesc Psychiatry 35:1449-1459
42. Faraone SV, Biederman J, Monuteaux MC (2000) Attention-deficit disorder and conduct disorder in girls: Evidence for a familial subtype. Biol Psychiatry 48:21-29
43. Kollins SH, Garrett ME, McClernon FJ, Lachiewicz AM, Morrissey-Kane E, Fitzgerald D, …Ashley-Koch AE (2009) Effects of postnatal parental smoking on parent and teacher ratings of ADHD and oppositional symptoms. J Nerv Ment Dis 197:442-449
44. Christiansen J, Chen W, Oades RD, Asherson P, Taylor EA, Lasky-Su J,…Faraone SV (2008) Co-transmission of conduct problems with attention-deficit/hyperactivity disorder: Familial evidence for a distinct disorder. J Neural Transm 115:163-175
45. Hassan A, Agha SS, Langley K, Thapar A (2011) Prevalence of bipolar disorder in children and adolescents with attention-deficit hyperactivity disorder. Br J Psychiatry 198:195-198
46. Hebrani P, Behdani F (2007) Influence of gender on familial aggregation of ADHD in relatives of probands with ADHD. Pak J Med Sci 23:610-613

47. Mikami AY, Pfiffner LJ (2008) Sibling relationships among children with ADHD. J Attention Disord 11:482-492

48. Green H, McGinnity A, Meltzer H, Ford T, Goodman R (2005) Mental health of children and young people in Great Britain, 2004. Office for National Statistics, Department of Health, & the Scottish Executive.

49. Lewinsohn PM, Hops H, Roberts RE, Seeley JR, Andrews, JA (1993) Adolescent psychopathology: I. Prevalence and incidence of depression and other DSM-III-R disorders in high school students. J Abnorm Psychol 102:133-144

50. Polancyzk G, de Lima MS, Horta BL, Biederman J, Rohde LA (2007) The worldwide prevalence of ADHD: A systematic review and metaregression analysis. Am J Psychiatry 164:942-948

51. Biederman J, Faraone SV, Knee D, Munir K (1990) Retrospective assessment of DSM-III attention deficit disorder in nonreferred individuals. J Clin Psychiat 51:102-106

52. Sobanski E, Banaschewski T, Asherson P, Buitelaar J, Chen W, Franke B, Faraone SV (2010) Emotional lability in children and adolescents with attention deficit/hyperactivity disorder (ADHD): Clinical correlates and familial prevalence. J Child Psychol Psychiatry 51:915-923

53. Nock MK, Kazdin AE, Hiripi E, Kessler RC (2006) Prevalence, subtypes, and correlates of DSM-IV conduct disorder in the National Comorbidity Survey Replication. Psychol Med 36:699-710

54. Biederman J, Faraone SV, Keenan K, Tsuang MT (1991) Evidence of familial association between attention deficit disorder and major affective disorders. Arch Gen Psychiatry 48:633-642

55. Geller B, Tillman R, Bolhofner K, Zimerman B, Strauss NA, Kaufman P (2006) Controlled, blindly rated, direct-interview family study of a prepubertal and early-

adolescent bipolar I disorder phenotype: Morbid risk, age at onset, and comorbidity. Arch Gen Psychiatry 63:1130-1138

56. Last CG, Hersen M, Kazdin A, Orvaschel H, Perrin S (1991) Anxiety disorders in children and their families. Arch Gen Psychiatry 48:928-934
57. Hill EM, Stoltenberg SF, Bullard KH, Li S, Zucker RA, Burmeister M (2002) Antisocial alcoholism and serotonin-related polymorphisms: Association tests. Psychiatr Genet 12:143-153
58. Nurnberger JI Jr, Wiegand R, Bucholz K, O'Connor S, Meyer ET, Reich T,…Porjesz B (2004) A family study of alcohol dependence: Coaggregation of multiple disorders in relatives of alcohol-dependent probands. Arch Gen Psychiatry 61:1246-1256
59. Hovens JG, Cantwell DP, Kiriakos R (1994) Psychiatric comorbidity in hospitalized adolescent substance abusers. J Am Acad Child Adolesc Psychiatry 33:476-483
60. Hasin DS, Stinson FS, Ogburn O, Grant BF (2007) Prevalence, correlates, disability, and comorbidity of DSM-IV alcohol abuse and dependence in the United States: Results from the National Epidemiologic Survey on Alcohol and Related Conditions. Arch Gen Psychiatry 64:830-842
61. Merikangas KR, He JP, Burstein M, Swanson SA, Avenevoli S, Cui L,…Swendsen J (2010) Lifetime prevalence of mental disorders in U.S. adolescents: Results form the National Comorbidity Survey Replication-Adolescent Supplement (NCS-A). J Am Acad Child Adolesc Psychiatry 49:980-989
62. Pine DS, Wasserman GA, Miller L, Coplan JD, Bagiella E., Kovelenku P,…Sloan RP (1998) Heart period variability and psychopathology in urban boys at risk for delinquency. Psychophysiology 35:521-529

63. Wasserman G, Miller LS, Pinner E, Jaramillo B (1996) Parenting predictors of early conduct problems in urban, high-risk boys. J Am Acad Child Adolesc Psychiatry 35:1227-1236

64. Seguin M, Manion I, Cloutier P, McEvoy L, Cappelli M (2003) Adolescent depression, family psychopathology and parent/child relations: A case control study. Can Child Adolesc Psychiatr Rev 12:2-9

65. Kelvin RG, Goodyer IM, Altham PM (1996) Temperament and psychopathology amongst siblings of probands with depressive and anxiety disorders. J Child Psychol Psychiatry 37:543-550

66. Ryan ND, Williamson DE, Iyengar S, Orvaschel H (1992) A secular increase in child and adolescent onset affective disorder. J Am Acad Child Adolesc Psychiatry 31:600-605

67. Karno M, Golding JM, Sorenson SB, Burnam A (1988) The epidemiology of obsessive-compulsive disorder in five US communities. Arch Gen Psychiatry 45:1094-1099

68. Reddy YC, Rao NP, Khanna S (2010) An overview of Indian research in obsessive compulsive disorder. Indian J Psychiatry 52:S200-S209

69. Boyd CP, Kostanski M, Gullone E, Ollendick TH, Shek DTL (2000) Prevalence of anxiety and depression in Australian adolescents: Comparisons with worldwide data. J Genet Psychol 161:479-492

70. Fulton BD, Scheffler RM, Hinshaw SP, Levine P, Stone S, Brown TT, Modrek S (2009) National variation of ADHD diagnostic prevalence and medication use: Health care providers and education policies. Psychiat Serv 60:1075-1083

71. Hopfer CJ, Stallings MC, Hewitt JK, Crowley TJ (2003) Family transmission of marijuana use, abuse, and dependence. J Am Acad Child Adolesc Psychiatry 42:834-841

72. Anderson KG, Tapert SF, Moadab I, Crowley TJ, Brown SA (2007) Personality risk profile for conduct disorder and substance use disorders in youth. Addict Behav 32:2377-2382

73. Sakai JT, Stallings MC, Crowley TJ, Gelhorn HL, McQueen MB, Ehringer MA (2010) Test of association between GABRA2 (SNP rs279871) and adolescent conduct/alcohol use disorders utilizing a sample of clinic referred youth with serious substance and conduct problems, controls and available first degree relatives. Drug Alcohol Depen 106:199-203

74. Waldman ID, Rowe DC, Abramowitz A, Kozel ST, Mohr JH, Sherman SL,…Stever C (1998) Association and linkage of the dopamine transporter gene and attention-deficit hyperactivity disorder in children: Heterogeneity owing to diagnostic subtype and severity. Am J Hum Genet 63:1767-1776

75. Altink ME, Arias-Vasquez A, Franke B, Slaats-Willemse DI, Buschgens CK, Rommelse NN,…Buitelaar JK (2008) The dopamine receptor D4 7-repeat allele and prenatal smoking in ADHD-affected children and their unaffected siblings: No gene-environment interaction. J Child Psychol Psychiatry 49:1053-1060

76. Milberger S, Biederman J, Faraone SV, Wilens T, Chu MP (1997) Associations between ADHD and psychoactive substance use disorders: Findings from a longitudinal study of high-risk siblings of ADHD children. Am J Addiction 6:318-329

77. Faraone SV, Chen WJ, Warburton R, Biederman K, Milberger S, Tsuang MT (1995) Genetic heterogeneity in attention-deficit hyperactivity disorder (ADHD): Gender, psychiatric comorbidity, and maternal ADHD. J Abnorm Psychol 104:334-345

78. Szatmari P, Boyle MH, Offord DR (1993) Familial aggregation of emotional and behavioral problems of childhood in the general population. Am J Psychiat 150:1398-1403

79. Biederman J, Faraone SV, Chen W (1993) Social Adjustment Inventory for Children and Adolescents: Concurrent validity in ADHD children. J Am Acad Child Adolesc Psychiatry 32:1059-1064
80. Faraone SV, Biederman J, Jetton JG, Tsuang MT (1997) Attention deficit disorder and conduct disorder: Longitudinal evidence for a familial subtype. Psychol Med 27:291-300
81. Chen W, Zhou K, Sham P, Franke B, Kuntsi J, Campbell D,…Asherson P (2008) DSM-IV combined type ADHD shows familial association with sibling trait scores: A sampling strategy for QTL linkage. Am J Med Genet Part B 147:1450-1460
82. Faraone SV, Biederman J, Mennin D, Russell R (1998a) Bipolar and antisocial disorders among relatives of ADHD children: Parsing familial subtypes of illness. Am J Med Genet 81:106-116
83. Faraone SV, Biederman J, Mennin D, Russell R, Tsuang MT (1998b) Familial subtypes of attention deficit hyperactivity disorder: A 4-year follow-up study of children from antisocial-ADHD families. J Child Psychol Psychiatry 39:1045-1053
84. Chauhan SP, Scardo JA, Hayes E, Abuhamad AZ, Berghella V (2010) Twins: Prevalence, problems, and preterm birth. Am J Obstet Gynecol 203:305-315
85. US Census Bureau (2004) Average number of children per family and per family with children, by state: 2000 Census. Retrieved Nov 15 from http://www.census.gov/population/socdemo/hh-fam/tabST-F1-2000.pdf
86. Griffiths C, Sin J (2013) Rethinking siblings and mental illness. The Psychologist 26:808-810
87. Bullock BM, Dishion TJ (2002) Sibling collusion and problem behaviour in early adolescence: Toward a process model for family mutuality. J Abnorm Child Psychol 30:143-153

88. Giletta M, Burk WJ, Scholte RHJ, Engels RCME, Prinstein MJ (2013) Direct and indirect peer socialization of adolescent nonsuicidal self-injury. J Res Adolesc 23:450-463
89. Quinn P, Wigal S (2004) Perceptions of girls and ADHD: Results from a national survey. MedGenMed 6:2
90. Kendler KS (2001) Twin studies of psychiatric illness: An update. Arch Gen Psychiatry 58:1005-1014
91. Beardselee WR, Versage EM, Giadstone TRG (1998) Children of affectively ill parents: A review of the past 10 years. J Am Acad Child Adolesc Psychiatry 37:1134-1141
92. Calhoun CD, Franklin JC, Adelman CB, Guerry JD, Hastings PD, Nock MK, Prinstein MJ (2012) Biological and cognitive responses to an in vivo interpersonal stressor: Longitudinal associations with adolescent depression. Int J Cognit Ther 5:283-299
93. Hardt J, Franke P (2007) Validity, reliability, and objectivity of the family history method in psychiatry: A meta analysis. Eur Psychiatry 22:49-58
94. Bristow K, Patten S (2002) Treatment-seeking rates and associated mediating factors among individuals with depression. Can J Psychiatry 47:660-665
95. Narrow WE, Kuhl EA (2011) Dimensional approaches to psychiatric diagnosis in DSM-5. J Ment Health Policy Econ 14:197-200
96. Ma N, Roberts R, Winefield H, Furber G (2014) A dimensional approach to the mental health of siblings of children with mental health problems: A 20-year systematic review. Manuscript submitted for publication.

Chapter 4. Paper Two

A Dimensional Approach to the Mental Health of Siblings of Children with Mental Health Problems: A 20-Year Systematic Review

(Manuscript Under Review)

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Clinical Psychology Review. Submitted, July 2014.

Statement of Authorship

Nylanda Ma (Candidate)

I was responsible for the design and implementation of the systematic review, including development of the search strategy, collection of the included studies, data extraction, data analysis, and writing the manuscript. I served as corresponding author and was responsible for manuscript submission, revisions, and responses to journal reviews.

Signed: Nylanda LT Ma _____ Date: 4/8/2014

Dr. Rachel Roberts, Prof. Helen Winefield, & Dr. Gareth Furber (Co-authors)

We provided ongoing supervision through the systematic review and there was ongoing collaboration between Nylanda Ma and ourselves in refining the direction of research and methodology. We commented on manuscript drafts, made suggestions on the presentation of the material in the manuscript, and provided guidance on journal revisions and responses. We hereby give our permission for the inclusion of this paper in Nylanda Ma's submission for the degree of Doctor of Philosophy from the University of Adelaide.

Signed: Dr. Rachel Roberts _____Date: 4/8/2014

Signed: Prof. Helen Winefield _____Date: 4/8/2014

Signed: Dr. Gareth Furber _____ Date: 4/8/2014

Abstract

A previous review on the prevalence of psychopathology in siblings of children with mental health problems (MHPs) suggested that these siblings are at increased risk of a broad range of psychopathologies. No disorder-specific risk could be identified. It may be that a different approach to understanding mental health is needed. The following paper aimed to explore if using a dimensional approach could provide greater clarity and insight into the mental health of these siblings by conducting a similar systematic review using dimensional data on the psychosocial functioning of siblings of children with MHPs. An electronic search of publications from 1990-2011 identified 29 articles. The findings show that while mean sibling scores on internalising problems, externalising problems, and social problems were in the normal range compared to standardised norms, siblings of children with MHPs scored poorer than control children and a greater proportion of siblings scored in the borderline/clinical range compared to children in the general population. Particular areas of functional impairments identified were delinquent behaviour, somatic complaints, anxious/depressed behaviour, and social problems. Effect sizes were moderate to large. Findings on symptom scores with categorical underpinnings were inconsistent. Implications for the debate between dimensional and categorical approaches are discussed and guidelines for future research are outlined. Implications for clinical treatment for siblings are highlighted.

Keywords: sibling, child, mental health, psychosocial functioning, systematic review, dimensional

Family systems and socioecological perspectives on child development hold that children and the systems or contexts they participate in have a reciprocal relationship (Bronfrenbrenner, 1979; Hoffman, 1981). That is, child development is influenced by the functioning of families, schools, and broader society and children, in turn, influence these systems. Based on these perspectives, the presence of mental health problems (MHPs) in family members will affect the functioning of the children in these families. This has been well established with research with parents of children with MHPs and with research with children of parents with MHPs. In such families, research has shown that MHPs impact on family relationships, the psychosocial functioning of family members, and the functioning of the family as a whole (Sawyer et al., 2000).

Similarly, according to theories on the development of psychopathology, genetic or biological, shared environment, and non-shared environment or individual differences factors interact in the development of MHPs (Sattler & Hoge, 2006). Exposure to risk factors within these domains has been linked to elevated risk of MHPs in children (Sattler & Hoge, 2006). Based on these theories then, in families with a member with MHPs, we would expect other family members to have an elevated risk of MHPs or psychosocial impairment given that they share genetic or biological and shared environmental risk factors. Familial aggregation of MHPs has been well established in research with parents of children with MHPs and in research with children of parents with MHPs (e.g. State, Lombroso, Pauls, & Leckman, 2000).

These theoretical frameworks suggest that siblings of children with MHPs would similarly be at risk for psychopathology or psychosocial impairment. Despite the significant role of siblings within the family system (Feinberg, Solmeyer, & McHale, 2012), little research has examined the mental health of these siblings. The authors conducted a systematic review on the prevalence of psychopathology for these siblings

(Ma, Roberts, Winefield, & Furber, 2014). This review found that siblings of children with MHPs have higher rates of psychopathology than control children. The data from the included studies from that review suggest that siblings of children with MHPs have a broad risk of psychopathology. That is, siblings had equally high risk of developing a range of psychopathologies. For example, siblings of children with attention deficit hyperactivity disorder (ADHD) had significantly higher rates of ADHD, conduct disorder (CD), oppositional defiant disorder (ODD), and certain types of anxiety disorders than control children. The majority of odds ratios from higher quality studies all fell within 2.0 – 4.0 suggesting that these siblings had a similar degree of elevated risk regardless of the disorder assessed. No clear specific patterns of risk for siblings could be identified based on the type of disorder assessed in the sibling, the type of MHP in the target child, or based on other moderators, such as age and gender. The findings of our previous review suggest that a different approach to understanding the mental health of siblings of children with MHPs may be needed. It may be that a categorical approach or current categorical measures are not sufficiently sensitive to illuminate specific patterns of risk for siblings of children with MHPs. Taking a different approach, and considering this alongside the categorical findings, may provide a more comprehensive understanding and may identify specific patterns of risk that could not be identified using categorical diagnostic data alone.

Several authors have suggested that using a dimensional approach to mental health can provide significant benefits and can provide additional insight beyond that based on categorical approaches (Lopez, Compton, Grant, & Breiling, 2007; Rutter, 2011). A dimensional approach holds that there are different dimensions of functioning, such as mood and cognitive processing, and it is different patterns across these dimensions of functioning that reflect psychiatric disorders (Harkavy-Friedman, 2009).

Every child or, in the case of this review, a particular population of children, would then have a particular profile of functioning that reflects the mental health problems facing that group. There are several different dimensional processes including broadly examining functioning across a range of areas or using existing diagnostic categorical structures but including continuous frequency and severity data, such as symptom counts (Harkavy-Friendman, 2009). A categorical approach, in contrast, follows the process of determining groups of symptoms that reflect psychiatric disorders (Harkavy-Friedman, 2009). A child would need to meet a set cut-off of symptoms to be classified as having a particular psychiatric or mental health problem. This approach can limit the amount of clinically useful information that can be obtained (Helzer et al., 2008). For example, two children both presenting with a diagnosis of depression may display different symptomatology. One child may present with suicidal ideation as a primary symptom whereas another child may present with primarily with neurobiological symptoms, such as poor sleep and concentration. These may lead to different treatment plans, such as psychotherapy versus medication. Furthermore, this approach follows a present versus absent dichotomy and may not be able to identify difficulties in functioning that do not necessarily fit within a single diagnostic category or may not necessarily reach the set cut-off. This might lead to inaccurate conclusions that a child, or in the case of this paper, that siblings do not experience significant mental health difficulties. For example, siblings may experience significant difficulties in peer relationships. Using a categorical approach, siblings may be considered absent of mental health difficulties because their symptoms do not meet the criteria for a particular psychiatric diagnosis. In contrast, a dimensional approach may be able to reveal and highlight significant peer relationship problems and recognise that this population is in need of support.

The main aim of this paper is to explore whether a dimensional approach to understanding mental health can provide greater insight and clarity into the risk of mental health difficulties for siblings of children with MHPs and illuminate specific patterns of risk. The following paper presents the findings of a systematic review of the dimensional data on the mental health of siblings of children with MHPs followed by a discussion of how these findings compare to those of the categorical data review.

Methods

The method used in this review has been described in detail elsewhere (Ma et al., 2014). Briefly, four electronic databases were searched from January 1990 to July 2011: Cumulative Index to Nursing and Allied Health Literature (CINAHL), PsycINFO, PubMed, and SciVerse Scopus using terms such as *sibling; first degree relative; child; emotional; behavioural.* Studies were included if they a) reported continuous data on the mental health of siblings of a target child with MHPs (e.g. internalising, externalising, social problems) and/or data on associated variables or covariates, b) reported primary data, c) were published in a peer-reviewed journal, and d) were published in English. As the primary research question in this review involved comparisons between siblings of children with MHPs and healthy children, studies were required to either include a matched control group or employ measures with standardised norm data. In this study, children (2-18 years) were considered to have MHPs if they had an established mental illness diagnosis, screened positive on diagnostic measures, had clinically elevated scores on psychological measures, had been referred to or were attending a mental health clinic or mental health treatment, or had been adjudicated for delinquency. The ‘target child’ was defined as the one who is the primary target of treatment or research and the ‘target sibling’ as the brothers and/or sisters of this child. Control children are similarly defined. It should be noted that based

on this definition and the data available, no causal links between the target child's MHPs and the mental health of the sibling can be inferred.

The initial search identified 4,387 records after removing duplicates. After screening titles and abstracts, 761 full-text articles were reviewed (see Figure 1.). The final pool of studies consisted of 29 articles. In cases where studies have recruited participants from the same population, only unique data points were extracted to ensure participants were not counted twice (e.g. sibling scores on different subscales of psychosocial measures). If two studies recruited the same participants and reported the same data, the data from the highest quality study were extracted. If the studies segregated the data by categories that were not pertinent to this review (e.g. sibling age or sex), the data were combined into a pooled sibling score. If studies recruited a control group, *t*-tests were conducted and Cohen's *d* was calculated or these data were extracted directly from the included studies (Cohen, 1988). If no control group was recruited, comparisons were made to standardised norm data and clinical cut-offs. It was deemed inappropriate to conduct statistical testing with normative data due to methodological differences (e.g. large differences in sample sizes) and imperfect matching on demographic characteristics (e.g. location, age, gender proportions). While this may be appropriate in some circumstances, given that the majority of studies in this field have multiple methodological issues, it was considered inappropriate to compound this by conducting methodologically problematic statistical testing.

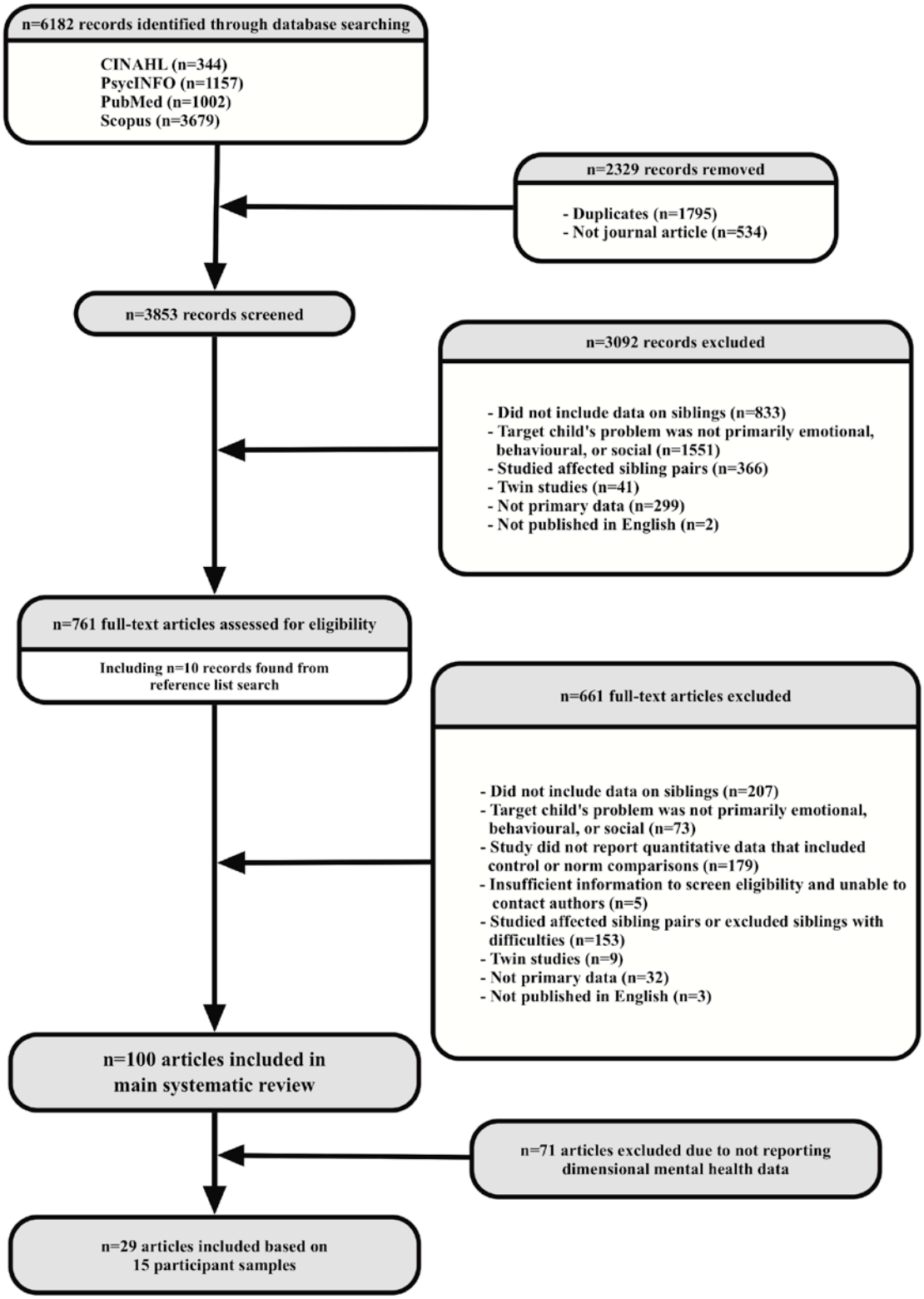

Figure 1. Flow of included studies.

The 95% confidence intervals around effect sizes were calculated using software developed by Durham University (available at http://www.cem.org/evidence-based-education/effect-size-calculator). A meta-analysis of the extracted data was not conducted due to methodological differences across studies (e.g. gender proportions, measures used, and the subscales assessed) and due to limited numbers of studies that included control groups and enabled effect size calculations. Thus, the following is a narrative synthesis with particular attention to consistent findings and methodological quality. To present a meaningful summary, data on covariates were only presented if they were explored by two or more studies.

Results

Characteristics of Included Studies

This review is based on 29 studies assessing approximately 8,916 target siblings of children with MHPs across six countries. Characteristics of the included studies are summarised in Table 1. The size of participant samples tended to be small (16 studies; $n<100$) with only seven studies assessing large sample sizes ($n>200$). Only 12 studies recruited a matched control group of either control children matched to the target child's characteristics (e.g. age, sex, socioeconomic status) or the siblings of control children. Target children ranged in age from 4-18 years. Target siblings ranged in age from 2-21 years.

Eleven studies assessed the mental health of siblings based on reports from multiple informants. Given that scores on psychological measures often differ across informants (Vermaes, van Susante, & van Bakel, 2012), studies that recruited multiple informants are more reliable and comprehensive and are considered of higher quality. Nineteen studies assessed multiple siblings in the target child's family. The remaining studies assessed only one sibling, typically the closest in age to the target child. Higher

quality studies are those that assessed multiple siblings as the latter method increases sampling error, particularly where siblings were not randomly selected. All but two studies used well-established and validated standardised psychological measures, most commonly the Child Behavior Checklist (CBCL; Achenbach & Rescorla, 2001). The remaining studies used psychiatric symptom scales based on diagnostic criteria (McDougall, Hay, & Bennett, 2006; Waldman et al., 1998). To the best of our knowledge, the reliability and validity of these scales have not been examined and these studies are considered to be of poorer quality. The following section presents the results of the synthesis organised by individual components of mental health.

Externalising Behaviour Problems

Seventeen studies assessed externalising behaviour problems on dimensional scales (Brotman et al., 2005b; Cohen, Barwick, Horodezky, & Isaacson, 1996; Christiansen et al., 2008; Copeland, Landry, Stanger, & Hudziak, 2004; Deal & MacLean, 1995; Dennis & Brotman, 2003; Dia & Harrington, 2006; Faraone, Biederman, Mennin, Gershon, & Tsuang, 1996; Hudson & Rapee, 2002; Hudziak, Copeland, Stanger, & Wadsworth, 2004; Kuntsi et al., 2010; Listug-Lunde, Zevenbergen, & Petros, 2008; Milberger, Faraone, Biederman, Testa, & Tsuang, 1996; Sobanski et al., 2010; Stallings et al., 1997; Waldman et al., 1998; Wasserman et al., 1996). On the CBCL total externalising, delinquent, and aggressive behaviour scales, with the exception of one study as described below, all sibling mean *T*-scores were in the normal range (*M*=46.3-58.9, *SD*=4.0-12.5). However, in the majority of sibling scores (21 of 23), there was a consistent shift to the right with target sibling scores falling in the upper end of the normal range (>50). Further, when compared to control sibling scores, target siblings had higher mean *T*-scores indicating more externalising problems or more severe externalising problems in siblings of children with MHPs.

Table 1

Summary of Included Studies

Reference	Target child MHP	Sibling		Measures	Key methodological strengths (S) and weaknesses (W)
		n	Age (years)		
Barrett et al. (2001)	OCD	5	8-14	MASC; CDI	(S): Assessed all siblings; Control group (SC; *n*=5) (W): Single informant (S); S; Very small sample; Only prepubertal targets and small age range (10-13 years)
Christiansen et al. (2008)	ADHD	1828	10.9	CRS; PACS	(S): Assessed all siblings; Multiple informants (P/T); Control group (only Kuntsi et al., 2010; CO; *n*=345); Large sample sizes
Kuntsi et al. (2010)		456	11.38		(W): No control group in remaining studies
Sobanski et al. (2010)		1827	10.4		
Müller et al. (2011a)		1446	5-17		
Müller et al. (2011b)					
Chen et al. (2008)		1135	--		
Cohen et al. (1996)	MHPs	100	4-12	CBCL	(S): Multiple informants (P/T); Only prepubertal siblings and targets (W): Assessed one sibling in same age range as target child; No controls
Copeland et al. (2004)	Attention	206	10.82	CBCL	(S): Multiple informants (P/S; Copeland et al., 2004); Large sample sizes
Hudziak et al. (2004)	and/or	183	6-18	CBCL	(W): Assessed one randomly selected sibling; Single informant (P; Hudziak et
Rettew et al. (2004)	aggression	165	6-18	CBCL	al., 2004 & Rettew et al., 2004); No control group

Table 1 (cont.)

Summary of Included Studies

Reference	Target child MHP	Sibling		Measures	Key methodological strengths (S) and weaknesses (W)
		n	Age (years)		
Deal & MacLean (1995)	MHPs	15	9-15	CBCL CDI; RCMAS	(S): Multiple informants in study; Controls (CO; *n*=15); Small sample (W): Assessed one sibling closest in age; One informant per measure; Only adolescent targets; Only hospitalised target adolescents; Only younger siblings
Dennis & Brotman (2003)	DEL	37	5.98	CBCL	(S): Assessed all siblings (only Brotman et al., 2005b) (W): Assessed one sibling closest to 4 years of age; Single informant (P); No control group; Small age range for siblings and only younger siblings;
Brotman et al. (2004)		92	3.94		
Brotman et al. (2005a)		84	3.95		
Brotman et al. (2005b)		99	2.75-5.25		
Dia & Harrington (2006)	ANX	65	9.9	CBCL	(W): Assessed one sibling closest in age to target; Single informant (P); No control group;
Faraone et al. (1996)	ADHD	169-174	--	CBCL; SAICA; SADS	(S): Assessed all siblings; Multiple informants (P/S); Control group (SC; *n*=120-143); Large sample size (except Chen et al., 1994) (W): Only male target children
Faraone et al. (1998)		219	--		
Chen et al. (1994)		27	--		
Milberger et al. (1996)		146	--		

Table 1 (cont.)

Summary of Included Studies

Reference	Target child MHP	Sibling *n*	Sibling Age (years)	Measures	Key methodological strengths (S) and weaknesses (W)
Hudson & Rapee (2002)	ANX	37	7-16	CDI; RCMAS CBCL	(S): Multiple informants in study (P/S); Control group (SC; *n*=20) (W): Assessed one sibling closest in age; One informant per measure; Small sample size
Kelvin et al. (1996)	DEP ANX	31	8-17	CGAS	(S): Assessed all siblings; Control group (CO; *n*=40) (W): Single informant (P); Small sample size
Listug-Lunde et al. (2008)	ADHD	41	11.29	CBCL; DBRS; CDI; MASC	(S): Multiple informants within study (P/S); Control group (SC; *n*=30) (W): Assessed one sibling; One informant per measure; Small sample
McDougall et al. (2006)	ADHD	69	3-21	Scale based on DSM-IV criteria	(S): Control group (CO; *n*=246); Large age range for siblings (W): Assessed one sibling closest in age; Single informant (P); Not well-established measure; Small sample size
Stallings et al. (1997)	SUD DEL	13	--	DIS; DICA; CIDI-SAM	(S): Assessed all siblings; Control group (SC; *n*=41) (W): Single informant (S); Very small sample; Only adolescent targets

Table 1 (cont.)

Summary of Included Studies

Reference	Target child MHP	Sibling		Measures	Key methodological strengths (S) and weaknesses (W)
		n	Age (years)		
Waldman et al. (1998)	Externalising problems	41	9.51	Emory Diagnostic Rating Scale	(S): Assessed all siblings; Control group (Locally representative twin population; *n*=756) (W): Single informant (P); Not well-established measure; Small sample
Wasserman et al. (1996) Pine et al. (1997)	DEL	112-126 34	6-10 8.4-10.0	CBCL	(S): Assessed all siblings (W): Single informant (P); No control group; Only male younger siblings; Small sample size (except Wasserman et al., 1996); Only male targets

Note. --: Not reported; AUS: Australia; UK: United Kingdom; US: United States of America; MHP: Mental health problem; ADHD: Attention deficit hyperactivity disorder; ANX: Anxiety disorder; DEL: Delinquency; DEP: Depressive disorders; OCD: Obsessive-compulsive disorder; SUD: Substance use disorders; SC: Siblings of control children matched to the target child's characteristics; CO: Control children matched to the target child's characteristics; P: Parent; T: Teacher; S: Sibling; CBCL: Child Behavior Checklist; CDI: Children's Depression Inventory; CIDI-SAM: Substance Abuse Model of the Composite International Diagnostic Interview; CGAS: Children's Global Assessment Scale; CRS: Conners' Rating Scales; DBRS: Disruptive Behavior Rating Scale; DIS: NIMH Diagnostic Interview; DICA; Diagnostic Interview for Children and Adolescents; MASC: Multidimensional Anxiety Scales for Children; PACS: Parental Account of Childhood Symptoms; RCMAS: Revised Children's Manifest Anxiety Scale; SADS: Schedule for Affective Disorders and Schizophrenia; SAICA: Social Adjustment Inventory for Children and Adolescents; NB: References contained within the same row drew on the same participant pool

This finding was also supported by categorical data. Two studies reported the proportion of target siblings that scored in the clinical range on the externalising scale (Brotman et al., 2005b; Wasserman et al., 1996). Across both studies, the reported proportion in siblings of children with MHPs was greater than that found in non-referred children (22-27% v 18%, respectively; Achenbach & Rescorla, 2001). A greater proportion of target siblings scoring in the clinical range may explain the shift in mean sibling scores towards the upper end of the normal range.

There were inconsistencies across studies regarding statistical significance however. Higher quality studies that recruited all available siblings, obtained data from multiple informants, and had large sample sizes (Faraone et al., 1996; Listug-Lunde et al., 2008) reported statistically significant differences between target and control siblings while poorer quality studies did not (Deal & MacLean, 1995; Hudson & Rapee, 2002). Regarding differences between the CBCL scales, consistently across studies and across types of target child difficulties, target siblings scored higher on the delinquent behaviour scale (e.g. stealing, substance use, rule-breaking; M=52.5-60.3, SD=6.2-9.8) than the aggressive behaviour scale (e.g. destructive, tantrums, bullying) and total externalising scale (M=50.1-58.9, SD=4.0-10.2). In addition, the only study that reported a mean T-score in the borderline/clinical range of the CBCL was on the delinquent behaviour scale (M=60.3, SD=9.8; Pine et al., 1997). Furthermore, effect sizes of the difference between target sibling and control sibling scores were larger for the delinquent behaviour scale (d=0.32-1.43) than the other two scales (d=0.25-0.47). All effect size confidence intervals from higher quality studies were wide, spanning from small to large, suggesting limited precision and considerable variation in the sibling data (e.g. 95% CI [0.16, 0.72] from delinquent behaviour problems; Faraone et al., 1996).

On measures of symptoms of externalising disorders (e.g. ADHD and CD), similar patterns arose (Christiansen et al., 2008; Copeland et al., 2004; Deal & MacLean, 1995; Faraone et al., 1996; Kuntsi et al., 2010; Listug-Lunde et al., 2008; Milberger et al., 1996; Sobanski et al., 2010; Stallings et al., 1997; Waldman et al., 1998). All target sibling scores were in the normal range but were higher than control sibling scores. Of the seven studies that included control siblings, only one did not report statistically significant differences between target and control siblings. This study was of poorer quality due to a very small sample size (n=15) and assessing only the sibling closest in age to the target child (Deal & MacLean, 1995). Effect size ranges for symptoms of ADHD and CD/ ODD overlapped considerably (d=0.21-0.71 and d=0.20-1.63, respectively) and there was considerable variability in effect sizes across studies. This variability may be due to differences in measures of symptomatology. For example, some studies assessed ADHD symptoms as one construct, while other studies examined hyperactive and inattention symptoms separately. Similarly, there was considerable variability apparent in effect size confidence intervals. This suggests that there is also substantial within-study variance in sibling data. Even in high quality studies with large sample sizes (n>100), effect size confidence intervals spanned from small to large (e.g. 95% CI [0.23, 0.72] for ADHD symptom scores; Milberger et al., 1996).

Internalising Behaviour Problems

Sixteen studies examined internalising behaviour problems in siblings of children with MHPs including somatic complaints, withdrawal, negative affect, and symptoms associated with internalising disorders, such as depression and anxiety (Barrett, Rasmussen, & Healy, 2001; Brotman, Gouley, Chesir-Teran, 2005a; Cohen et al., 1996; Copeland et al., 2004; Deal & MacLean, 1995; Dia & Harrington, 2006;

Faraone et al., 1996; Hudson & Rapee, 2002; Hudziak et al., 2004; Listug-Lunde et al., 2008; McDougall et al., 2006; Milberger et al., 1996; Pine et al., 1997; Stallings et al., 1997; Waldman et al., 1998; Wasserman et al., 1996). Only one type of behaviour rating scale that is not specific to individual internalising disorders was used. On the CBCL, similar to externalising problems, all sibling mean *T*-scores were in the normal range (*M*=51.1-59.0, *SD*=5.5-14.9). Again, there was a right shift with target sibling scores falling in the upper end of the normal range. When compared to control siblings, target siblings had higher scores than control siblings indicating that target siblings have more or more severe internalising problems. As with externalising problems, Wasserman and colleagues (1996) found a higher proportion of target siblings scored in the borderline/clinical range on the internalising scale than is reported for non-referred children (22-25% v 18%, respectively; Achenbach & Rescorla. 2001). In contrast, Brotman and colleagues (2005a) found that only 16% of their sample of target siblings scored in the borderline/clinical range. It should be noted however, that they recruited only pre-school aged siblings and the above norms are for children aged 6-18 years. Of the four studies that included a control group, three reported statistically significant findings (Deal & MacLean, 1995; Faraone et al., 1996; Listug-Lunde et al., 2008). The remaining study was poor in quality with a small sample size and assessed only the sibling closest in age to the target child (Hudson & Rapee, 2002).

Comparing across the individual internalising scales, larger effect sizes, ranging from moderate to large, were reported for the somatic complaints and anxious/depressed subscales (*d*=0.39-1.24) than for the withdrawn and total internalising scale (*d*=0.29-0.80). Similarly, mean sibling *T*-scores were higher on the former subscales than the latter. Siblings scored highest on the somatic complaints subscale, however the difference between means between somatic complaints and

anxious/depressed was small. Again, effect size confidence intervals were wide suggesting considerable within-study variation. Even within the study with the largest sample size (Faraone et al., 1996), the effect size confidence intervals spanned from negligible to moderate (e.g. 95% CI [0.06, 0.59] for withdrawn behaviours).

The remaining measures employed in the included studies were scales based specifically on the symptoms of individual internalising disorders, such as depression and anxiety disorders. The measures used were the Children's Depression Inventory (CDI; Kovacs, 1992), the Multi-dimensional Anxiety Scale for Children (MASC; March, 1997), the Revised Children's Manifest Anxiety Scale (RCMAS; Reynolds & Richmond, 1990), and diagnostic interviews and rating scales. As with externalising problems, more variability was found when disorder-specific measures were used. First, although the majority of mean target sibling scores were in the normal range, there were some exceptions. On the MASC and RCMAS, target siblings scored in the above average range for social anxiety and general anxiety (Barrett et al., 2001; Deal & MacLean, 1995). It should be noted however that both of these studies has important methodological limitations including very small samples ($n \leq 15$). Thus, the reliability of these findings are limited. Second, although the majority of studies reported higher scores for target siblings than control siblings, there were some exceptions. On the CDI, siblings had lower total depression scores and on the MASC, target siblings had lower separation anxiety scores (Barrett et al., 2001; Hudson & Rapee, 2002). Again, these studies had important methodological limitations including very small samples ($n<40$) and/or assessment of only the sibling closest in age to the target child.

Regarding statistical significance, on measures of depression symptoms, only one of five studies that included control siblings reported a statistically significant difference between target and control siblings on depression symptoms. This study

again had a very small sample size (*n*=13) and was poorer in quality compared to most of the other studies that did not report statistically significant findings (e.g. Listug-Lunde et al., 2008 & Milberger et al., 1996). On measures of anxiety disorder symptoms, there was considerable variation across studies on what specific anxiety disorders were assessed. Thus, it was difficult to ascertain specific patterns as typically only one or two studies assessed the same anxiety disorder and only one of the studies assessing anxiety symptoms were considered high quality. With these caveats in mind, target siblings and control siblings significantly differed most consistently on measures of generalised anxiety and separation anxiety. Similarly with effect sizes, there was considerable variability. On measures of depression and anxiety symptoms, effect sizes ranged from minimal to large (*d*=0.02-1.55). Large effect sizes were most consistently reported for generalised anxiety (*d*=0.88-1.50). However, these were reported in studies of poorer quality. Effect size confidence intervals from the only high quality study spanned from minimal to moderate (e.g. 95% CI [0.08, 0.57] for separation anxiety; Milberger et al., 1996).

Social Problems

Five studies assessed social problems in siblings of children with MHPs (Barrett et al., 2001; Deal & MacLean, 1995; Faraone et al., 1996; Faraone, Biederman, Mennin, Russell, & Tsuang, 1998; Hudziak et al., 2004) using the CBCL, CDI, and the Social Adjustment Inventory for Children and Adolescents (SAICA; John, Gammon, Prusoff, & Warner, 1987). The findings on social problems were consistent across all five studies. All sibling mean scores were in the normal range but there was evidence of a right shift with sibling scores falling in the upper end of the normal range. In comparison to control siblings, target siblings had higher scores on all scales. There was only one exception: Target siblings had equal scores with control siblings on boy-girl

relationships and problems with the opposite sex on the SAICA (Faraone et al., 1996). Three studies included a matched control group and all studies reported statistically significant differences between target and control sibling scores. On the SAICA, only school behaviour problems and peer problems were statistically significant while spare-time activities and boy-girl relationships were not (Faraone et al., 1996). Effect sizes ranged from small to large (*d*=0.18-0.95). Faraone and colleagues (1996) conducted the only study that measured several aspects of social behaviour. The largest effect size reported in their study was for school behaviour problems (*d*=0.65, 95% CI [0.41, 0.87]). Effect size estimates from other studies with controls were on the total social problems scale of the CBCL and were in the moderate to large range (*d*=0.40-0.95; Deal & MacLean, 1995; Faraone et al., 1996). However, the effect size confidence intervals were wide and spanned from small to large (e.g. 95% CI [0.25, 0.81]; Faraone et al., 1996), suggesting limited precision and considerable within-study variation.

Global functioning

Seven studies reported data on the global psychosocial functioning of siblings of children with MHPs (Cohen et al., 1996; Deal & MacLean, 1995; Dia & Harrington, 2006; Faraone et al., 1996; Kelvin, Goodyer, & Altham, 1996; Müller et al., 2011b; Rettew, Copeland, Stanger, & Hudziak, 2004). Five of these studies employed the standardised rating scales CBCL and CRS and reported sibling scores on the total behaviour problems scale or the total across all subscales. Mean target sibling scores were in the normal range across all five studies. Again, there was a consistent shift with sibling scores falling in the upper end of the normal range. Deal and MacLean (1995) conducted the only study that recruited control siblings. Target siblings of children with a range of MHPs had significantly higher scores on the total behaviour problems of the CBCL than control siblings to a large degree (*d*=1.15, 95% CI [0.31, 1.84]). However,

the effect size confidence interval was wide, spanning from small to large, indicating poor precision, likely in part due to a very small sample size (*n*=15).

Two studies assessed target siblings' global psychosocial functioning using the Global Assessment of Functioning scale (GAF; Diagnostic and Statistical Manual, 4th edition [DSM-IV]; American Psychiatric Association [APA], 1994) and the Children's Global Assessment Scale (CGAS; Shaffer et al., 1983). On the GAF, target siblings' mean score fell in the 'some mild symptoms or some difficulty in functioning' category (*M*=61.3-62.0, *SD*=9.7-12.4; Faraone et al., 1996). Target siblings had significantly poorer global functioning scores compared to control siblings. The effect was small to moderate (*d*=0.34-0.61). Again, effect size confidence intervals were wide, despite large sample sizes (*n*=169-174) suggesting considerable variability in sibling scores (95% CIs [0.11, 0.54] and [0.37, 0.83]). On the CGAS, 51.6% of target siblings experienced minimal impairment, 32.3% had mild impairment (i.e. difficulties in some situations or areas), and 16.1% had moderate impairment (i.e. obvious difficulties across several situations; Kelvin et al., 1996). It should be noted that Kelvin and colleagues (1996) found that the presence of a mental health diagnosis in target siblings did not directly relate to impairment scores. Four siblings showed signs of impairment without a diagnosis and two with a diagnosis showed minimal impairment. This data shows a discontinuity between dimensional data based on functioning across multiple areas and categorical diagnostic data.

Variables Associated with the Mental Health of Siblings

Very few studies examined variables associated with the mental health of siblings of children with MHPs. The findings across studies were varied and/or inconsistent and no firm conclusions could be formed. Therefore, an extensive discussion is not warranted and only a brief summary of the findings is presented here.

Gender. Six studies assessed the relationship between the demographic characteristics of the target family and the target siblings' mental health. On general behaviour rating scales, two studies reported more internalising, externalising, attention, and social problems in female siblings than male siblings (Chen, Faraone, Biederman, & Tsuang, 1994; Dennis & Brotman, 2003). However, the differences were small and score ranges for male and female siblings overlapped considerably. Dia and Harrington (2006) reported no significant gender effect on the target siblings' internalising and externalising behaviour problems scores. Similar to the findings reported in the above sections, on measures of disorder-specific symptomatology, the findings were varied. Male siblings were reported to have higher ADHD, CD, and separation anxiety symptom scores, and poorer global functioning compared to female siblings (Milberger et al., 1996; Müller et al., 2011a, 2011b). However, female siblings had more agoraphobia, simple phobia, social phobia, and panic disorder symptoms. Male and female siblings had approximately equal depression and overanxious disorder symptom scores.

Severity of target child's symptoms. Three studies explored the severity of the target child's MHP as a possible covariate. Listug-Lunde and colleagues (2008) found that siblings of children with high severity ADHD had significantly higher scores on the CBCL, CDI, and had more inattention symptoms than siblings of children with low severity ADHD. No significant difference was found for MASC scores or the hyperactive symptom scores. Sobanski and colleagues (2010) reported higher emotional lability scores for siblings of children with ADHD and high emotional lability scores compared to those of children with ADHD and low emotional lability scores. Last, Dia and Harrington (2006) found that siblings of children with anxiety disorders that had been in treatment for a longer period of time had significantly higher total behaviour

problem scores compared to those of children who had been in treatment for a shorter time. However, no significant difference was found on the internalising and externalising behaviour problem scales.

Family variables. Three studies examined family history of MHPs as a potential covariate of sibling scores on psychological measures. Faraone and colleagues (1998) found that a family history of antisocial disorders either in the parents or the target child with a primary diagnosis of ADHD was significantly related to greater impairments in global functioning and more delinquent and aggressive behaviour problems in target siblings. These findings remained significant after controlling for CD in target children, which suggests that parental antisocial disorders alone are significant covariates. No significant family history effect was found for attention or internalising problems. Brotman and colleagues similarly found that parental psychopathology was linked to more conduct problems in target siblings (Brotman, Gouley, O'Neal, & Klein, 2004). However, the finding was no longer significant once other risk factors were included in the model (e.g. biological and sociocultural risk factors). Last, Dia and Harrington (2006) found no significant parental psychopathology effect for sibling externalising problems, internalising problems, and total behaviour problems.

Five studies examined the associations between family relationships and family environment factors and target siblings' scores on psychological measures. Wasserman and colleagues (1996) found that parent-sibling conflict (e.g. parental rejection, punishment) and emotional support (e.g. parent-sibling communication) were significantly correlated with target siblings' externalising and internalising problems. Both had incremental validity and explained 10% and 13% of the variance in sibling scores, respectively. Parental monitoring was not statistically significant after controlling for conflict and support. Pine and colleagues (1997) similarly found

significant relationships between target siblings' scores on externalising and aggression scales and parental emotional responsivity to the sibling but no significant relationship for parental limit setting. Brotman and colleagues (2004) also found significant correlations between parenting and siblings' externalising problems. However, after including other risk domains (biological and sociocultural), parenting risk factors were no longer significant correlates. It should be noted that these studies focused on younger brothers of male target children with delinquent behaviour problems and on minority families with low-income backgrounds. Thus, generalizability is limited.

Low parent education levels were significantly related to externalising and conduct problems in siblings of children with delinquent behaviour problems (Brotman et al., 2004; Wasserman et al., 1996). Correlations ranged from negligible to moderate (r= 0.17-0.35). Occupational status and family composition (including family size and single parent households) were not significant covariates (Brotman et al., 2004; Wasserman et al., 1996).

Discussion

Summary and Implications

This systematic review of 29 articles allows several conclusions to be made regarding the mental health of siblings of children with MHPs. Firstly, regarding overall patterns of risk, on general (i.e. not disorder-specific) behaviour rating scales (i.e. CBCL, Conners' Rating scales, SAICA), target sibling mean *T*-scores were most consistently in the upper end of the normal range across externalising, internalising, and social problems. This shift points to more or more severe mental health difficulties for siblings of children with MHPs compared to children in the general population. There are two possible explanations for this finding. It may be that while most siblings are not experiencing significant difficulties, a substantial proportion of siblings, greater than

that seen in the general population, experience clinically significant impairments, which increases the overall group mean. This hypothesis is supported by the proportional data outlined in the results. Alternatively, it may be that the entire population of siblings are experiencing more or more severe mental health difficulties than seen in the general population. That is, the increase in the group mean is not due to a cluster of siblings experiencing significant clinical difficulties but that all siblings experience greater subclinical or clinical difficulties than those in the general population, shifting the entire group mean higher. There is some data to support this hypothesis. Chen and colleagues (2008) found no evidence of a distinct cluster of siblings that scored higher on rating scales, but that the entire distribution of sibling scores were shifted towards the upper end. This finding suggests that all siblings experience mental health difficulties to a greater degree than found in children in the general population and attention should be given to the entire population, not just those experiencing clinically significant difficulties. Although there are few studies that explore these two possibilities, the research that has been conducted supports both the notion that a substantial proportion of siblings experience clinically significant difficulties and are in need of some form of mental health treatment and that all or the majority of siblings are also experiencing difficulties to a greater degree than children in the general population. Clinicians should be aware of this increased risk of mental health difficulties for siblings of children with MHPs, enquire about the mental health of siblings, and advise parents to monitor siblings for signs of difficulties.

In relation to the main aim of this paper, the aforementioned findings in this review have extended those from the review of the categorical data (Ma et al., 2014). The latter highlighted that siblings of children with MHPs are at increased risk of displaying or developing MHPs and that attention should be paid to these siblings to

allow early intervention and mental health treatment. The findings of this review however, highlight the possibility that all siblings may be experiencing more mental health difficulties than children in the general population and that they may be experiencing difficulties across a range of psychosocial domains. The absent/present dichotomy underpinning categorical data could not account for siblings who do not meet the criteria for psychopathology but are, nonetheless, experiencing psychological difficulties (e.g. significant social problems). By examining data presented on a continuum and by assessing a range of components of mental health, this dimensionally driven review was able to explore the mental health of the entire population of siblings and consider siblings that may be experiencing mental health difficulties outside the limits of psychiatric diagnostic categories. Furthermore, the findings in this paper suggest that, in addition to facilitating early mental health intervention for siblings that are displaying MHPs, a population-level strategy may be useful, such as support programs or universal prevention programs. This would enable siblings that are presenting with subclinical difficulties to also receive support and may prevent the development of more severe psychopathology in the future.

Secondly, in relation to specific risk profiles for siblings of children with MHPs, this systematic review allows several, albeit tentative, conclusions to be formed. Examination of individual subscales of the CBCL revealed particular areas of impairment for siblings of children with MHPs: Delinquent behaviour, somatic complaints, anxious/depressed behaviour, and social problems. This was consistently found regardless of the diagnosis of the target child. It is unclear why these areas may be particular difficulties for siblings. Sufficient theoretical frameworks or etiological data do not exist to explain these findings. For example, while theories have been developed to explain shared delinquency in target children and their siblings (Feinberg

et al., 2012), these theories rely on pathways such as modelling that apply only where the target child similarly has delinquent behaviour problems. As found in this review, delinquent behaviour problems were not limited to siblings of children with externalising disorders. Future research should explore possible explanatory mechanisms common to siblings of children with a range of MHPs that would account for these particular patterns of impairments in functioning.

In relation to the main aim of this paper, using a dimensional approach allowed identification of specific areas of risk for siblings of children with MHPs while the categorical approach could not. The findings showed that siblings have greater impairments in areas that cross diagnostic boundaries, which may explain why the findings from the previous review did not reveal conclusive patterns. For example, there is no diagnostic category that neatly incorporates all of the particular areas of risk identified in this review. These areas cross several diagnostic categories such as CD, a range of anxiety disorders, and social problems can be present in a variety of diagnoses including CD, ODD, ADHD, anxiety disorders and so on (APA, 1994). Given this risk profile for siblings, one would not expect to find siblings to be at greater risk of one particular psychiatric disorder or cluster of disorders. Thus, this review has provided insight into why no clear patterns were identified in our previous review, has identified key areas of impairment for siblings, and has shown that siblings experience greater impairments across a range of areas relating to mental health, not only an increased risk of developing psychopathology.

However, significant gaps and limitations in our knowledge remain. Firstly, effect size confidence intervals were consistently wide across all studies. Importantly, this also occurred in studies that had large sample sizes. This suggests that the effect size confidence intervals were wide due to considerable within-study and within-group

variation (Higgins & Green, 2011). That is, it is likely that there is considerable variation in the mental health and psychosocial functioning of siblings of children with MHPs. As a result, it is difficult to form any conclusions about effect size estimates as wide confidence intervals suggest that these estimates are limited in precision and the true population effect size may be anywhere between minimal and large. It is unclear what factors may explain this variance and research on moderators is lacking. Research focused on exploring within-study and within-group variation is greatly needed.

Secondly, as discussed above, several areas of functioning were highlighted as particular areas of difficulty for siblings of children with MHPs. However, these areas are quite broad and cover each of the major components of mental health (i.e. emotional, behavioural, and social functioning). Because of this, the implications and clinical guidelines that can be inferred from these findings are limited. For example, parents and clinicians would need to be mindful of difficulties in almost every area of functioning and researchers would need to focus on explanatory mechanisms relating to each of these areas of functioning. This essentially negates the utility of identifying these areas of risk. However, it does highlight the potential of dimensional approaches. Using a dimensional approach was able to highlight specific patterns of risk, albeit quite broad patterns, where a categorical approach could not. It could be argued that with more comprehensive and refined dimensional measures, more specific areas of risk could be identified that would be of greater use to clinicians, parents, and researchers. As can be seen from studies that used the SAICA, an extensive measure of social problems that includes 77 items and several subscales specific to areas of social functioning (John et al., 1987), the use of a more comprehensive and detailed measure allowed a specific area of risk to be identified, namely school behaviour problems, such as difficulty in relating with other students. This could lead to more targeted

intervention and research. For example, school-based prevention programs for siblings could be implemented to encourage prosocial behaviours and clinicians could closely collaborate with the child's teacher to formulate school-based intervention, such as reward charts. Researchers could explore etiological pathways specifically relating to the school setting. For example, it may be that siblings of children with MHPs are also at increased risk of learning disorders that are linked to decreased social competence in the school setting (Nowicki, 2003). It may also be that siblings of children with MHPs experience significant home life disruptions (Ma, Roberts, Winefield, & Furber, in press) leading to increased stress, sleep difficulties, and poor concentration, which may impact on siblings' functioning at school. Thus, while the current state of evidence using a dimensional approach has highlighted some areas of risk for siblings, improvements and advancements in dimensional measures will likely allow more specific conclusions to be formed in the future. We recommend that future researchers use more comprehensive dimensional measures or those that can provide detailed information on areas of functioning, such as the SAICA, when assessing siblings of children with MHPs.

In contrast, disorder-specific dimensional measures may be less informative. On disorder-specific symptom measures, the findings were varied and often inconsistent, particularly with regard to effect size and statistical significance. This variability may be an artefact of variations in measurement instruments used across studies. However, it may also be related to the particular areas of impairment as described above. These areas of impairment are not specific to a particular mental health diagnosis or even to a group of mental health diagnoses, such as internalising and externalising disorders. Thus, it is not surprising that inconsistencies arise when attempting to measure disorder-specific symptoms. In relation to the main aim of this paper, findings on

disorder-specific symptoms did not provide additional insight than that found in our categorical data review. This suggests that dimensional measures that are based in categorical understandings of mental health (i.e. categories of psychiatric disorders) may be less fruitful than dimensional measures based on continuums of psychosocial functioning and mental health.

In sum, using a dimensional approach to understanding the mental health of siblings of children with MHPs provided additional insight and clarity and extended the findings from the previous review of categorical data. Importantly, significant insights can also be obtained from combining data from both approaches. First, a similar pattern of a broad risk of mental health difficulties for siblings was found across both reviews. Siblings of children with MHPs were at increased risk of psychopathology, and although there was a trend for shared internalising and externalising disorders between the target child and sibling, siblings were similarly at risk of developing a broad range of psychopathologies. Similarly here, although siblings of children with MHPs had greater impairment in particular areas of functioning, they had greater impairments compared to control children and norms across all areas of functioning. Thus, the findings of both reviews reciprocally provide support for the one another and collectively provide strong evidence of a broad risk of mental health difficulties for siblings of children with MHPs.

Second, although target sibling mean *T*-scores were in the normal range, when target sibling scores were compared to control sibling scores, the majority of studies reported statistically significant differences and all higher quality studies reported statistically significant differences. Furthermore, the mean scores were all in the upper end of the normal range. If clinicians and researchers relied on the categorical aspect of the above measures, they might conclude that siblings of children with MHPs do not

experience significant difficulties as their scores fall within the normal range. However, when compared to control sibling scores and upon closer examination of the mean scores, we can see that target siblings do experience more or more severe mental health difficulties. On the other hand, interpreting dimensional data without the context provided by the categorical data (i.e. what is considered normal versus clinically significant) would provide little insight into the functioning of siblings. A researcher or clinician would have a score or figure without any indication of its significance and its clinical interpretation or meaning. This highlights the utility and necessity of considering both dimensional and categorical data when assessing siblings of children with MHPs.

Last, global functioning findings from Kelvin and colleagues' (1996) study, as discussed above, show that categorical data (i.e. presence of diagnosis) and dimensional data (i.e. global functioning score) can provide different results and that using one stream only may skew our understanding of that individual. Thus, combining categorically driven data and dimensionally driven data can provide greater clarity, additional insight, and stronger evidence than categorical or dimensional data alone. Similar to what many other authors have suggested (e.g. Rutter, 2011), we argue that it is essential to obtain both categorical and dimensional data in both research and clinical settings and to provide due to consideration to each when assessing siblings of children with MHPs.

Methodological Limitations of Sibling Studies

In addition to the paucity of research in this field, several methodological issues in the included studies limited the conclusions that could be formed in this reviews. First, sample sizes tended to be small with a third of the studies recruiting less than 50 participants. As a result, reliability was reduced and power was reduced, which may

have contributed to inconsistencies across studies regarding statistical significance. Second, only five studies obtained data from multiple informants on the same measure. Research has shown that scores vary across informants and it is therefore important to obtain data from multiple informants (Vermaes et al., 2012). It is particularly important to obtain data across settings (e.g. school and home; Sattler & Hoge, 2006), however only two studies reported separate data for teachers and parents. Third, approximately half of the included studies assessed only one sibling in the family. More problematically, the vast majority of these obtained data on only the sibling closest in age to the target child. It may be that siblings closer in age to the target child are more or less likely to have mental health difficulties or MHPs than other siblings in the family. Last, only three studies assessed the proportion of target siblings with scores in the borderline/clinical range. The vast majority reported only mean scores that, as highlighted above, can give the appearance of normal functioning in target siblings despite a large proportion experiencing significant difficulties.

We recommend future research in this area be conducted according to the following guidelines. Given effect sizes tended to be small to moderate, ideally 200 participants but at least 100 participants should be recruited to ensure sufficient power based on Cohen's power guidelines (Cohen, 1992) and to increase reliability. Data on all available siblings, or on at least one randomly selected sibling, should be assessed to reduce sampling error. Further, when using standardised measures, it is essential that the proportion of siblings scoring in the borderline/clinical range be reported alongside mean scores. Mean scores should also be closely examined and researchers should not base their interpretations on the categorical aspects of the measure only (i.e. normal v clinical range). Lastly, a matched control group of siblings of children without MHPs

should be recruited. This will enable statistical testing and effect size calculations and will justify future meta-analytic syntheses.

Limitations of this Review

Firstly, effect sizes and other statistical testing were not conducted for studies that did not include a matched control samples. The reasons for this were outlined in the methods section. However, this limited the number of effect sizes analysed. Further, it resulted in the effect sizes being solely based on comparisons with control groups. Research shows that effect sizes significantly vary across studies using normative samples as comparison groups compared to those using matched control groups (Vermaes et al., 2012). Effect sizes in studies using control groups were significantly lower than those found from studies using normative samples (Vermaes et al., 2012). Therefore, the findings from this review may underestimate effect sizes. Secondly, as previously discussed, a meta-analysis was not conducted. Lastly, the included studies focused primarily on problem-oriented scales and as a result, this review has focused on siblings that are experiencing mental health difficulties. This problem-oriented approach can overlook the adaptive traits in a population, such as resilience, and can overlook protective factors that significantly influence the development of MHPs. As discussed above, a substantial proportion of siblings are not experiencing significant difficulties despite facing numerous challenges associated with having a brother or sister with MHPs and despite likely exposure to numerous psychopathology risk factors. We believe that it is important to acknowledge this and for future research to explore this subset of siblings and highlight potential protective factors.

Future Directions

This review has highlighted several important methodological issues in the existing literature. High quality research is greatly needed. High quality research on

covariates is also particularly important as it will highlight relevant etiological pathways and what treatment strategies may be particularly beneficial for siblings of children with MHPs. It would also highlight risk and protective factors to guide clinicians' assessments of siblings. Research on what may account for why some siblings experience clinically significant difficulties while others do not is highly recommended. Furthermore, while this paper, in combination with the previous categorical review, has highlighted the increased risk of mental health difficulties for siblings of children with MHPs, there is a key area of childhood functioning that has not been considered, namely family relationships and functioning. Family relationships and the family system are integral parts of a child's life and it is essential to assess these areas to gain a comprehensive understanding of the wellbeing of siblings of children with MHPs (Bronfrenbrenner, 1979; Hoffman, 1981). Last, while this review shows that siblings of children with MHPs are at increased risk for mental health difficulties, it is not yet known if siblings typically receive any form of support or intervention. Research on this topic and research on the effectiveness of existing programs for siblings is greatly needed.

References

Achenbach, T.M., & Rescorla, L.A. (2001). *Manual for the ASEBA School-Age forms & profiles.* Vermont: University of Vermont, Research Center for Children, Youth, & Families.

American Psychiatric Association [APA] (1994). *Diagnostic and statistical manual for mental disorders* (4th ed.). Washington, DC: American Psychiatric Association.

Barrett, P., Rasmussen, P., & Healy, L. (2001). The effects of childhood obsessive-compulsive disorder on sibling relationships in late childhood and early adolescents: Preliminary findings. *Australian Educational & Developmental Psychologist, 7,* 82-102.

Bronfrenbrenner, U. (1979). *The ecology of human development.* Massachusetts, U.S.A.; Harvard University Press.

Brotman, L.M., Gouley, K.K., O'Neal, C., & Klein, R.G. (2004). Preschool-aged siblings of adjudicated youths: Multiple risk factors for conduct problems. *Early Education & Development, 15*, 387-406. doi:10.1207/s15566935eed1504_3

Brotman, L.M., Gouley, K.K., & Chesir-Teran, D. (2005a). Assessing peer entry and play in preschoolers at risk for conduct problems. *Journal of Clinical Child & Adolescent Psychology, 34*, 671-680. doi:10.1207/s15374424jccp3404_9

Brotman, L.M., Gouley, K.K., Chesir-Teran, D., Dennis, T., Klein, R.G., & Shrout, P. (2005b). Prevention for preschoolers at high risk for conduct problems: Immediate outcomes on parenting practices and child social competence. *Journal of Clinical Child & Adolescent Psychology, 34*, 724-734. doi:10.1207/s15374424jccp3404_14

Chen, W.J., Faraone, S.V., Biederman, J., & Tsuang, M.T. (1994). Diagnostic accuracy of the Child Behavior Checklist scales for attention-deficit hyperactivity disorder: A

receiver-operating characteristic analysis. *Journal of Consulting and Clinical Psychology, 62*, 1017-1025. doi:10.1037/0022-006x.62.5.1017

Chen, W., Zhou, K., Sham, P., Franke, B., Kuntsi, J., Campbell, D…Asherson, P. (2008). DSM-IV combined type ADHD shows familial association with sibling trait scores: A sampling strategy for QTL linkage. *American Journal of Medical Genetics(B), 147,* 1450-1460. doi:10.1002/ajmg.b.30672

Christiansen, J., Chen, W., Oades, R.D., Asherson, P., Taylor, E.A., Lasky-Su, J.,…Faraone, S.V. (2008). Co-transmission of conduct problems with attention-deficit/hyperactivity disorder: Familial evidence for a distinct disorder. *Journal of Neural Transmission, 115,* 163-175. doi:10.1007/s00702-007-0837-y

Cohen, J. (1988). *Statistical power analysis for the behavioral sciences* (2nd ed.). New Jersey: Erlbaum.

Cohen, J. (1992). A power primer. *Psychological Bulletin, 112*, 155-159. doi:10.1037/0033-2909.11.1.155

Cohen, N.J., Barwich, M.A., Horodezky, N., & Isaacson, L. (1996). Comorbidity of language and socio-emotional disorders: Comparisons of psychiatric outpatients and their siblings. *Journal of Clinical Child Psychology, 25,* 192-200. doi:10.1207/s15374424jccp2502_8

Conners, C.K. (1997). *Conners' Rating Scales-Revised: Technical manual.* New York: Multi-Health Systems Inc.

Copeland, W., Landry, K., Stanger, C., & Hudziak, J.J. (2004). Multi-informant assessment of temperament in children with externalizing behavior problems. *Journal of Clinical Child & Adolescent Psychology, 33,* 547-556. doi:10.1207/s15374424jccp3303_12

Deal, S. N., & MacLean, W. E., Jr. (1995). Disrupted lives: Siblings of disturbed adolescents. *American Journal of Orthopsychiatry, 65*, 274-281. doi:10.1037/h0079616

Dennis, T., & Brotman, L.M. (2003). Effortful control, attention, and aggressive behavior in pre-schoolers at risk for conduct problems. *Annals of the New York Academy of Sciences, 1008,* 252-255. doi:10.1196/annals.1301.026

Dia, D.A., & Harrington, D. (2006). Research note: What about me? Siblings of children with an anxiety disorder, *Social Work Research, 30,* 183-188. doi:10.1093/swr/30.3.183

Faraone, S.V., Biederman, J., Mennin, D., Gershon, J., & Tsuang, M.T. (1996). A prospective four-year follow-up study of children at risk for ADHD: Psychiatric, neuropsychological, and psychosocial outcome. *Journal of the American Academy of Child & Adolescent Psychiatry, 35,* 1449-1459. doi:10.1097/00004583-199611000-00013

Faraone, S.V., Biederman, J., Mennin, D., Russell, R., & Tsuang, M.T. (1998). Familial subtypes of attention deficit hyperactivity disorder: A 4-year follow-up study of children from antisocial-ADHD families. *Journal of Child Psychology & Psychiatry, 39,* 1045-1053. doi:10.1017/s0021963098002996

Feinberg, M.E., Solmeyer, A.R., & McHale, S. (2012). The third rail of family systems: Sibling relationships, mental and behavioral health, and preventive intervention in childhood and adolescence. *Clinical Child & Family Psychology Review, 15*, 43-57. doi:10.1007/s10567-011-0104-5

Harkavy-Friedman, J. (2009). Dimensional approaches in diagnostic classification: Refining the research agenda for DSM-V. *American Journal of Psychiatry, 166,* 118-119. doi:10.1176/appi.ajp.2008.08060939

Higgins, J. P. T., & Green, S. (Eds.). (2011). *Cochrane handbook for systematic reviews of interventions* (Version 5.1.0): The Cochrane Collaboration.

Hoffman, L. (1981). *Foundations of family therapy: A conceptual framework for systems change.* New York: Basic Books.

Hudson, J. L., & Rapee, R. M. (2002). Parent-child interactions in clinically anxious children and their siblings. *Journal of Clinical Child & Adolescent Psychology, 31*, 548-555. doi:10.1375/bech.22.4.220

Hudziak, J.J., Copeland, W., Stanger, C., & Wadsworth, M. (2004). Screening for DSM-IV externalizing disorders with the Child Behavior Checklist: A receiver-operating characteristic analysis. *Journal of Child Psychology & Psychiatry, 45,* 1299-1307. doi:10.1111/j.1469-7610.2004.00314.x

John, K., Gammon, G.D., Prusoff, B.A., & Warner, V. (1987). The Social Adjustment Inventory for Children and Adolescents (SAICA): Testing of a new semistructured interview. *Journal of the American Academy of Child & Adolescent Psychiatry, 26,* 898-911. doi:10.1097/00004583-198726060-00015

Kelvin, R.G., Goodyer, I.M., & Altham, P.M. (1996). Temperament and psychopathology amongst siblings of probands with depressive and anxiety disorders. *Journal of Child Psychology & Psychiatry, 37,* 543-550. doi:10.1111/j.1469-7610.1996.tb01440.x

Kovacs, M. (1992). *Children's Depression Inventory manual.* New York: Multi-Health Systems Inc.

Kuntsi, J., Wood, A.C., Rijsdijk, F., Johnson, K.A., Andreou, P., Albrecht, B.,…Asherson, P. (2010). Separation of cognitive impairments in attention-deficit/hyperactivity disorder into 2 familial factors. *Archives of General Psychiatry, 67,* 1159-1167. doi:10.1001/archgenpsychiatry.2010.139

Listug-Lunde, L., Zevenbergen, A.A., & Petros, T.V. (2008). Psychological symptomatology in siblings of children with ADHD. *Journal of Attention Disorders, 12,* 239-247. doi:10.1177/1087054708316253

Lopez, M.F., Compton, W.M., Grant, B.F., & Breiling, J.P. (2007). Dimensional approaches in diagnostic classification: A critical appraisal. *International Journal of Methods in Psychiatric Research, 16,* S6-S7. doi:10/1002/mpr.213

McDougall, M.R., Hay, D.A., & Bennett, K.S. (2006). Having a co-twin with attention-deficit hyperactivity disorder. *Twin Research & Human Genetics, 9,* 148-154.

Ma, N., Roberts, R., Winefield, H., & Furber, G. (2014). The prevalence of psychopathology in siblings of children with mental health problems: A 20-year systematic review. *Child Psychiatry and Human Development.* Advance online publication. doi: 10.1007/s10578-014-0459-1

Ma, N., Roberts, R., Winefield, H., & Furber, G. (in press). Utility of qualitative metasynthesis: Advancing knowledge on the wellbeing and needs of siblings of children with mental health problems. *Qualitative Psychology*.

March, J.S. (1997). *Multidimensional Anxiety Scale for Children: Technical manual.* New York: Multi-Health Systems Inc.

Milberger, S., Faraone, S.V., Biederman, J., Testa, M., & Tsuang, M.T. (1996). New phenotype definition of attention deficit hyperactivity disorder in relatives for genetic analyses. *American Journal of Medical Genetics, 67,* 369-377.

Müller, U.C., Asherson, P., Banaschewski, T., Buitelaar, J.K., Ebstein, R.P., Eisenberg, J.,…Steinhausen, H. (2011a). The impact of study design and diagnostic approach in a large multi-centre ADHD study: Part 1: ADHD symptom patterns. *BioMed Central Psychiatry, 11,* 1-17. doi:10.1186/1471-244X-11-54

Müller, U.C., Asherson, P., Banaschewski, T., Buitelaar, J.K., Ebstein, R.P., Eisenberg, J.,…Steinhausen, H. (2011b). The impact of study design and diagnostic approach in a large multi-centre ADHD study: Part 2: Dimensional measures of psychopathology and intelligence. *BioMed Central Psychiatry, 11,* 1-17. doi:10.1186/1471-244X-11-55

Nowicki, E.A. (2003). A meta-analysis of the social competence of children with learning disabilities compared to classmates of low and average to high achievement. *Learning Disability Quarterly, 26,* 171-188. doi: 10.2307/1593650

Pine, D.S., Coplan, J.D., Wasserman, G.A., Miller, L.S., Fried, J.E., Davies, M.,…Parsons, B. (1997). Neuroendocrine response to fenfluramine challenge in boys: Associations with aggressive behavior and adverse rearing. *Archives of General Psychiatry, 54,* 839-846.

Rettew, D.C., Copeland, W., Stanger, C., & Hudziak, J.J. (2004). Associations between temperament and DSM-IV externalizing disorders in children and adolescents. *Journal of Developmental & Behavioral Pediatrics, 25,* 383-391.

Reynolds, C.R. & Richmond, B.O. (1990). *Revised Children's Manifest Anxiety Scale [RCMAS]: Manual* (3rd printing). California: Western Psychological Services.

Rutter, M. (2011). Research review: Child psychiatric diagnosis and classification: Concepts, findings, challenges and potential. *Journal of Child Psychology and Psychiatry, 52,* 647-660. doi:10.1111/j.1469-7610.2011.02367.x

Sattler, J.M. & Hoge, R.D. (2006). *Assessment of children: Behavioral, social, and clinical foundations* (5th ed.). San Diego: Jerome M. Sattler, Publisher, Inc.

Sawyer, M. G., Arney, F. M., Baghurst, P. S., Clark, J. J., Graetz, B. W., Kosky, R. J., . . . Zubrick, S. R. (2000). *The mental health of young people in Australia*. Australia: Commonwealth Department of Health and Aged Care.

Shaffer, D., Gould, M.S., Brasic, J., Ambrosini, P., Fisher, P., Bird, H., & Aluwahlia, S. (1983). A Children's Global Assessment Scale (CGAS). *Archives of General Psychiatry, 40,* 1228-1231. doi:10.1001/archpsyc.1983.01790100074010

Sobanski, E., Banaschewski, T., Asherson, P., Buitelaar, J., Chen, W., Franke, B., . . . Faraone, S. V. (2010). Emotional lability in children and adolescents with attention deficit/hyperactivity disorder (ADHD): Clinical correlates and familial prevalence. *Journal of Child Psychology & Psychiatry, 51*, 915-923. doi:10.1111/j.1469-7610.2010.02217x

Stallings, M.C., Cherny, S.S., Young, S.E., Miles, D.R., Hewitt, J.K., & Fulker, D.W. (1997). The familial aggregation of depressive symptoms, antisocial behavior, and alcohol abuse. *American Journal of Medical Genetics, 74,* 183-191.

State, M.W., Lombroso, P.J., Pauls, D.L., & Leckman, J.F. (2000). The genetics of childhood psychiatric disorders: A decade of progress. *Journal of the American Academy of Child & Adolescent Psychiatry, 39,* 946-962. doi:10.1097/00004583-200008000-00006

Vermaes, I.P., van Susante, A.M., & van Bakel, H.J. (2012). Psychological functioning of siblings in families of children with chronic health conditions: A meta-analysis. *Journal of Pediatric Psychology, 37,* 166-184. doi:10.1093/jpepsy/jsr081

Waldman, I.D., Rowe, D.C., Abramowitz, A., Kozel, S.T., Mohr, J.H., Sherman, S.L., . . . Stever, C. (1998). Association and linkage of the dopamine transporter gene and attention-deficit hyperactivity disorder in children: Heterogeneity owing to diagnostic subtype and severity. *American Journal of Human Genetics, 63*, 1767-1776. doi:10.1086/302132

Wasserman, G., Miller, L.S., Pinner, E., & Jaramillo, B. (1996). Parenting predictors of early conduct problems in urban, high-risk boys. *Journal of the American Academy*

of Child & Adolescent Psychiatry, 35, 1227-1236. doi:10.1097/00004583-199609000-00020

Chapter 5. Paper Three

The Quality of Family Relationships for Siblings of Children with Mental Health Problems: A 20-Year Systematic Review

(Manuscript Under Review)

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Statement of Authorship

Nylanda Ma (Candidate)

I was responsible for the design and implementation of the systematic review, including development of the search strategy, collection of the included studies, data extraction, data analysis, and writing the manuscript. I served as corresponding author and was responsible for manuscript submission, revisions, and responses to journal reviews.

Signed: Nylanda LT Ma _____ Date: 4/8/2014

Dr. Rachel Roberts, Prof. Helen Winefield, & Dr. Gareth Furber (Co-authors)

We provided ongoing supervision through the systematic review and there was ongoing collaboration between Nylanda Ma and ourselves in refining the direction of research and methodology. We commented on manuscript drafts, made suggestions on the presentation of the material in the manuscript, and provided guidance on journal revisions and responses. We hereby give our permission for the inclusion of this paper in Nylanda Ma's submission for the degree of Doctor of Philosophy from the University of Adelaide.

Signed: Dr. Rachel Roberts _____ Date: 4/8/2014

Signed: Prof. Helen Winefield _____ Date: 4/8/2014

Signed: Dr. Gareth Furber _____ Date: 4/8/2014

Abstract

While it is well recognised that family relationships have a significant impact on child mental health and vice versa, little research has examined the impact of living with a child with mental health problems (MHPs) on family relationships for their siblings. This reports aims to synthesise the existing literature and comment on the current state of evidence. An electronic search of publications from 1990-2011 was conducted and 22 studies were included. The findings show that families of children with MHPs have less positive (e.g. less supportive) and more negative (e.g. more conflictual, aggressive) sibling relationships and more negative parent-sibling relationships compared to control families. Exceptions were sibling relationships in families of children with conduct disorder (more positive) and anxiety disorders (did not significantly differ from controls). Limitations of the existing literature include the types of measures used and the use of single informants. Methodological guidelines for future studies are outlined.

Keywords: sibling, child, mental health problem, family relationships, systematic review

According to family systems theories and socio-ecological theories, there is a reciprocal relationship between family functioning and individual level functioning. Specifically, a systems perspective maintains that individuals and the systems, such as schools or communities, that they belong to and form part of are inherently interrelated (Hoffman, 1981). As a result, changes in one system (e.g. family) are associated with changes in all other systems (Hoffman, 1981). Similarly, Bronfenbrenner's (1979) socio-ecological perspective holds that individual development is influenced by multiple systems and the individual, in turn, influences those systems. Applying this to child mental health, behaviour problems in the child are likely to affect family functioning and the quality of family functioning is likely to affect the frequency and intensity of the child's behaviour problems. For example, if a child displays aggressive, noncompliant behaviour, the parents may be more likely to use more harsh and punitive parenting strategies which, in turn, may increase the frequency of aggressive, noncompliant behaviour in the child.

There is a wealth of research supporting these theories, particularly the dynamics between children with mental health problems (MHPs) and their parents. Research has found that impaired family relationships impact on the wellbeing of children. For example, children from families where the parents are hostile towards each other have poorer emotional wellbeing (Baxter, Weston, & Qu, 2011). Further, research has shown that poorer wellbeing in family members impacts on family relationships. For example, parents of children with MHPs report poorer family cohesion and increased likelihood of marital relationship breakdown and parental psychological distress (Slowik, Wilson, Loh, & Noronha, 2004).

Two family relationships that have been neglected in this literature include the relationship between the child with MHPs and their siblings, and the relationship

between siblings of children with MHPs and their parents. This is despite the significant role siblings play in shaping family dynamics (Cox, 2010). Sibling relationships are different from parent-child relationships. For example, siblings tend to spend more time together and have a smaller age gap than relationships than between parents and children (Cox, 2010; Feinberg, Solmeyer, & McHale, 2012). Siblings also contribute significantly to each other's development, for example, through contributing to the development of social skills and understanding and identity formation (Feinberg, et al., 2012; Kramer, 2010). Furthermore, poor sibling relationships are predictive of poorer adult outcomes (Gully, Dengerink, Pepping, & Bergstrom, 1981). In particular, several authors have suggested that the quality of childhood sibling relationships is predictive of the quality of adult relationships, including marital relationships (Cicirelli, 1982). For example, experiences of sibling violence in adolescence have been shown to significantly predict becoming victim to or becoming a perpetrator of intimate partner violence in adulthood, beyond the effects of parental violence in childhood (Noland, Liller, McDermott, Coulter, & Seraphine, 2004). Adult relationship problems such as these can have further detrimental effects on the mental and physical health of offspring (Campbell & Lewandowski, 1997). In this way, sibling relationships can have a long-term impact on individuals, can have detrimental intergenerational effects, and may contribute to a perpetuating cycle of maladaptive relationship formation. This highlights the need for research into the quality of sibling relationships, particularly with a population that is likely to be at high risk of impairments in family relationships and functioning such as siblings of children with MHPs.

In this paper, we sought to synthesise the available literature on the quality of family relationships for siblings in families with a child with MHPs. Specifically, we sought studies that examined the relationships between children with MHPs and their

siblings and between siblings of children with MHPs and their parents. This review had two main aims: a) clarify the current state of evidence on the quality of family relationships for siblings of children with MHPs, and b) assess the methodologies used across studies in this field.

Methods

The method used in this review has been described in detail elsewhere (Ma, Roberts, Winefield, & Furber, 2014a,b). Briefly, four electronic databases were searched from January 1990 to July 2011: Cumulative Index to Nursing and Allied Health Literature (CINAHL), PsycINFO, PubMed, and SciVerse Scopus using terms such as *sibling; first degree relative; child; emotional; behavioural.* Appendix A provides a full list of search terms used in this systematic review. As this review is part of a larger systematic review on multiple aspects of the mental health and wellbeing of siblings of children with MHPs, the list of search terms was designed to be inclusive and did not specify search terms relating to the outcome in siblings measured (e.g. *family functioning*). This has significant benefits. Given that multiple terms can be used to describe family relationships (e.g. family relationships, parent-child relations, home environments), a search strategy that does not include parameters about outcome terms is likely to capture a greater number of potentially relevant studies.

Studies were included if they a) reported quantitative data on the quality of family relationships for siblings of a target child with MHPs (i.e. either reported sibling relationship data or parent-sibling relationship data) and/or data on moderators of family relationship quality, b) reported primary data, c) were published in a peer-reviewed journal, and d) were published in English. Data on overall family environment with no specific reference to siblings was not included. In this study, children (2-18 years) were considered to have MHPs if they had an established mental illness

diagnosis, screened positive on diagnostic measures, had clinically elevated scores on psychological or mental health measures (e.g. behavioural checklists), had been referred to or were attending a mental health clinic or mental health treatment, or had been adjudicated for delinquency. Throughout this review, children with MHPs and their families are referred to using the designation 'target' and control families (containing children without MHPs) using 'control'.

The initial search identified 4,387 records after removing duplicates (see Figure 1). After screening titles and abstracts, 761 full-text articles were reviewed. The final pool of studies consisted of 22 articles. Where possible, *t*-tests were conducted and Cohen's *d* was calculated or these statistics were extracted directly from the included studies (Cohen, 1988).

A meta-analysis of effect sizes was not conducted due to limited number of studies, informant effects, and conceptual differences in relationship factors measures. For example, while self-reported warmth/closeness and observed affectionate gestures are similar, the concepts are not matched closely enough to warrant synthesising them statistically. Warmth and closeness can be represented by several behaviours, such as prosocial behaviour and shared interests, and affection represents only one aspect of warmth and closeness (Furman & Buhrmester, 1985).

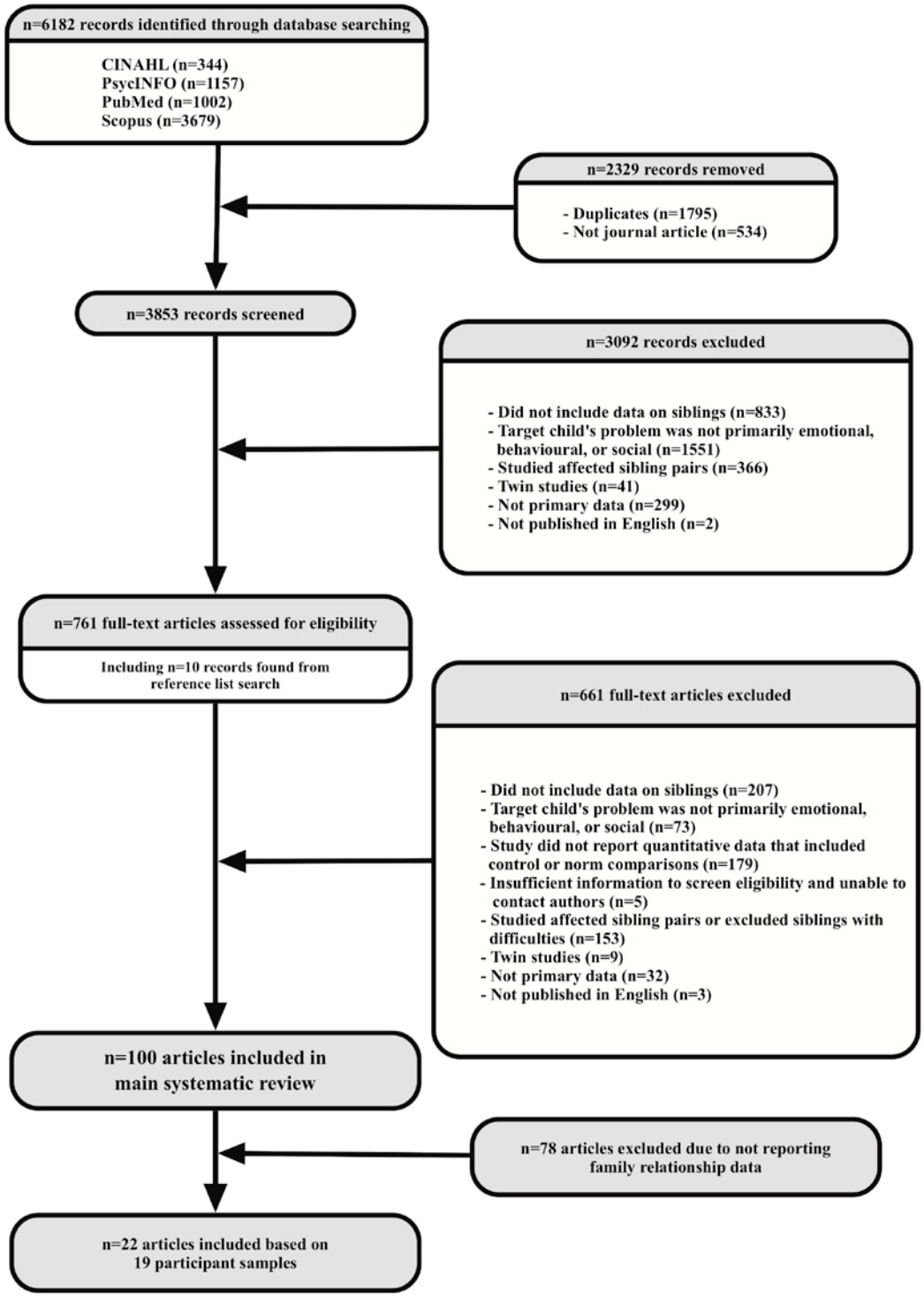

Figure 1. Flow of included studies.

Even if we assumed that these concepts could be synthesised, in relationship research, because perceptions of relationship quality typically differ across informants, it is essential to control for informant effects using subgroup meta-analytic techniques (Holmbeck, Li, Schurman, Friedman, & Coakley, 2002; Serot & Teevan, 1961). However, there were too few studies to warrant this approach (Valentine, Pigott, & Rothstein, 2010) with typically only one or two studies reporting data from a particular informant. Thus, the following is a narrative synthesis with particular attention on consistent findings and methodological quality.

Results

Characteristics of Included Studies

This review is based on 22 studies assessing approximately 3,142 participants across six countries, as summarised in Table 1. Participants were target children with MHPs, their parents, and/or their siblings. The ages of target children and siblings ranged from 3-18 and 4-18 years, respectively. The majority of these studies were conducted in Western countries (21 studies) and recruited target children and their families from mental health treatment programs (16 studies) and/or from the general population (10 studies). The size of participant samples tended to be very small (10 studies with $n<50$) with only five studies assessing large sample sizes ($n>100$). Studies with larger sample sizes were considered to be of higher quality due to increased generalizability and reliability. The majority of studies (17 studies) recruited a control group of either control children matched to the target child's characteristics (e.g. age, sex) or family members of these children (e.g. siblings of control children).

Table 1

Characteristics of Included Studies

Reference	Country	Target child			Sibling		Control	Measure (Informant)
		MHP	*n*	Age(y)/%Male	*n*	Age(y)/%Male	*n*	
Biederman et al. (1993)	US	ADHD	140	6-17 / 100%	--	-- / --		SAICA: 9 sibling items, 10 parent-sibling items (P/C)
Faraone et al. (1996)								
Faraone et al. (1998)			--		169-174		129-143	(P/S)
Greene et al. (2001)			99	6-17 / 100%				(P/C)
			227	6-17 / 50%			129-143	(P/C)
Geller et al. (2000)	US	ADHD	93	7-16 / 79%	--		94	PSS: 1item (P/C)
		Bipolar	81	/ 61%				
Mikami & Pfiffner (2008)	US	ADHD	77	5-11/ 82%	77	4-18 / 57%	14	SRQ: 48 items (P/C/S)
Tseng et al. (2011)	Taiwan	ADHD	41	9.7 (2.4) / 78%	--		204	SAICA: 9 sibling items, 10 parent-child items (P)
		ODD	14	10.6 (2.4) / 71%				
		ADHD+ODD	47	11.1 (2.7) / 87%				

Table 1 (cont.)

Characteristics of Included Studies

Reference	Country	Target child			Sibling		Control	Measure (Informant)
		MHP	*n*	Age(y)/%Male	*n*	Age(y)/%Male	*n*	
Dadds et al. (1992)	AUS	CD	27	9.6 (2.3) / 88%	30	8.3 (3.8) / 56%	16	Family Observation Schedule: 5 sibling items, 5 parent-sibling items (O)
		DEP	18	10.3 (2.3) / 87%	24	9.8 (3.1) / 43%		
		CD+DEP	12	10.5 (2.2) / 88%	16	10.4 (3.5)/ 29%		
Daniels & Moos (1990)	US	CD	58	12-18 / 63%	--		38	Life stressors and social resources inventory: 6 items (C)
		DEP	49	/ 39%				
Green et al. (2002)	US	ODD	643	10.6(3.4) / 73%	--			SAICA: 9 sibling items, 10 parent-child items (P)
		ODD+CD	262	10.8(3.7) / 80%				
		Other MHPs	487	10.7(3.5) / 70%				
Schachar & Wasmuth (1990)	Canada	CD	22	7-11 / 100%	--		20	Semi-structured interview: 3 items (P/C)
		ODD	21					
Slomkowski et al. (1997)	US	Delinquency	29	15.4 (1.4) / 100%	29	8.4 (1.5)/ 100%	16	Social Interaction Between Siblings: 87 items (P)
Geller et al. (2001)	US	DEP	72	6-12 / --			28	Longitudinal Interval Follow-up Evaluation: 1 item (C)

Table 1 (cont.)

Characteristics of Included Studies

Reference	Country	Target child			Sibling		Control	Measure (Informant)
		MHP	*n*	Age(y)/%Male	*n*	Age(y)/%Male	*n*	
Puig-Antich et al. (1993)	US	DEP	62	14.7(1.8) / 44%			38	PSS: 1 item (P/C)
Barrett et al. (2001)	AUS	OCD	4	10-13 / 25%	5	8-12 / 40%	5	SRQ: 48 items (S)
Hudson & Rapee (2002)	AUS	ANX	37	7-16 / --	37	7-16 / --	20	Observations of parent-child interaction: 5 items (O)
Lindhout et al. (2003)	NL	ANX	24	8-13 / 56%	--		25	Sibling Relationship Inventory: 17 items (C)
Toro et al. (1992)	Spain	OCD	72	5-18 / 65%	--		72	Parent descriptions: 1 item (P)
Deal & MacLean (1995)	US	Range of MHPs	15	12-18 / --	15	9-15 / 53%	15	SRQ: 48 items (S) Structured interview: 4 items (S)
Donenberg & Baker (1993)	US	Externalising	22	3.5-6 / 73%	--		22	Family Impact Questionnaire: 9 items (P)
Dumas (1996)	Canada	Externalising	22	3-10 / 64%	--		11	INTERACT Coding System: 5 scales (O)
Stormont-Spurgin & Zentall (1995)	US	Externalising	48	3-5 / 100%	--		15	Qualitative interview: 1 item (P)

Note. ADHD: Attention deficit hyperactivity disorder; ANX: Anxiety disorders; CD: Conduct disorder; DEP: Depression; OCD: Obsessive-compulsive disorder; ODD: Oppositional defiant disorder; MHP: Mental health problem; PSS: Psychosocial Schedule for School Age Children; SAICA: Social Adjustment Inventory for Children and Adolescents; SRQ: Sibling Relationship Questionnaire; P: Parent; C: Target child; S: Sibling; O: Independent observer; AUS: Australia; NL: Netherlands; US: United States of America

Approximately half of the included studies assessed family relationships using only one informant (10 studies). Very few obtained data from target siblings (four studies) or from all family members (one study). Given that perceptions of family relationships differ significantly across family members, higher quality studies are those that obtain data from multiple informants and provide a more reliable and comprehensive picture of family relationships (Dancyger, Formari, Scionti, Wisotsky, & Sunday, 2005). Observational studies of all family members are considered high quality, though are limited by only assessing observed behaviours. That is, these studies could not assess relationship aspects that are not easily or entirely expressed in observable behaviours (see above example regarding warmth/closeness). As previously highlighted, perceptions are an important aspect to consider when assessing relationship quality and purely observational studies cannot provide such data.

Studies also varied in the type of measure used, ranging from specifically designed family relationship measures to general psychosocial functioning measures. Quality was defined by the following in descending order: Comprehensive family measures assessing multiple relationship components (e.g. warmth, dominance, conflict), general psychosocial measures including five or more items per scale and/or multiple outcome scales (e.g. problems, such as conflict, and positives, such as shared activity, in relationships), and psychosocial measures including only one item per scale and/or only one outcome scale. General psychosocial measures are scales that have been developed to measure a range of psychosocial outcomes. The Social Adjustment Inventory for Children and Adolescents (SAICA; John, Gammon, Prusoff, & Warner, 1987), for example, was designed to measure a range of social behaviours including school behaviour, peer relationships, and family relationships. Because these measures do not focus on relationship components, they are often less comprehensive and

measure only a few aspects of relationship quality. For example, in the SAICA, the only positive sibling relationship components measured are shared activity, friendliness and affection, and talking with siblings. While this covers several areas of what children consider to be a good aspect of sibling relationships (Furman & Buhrmester, 1985), it does not include other important areas such as emotional support. As such, data from these measures may not provide a complete and accurate picture of an aspect of relationship quality, such as warmth and closeness, because not all components relating to that aspect are measured. Thus, studies that use these measures are considered to be of lower quality than those that use comprehensive relationship measures. For similar reasons, those that use psychosocial measures that include only one item or one outcome scale are considered to be of lower quality as they do not assess a comprehensive range of behaviours and perceptions that impact on the perceived quality of relationships.

The following sections present the results of the narrative synthesis structured by type of family relationship, type of relationship component (i.e. positive or negative as loosely defined by past research; Furnam & Buhrmester, 1985), and by informant type.

Sibling Relationships

Positive relationship components. Nine studies examined positive components in sibling relationships, such as warmth, closeness, and shared activities. Two studies examined target children's positive perceptions of sibling relationships (Daniels & Moos, 1990; Lindhout et al., 2003). Both of these studies were considered lower quality due to recruiting small samples (n=24-58). Target children with anxiety disorders and affective disorders viewed their relationship with their sibling as less affectionate and as having fewer resources, such as emotional support, compared to control children. In

contrast, target children with conduct disorder (CD) viewed the sibling relationship as more positive, specifically as providing greater sibling resources, than control children. All findings did not reach statistical significance. Effect sizes could only be calculated for Lindhout and colleagues' (2003) data as Daniels and Moos (1990) did not report standard deviation data. Target children with anxiety disorders perceived less affection in the sibling relationship compared to control children to a small to moderate degree (*d*=0.34, 95% CI [-0.24, 0.89]).

Two studies assessed sibling's perceptions of the positive aspects of sibling relationships (Barrett, Rasmussen, & Healy, 2001; Deal & MacLean, 1995). These studies were also considered to be lower in quality due to recruiting extremely small sample sizes (*n*=5-15). Siblings of children with obsessive-compulsive disorder (OCD) and those of children with a range of MHPs reported less warmth/closeness, intimacy, similarity (e.g. in interests or personality, and admiration in their sibling relationships than controls). There was one exception: Siblings of children with a range of MHPs had approximately equal ratings of warmth and closeness as control siblings (*M*=21.5 and 21.4, respectively; Deal & MacLean, 1995). All of these findings did not reach statistical significance. Effect sizes could not be calculated from Barrett and colleagues' (2001) study, as they did not report standard deviation data. In Deal and MacLean's (1995) study, effect sizes (not including those for warmth and closeness) were minimal to small (*d*=0.09-0.23). Larger effect sizes were reported for intimacy and similarity (e.g. in interests and personality) compared to those for admiration of sibling and admiration by sibling. Effect size confidence intervals were wide suggesting limited precision in effect size estimates (e.g. *d*=0.23, 95% CI [-0.50, 0.93]; Deal & MacLean, 1995).

One study examined parental perceptions of sibling relationships (Slomkowski, Wasserman, Schaffer, Rende, & Davies, 1997). This study was considered to be lower in quality due to a small sample size (n=29). However, a comprehensive relationship measure was used, the Social Interaction Between Siblings measure, that included 87 items (SIBS; Slomkowski et al., 1997). Parents of children with delinquent behaviour problems reported significantly more positive qualities (e.g. warmth and admiration) in the sibling relationship compared to control parents to a large degree (d=0.73, 95% CI [0.24, 1.24]). However, the effect size confidence interval was wide, spanning from small to large, suggesting limited precision in the effect size estimate, likely due to a small sample size.

Four studies examined the positive aspects of sibling relationships using multiple informants and/or observations of all family members. The three self-report studies were with target children with attention-deficit hyperactivity disorder (ADHD). These studies were of good quality employing multiple informants, a measure with more than five items per scale, and large sample sizes (n=140-174). Based on reports from parents, target boys with ADHD, and their siblings, sibling relationships in these families had significantly greater impairments in sibling activities (i.e. play, affection, talking) than control children (Biederman, Faraone, & Chen, 1993; Faraone, Biederman, Mennin, Gershon, & Tsuang, 1996). Effect sizes ranged from small to moderate (d=0.29-0.53). The effect size confidence interval for the small effect size ranged from minimal to moderate (95% CI [0.06, 0.51]) and for the moderate effect size ranged from small to large (95% CI [0.30, 0.75]). Thus, the effect size confidence intervals were still considerably wide and suggest that the plausible range of effect sizes includes minimal to large effects. These studies had large sample sizes suggesting that the confidence intervals were wide due to considerable within-study and within-group

variation. Mikami and Pfiffner (2008) similarly found that the target child-sibling relationship was characterised by less warmth and closeness compared to control children. However, the difference was not statistically significant. Lack of statistical significance may be due to smaller sample size relative to the studies above (n=77). Based on a small to moderate effect size, a minimum sample size of 100 participants would be needed, according to Cohen's power calculations (Cohen, 1992). Thus, it is likely that Mikami and Pfiffner's (2008) study lacked sufficient power to detect statistically significant differences.

Last, Dumas (1996) assessed the frequency of observed positive behaviours between the sibling and target child with externalising behaviour problems. The frequency of approval (e.g. of conduct), positive affect (e.g. affectionate gestures), and compliant behaviours between the target child and sibling was less than that observed in control families. The difference between target and control families in the frequency of positive affect was statistically significant while differences in approval and compliant behaviours were not. The associated effect size was large, however the confidence interval was wide suggesting limited precision and reliability in this effect size estimate, likely due to a small sample size (d=1.15, 95% CI [0.32, 1.86]). Effect sizes for frequency of approval and compliant behaviours were in the moderate range (d=0.43 and 0.44, respectively) with wide confidence intervals also (95%CIs [-0.33, 1.13] and [-0.32, 1.14], respectively). Individual level analyses revealed that siblings were significantly more compliant with the target child's requests and instructions than vice versa. No significant differences in individual level analyses were found for positive affect and approval.

In sum, across all nine studies, sibling relationships in families of children with MHPs were less positive than in control families. However, few studies reported

statistically significant findings. Those that did were higher quality studies that recruited large sample sizes (n>100) and had sufficient statistical power, obtained data from multiple informants, or those that obtained independent observational data on all family members. Based on higher quality studies, effect sizes ranged from small to moderate. However, effect size confidence intervals were wide, spanning from small to large. Thus, it is difficult to estimate a precise effect size estimate. There was one consistent exception to these findings: Parents and target children with CD or delinquent behaviour problems rated sibling relationships as more positive than control families (Daniels & Moos, 1990; Slomkowski et al., 1997). While these studies were of poorer quality, recruiting small sample sizes and obtaining data from only one informant, the consistency in this finding should also be considered. With regards to informant effects, effect sizes were larger for parent report and target child report compared to sibling self-reports. However, given the limited number of studies and without direct statistical comparisons, no firm conclusions can be drawn.

Negative relationship components. Fifteen studies assessed the negative qualities of sibling relationships, almost twice as many studies than those that examined positive sibling relationship components (nine studies). Three studies assessed target children's perceptions of the negative aspects of sibling relationships (Daniels & Moos, 1990; Geller, Zimerman, Williams, Bolhofner, & Craney, 2001; Lindhout et al., 2003). All of these studies were considered to be lower quality due to small sample sizes and/or the use of general psychosocial measures. Daniels and Moos (1990) and Lindhout and colleagues (2003) found that target children rated their sibling relationships as more hostile and as having a greater number of associated stressors, such as criticalness, than control children. This was found for target children with CD, affective disorders, and anxiety disorders. However, the findings did not reach

statistical significance. As previously discussed, these studies did not recruit sufficiently large sample sizes to obtain sufficient statistical power to detect statistically significant differences. The effect size obtained from Lindhout and colleagues' (2003) study was in the moderate range (d=0.44, 95% CI [-0.14, 0.99]).

Geller and colleagues (2001) conducted a 10-year follow-up study of children with depression. These children reported greater impairment in their sibling relationships, such as avoidance and arguments, in the past five years than control children. However, only ratings from targets with a mood or substance use disorder in the past 12 months significantly differed from controls. Though it should be noted that targets without these disorders in the past 12 months also reported greater impairment in sibling relationships than controls, albeit not to a statistically significant degree after correcting for multiple comparisons (p<0.05; corrected alpha value set at p< 0.0001). The effect for those with these disorders in the past 12 months was large and the confidence interval, though wide, was consistently in the large range (d=1.25, 95% CI [0.74, 1.75]). The same pattern was found when expanding the time period to the past five years. Only targets that had a mood or substance use disorder in the past five years significantly differed from controls. The effect size was large but the confidence interval was wide suggesting limited precision (d=1.12, 95% CI [0.61, 1.60]). When considering these findings, it should be noted that, in addition to the aforementioned methodological limitations, Geller and colleagues (2001) used a general psychosocial measure (see Table 1) that included only one item relating to sibling relationships. Thus, these findings should be interpreted with caution.

Three studies assessed parental perceptions of the negative aspects of sibling relationships (Slomkowski et al., 1997; Stormont-Spurgin & Zentall, 1995; Tseng, Kawabata, & Gau, 2011). Each of these studies was considered to be of lower quality

with small sample sizes and obtaining data from a single informant. Parents of target children reported more negative sibling relationships than control parents. This was found with parents of children with ADHD, oppositional defiant disorder (ODD), delinquent behaviour problems, and mixed externalising difficulties and included negative components such as aggression, bullying, blaming, and negative influence or dominance over each other. Findings in terms of statistical significance were varied. First, Tseng and colleagues (2011) found that ratings of sibling relationship problems from parents of children with ADHD did not significantly differ from controls, but those from parents of children with ODD and of children with ADHD and ODD did. The effect size for the former was small to moderate (d=0.32, 95% CI [-0.02, 0.66]). Effect sizes for statistically significant findings were large (d=1.04 and 1.37, respectively). The effect size confidence interval for the difference between parents of children with ADHD and ODD and controls was relatively narrow and the range of plausible values was contained to large effect sizes (95% CI [1.03, 1.70]). This suggests that we can be reasonably confident in concluding that there is a significant difference between these groups to a large degree. In contrast, the effect size confidence interval for the difference between parents of children with ODD and controls was wide, spanning from moderate to large, suggesting less precision in the effect size estimate (95% CI [0.48, 1.58]). These findings suggest that children with comorbid externalising difficulties may have greater impairments in their sibling relationships than those with symptoms of one type of externalising difficulty.

Second, Slomkowski and colleagues (1997) found the parents of children with delinquent behaviour problems reported more negative qualities (e.g. destroying their sibling's property) in the sibling relationship and more negative influence (e.g. going along with the sibling's 'bad' act) than control parents. The effect size for negative

qualities was minimal, the confidence interval was wide, and the difference was not statistically significant (*d*=0.07, 95% CI [-0.46, 0.54]). The effect size for negative influence was large, the difference was statistically significant, and the confidence interval, though wide, was consistently in the large range (*d*=1.33, 95% CI [0.70, 1.79]).

Last, Stormont-Spurgin and Zentall (1995) compared parent reports of retaliatory aggression between the target child and their sibling from parents of children with hyperactivity, with aggression, with hyperactivity and aggression, and of control children. They found a significant difference across the three groups. Parents of children with hyperactivity and with hyperactivity and aggression reported higher scores (14.58% and 31.25%, respectively) than those with children with aggression only and control parents (8.33% for both groups). It should be noted that these data were obtained from a single item in a qualitative interview, thus the reliability and validity of these findings are limited.

As can be seen in the above discussion, findings regarding statistical significance were varied. Because each of these studies assessed a different type of negative relationship component, it is difficult to ascertain any clear pattern as to why some differences were statistically significant and others were not. Further, each of these studies were similarly of poor quality, thus no conclusions could be formed as to which findings may be more reliable. Two consistent findings across the studies were that parents of children with MHPs perceived more negative sibling relationships than control parents, though whether this reached statistical significance varied across and within studies. When statistically significant differences were found, the effect size was typically, and with reasonable confidence based on confidence intervals, a large effect size.

Two studies obtained sibling self-reports on the negative aspects of target child-sibling relationships (Barrett et al., 2001; Deal & MacLean, 1995). As highlighted previously, both of these studies are considered to be poor quality due to extremely small sample sizes (*n*=5-15). Both studies found that siblings of children with anxiety disorders and a range of MHPs perceived more conflict in the target child-sibling relationship than control children. However, in both studies, the difference was not statistically significant. The effect size calculated from Deal and MacLean's (1995) study was in the moderate range with a wide confidence interval likely due to a small sample size (*d*=0.50, 95% CI [-0.25, 1.20]). Similarly, on the relative power/dominance subscale of the Sibling Relationship Questionnaire (SRQ; Furnam & Buhrmester, 1985) that assesses the sibling's nurturance of and dominance over the target child relative to nurturance and dominance by the target child, both studies reported negative scores on this subscale. This indicates that target siblings felt more dominated by the target child and had less power in the relationship. However, when compared to control scores, the studies reported inconsistent findings. Barrett and colleagues (2001) found that target siblings and control siblings reported the same degree of dominance. Deal and MacLean (1995) found that siblings of children with MHPs reported feeling more dominated by the target child than control siblings. However, the difference was not statistically significant and the effect was small with a wide confidence interval (*d*=0.22, 95% CI [-0.51, 0.93]).

Seven studies obtained data from multiple informants via self-report or observations of all family members (Biederman et al., 1993; Dumas, 1996; Faraone et al., 1996; Geller, Bolhofner, Craney, Williams, DelBello, & Gundersen, 2000; Mikami & Pfiffner, 2008; Schachar & Wachsmuth, 1990; Puig-Antich, Kaufman, Ryan, & Williamson, 1993). All of the six self-report studies found that sibling relationships in

families of children with MHPs were more negative than in control families. This was found in families of children with ADHD, CD, ODD, and affective disorders on a range of negative relationship components including avoidance, conflict, bullying, fighting, and criticalness. Each of these seven studies reported statistically significant differences between target families and control families. There were two exceptions. Geller and colleagues (2000) found that siblings of children with bipolar disorder had significantly more sibling relationship problems than control siblings while siblings of children with ADHD did not. Schachar and Wachsmuth (1990) found that siblings of children with ODD had significantly more sibling relationship problems while those of children with CD did not. These were the only two studies that included data on target children with these disorders. As such, it is unclear how reliable and accurate these findings are and whether there are real differences in sibling relationship quality depending on the diagnosis of the target child.

With regard to effect sizes, effect sizes could only be calculated for two of the seven studies due to insufficient data (Faraone et al., 1996; Puig-Antich et al., 1993). The calculated effect sizes ranged from small to moderate (d=0.27-0.51) and were found on broad sibling relationship problem scales. The related effect size confidence intervals were all wide and spanned several interpretive ranges (95% CIs [0.03, 0.49], [0.17, 0.69], and [0.09, 0.91]).

Last, Dumas (1996) obtained observational data on all family members and assessed the frequency of observed negative behaviours between target children with externalising behaviour problems and their sibling. Target children and siblings were more aggressive towards each other than controls but made fewer attempts to control each other than control children. Both findings were non-significant with small effect sizes (d= 0.10-0.11). Both the confidence intervals for attempts to control each other

(95% CI [-0.82, 0.63]) and for aggression (95% CI [-0.62, 0.83]) were extremely wide. Individual level analyses revealed that target children were significantly more aggressive towards their siblings even though their sibling was not significantly more aggressive towards them. Target children and their siblings did not significantly differ in the frequency of attempts to control one another.

In sum, across all fifteen studies, the target child-sibling relationship was more negative than in control families, with the exception of dominance and control, as reported in Barrett and colleagues (2001) and Dumas' (1996) study above, and in retaliatory aggression for target children with aggression only, as reported in Stormont-Spurgin and Zentall's (1995) study above. Statistical significance varied considerably across lower quality studies. However, all higher quality studies that obtained data from multiple informants and/or had large sample sizes reported statistically significant differences between target families and control families (with the two exceptions noted above). Based on higher quality studies, the effect size is small to moderate. However, the confidence intervals were considerably wide suggesting limited confidence and precision in these effect size estimates and likely considerable within-study and within-group variance. There were two cases where the effect size confidence intervals, though wide, were contained within one interpretive category. That is, although the confidence interval was wide, the range of plausible values was always in the large range. However, both were considered to be of lower quality, with small sample sizes (n<50). Thus, the reliability and validity of these findings are unclear and they should be interpreted with caution. With regards to informant effects, effect sizes were larger for parent report and target child report compared to sibling self-reports (moderate to large versus small to moderate, respectively). However, given the limited number of studies and without direct statistical comparisons, no firm conclusions can be drawn.

Parent-Sibling Relationships

Positive relationship components. Only two studies examined the positive aspects of parent-sibling relationships (Dadds et al., 1992; Faraone et al., 1996). Faraone and colleagues (1996) obtained data from siblings and parents of boys with ADHD on the frequency of shared activities, how friendly or affectionate the sibling is to the parent, and if the sibling talks with the parent. Target siblings and mothers had significantly greater impairments in these aspects than controls. The effect sizes were small (d=0.30 and 0.31) and effect size confidence intervals were considerably wide, spanning from minimal to moderate (95% CIs [0.07, 0.53] and [0.01, 0.46], respectively). Target siblings and fathers had equal or poorer scores on positive relationship aspects than controls. For the latter, the difference was non-significant with a small effect size (d=0.23). The effect size confidence interval mirrored that found for mothers and siblings (95% CI [0.08, 0.53]). The second study assessed the frequency of positive behaviours between siblings and parents in three groups of target children (CD, depression, and CD and depression; Dadds, Sanders, Morrison, & Rebgetz, 1992). Based on independent observations, parents and siblings of target children with CD, depression, and CD and depression less frequently displayed positive behaviours towards one another (e.g. praise) compared to controls. There was one exception: Mothers and siblings displayed more positive behaviours in families of children with CD and depression compared to controls. However, all findings did not reach statistical significance. Effect sizes ranged from small to large (d=0.11-0.95). Moderate to large effect sizes were found for mother-sibling positive and father-sibling positive behaviours in CD families and for mother-sibling positive behaviours in CD and depression families. However, all effect size confidence intervals were extremely wide

suggesting limited precision in these effect size estimates (e.g. 95% CI [-0.29, 1.59] for mother-sibling positive in CD families).

In sum, parent and sibling relationships in target families did not significantly differ from control families in positive relationship components. However, the majority of findings (8 of 10) were in the direction of less positive parent-sibling relationships in target families compared to control families (see exceptions above). There was significant variability in effect size estimates and all confidence intervals were wide suggesting limited precision. Larger effect sizes were found for mother-sibling relationships across both studies. However, no firm conclusions can be made given wide confidence intervals.

Negative relationship components. Six studies examined the negative aspects of parent-sibling relationships in families of children with MHPs. Only one study obtained data from the perspective of the target child and had a very small sample size (n=24). Thus, the findings should be interpreted with caution. Lindhout and colleagues (2003) found that target children with anxiety disorders reported significantly more unjust or unfair parental differential treatment in favour of their sibling than control children. The effect size was moderate to large (d=0.67, 95% CI [0.07, 1.22]). However, the effect size interval was extremely wide spanning from minimal to large.

Two studies obtained data from siblings and both were of lower quality having extremely small sample sizes (n=5-15; Barrett et al., 2001; Deal & MacLean, 1995). Both studies measured parental differential treatment using the SRQ (Furnam & Buhrmester, 1985). The studies reported inconsistent findings. Barrett and colleagues (2001) found no significant difference between target siblings and control siblings' perceptions of parental differential treatment. Siblings believed that their parents favoured them over the target child. In contrast, Deal and MacLean (1995) found that

siblings of children with MHPs believed that their parents favoured the target child. Examining maternal and paternal partiality separately, they found that siblings believed their mothers did not display any favouritism but believed that their fathers favoured the target child. Thus, parental differential treatment was entirely accounted for by siblings' beliefs about their father's behaviours. Relative to control children, siblings of children with MHPs perceived a significantly greater degree of parental differential treatment and paternal differential treatment than control siblings. No significant difference was found for maternal differential treatment. Effect sizes could not be calculated because the scale is structured such that the scores are not interpreted in the conventional sense where higher integers represent more of the particular outcome. Difference from the zeropoint (i.e. 6.0 for parental differential treatment and 3.0 for maternal and paternal partiality) can be used for statistical comparisons, however effect sizes could not be calculated from this as the standard deviation data referred to mean scores not difference from the zeropoint.

Faraone and colleagues' (1996) study was the only study to obtain self-report data from multiple informants. Based on reports from siblings and parents of boys with ADHD, target siblings had significantly more parent relationship problems (e.g. noncompliance, violence towards parents) than control siblings. Effect sizes were in the moderate range (d=0.38 and 0.49). Effect size confidence intervals were wide and spanned from small to large suggesting limited precision in the effect size estimates and considerable variability in the within-study and within-group data (95% CIs [0.15, 0.61] and [0.26, 0.71], respectively).

Last, two studies reported observational data on the frequency of negative behaviours between parents and siblings. Hudson and Rapee (2002) focused on parental intrusive involvement (e.g. providing unsolicited help) in tasks completed by siblings of

children with anxiety disorders. They found that mothers of target siblings more often displayed overinvolvement behaviours in sibling activities than control mothers. The difference was statistically significant with a moderate effect size (*d*=0.64, 95% CI [0.09, 1.21]). Fathers of target siblings similarly displayed more overinvolvement than control fathers, however the difference was not statistically significant. The effect size was small (*d*=0.19, 95% CI [-0.35, 0.74]). In both cases, the effect size confidence interval was extremely wide suggesting limited precision and considerable variability in the data. Dadds and colleagues (1992), as previously described, assessed the frequency of negative parent-sibling behaviours (e.g. use of an aversive tone, behaviours causing pain or discomfort, aversive content in speech). In all three target groups (CD, depression, and CD and depression), mothers and fathers displayed more frequent aversive behaviours towards the target sibling than control families. However, only mothers of children with CD were significantly more aversive towards target siblings than control mothers. Effect sizes ranged from minimal to large (*d*=0.07-1.40). Large effect sizes were noted for mother-sibling aversive behaviours across all three comparison groups. However, confidence intervals for all effect sizes were considerably wide suggesting limited precision and considerable variability in the data (e.g. 95% CI [-0.27, 0.97] for father-sibling aversive in families of children with CD). For the statistically significant difference seen in mother-sibling aversive behaviour in families of children with CD, the effect size was large, and although the confidence interval was wide, it contained only large effect sizes (*d*=1.40, 95% CI [0.69, 2.06]).

In sum, the only high quality study that obtained data from multiple informants and recruited a sufficiently large sample size (Faraone et al., 1996) reported significantly more problems in the parent-sibling relationship. Both of the observational studies that were considered of higher quality reported at least one statistically

significant difference in their study. The findings suggest that there is a greater difference between target families and control families in terms of mother-sibling relationships, but not in father-sibling relationships. Effect sizes support this with larger effect sizes seen for former than the latter. However, the effect size confidence intervals overlapped considerably. The findings from the self-report, single informant studies, all three of which examined parental differential treatment were inconsistent. It is unclear what may account for these differences, at this point, as all of these studies were similar in quality. There is insufficient data on this topic to examine the role of other study characteristics, such as the diagnosis of the target child, which might explain the inconsistencies.

Variables Associated with Family Relationship Quality

Few studies explored moderators of family relationship quality and very few moderators were examined by more than two studies. Thus, an in-depth analysis of these findings is not warranted and they will be only briefly outlined in the section below. Demographic factors were not significantly associated with the quality of family relationships, including age and gender of the target child and sibling, number of siblings, parent income and family SES (Donenberg & Baker, 1993; Greene et al., 2001; Hudson & Rapee, 2002; Mikami & Pfiffner, 2008). An informant effect was found in several studies (Daniels & Moos, 1990; Faraone et al., 1996; Faraone, Biederman, Mennin, & Russell, 1998; Mikami & Pfiffner, 2008). However, the findings were inconsistent regarding the exact nature of the effect. For example, some studies reported significant differences across informants on ratings of the positive, but not negative aspects of sibling relationships while others reported the opposite effect. Several studies explored the mental health and psychosocial functioning of parents as moderators of family relationship quality, however very few measures of functioning

were used by more than one study (Donenberg & Baker, 1993; Hudson & Rapee, 2002; Faraone et al., 1998). These preliminary works suggest that certain types of mental health issues in parents are associated with greater impairment in sibling relationships (e.g. anxiety, parenting stress), while others are not (e.g. parental depression). Preliminary findings also showed that greater impairments in the current psychosocial functioning (including the presence of comorbid MHPs) of the target child were associated with more impaired sibling relationships (Biederman et al., 1993; Donenberg & Baker, 1993; Greene et al., 2001; Greene, Biederman, Zerwas, Monuteaux, Goring, & Faraone, 2002; Mikami & Pfiffner, 2008; Tseng et al., 2011).

Across this narrative, study quality has moderated the findings, particularly when considering statistical significance. That is, higher quality studies most often report statistically significant differences, while lower quality studies most often do not. This has been the focus of this narrative as study quality is an essential consideration for readers when interpreting study findings. Considering the patterns found in statistical significance from a different perspective, another possible explanation emerges. It may be that family relationship quality differs depending on the diagnosis of the target child. On the positive aspects of sibling relationships, significant findings were only reported for target children with ADHD, CD, delinquency, and externalising behavioural problems. Findings for target children with depression and anxiety disorders were consistently non-significant compared to controls. On the negative aspects of sibling relationships, similarly, findings for target children with anxiety disorders were consistently non-significant when compared to controls. In support of this, within-study comparisons have noted that anxiety disorders in the target child were associated with the least impairment in sibling relationships compared to other types of MHPs (Greene et al., 2001, 2002; Toro, Cervera, Osejo & Salamero, 1992). In addition,

Greene and colleagues (2002) found that there was a trend for reduced odds of sibling relationship problems for target children with ODD+anxiety disorders compared to ODD alone (Adjusted Odds Ratio=-0.11; Greene et al., 2002)

While these patterns are possible explanatory mechanisms for the variations in findings noted in this review, it should be noted that the pattern regarding target children with anxiety disorders is directly related to study quality. Both of the studies that focused on target children with anxiety disorders (Barrett et al., 2001; Lindhout et al., 2003) and reported data on sibling relationship quality were considered to be poor quality with very small sample sizes (*n*=5-24), insufficient statistical power, and obtained data from a single informant. Thus, both explanations have some support and it is unclear, at this stage, if one or both of these explanations account for the inconsistencies across studies regarding statistical significance.

Another trend emerged relating to the target child's diagnosis that should be highlighted. Families of children with CD and delinquent behaviour problems were the only target families that reported more positive sibling and parent-sibling relationships relative to controls. No other differences between target child groups on sibling relationship measures were found within studies (Daniels & Moos, 1990; Geller et al., 2000; Schachar & Wachsmuth, 1990) or across the studies in this review.

Discussion

Summary of Findings

This systematic review allows several conclusions to be made about the state of evidence on the quality of family relationships for siblings of children with MHPs compared to siblings in control families. First, based on higher quality studies that employed multiple informants and/or higher quality measures, target child-sibling relationships were significantly less positive (e.g. warmth, shared activities) and more

negative (e.g. bullying, conflict) than control sibling relationships. Effect sizes ranged from small to large with the majority of findings from higher quality studies reporting effect sizes in the small to moderate range. However, even in studies with large sample sizes, the effect size confidence intervals were considerably wide. This suggests that there is considerable within-study variation (Higgins & Green, 2011). That is, the quality of family relationships varies considerably across siblings and families. The findings on moderators of family relationship qualities suggest several variables that may explain this variation. For example, several informant effects were noted, such as reports from target children and parents tended to yield larger effect sizes than data from sibling self-reports. It may be that having a child with MHPs creates a family dynamic wherein the sibling tends to downplay the difficulties they experience so as to not burden their already overburdened parents (Abrams, 2009). However, as previously argued, there is insufficient evidence at this time to form any conclusions on what variables explain the variation seen across siblings and families.

The existing literature on the quality of target child-sibling relationships and moderators of relationship quality also do not explain why these relationships differ in quality from control families. Socio-ecological and family systems theories suggest that the target child's MHPs will have an impact on the family system and the relationships within that system (Bronfenbrenner, 1979; Hoffman, 1981). However, the question remains as to how this impacts on the family system and via what processes. Findings from a recent systematic review of the qualitative literature on the experiences of siblings of children with MHPs may provide insight into how MHPs impact on the family system and family relationships (Ma, Roberts, Winefield, & Furber, in press). For example, siblings of children with MHPs describe often having to take on a caregiving or third parent role with the target child, including monitoring, ensuring the

child completes daily tasks, preventing the child from acting inappropriately, and covering up for misbehaviour. These caregiving responsibilities were viewed as burdensome and could lead to feelings of resentment towards the target child resulting in a greater frequency of sibling conflict. Furthermore, acting as a third parent may decrease positivity in the sibling relationship (e.g. supportiveness) as the sibling may have to discipline the target child. These findings suggest several ways in which the target child's MHPs could have a direct impact on the quality of sibling relationships.

However, the target child's MHPs may also have an indirect impact. According to developmental psychopathology frameworks, because siblings and target children have similar genetic backgrounds and are likely raised in the same family environment, siblings are more likely to experience difficulties in psychosocial functioning and display psychopathology than siblings of children without MHPs (Parritz & Troy, 2011). The existing empirical literature supports this. For example, siblings of children with MHPs have significantly more social problems than control siblings (Ma et al., 2014a). Thus, target siblings may have difficulties navigating social situations due to less developed or limited social skills, including skills such as conflict resolution. These difficulties may also extend to the sibling relationship dynamic and create a dynamic with greater conflict and less support or warmth. Preliminary research also supports these pathways. For example, in families with target children with ADHD, if their sibling also meets criteria for ADHD, there were significantly more sibling relationship problems than if their sibling did not have ADHD (Faraone et al., 1996). While research and theoretical frameworks in related areas suggest a range of pathways and variables that may explain why sibling relationships in families of children with MHPs differ from those in control families, very few studies have examined these variables directly

with family relationship quality data and it is not yet known to what extent these factors explain variation in sibling relationship quality seen across siblings and families.

Second, although fewer studies examined the quality of the parent-sibling relationship in families of children with MHPs, several conclusions can be formed. First, parent-sibling relationships in target families did not significantly differ from control families on positive relationship components, such as praise, affection, and spending time with each other. However, the majority of raw differences suggest that parent-sibling relationships in target families are less positive than in control families to a small degree, with larger effect sizes reported for mother-sibling relationships than father-sibling relationships. Only two studies explored this aspect of parent-sibling relationships, thus it is difficult to draw any firm conclusions from the existing data. Second, based on higher quality studies, siblings of target children had significantly more relationship problems with their parents than control siblings. Though the findings suggest that this is only for mother-sibling relationships, not father-sibling relationships. Based on higher quality studies, effect sizes for the former were in the moderate to large range and the small to moderate range for the latter. Similarly to findings on sibling relationship quality, the effect size confidence intervals were wide, even in studies with large sample sizes. Thus, there is significant variation in parent-sibling relationship quality within-study. Similarly to sibling relationship findings, research on moderators of parent-sibling relationships is lacking and no firm conclusions can be made at this time.

These findings suggest that there are broad level family processes occurring in these families beyond dyadic relationships. That is, having a target child with MHPs in the family is related to more negative parent-sibling interactions even though the target child is not a direct participant in the parent-sibling relationship. Existing research

supports this with less family environment cohesion and expressiveness in target families (Faraone, Biederman, Mennin, Russell, & Tsuang, 1998). Findings from our qualitative synthesis also support the operation of broad level family processes (Ma et al., in press). To extend the example given above regarding sibling relationships, the increased caregiving responsibilities also impacted on the parent-sibling relationship. Siblings reported receiving little recognition or reward for undertaking these caregiving tasks. These tasks were dictated to siblings and they had little input in decision-making. This left siblings feeling powerless, helpless, and invalidated and is likely to create challenges within the parent-sibling dyad. As with sibling relationships however, there are other factors not directly related to the impact of the target child's MHP on the family system that may account for these findings. Children with MHPs are more likely to have parents with MHPs and parents that display maladaptive parenting styles, such as harsh discipline strategies (Parritz & Troy, 2011). These factors would likely directly impact the quality of the parent-sibling relationship. Findings from the included studies in this review suggest that certain types of psychopathology in parents are associated with family relationship quality while others are not. However, too few studies examined this variable to form firm conclusions. More research is needed to confirm and extend this preliminary research.

Emergent Trends

In addition to the above findings, two patterns regarding the type of MHP in the target child were noted that might explain variations in findings across studies. Relationship quality of families of children with CD and anxiety disorders seem to differ from those of children with other types of MHPs. Firstly, sibling relationship quality in families of children with anxiety disorders consistently did not significantly differ from control families. Accommodation around the target child's anxiety

symptoms may explain these findings. Research with families of children with anxiety disorders show a high prevalence of accommodation in these families where family members alter their behaviours to adapt around the target child's behaviours (e.g. modify family routines; Barrett et al., 2001). According to family systems theory, this is likely an attempt to maintain balance or homeostasis among systems (Hoffman, 1981) and is likely to reduce sibling conflict relating to the target child's behaviour. Thus, we would expect, and as confirmed by the existing literature, sibling relationship quality in families of children with anxiety disorders to be similar to that of families of children without MHPs. Furthermore, although limited research has explored the experiences of families of children with anxiety disorders, what has been done suggests that the impact is mostly practical (e.g. delays and modifications to family routines; Barrett et al., 2001) rather than impacting on emotional or interpersonal issues. This was also confirmed in our qualitative synthesis with siblings of children with MHPs (Ma et al., in press). Thus, it would be expected that target children with anxiety disorders would have less impairment in family relationships. However, as previously discussed, studies conducted with families of children with anxiety disorders were also all considered to be lower in quality. Methodological limitations, particularly small sample size and insufficient power, may also explain the non-significant findings reported for families of children with anxiety disorders.

Secondly, families of children with CD or delinquent behaviour were the only target families that reported more positive sibling and parent-sibling relationships relative to controls. However, they also had more negative family relationships than controls, as similarly found for other types of MHPs. Deviancy training may explain these findings (Bullock & Dishion, 2002). While sibling relationships have been linked with several types of MHPs, they have been particularly emphasised in the development

and maintenance of conduct problems and delinquency (Feinberg et al., 2012; Sakai et al., 2010; Wasserman, Miller, Pinner, & Jaramillo, 1996). Because research has shown that siblings of children with CD or delinquent behaviour problems often exhibit similar behaviour problems as the target child, researchers have argued that families of children with CD or delinquent behaviour problems have an underlying dynamic of deviancy training (Bullock & Dishion, 2002). That is, the sibling relationship acts as a training ground for shared delinquent behaviour between the target child and sibling (Bullock & Dishion, 2002). For example, the target child models delinquent behaviour, increases the sibling's exposure to delinquent peers, colludes with the sibling to undermine authorities and rules, and positively reinforces the sibling's delinquent behaviours (Bullock & Dishion, 2002; Feinberg et al., 2012). One of the key relationship dynamics that underpin deviancy training is positivity in the sibling relationship (Bullock & Dishion, 2002). Siblings and target children need to spend time together, share activities, be supportive of each other's actions, and admire one another for these training processes to operate. Researchers also argue that deviancy training operates through the negative aspects of sibling relationships. Negative sibling interactions may provide opportunities to practice antisocial behaviours such as lying, fighting, and destruction of sibling property (Slomkowski et al., 1997). Numerous research studies have supported this dynamic of deviancy training in families of children with delinquent behaviour problems (see Feinberg et al., 2012; 2013 for a summary). Thus, deviancy training likely explains the relationship dynamics found in this review, namely that sibling relationships are more positive but also more negative in families of children with CD or delinquent behaviour problems.

Methodological Limitations of Existing Research

This review also highlights several methodological issues in the existing literature. The two main issues relate to the measures used and the number of informants. First, most of the included studies employed general psychosocial measures that provided little data on relationship quality. These measures typically provided a combined positive and negative score for relationship quality. This provides little information on the exact nature of these relationships. As a result, no conclusions can be formed beyond greater impairments in relationships overall. This is in contrast to more comprehensive relationship measures that provide data on numerous relationship components, such as hostility, dominance, and rivalry.

Second, half of the included studies obtained data from a single informant. Ratings on family functioning and family relationships have been widely shown to vary across informants and patterns in the data relating to informant effects were also noted in this review (e.g. Dancyger et al., 2005; Mikami & Pfiffner, 2008). Thus, it is difficult to obtain reliable estimates of family relationship quality when only one informant is used. Further, very few studies obtained data directly from siblings. As a result, we have very little data on the quality of family relationships from the siblings' perspective. This further highlights the tendency to overlook siblings in family research. A related issue is that when multiple informants were employed, studies often combined the data into a single score but did not describe how this was done. Further, by combining the data, information was lost on differences in perceptions across family members.

Implications

The findings of this review have several important implications. First, there is a need for increased awareness by parents and clinicians of the family relationships of

children with MHPs. Given the associations between family relationship problems and child and adult psychosocial outcomes and the increased relationship problems found for siblings of children with MHPs, clinicians and parents need to be aware of any problems to allow early intervention (Fosco, Caruthers, & Dishion, 2012). Importantly, as the existing literature highlights, it is essential that clinicians discuss these issues with all family members as their perceptions of family relationships may differ.

Second, the findings of this review and existing theories suggest that clinicians and treatment services should consider the role of siblings in treatment. According to the previously discussed theories, while problems in one system may be associated with problems in another, it would also hold that improvements in one would be associated with improvements in the other. Therefore, including siblings in treatment may help to improve family relationships, which may result in enhanced wellbeing for the sibling, the target child, and the entire family system (Lewis & Karen, 1990; Barrett et al., 2001). Preliminary research has shown improvements in internalising behaviour problems, self-control, social and academic competence in children and decreases in maternal depression for consumers of a program designed to enhance sibling relationships and improve strategies for parenting sibling dyads (Feinberg, Solmeyer, Hostetler, Sakuma, Jones, & McHale, 2013).

Due to insufficient data, this review has few implications for existing theories and models. Very few studies examined pathways suggested by impact of illness frameworks, such as those highlighted in the qualitative literature (e.g. perceptions of burden relating to caregiving), and developmental psychopathology frameworks. In addition to the aforementioned theories, Brody (1998) developed a heuristic model, based on theoretical and empirical works, of the interrelationship between family experiences, parent-child interactions, and sibling relationships. Brody argues that

sibling relationship quality is determined by multiple factors, such as individual interpersonal styles, parent-child relations, and other family processes. He particularly emphasises the role of parental differential treatment, how parents manage sibling conflict, and parent-child interactions in determining sibling relationship quality. Sibling relationships, in turn, also influence these family processes. Very few studies in this review examined mediators of family relationship quality, such as those outlined in Brody's model, and those examined were limited in scope. As such, this review cannot comment on theorised pathways between family processes and family relationship quality. Theoretically-driven research is greatly needed.

Last, importantly, this review has highlighted several methodological issues in the existing literature. To address this, we suggest that authors implement the following methodological guidelines. First, data should be obtained from multiple informants, ideally from all family members. Taking into account possible practical limitations and given that the existing literature suggests that the greatest differences in reports are observed between parents and children (Mikami & Pfiffner, 2008), we recommend obtaining data from parents and at least one child, preferably the sibling. Second, we recommend that data from different informants should be segregated and compared. In areas where perceptions are important, examining reports from individual informants have more utility than a combined single score (Holmbeck et al., 2002). Third, comprehensive relationship measures should be used where multiple relationship components are measures. The SRQ (Furman & Buhrmester, 1985) is a well-established, widely used measure with good reliability and demonstrated validity (e.g. Derkman, Scholte, Van der Veld, & Engels, 2010). Given that this is the most commonly used comprehensive measure across the included studies, we recommend its

use to increase consistency and comparability across studies. Last, where possible, data on theoretically- or empirically-driven mediators should be obtained.

Limitations

There are several limitations of this review that should be considered alongside the results. First, the heterogeneity of included studies and limited number of studies in this area precluded conducting a formal meta-analysis. This limits the conclusions that can be drawn. Second, to provide structure to the results, the findings were grouped into positive and negative aspects of relationships. In doing so, an in-depth discussion of specific relationship components was not conducted. This was partly due to few studies reporting specific data. For example, only three studies examined parental differential treatment and only one study explored similarity and admiration in sibling relationships. Thus, we were unable to form any conclusions regarding specific relationship components and compare effect sizes across specific relationship components. Last, it is difficult to make general conclusions about the overall quality of sibling relationships, as there are no guidelines for what constitutes an ideal relationship. While it seems logical that less conflict is more desirable than higher levels of conflict, authors have argued that low levels of conflict may not necessarily be more desirable as conflict within relationships provides an opportunity for skill development, such as problem solving and conflict resolution (Kramer, 2010).

Future Directions

The key recommendation for future research is to conduct robust, comprehensive research with siblings of children with MHPs, as previously discussed. Research with families of children with CD and anxiety disorders and how family dynamics may differ in these families may be of particular interest. Another area of interest would be to examine family relationship quality as a mediator of MHPs.

Siblings of children with MHPs have been shown to be at increased risk of MHPs themselves (Ma et al., 2014b). Does family relationship quality mediate this risk? Last, additional research on the quality of family relationships for siblings of children with MHPs in adulthood is needed. Given that, in many cases, childhood MHPs in the target child persist into adulthood, siblings of children with MHPs may continue to experience disruptions in family relationships in adulthood. These disruptions may also extend to relationships specific to adulthood and those outside the childhood family system, such as marital relationships and parenthood. Thus, research in this area is greatly needed given the possible long-term detrimental impact on siblings.

References

Barrett, P., Rasmussen, P., & Healy, L. (2001). The effects of childhood obsessive-compulsive disorder on sibling relationships in late childhood and early adolescents: Preliminary findings. *Australian Educational and Developmental Psychologist, 7,* 82-102.

Baxter, J., Weston, R., & Qu, L. (2011). Family structure, co-parental relationship quality, post-separation paternal involvement and children's emotional wellbeing. *Journal of Family Studies 17*, 86-109.

Biederman, J., Faraone, S.V., & Chen, W. (1993). Social Adjustment Inventory for Children and Adolescents: Concurrent validity in ADHD children. *Journal of American Academy of Child & Adolescent Psychiatry, 32,* 1059-1064. doi:10.1097/00004583-199309000-00027

Brody, G.H. (1998). Sibling relationship quality: Its causes and consequences. *Annual Review of Psychology, 49*, 1-24. doi:10.1146/annurev.psych.49.1.1

Bronfrenbrenner, U. (1979). *The ecology of human development.* Cambridge, M.A.: Harvard University Press.

Bullock, B.M., & Dishion, T.J. (2002). Sibling collusion and problem behavior in early adolescence: Toward a process model for family mutuality. *Journal of Abnormal Child Psychology, 30,* 143-153. doi:10.1023/A:1014753232153

Campbell, J.C., & Lewandowski, L.A. (1997). Mental and physical health effects of intimate partner violence on women and children. *Psychiatric Clinics of North America, 20,* 353-374. doi: 10.1016/S0193-953X(05)70317-8

Cicirelli, V.G. (1982). Sibling influence throughout the lifespan. In M.E. Lamb & B. Sutton-Smith (Eds). *Sibling relationships: Their nature and significance across the lifespan*. New York: Lawreunce Erlbaum Associates, Inc.

Cohen, J. (1988). *Statistical power analysis for the behavioral sciences* (2nd ed.). New Jersey: Erlbaum.

Cox, M. J. (2010). Family systems and sibling relationships. *Child Development Perspectives, 4*, 95-96. doi: 10.1111/j.1750-8606.2010.00124.x

Dadds, M. R., Sanders, M. R., Morrison, M., & Rebgetz, M. (1992). Childhood depression and conduct disorder: II. An analysis of family interaction patterns in the home. *Journal of Abnormal Psychology, 101*, 505-513. doi:10.1037/0021-843X.101.3.505

Daniels, D., & Moos, R. (1990). Assessing life stressors and social resources among adolescents: Applications to depressed youth. *Journal of Adolescent Research, 5*, 268-289. doi:10.1177/074355489053002

Dancyger, I., Fomari, V., Scionti, L., Wisotsky, W., & Sunday, S. (2005). Do daughters with eating disorders agree with their parents' perception of family functioning? *Comprhensive Psychiatry, 46,* 135-139. doi:10.1016/j.comppsych.2004.07.024

Deal, S. N., & MacLean, W. E., Jr. (1995). Disrupted lives: Siblings of disturbed adolescents. *American Journal of Orthopsychiatry, 65*, 274-281. doi:10.1037/h0079616

Derkman, M.M.S., Scholte, R.H.J., Van der Veld, W.M., & Engels, R.C.M.E. (2010). Factorial and construct validity of the Sibling Relationship Questionnaire. *European Journal of Psychological Assessment, 26,* 277-283. doi:10.1027/1015-5759/a000037

Donenberg, G., & Baker, B. L. (1993). The impact of young children with externalizing behaviors on their families. *Journal of Abnormal Child Psychology, 21*, 179-198. doi: 10.1007/BF00911315

Dumas, J. E. (1996). Why was this child referred? Interactional correlates of referral status in families of children with disruptive behavior problems. *Journal of Clinical Child Psychology, 25*, 106-115. doi:10.1207/s15374424jccp2501_12

Faraone, S.V., Biederman, J., Mennin, D., Gershon, J., & Tsuang, M.T. (1996). A prospective four-year follow-up study of children at risk for ADHD: Psychiatric, neuropsychological, and psychosocial outcome. *Journal of the American Academy of Child & Adolescent Psychiatry, 35,* 1449-1459. doi:10.1097/00004583-199611000-00013

Faraone, S.V., Biederman, J., Mennin, D., & Russell, R. (1998). Bipolar and antisocial disorders among relatives of ADHD children: Parsing familial subtypes of illness. *American Journal of Medical Genetics, 81,* 108-116. doi:10.1002/(SICI)1096-8628(19980207)81:1<108::AID-AJMG18>3.0.CO;2-N

Faraone, S.V., Biederman, J., Mennin, D., Russell, R., & Tsuang, M.T. (1998). Familial subtypes of attention deficit hyperactivity disorder: A 4-year follow-up study of children from antisocial-ADHD families. *Journal of Child Psychology & Psychiatry, 39,* 1045-1053. doi:10.1017/s0021963098002996

Feinberg, M.E., Solmeyer, A.R., Hostetler, M.L., Sakuma, K.L., Jones, D., & McHale, S. (2013). Siblings are special: Initial test of a new approach for preventing youth behavior problems. *Journal of Adolescent Health, 53,* 166-173. doi: 10.1016/j.jadohealth.2012.10.004.

Feinberg, M. E., Solmeyer, A. R., & McHale, S. (2012). The third rail of family systems: Sibling relationships, mental and behavioral health, and preventive intervention in childhood and adolescence. *Clinical Child and Family Psychology Review, 15*, 43-57. doi:10.1007/s10567-011-0104-5

Fosco, G. M., Caruthers, A. S., & Dishion, T. J. (2012). A six-year predictive test of adolescent

family relationship quality and effortful control pathways to emerging adult social and emotional health. *Journal of Family Psychology, 26*, 565-575. doi: 10.1037/a0028873

Furman, W. & Buhrmester, D. (1985). Children's perceptions of the qualities of the sibling relationships. *Child Development*, 56, 448-461. doi: 10.2307/1129733.

Geller, B., Bolhofner, K., Craney, J.L., Williams, M., DelBello, M.P., & Gundersen, K. (2000). Psychosocial functioning in prepubertal and early adolescent bipolar disorder phenotype. *Journal of the American Academy of Child and Adolescent Psychiatry, 39,* 1543-1548. doi:10.1097/00004583-200012000-00018

Geller, B., Zimerman, B., Williams, M., Bolholfner, K., & Craney, J.L. (2001). Adult psychosocial outcome of prepubertal major depressive disorder. *Journal of the American Academy of Child and Adolescent Psychiatry, 40,* 673-677. doi:10.1097/00004583-200106000-00012

Greene, R.W., Biederman, J., Faraone, S.V., Monuteaux, M.C., Mick, E., DuPre, E.P….Goring, J.C. (2001). Social impairment in girls with ADHD: Patterns, gender comparisons, and correlates. *Journal of American Academy of Child and Adolescent Psychiatry, 40,* 704-710. doi:10.1097/00004583-200106000-00016

Greene, R.W., Biederman, J., Zerwas, S., Monuteaux, M.C., Goring, J.C., & Faraone, S.V. (2002). Psychiatric comorbidity, family dysfunction, and social impairment in referred youth with oppositional defiant disorder. *American Journal of Psychiatry, 159,* 1214-1224. doi:10.1176/appi.ajp.159.7.1214

Gully, K. J., Dengerink, H. A., Pepping, M., & Bergstrom, D. (1981). Research note: Sibling contribution to violent behavior. *Journal of Marriage & Family, 43*, 333-337.

Higgins, J. P. T., & Green, S. (Eds.). (2011). *Cochrane handbook for systematic reviews of interventions* (Version 5.1.0): The Cochrane Collaboration.

Hoffman, L. (1981). *Foundations of family therapy: A conceptual framework for systems change.* New York: Basic Books.

Hudson, J. L., & Rapee, R. M. (2002). Parent-child interactions in clinically anxious children and their siblings. *Journal of Clinical Child and Adolescent Psychology, 31*, 548-555. doi:10.1375/bech.22.4.220

Holmbeck, G.N., Li, S.T., Schurman, J.V., Friedman, D., & Coakley, R.M. (2002). Collecting and managing multisource and multimethod data in studies of pediatric populations. *Journal of Pediatric Psychology, 27,* 5-18. doi:10.1093/jpepsy/27.1.5

John, K., Gammon, G.D., Prusoff, B.A., & Warner, V. (1987). The Social Adjustment Inventory for Children and Adolescents (SAICA): Testing of a new semistructured interview. *Journal of the American Academy of Child and Adolescent Psychiatry, 26,* 898-911. doi:10.1097/00004583-198726060-00015

Kramer, L. (2010). The essential ingredients of successful sibling relationships: An emerging framework for advancing theory and practice. *Child Development Perspectives, 4*, 80-86. doi: 10.1111/j.1750-8606.2010.00122.x

Lewis, K., & Karen, G. (1990). Siblings: A hidden resource in therapy. *Journal of Strategic and Systemic Therapies, 9*, 39-49.

Lindhout, I. E., Boer, F., Markus, M. T., Hoogendijk, T. H., Maingay, R., & Borst, S. R. (2003). Sibling relationships of anxiety disordered children-a research note. *Journal of Anxiety Disorders, 17*, 593-601. doi:10.1016/S0887-6185(02)00230-X

Lukens, E., Puig-Antich, J., Behn, J., Goetz, R., Tabrizi, M., & Davies, M. (1983). Reliability of the psychosocial schedule for school-age children. *Journal of the*

American Academy of Child Psychiatry, 22, 29-39. doi:10.1097/00004583-198301000-00005

Ma, N., Roberts, R., Winefield, H., & Furber, G. (2014a). *A dimensional approach to the mental health of siblings of children with mental health problems: A 20-year systematic review.* Manuscript submitted for publication.

Ma, N., Roberts, R., Winefield, H., & Furber, G. (2014b). The prevalence of psychopathology in siblings of children with mental health problems: A 20-year systematic review. *Child Psychiatry and Human Development.* Advance online publication. doi: 10.1007/s10578-014-0459-1

Ma, N., Roberts, R., Winefield, H., & Furber, G. (in press). Utility of qualitative metasynthesis: Advancing knowledge on the wellbeing and needs of siblings of children with mental health problems. *Qualitative Psychology.*

Mikami, A.Y., & Pfiffner, L.J. (2008). Sibling relationships among children with ADHD. *Journal of Attention Disorders, 11,* 482-492. doi:10.1177/1087054706295670

Noland, V.J., Liller, K.D., McDermott, R.J., Coulter, M.L., & Seraphine, A.E. (2004). Is adolescent sibling violence a precursor to college dating violence? *American Journal of Health Behavior, 28,* S13-S23.

Parritz, R.H., & Troy, M.F. (2011). *Disorders of childhood: Development and psychopathology* (2nd ed.). USA: Wadsworth, Cengage Learning.

Puig-Antich, J., Kaufman, J., Ryan, N. D., & Williamson, D. E. (1993). The psychosocial functioning and family environment of depressed adolescents. *Journal of the American Academy of Child and Adolescent Psychiatry, 32*, 244-253. doi:10.1097/00004583-199303000-00003

Sakai, J.T., Stallings, M.C., Crowley, T.J., Gelhorn, H.L., McQueen, M.B., & Ehringer, M.A. (2010). Test of association between GABRA2 (SNP rs279871) and adolescent conduct/alcohol use disorders utilizing a sample of clinic referred youth with serious substance and conduct problems, controls and available first degree relatives. *Drug & Alcohol Dependence, 106*, 199-203. doi:10.1016/j.drugalcdep.2009.08.015

Schachar, R. J., & Wachsmuth, R. (1990). Oppositional disorder in children: A validation study comparing conduct disorder, oppositional disorder and normal control children. *Journal of Child Psychology and Psychiatry, 31*, 1089-1102. doi:10.1111/j.1469-7610.1990.tb00848

Serot, N.M., & Teevan, R.C. (1961). Perception of the parent-child relationship and its relation to child adjustment. *Child Development, 32,* 373-378. doi:10.2307/1125951

Slomkowski, C., Wasserman, G., Schaffer, D., Rende, R., & Davies, M. (1997). A new instrument to assess sibling relationships in antisocial youth: The social interaction between siblings (SIBS) interview: A research note. *Journal of Child Psychology and Psychiatry, 38*, 253-256. doi:10.1111/j.1469-7610.1997.tb01858.x

Slowik, M., Wilson, S.W., Loh, E.C.C., & Noronha, S. (2004). Service innovations: Developing a parent/carer support group in an in-patient adolescent setting. *Psychiatric Bulletin, 28,* 177-179. doi:10.1192/pb.28.5.177

Stormont-Spurgin, M., & Zentall, S. S. (1995). Contributing factors in the manifestation of aggression in preschoolers with hyperactivity. *Journal of Child Psychology and Psychiatry, 36*, 491-509. doi:10.1111/j.1469-7610.1995.tb01305.x

Toro, J., Cervera, M., Osejo, E., & Salamero, M. (1992). Obsessive-compulsive disorder in childhood and adolescence: A clinical study. *Journal of Child Psychology & Psychiatry, 33*, 1025-1037. doi:10.1111/j.1469-7610.1992.tb00923.x

Tseng, W.L., Kawabata, Y., & Gau, S.S. (2011). Social adjustment among Taiwanese children with symptoms of ADHD, ODD, and ADHD comorbid with ODD. *Child Psychiatry and Human Development, 42*, 134-151. doi:10.1007/s10578-010-0204-3

Valentine, J.C., Pigott, T.D., & Rothstein, H.R. (2010). How many studies do you need? A primer on statistical power for meta-analysis. *Journal of Educational and Behavioral Statistics, 35,* 215-247. doi: 10.3102/1076998609346961

Wasserman, G., Miller, L.S., Pinner, E., & Jaramillo, B. (1996). Parenting predictors of early conduct problems in urban, high-risk boys. *Journal of the American Academy of Child & Adolescent Psychiatry, 35*, 1227-1236. doi:10.1097/00004583-199609000-0002

Chapter 6. Paper Four

The Utility of Qualitative Metasynthesis: Advancing Knowledge on the Wellbeing and Needs of Siblings of Children with Mental Health Problems

(ACCEPTED FOR PUBLICATION)

Nylanda Ma, University of Adelaide

Rachel Roberts, University of Adelaide

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Gareth Furber, University of South Australia

Qualitative Psychology (in press). Accepted July 17, 2014.

Statement of Authorship

Nylanda Ma (Candidate)

I was responsible for the design and implementation of the systematic review, including development of the search strategy, collection of the included studies, data extraction, data analysis, and writing the manuscript. I served as corresponding author and was responsible for manuscript submission, revisions, and responses to journal reviews.

Signed: Nylanda LT Ma _____ Date: 4/8/2014

Dr. Rachel Roberts, Prof. Helen Winefield, & Dr. Gareth Furber (Co-authors)

We provided ongoing supervision through the systematic review and there was ongoing collaboration between Nylanda Ma and ourselves in refining the direction of research and methodology. We commented on manuscript drafts, made suggestions on the presentation of the material in the manuscript, and provided guidance on journal revisions and responses. We hereby give our permission for the inclusion of this paper in Nylanda Ma's submission for the degree of Doctor of Philosophy from the University of Adelaide.

Signed: Dr. Rachel Roberts _____Date: 4/8/2014

Signed: Prof. Helen Winefield _____Date: 4/8/2014

Signed: Dr. Gareth Furber _____ Date: 4/8/2014

Ma, N., Roberts, R., Winefield, H. & Furber, G. (2014) The utility of qualitative metasynthesis: advancing knowledge on the wellbeing and needs of siblings of children with mental health problems. *Qualitative Psychology, In press*

Chapter 7. Preamble for the Primary Research Study

7.1 Overview of Chapter

This chapter aims to provide a link between the primary research study and the systematic review and to provide additional methodological details that were not included in the paper. The methodology, main findings, and contributions of this work are discussed in sufficient detail in the following papers and are therefore not repeated in this chapter. Instead, the chapter focuses on the research process and how this study leads on from the systematic review.

7.2 Rationale for Primary Research Study

The main aims of this thesis included conducting primary research to explore areas that had not been sufficiently addressed in the existing research. I wanted this research to have particular relevance to current child clinical psychology practice, clinicians, and mental health services. As such, I decided to conduct this research with the main public mental health service for children and adolescents in Australia – the Child and Adolescent Mental Health Services (CAMHS) organisation. I formed a collaborative relationship with the research team in CAMHS in South Australia, in particular with Dr. Gareth Furber. He was planning an externally funded study (Furber, Segal, Leach & Cocks, 2013), in collaboration with the University of South Australia, and was willing to simultaneously conduct research on siblings of children with MHPs.

Based on a preliminary survey of the existing literature, I noted that no studies had explored if siblings of children with MHPs were receiving any form of support or treatment. There had been multiple studies suggesting that siblings of children with MHPs are at greater risk of MHPs, yet the question remained if these siblings were

receiving appropriate treatment. It may be that the majority of the research in this field has focused on the theoretical implications of this work. Because siblings are often researched within the context of understanding the genetic and family environment contributions to psychopathology, much of the focus has been on aetiology and theory and there has been less consideration of real-world clinical implications. Do mental health services and clinicians attend to the mental health of siblings? Are they equipped to assess siblings and facilitate appropriate treatment or support? Are siblings accounting for a substantial proportion of clientele for mental health services? Is there a place for the implementation of prevention or whole-family treatment programs from a cost-effectiveness standpoint? These are all important questions with significant implications for clinical practice. For these reasons, I chose to explore the treatment utilisation of siblings of children with MHPs and this became the first research direction for the primary study: To examine the rate of MHPs in siblings of children attending a child and adolescent mental health clinic and measure the rate of treatment utilisation in these siblings. As discussed in more detail in paper 5, items designed to answer these questions were included in the parallel study referenced above, which involved conducting telephone interviews with caregivers of current clients of CAMHS.

When data collection was complete, from an initial examination of the primary research data with CAMHS, it was apparent that there was a high rate of MHPs in siblings, as identified by caregivers, and a high rate of treatment utilisation or treatment demand. This pointed to the need for research into how clinicians can identify siblings at risk for MHPs to facilitate early intervention and treatment. At this point, I had also completed the first analyses of the data from the systematic review. It was apparent from the systematic review that there were significant gaps in our understanding of predictors of the mental health of siblings, particularly in terms of clinically

representative samples. Thus, additional research on predictors of MHPs in siblings is needed.

As the primary research study was conducted simultaneously with the parallel study, I had access to data on variables that could potentially moderate the mental health of siblings. Specifically, data on the psychosocial functioning, daily functioning, and treatment length of the target child were obtained from caregivers. In addition, demographic data on the age and gender of siblings and target children were available. This provided the opportunity to explore birth order, age difference, and gender composition of the target child-sibling dyad as potential moderators. Very little existing research had explored these variables, particularly in terms of predicting the presence of MHPs in siblings of children with MHPs. Thus, I was able to conduct primary research that addressed significant gaps in the existing literature and that could provide a significant contribution to the literature.

The findings of this primary research could also have significant implications for research trends noted in the systematic review. First, I have argued previously that findings from twin studies may not generalise to non-twin siblings and the need for a synthesis of research with non-twin siblings. If birth order or age-spacing are significant predictors, this would support this argument. These factors are not present among twin siblings as they are, for all intents and purposes, the same age. Twin research then cannot be generalised to non-twin siblings as there is a key difference between the groups of significance to understanding the mental health of siblings. Furthermore, this would point to the value of researching non-twin siblings in that it can reveal predictors of mental health that cannot be researched in the twin sibling context. Second, one of the key methodological limitations of this field is in the manner in which the mental health of siblings is assessed. As noted in the papers, many studies recruited only one

sibling per family and oftentimes, recruited the sibling closest in age to the target child. If age-spacing is a significant predictor, this would have significant implications for the validity of the findings of those studies and highlight the need for future research to assess all available siblings or recruit one sibling through random selection.

Thus, the primary research study addresses key gaps and builds on the findings of the systematic review to advance our understanding of the mental health and wellbeing of siblings of children with MHPs. In addition, it feeds back into the systematic review by exploring the validity of arguments made in the systematic review (i.e. twin studies and age-spacing above). In this way, the primary research study is a well-suited companion to the systematic review and works well as the final piece of work in this thesis by combining past and current research and encouraging future research. The findings of this study are reported in two separate papers based on journal editor feedback. The following paper is a brief report describing the rate of MHPs and treatment utilisation of siblings of children with MHPs. Paper 6 reports on the relationship between the severity of the target child's MHPs, sibling age and gender, birth order, age difference, and gender composition of the target child-sibling pair, and the presence of caregiver-identified MHPs in siblings.

Chapter 8. Paper Five

Caregiver Perceptions of Mental Health Problems and Treatment Utilisation in Siblings of Children with Mental Health Problems

(Manuscript Under Review)

Nylanda Ma, University of Adelaide

Gareth Furber, University of South Australia

Rachel Roberts, University of Adelaide

Helen Winefield, University of Adelaide

Journal of Mental Health. Submitted, July 2014.

Statement of Authorship

Nylanda Ma (Candidate)

I was responsible for the design of the survey questions relating to siblings, for applying for and obtaining ethics approval from the University of Adelaide, for data analysis, and for writing the manuscript. As this project was conducted with a child and adolescent mental health service with confidentiality policies and was part of a parallel study, I was unable to complete data collection and implement the project within the CAMHS organisation. I was responsible for liaising with the research team responsible

for data collection. I served as corresponding author and was responsible for manuscript submission, revisions, and responses to journal reviews.

Signed: Nylanda LT Ma _____ Date: 4/8/2014

Dr. Gareth Furber (Co-author)

I was responsible, in collaboration with the research team at CAMHS and the University of South Australia, for the design of the parallel study, ethics applications with these institutions, implementation of the study within CAMHS, and supervising data collection and entry by independent research assistants. I provided ongoing supervision in the analysis of the data and writing of the manuscript. I commented on manuscript drafts, made suggestions on the presentation of the material in the manuscript, and provided guidance on journal revisions and responses. I give my permission for the inclusion of this paper in Nylanda Ma's submission for the degree of Doctor of Philosophy from the University of Adelaide.

Signed: Dr. Gareth Furber _____ Date: 4/8/2014

Dr. Rachel Roberts & Prof. Helen Winefield (Co-authors)

We provided ongoing supervision through the implemention of the research project and there was ongoing collaboration between Nylanda Ma and ourselves in refining the direction of research, methodology, and data analyses. We commented on manuscript drafts, made suggestions on the presentation of the material in the manuscript, and provided guidance on journal revisions and responses. We hereby give our permission for the inclusion of this paper in Nylanda Ma's submission for the degree of Doctor of Philosophy from the University of Adelaide.

Signed: Dr. Rachel Roberts _____Date: 4/8/2014

Signed: Prof. Helen Winefield _____Date: 4/8/2014

Abstract

Background: Siblings of children with mental health problems (MHPs) have been found to have higher rates of psychopathology and impaired psychosocial functioning compared to control children. It is not yet known how these siblings are managed within the clinical service context and if they are receiving appropriate treatment. Aims: The following brief report describes a pilot study at a child and adolescent mental health service (CAMHS) clinic in Australia aimed to explore this issue. Methods: Two hundred caregivers of children receiving treatment at CAMHS were interviewed about the mental health and treatment utilisation of their siblings. Results: The findings revealed a high rate of caregiver-identified MHPs in siblings (34.1%) and a high rate of treatment utilisation (85.7%). Conclusions: The findings suggest that, for the vast majority, when siblings of children with MHPs are identified by their caregivers as having MHPs, they are receiving appropriate support and treatment. Implications for mental health service costs are discussed and recommendations for future research are outlined.

Keywords: siblings; child; mental health services; treatment utilisation

Siblings of children with mental health problems (MHPs) have been shown to have higher rates of psychopathology, poorer psychosocial functioning, greater impairment in family relationships, and to experience a range of challenges associated with having a brother or sister with MHPs (Ma, Roberts, Winefield, & Furber, 2014a,b,c,in press). Several family and childhood mental health researchers and experts have argued that despite these findings, siblings tend to be overlooked in research and clinical practice (Cox, 2010; Feinberg, Solmeyer, & McHale, 2012). In the context of research, this does seem to be the case. Few studies have examined the mental health and wellbeing of siblings of children with MHPs, the majority have important methodological limitations, and significant gaps remain in the literature, particularly regarding predictors and causal mechanisms (Ma, Roberts et al., 2014a,b). However, little research has examined if siblings are also neglected within the clinical practice and service context. For example, do siblings typically receive appropriate clinical support or treatment? No known studies have explored this question.

The following brief report describes the findings from a pilot study designed to explore the treatment of siblings within the clinical service context. More specifically, how many siblings of children receiving mental health treatment are identified by caregivers as experiencing MHPs? What supports or treatment, if any, are these siblings receiving? Based on past research and opinions from researchers and experts in the field, it was expected that a greater proportion of siblings of children with MHPs would be identified as experiencing MHPs compared to the general population. It was expected that the majority of these siblings would not be receiving clinical support or treatment.

Methods

Design

Data collection for this study was conducted simultaneously with a study assessing the validity of a quality of life measure in a clinical child population (Furber, Segal, Leach, & Cocks, 2013). The methodology used is described in detail elsewhere (Furber et al., 2013; Ma, Furber, Roberts, & Winefield, 2014). Briefly, participants were parents or other caregivers of children aged 5-17 years who were registered as current clients (referred to as the 'target child') of a child and adolescent mental health service (CAMHS) clinic. Obtaining data from caregivers is common practice both in CAMHS and sibling research (Furber et al., 2013; Ma, Furber et al., 2014), where seeking self-report from children can be compromised by age and comprehension issues. Further, as caregivers are the primary gatekeepers to seeking treatment for children, their perceptions of MHPs are essential to understanding and assessing treatment seeking and utilisation (Logan & King, 2001). We, therefore, chose to use caregiver report to assess the prevalence of MHPs in siblings.

Potential participants were identified from the electronic clinical records of the CAMHS service. If participants had more than one child registered as a current client, a coin toss method was used to select the target child. Each participant provided consent and completed a telephone survey instrument. This study was approved by CAMHS (#384.11) and University ethics committees (#25739; #11/75).

Measures

A telephone survey instrument for parents or caregivers was developed. First, caregivers were asked if the target child had any siblings. If so, demographic data were obtained on the age and gender of the target child and siblings. Caregivers were then

asked if siblings had any past or current emotional and behavioural difficulties. If so, caregivers were asked if any support services (e.g. counselling or medication) had been received or if treatment was being sought for siblings.

Half way through the data collection process (*n*=115), it became apparent that the questions were not yielding the intended data. Caregivers were reporting data on siblings as one unit and detailed data were not obtained for each of the target child's siblings. The survey questions were revised for the remainder of the data collection (*n*=85). Caregivers were asked if the target child had any siblings and, if so, how many. Caregivers were then asked for detailed data on each of the siblings including demographic data, emotional and/or behavioural difficulty data, and support data.

Data Analysis

Data analysis was conducted using SPSS version 21. A chi-square test was used to compare the rate of caregiver-identified emotional and behavioural difficulties in siblings to local general population rates of MHPs in children based on caregiver report rating scales (i.e. parent version of the Child Behavior Checklist; Achenbach & Rescorla, 2001; Sawyer et al., 2000).

Results

Sample Characteristics

A total of 900 participants met the inclusion criteria. The participants were placed on a randomised list and were contacted sequentially until 200 eligible participants agreed to undertake the survey. A total of 407 caregivers were approached; 150 were not contactable, 37 declined to be interviewed, 14 were discovered not to meet criteria, and 6 interviews were not complete. The final sample of 200 caregivers interviewed included mothers (87.0%), fathers (8.5%), and other caregivers (4.5%), such as extended family. Target children (52.5% male) ranged in age from 5-17 years

with a mean age of 11.8 (*SD*=3.1) years. The majority of target children were receiving treatment from CAMHS for the first time (74.5%). The mean length of time for the current treatment period was 12.0 (*SD*=16.6) months. Based on clinically elevated caregiver-rated Strengths and Difficulties Questionnaire (SDQ; Goodman, 2007) scores, 60.0%, 51.5%, 51.0%, and 50.5% of target children had emotional, conduct, hyperactivity-inattention, or peer/social problems, respectively. A large proportion of target children had clinically elevated scores on two or more of these scales (67.0%) and on three or more of these scales (43.0%). Notably, 18.0% of target children did not have clinically elevated scores. Specific diagnostic information on the target child was not available. The majority of target children had at least one sibling (92.5%). Siblings (62.1% male) ranged in age from 1-35 years with a mean age of 13.5 years (*SD*=8.5).

Rate of Caregiver-Identified Emotional and/or Behavioural Difficulties in Siblings

Data on all available siblings aged 4 years and up (*n*=164) were obtained from 85 caregivers. Of these siblings, 34.1% (*n*=56) were identified by caregivers as having past or current emotional and/or behavioural difficulties or MHPs. For siblings aged 4-17 years (*n*=111), 38.7% were identified by caregivers as having past or current MHPs. Compared to local general population prevalence rates (parents of children aged 4-17 years, *n*=521,889; Sawyer et al., 2000), a significantly greater proportion of siblings of children with MHPs were identified by caregivers as having MHPs compared to children in the general population (38.7% v 14.1%, respectively; $\chi^2(1)=55.61$, $p<0.001$). Siblings of children with MHPs were 3.85 (95% CI [2.63, 5.64]) times more likely to be identified as having MHPs than children in the general population. Detailed data on the types of MHPs reported by caregivers were obtained for 55 siblings. Rates of emotional difficulties (e.g. depression, anxiety) and behavioural difficulties (e.g. conduct problems and anger) were similar (31% and 33%, respectively) and were the most

commonly reported difficulties by caregivers. Caregivers also described difficulties due to environmental and family factors, such as grief and marital breakdown (15%), and other types of difficulties, such as drug and alcohol problems (11%). Approximately 11% of siblings were reported to be experiencing more than one type of difficulty.

Support or Treatment Sought for Siblings

The majority of siblings (85.7%) identified by caregivers as displaying MHPs were currently receiving, on the waitlist for, or in the past had received some form of support or mental health treatment. Of target children with at least one sibling with MHPs, 80.0% had at least one sibling receiving or on the waitlist to receive treatment. The vast majority of those who were receiving or seeking support utilised counselling or psychological treatment (89.5%). Other types of support used were medication, combined medication and psychological treatment, and other allied health professionals and services, such as social workers. Of the eight siblings that had not or were not receiving any support or treatment, five were female, seven were prepubertal, seven were younger than the target child, and five displayed difficulties with anger.

Discussion

Two key findings arose from this pilot study. First, caregivers of children receiving mental health treatment identified a substantial proportion of siblings as also experiencing emotional and/or behavioural difficulties or MHPs. This proportion was significantly higher than the rate of MHPs in children in the general population. Approximately 1 in 3 (34.1%) siblings of children receiving mental health treatment were identified by caregivers as displaying emotional and behavioural difficulties. This rate is comparable to international and national findings from studies in child mental health services, which reported rates of any MHP in siblings between 26% and 42% (Barnett & Hunter, 2012; Cohen et al., 1996; James & Vereker, 1996; Ryan et al.,

1992). The odds ratio found in this pilot study is also consistent with international reports from higher quality studies that conducted extensive diagnostic assessments of siblings (OR=2.0-4.0; Ma, Roberts et al., 2014b). Thus, although this pilot study was based on caregiver report of emotional and behavioural difficulties only, the findings match those based on well established, standardised assessment measures and is consistent with findings from international studies.

Secondly, this study found a high rate of treatment utilisation by siblings identified by caregivers as experiencing MHPs. Thus, this preliminary study suggests that siblings are not being neglected in the real-world clinical service context. When caregivers identify a sibling as having MHPs, the vast majority of these siblings receive some form of treatment or support. However, this study and these findings are best viewed as a preliminary step to understanding the treatment of siblings in clinical practice. There are several gaps and issues that need further consideration.

First, this study only examined treatment utilisation in cases where siblings were identified as having emotional and/or behavioural difficulties. There is extensive literature that discusses the environmental stressors, relationship difficulties, and daily challenges that siblings of children with MHPs face (Ma, Roberts et al., in press). These siblings may not display clinically significant difficulties but are nonetheless in need of support. It is unclear if siblings of children with MHPs are routinely offered or attend support programs even when they are not identified as experiencing significant difficulties by caregivers.

Second, while the vast majority of siblings that were recognised by caregivers as experiencing significant difficulties received support or treatment, many siblings may be experiencing difficulties that caregivers are unaware of. There is extensive literature that shows that siblings of children with MHPs often downplay the difficulties they are

experiencing and that parents often minimise or overlook these difficulties (Ma, Roberts et al., in press). It is unclear if these siblings are receiving any support or are seeking support from sources that caregivers may be unaware of, such as online support groups.

Third, while it is encouraging that the vast majority of siblings are receiving treatment, there is a potential workload consequence for mental health services. Considering the above findings from another perspective, almost 1 in every 2 children (47.5%) seen by CAMHS had at least one sibling identified by caregivers as displaying MHPs. Given the high frequency of treatment seeking for siblings, MHPs in siblings place a significant strain on mental health agencies. If 1 in every 2 children seen have at least one sibling with MHPs and 4 in 5 of these have at least one sibling receiving or waiting to receive treatment, this results in approximately an additional 38 sibling clients (47.5% x 80.0%=38.0%) utilising treatment services per 100 target children. Thus, emotional and behavioural difficulties in siblings may account for a significant portion of service utilisation and result in an up to 38% increase in client load and cost for mental health services. This suggests that there might be significant benefits to implementing prevention programs for siblings once the target child presents for treatment and/or to implementing family-inclusive treatment protocols. It is unclear from this pilot study how multiple children with MHPs within a family are managed in child mental health services and the associated costs. Future research should explore common clinic practices in regards to siblings and evaluate the potential costs and effectiveness of the strategies used.

Limitations

There are several limitations of this study that should be considered alongside the results. Firstly, recruiting from real-world clinical child and adolescent services led

to several limitations including reliance on caregiver reports. However, these are balanced by the advantages of recruiting from real-world settings and this method has been found to be highly reliable when assessing 'any mental disorder' (Hardt & Franke, 2007). Further, this method gives consideration to caregivers' perceptions of siblings. Because caregivers are gatekeepers to siblings' utilisation of support and mental health services, it is useful to obtain data on their perceptions of MHPs in siblings (Logan & King, 2001). Second, rates of support service utilisation by siblings may be overestimated given the self-referred nature of the sample. Caregivers that have already accessed services for their children before may be more likely to seek support for other family members compared to community-based samples. Further, the questions around supports used were not worded in a way that would encourage caregivers to describe all supports that had been used. Caregivers were asked if any supports had been received or sought but not to list each type of support that have been received or sought. Caregivers may have only mentioned one type of support that siblings use. Thus, this study represents only a preliminary exploration of these issues and is need of replication and refinement in methodology. Last, the findings on risk in siblings of children with MHPs compared to the general population are limited by imperfect matching on key variables, including gender, across these groups.

Future Research

As previously described, there are still large gaps in sibling research. More research is needed to explore if siblings of children with MHPs who are not identified by caregivers as experiencing MHPs are receiving appropriate support or treatment. Further exploration is needed to assess common clinical practices in regards to siblings and to inform best practice guidelines for working with families with multiple children who are experiencing MHPs. Cost analyses would also be valuable to assess if the

implementation of prevention programs or family-inclusive treatment in child and adolescent mental health services would be beneficial.

References

Achenbach, T.M., & Rescorla, L.A. (2001). *Manual for the ASEBA School-Age forms & profiles.* Vermont: University of Vermont, Research Center for Children, Youth, & Families.

Barnett, R.A., & Hunter, M. (2012). Adjustment of siblings of children with mental health problems: Behaviour, self-concept, quality of life, and family functioning. *Journal of Child and Family Studies, 21,* 262-272. doi: 10.1007/s10826-011-9471-2

Cohen, N.J., Barwich, M.A., Horodezky, N., & Isaacson, L. (1996). Comorbidity of language and socio-emotional disorders: Comparisons of psychiatric outpatients and their siblings. *Journal of Clinical Child Psychology, 25,* 192-200.

Cox, M.J. (2010). Family systems and sibling relationships. *Child Development Perspectives, 4,* 95-96. doi: 10.1111/j.1750-8606.2010.00124.x

Feinberg, M.E., Solmeyer, A.R., & McHale, S. (2012). The third rail of family systems: Sibling relationships, mental and behavioral health, and preventive intervention in childhood and adolescence. *Clinical Child & Family Psychology Review, 15*, 43-57.

Furber, G., Segal, L., Leach, M., & Cocks, J. (2013). Mapping scores from the Strengths and Difficulties Questionnaire (SDQ) to preference-based utility values. *Quality of Life Research, 23,* 403-411. doi: 10.1007/s11136-013-0494-6

Goodman, R. (2007). The strengths and difficulties questionnaire: A research note. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 38*, 581-586.

Hardt, J., & Franke, P. (2007). Validity, reliability, and objectivity of the family history method in psychiatry: A meta analysis. *European Psychiatry, 22,* 49-58.

James, A.C. & Vereker, M. (1996). Family therapy for adolescents diagnosed as having borderline personality disorder. *Journal of Family Therapy, 18,* 269-283.

Logan, D.E., & King, C.A. (2001). Parental facilitation of adolescent mental health service utilization: A conceptual and empirical review. *Clinical Psychology: Science and Practice, 8,* 319-333. doi: 10.1093/clipsy.8.3.319

Ma, N., Furber, G., Roberts, R., & Winefield, H. (2014). *The role of birth order, age difference, and symptom severity in the mental health of siblings of children with mental health problems.* Manuscript submitted for publication.

Ma, N., Roberts, R., Winefield, H., & Furber, G. (2014a). *A dimensional approach to the mental health of siblings of children with mental health problems: A 20-year systematic review.* Manuscript submitted for publication.

Ma, N., Roberts, R., Winefield, H., & Furber, G. (2014b). The prevalence of psychopathology in siblings of children with mental health problems: A 20-year systematic review. *Child Psychiatry and Human Development.* Advance online publication. doi: 10.1007/s10578-014-0459-1

Ma, N., Roberts, R., Winefield, H., & Furber, G. (2014c). *The quality of family relationships for siblings of children with mental health problems: A 20-year systematic review.* Manuscript submitted for publication.

Ma, N., Roberts, R., Winefield, H., & Furber, G. (in press). The utility of qualitative metasynthesis: Advancing knowledge on the wellbeing and needs of siblings of children with mental health problems. *Qualitative Psychology.*

Ryan, N.D., Williamson, D.E., Iyengar, S., Orvaschel, J., Reich, T., Dahl, R.E., & Puig-Antich, J. (1992). A secular increase in child and adolescent onset affective disorder. *Journal of the American Academy of Child & Adolescent Psychiatry, 31,* 600-605.

Sawyer, M. G., Arney, F. M., Baghurst, P. S., Clark, J. J., Graetz, B. W., Kosky, R. J., . . . Zubrick, S. R. (2000). *The mental health of young people in Australia*. Australia: Mental Health and Special Programs Branch, Commonwealth Department of Health and Aged Care.

Chapter 9. Paper Six

The Role of Birth Order, Age Difference, and Symptom Severity in the Mental Health of Siblings of Children with Mental Health Problems

(Manuscript Under Review)

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Child and Adolescent Mental Health. Submitted, May 2014.

Statement of Authorship

Nylanda Ma (Candidate)

I was responsible for the design of the survey questions relating to siblings, for applying for and obtaining ethics approval from the University of Adelaide, for data analysis, and for writing the manuscript. As this project was conducted with a child and adolescent mental health service with confidentiality policies and was part of a parallel study, I was unable to complete data collection and implement the project within the CAMHS organisation. I was responsible for liaising with the research team responsible

for data collection. I served as corresponding author and was responsible for manuscript submission, revisions, and responses to journal reviews.

Signed: Nylanda LT Ma _____ Date: 4/8/2014

Dr. Gareth Furber (Co-author)

I was responsible, in collaboration with the research team at CAMHS and the University of South Australia, for the design of the parallel study, ethics applications with these institutions, implementation of the study within CAMHS, and supervising data collection and entry by independent research assistants. I provided ongoing supervision in the analysis of the data and writing of the manuscript. I commented on manuscript drafts, made suggestions on the presentation of the material in the manuscript, and provided guidance on journal revisions and responses. I give my permission for the inclusion of this paper in Nylanda Ma's submission for the degree of Doctor of Philosophy from the University of Adelaide.

Signed: Dr. Gareth Furber _____ Date: 4/8/2014

Dr. Rachel Roberts & Prof. Helen Winefield (Co-authors)

We provided ongoing supervision through the implemention of the research project and there was ongoing collaboration between Nylanda Ma and ourselves in refining the direction of research, methodology, and data analyses. We commented on manuscript drafts, made suggestions on the presentation of the material in the manuscript, and provided guidance on journal revisions and responses. We hereby give our permission for the inclusion of this paper in Nylanda Ma's submission for the degree of Doctor of Philosophy from the University of Adelaide.

Signed: Dr. Rachel Roberts _____Date: 4/8/2014

Signed: Prof. Helen Winefield _____Date: 4/8/2014

Abstract

BACKGROUND: Siblings of children with mental health problems (MHPs) have been shown to be at significantly greater risk of psychopathology, impaired psychosocial functioning, and impaired family relationships compared to control children. Very little research has examined risk factors. This study aimed to explore the relationship between the severity of the presenting or target child's symptoms, sibling age and gender, birth order, and age difference, and mental health difficulties in siblings of children with MHPs. METHOD: Two hundred caregivers of target children receiving mental health treatment reported on the rates of MHPs in their siblings, the severity of the target child's symptoms, and the gender and ages of the target child and their siblings. RESULTS: Siblings identified by caregivers as having MHPs were younger and closer in age to the target child than those without MHPs. Severity of symptoms of the target child and gender did not significantly differentiate siblings with MHPs and those without MHPs. CONCLUSIONS: Clinicians should pay particular attention to and assess the mental health of younger siblings and those closer in age to the child presenting for treatment. Recommendations for future research are discussed.
Keywords: child; adolescent; siblings; child mental disorders; mental health services

Key practitioner message:

- Siblings of children with mental health problems (MHPs) have been found to be at increased risk of mental health problems themselves.
- However, little research has examined potential risk factors for mental health difficulties in siblings.
- Based on caregiver interviews, siblings identified as having MHPs were more likely to be younger than the target child and closer in age to the target child

compared to those not identified as having MHPs. Sibling gender and the gender composition of the target child-sibling pair was not predictive. Similarly, severity of the target child's difficulties (including psychosocial functioning and daily functioning) was not predictive.

- Clinicians should pay particular attention to and assess the mental health of the younger siblings of children presenting for treatment and those closer in age to the child.
- Clinicians should assess the mental health of siblings regardless of the severity of the target child's symptoms.

In a recent pilot study, the rate of caregiver-identified mental health problems (MHPs) in siblings of a random sample of clients attending a child and adolescent mental health service (CAMHS) was assessed. Caregivers identified a significantly greater proportion of siblings as having MHPs than found in children in the general population (OR=3.85; Ma, Furber, Roberts, Winefield, 2014) with almost 1 in 3 siblings identified as having MHPs. Similar rates of MHPs in siblings have been reported across the existing literature (see review by Ma, Roberts, Winefield, & Furber, 2014b). The vast majority of these siblings had received, were receiving, or were on the waitlist for treatment services. Thus, siblings of children with MHPs represent a high-risk group with a high demand for treatment services.

Very few studies have explored predictors or moderators of sibling mental health (Ma, Roberts et al., 2014a,b). The majority of these studies have focused on target children with isolated disorders, which is not the typical presentation in child mental health services. Most children present for treatment for a range of symptoms or difficulties, present with significant comorbidities, and/or present with significant environmental stressors (e.g. family stress) and their difficulties may not neatly fit within a single diagnostic category (Bird, Gould, & Staghezza, 1993; Verhulst & van der Ende, 1997; Yeh & Weisz, 2001). To guide clinical practice and the development of theoretical frameworks relevant to clinical treatment and practice, sibling research based on representative clinical samples is needed.

This study aimed to address this gap in the literature by exploring potential predictors of MHPs in siblings of a random sample of clients receiving mental health treatment (‘target children’). Data from the pilot study, described above, were reanalysed with a focus on variables that may predict or moderate the presence of MHPs in siblings of children with MHPs drawing on data obtained from a study

conducted parallel to the pilot study (Furber, Segal, Leach, & Cocks, 2013). The variables examined were the severity of the target child's MHPs (e.g. psychosocial functioning, daily functioning, and length of treatment), sibling age and gender, and the age and gender composition of the target child-sibling dyad. This study is the first known research to explore the role of these variables, particularly age and gender composition, in a representative clinical sample in predicting the presence of MHPs in siblings. The following report describes the findings of this exploratory study.

Methods

The following section describes the methods used in the pilot study and parallel study from which the data presented in this paper was obtained.

Design

Data collection for this study was conducted simultaneously with a study assessing the validity of a quality of life measure with the CAMHS population (Furber et al., 2013). Participants were parents or other caregivers of children aged 5-17 years who were registered as current CAMHS clients. CAMHS is one of the largest government-funded mental health services in Australia and one of the main referral options used by parents, schools, and general practitioners. CAMHS is a multi-disciplinary organisation that includes psychiatrists, psychologists, nurses, and social workers. Obtaining data from caregivers is common practice both in CAMHS and sibling research (Furber et al., 2013; Ma, Roberts et al., 2014a), where seeking self-report from children can be compromised by age and comprehension issues. Further, as caregivers are the primary gatekeepers to seeking treatment for children, their perceptions of MHPs are essential to understanding and assessing treatment demand (Logan & King, 2001). Thus, we chose to obtain data from caregivers for this study.

Current client status was defined as having an open episode of care and a recorded contact within the last six weeks. In this study, the child currently receiving treatment is referred to as the 'target child' and their sibling(s) as the 'target sibling'. Caregivers without a recorded telephone number, who had specific 'no contact' instructions in the electronic clinical record, were foster carers, or whose child was the subject of current guardianship or family court orders, were excluded.

Potential participants were identified from the electronic clinical records of the CAMHS service. If participants had more than one child registered as a current client, a coin toss method was used to select the target child. Each participant provided consented and completed a telephone survey instrument. This study was approved by CAMHS (#384.11) and University ethics committees (#25739; #11/75).

Measures

A telephone survey instrument was developed. First, caregivers were asked if the target child (i.e. CAMHS client) had any siblings. If so, demographic data were obtained on the age and gender of the target child and siblings and data on the number of treatment episodes the target child had experienced. Caregivers were then asked if siblings had any past or current emotional and behavioural difficulties. If so, caregivers were asked if any support services (e.g. counselling or medication) had been received or if treatment was being sought for siblings.

Half way through the data collection process (n=115), it became apparent that the questions were not yielding the intended data. Caregivers were reporting data on siblings as one unit and detailed data were not obtained for each of the target child's siblings. The survey questions were revised for the remainder of the data collection (n=85). Caregivers were asked if the target child had any siblings and, if so, how many.

Caregivers were then asked for detailed data on each of the siblings including demographic data, emotional and/or behavioural difficulty data, and support data.

Second, psychosocial data on the target child were obtained using the caregiver-administered Strengths and Difficulties Questionnaire (SDQ; Goodman, 2007). The SDQ (Goodman, 2007) consists of 25 items, each describing a psychosocial attribute, which the caregiver indicates as being 'very true', 'somewhat true', or 'not true' of the target child over the last six months. Total scores and subscale scores (emotional, conduct, hyper, peer problems, and prosocial) were calculated with higher scores indicating poorer functioning for the problem scales and better functioning for the prosocial subscale. The SDQ has been psychometrically evaluated and has demonstrated reliability, sensitivity to detecting psychiatric diagnoses, and sensitivity to change (Goodman, 2001; Goodman & Goodman, 2009; Hawes & Dadds, 2004). The use of this measure is mandated for use in Australian CAMHS clinics (Furber et al., 2013).

Third, daily functioning data were obtained using the Child Health Utility-9D scale. The CHU-9D (Stevens, 2009; 2011) is a 9-item preference-based multi-attribute utility instrument designed for use in children aged 7-11 years. The measure focuses on the impact of health issues on quality of life and daily functioning. The items assess the child's functioning 'today' across several domains using a five-response format with the higher order responses indicating that the target child had more difficulties functioning in that domain. Data were obtained on the target child's ability to do schoolwork, complete daily routine tasks (e.g. eating, getting dressed), and on any sleeping difficulties. For the purpose of analysis, target children who were rated as having 'many problems' or 'can't do the task' by caregivers were defined as experiencing difficulties in functioning. Those that were rated as having 'no', 'few', or

'some' problems with the task were defined as not experiencing significant functional difficulties. The CHU-9D has been found to be a valid and practical measure for use with clinical child populations and has been psychometrically evaluated with Australian populations (Ratcliffe, Stevens, Flynn, Brazier, & Sawyer, 2012a,b; Stevens, 2011). In addition to the CHU-9D, caregivers were asked to indicate if the target child had missed school due to difficulties associated with their MHPs (e.g. truancy, suspension due to behavioural problems). This adds an additional measure of daily functioning in the school setting.

Last, length of treatment was measured in two ways. Caregivers reported on the length of time for the current treatment episode. Caregivers also reported on any past contact or treatment episodes with CAMHS.

Data Analysis

Data analysis was conducted using SPSS version 21. Data were screened and cleaned. For analysis purposes, a 'no problem' approach for the target child's psychosocial data was taken where missing values (0.4% of all data items) were replaced with the equivalent value for no problem. Independent *t*-tests and chi-square tests were used to examine the severity of the target child's MHPs, sibling age and gender, birth order, and age difference as predictors or covariates. Birth order was calculated by comparing the sibling's age to the target child's age and siblings were classified as either 'younger' or 'older' than the target child. In cases where the target child and sibling were of the same age, these cases were excluded from birth order analyses. Age difference was calculated by subtracting the sibling's age from the target child's age in years. Directionality (i.e. positive or negative) was ignored as this concept was measured via birth order calculations. *P*-values for all statistical analyses

were adjusted for multiple comparisons using the Bonferroni correction. Cohen's *d* (Cohen, 1988) and odds ratios were calculated for effect size estimates.

Results

Sample Characteristics

A total of 900 participants met the inclusion criteria. The participants were placed on a randomised list and were contacted sequentially until 200 eligible participants agreed to undertake the survey. A total of 407 caregivers were approached; 150 were not contactable, 37 declined to be interviewed, 14 were discovered not to meet criteria, and 6 interviews were not complete. The final sample of 200 caregivers interviewed included mothers (87.0%), fathers (8.5%), and other caregivers (4.5%), such as extended family. Target children (52.5% male) ranged in age from 5-17 years with a mean age of 11.8 (*SD*=3.1) years. The majority of target children were receiving treatment from CAMHS for the first time (74.5%). The mean length of time for the current treatment period was 12.0 (*SD*=16.6) months. Based on clinically elevated caregiver-rated SDQ scores, 60.0%, 51.5%, 51.0%, and 50.5% of target children had emotional, conduct, hyperactivity-inattention, or peer/social problems, respectively. A large proportion of target children had clinically elevated scores on two or more of these scales (67.0%) and on three or more of these scales (43.0%). Notably, 18.0% of target children did not have clinically elevated scores. Specific diagnostic information on the target child was not available. The majority of target children had at least one sibling (92.5%). Siblings (62.1% male) ranged in age from 1-35 years with a mean age of 13.5 years (*SD*=8.5). Caregivers identified 56 siblings as having MHPs (34.1%) and 108 siblings were not identified as having MHPs (see Ma, Furber et al., 2014).

Age and Gender

As shown in Table 1, sibling gender and the gender composition of the target child-sibling pair were not significantly associated with MHPs in siblings. In regards to age variables, siblings with MHPs were significantly younger in age than those without MHPs with an associated moderate effect size. The effect size confidence interval spanned from small to large. Siblings with MHPs had double the odds of being younger than the target child than siblings without MHPs. After applying the Bonferroni correction (alpha value set at $p<0.01$), this finding did not reach statistical significance. Siblings with MHPs were also closer in age to the target child than siblings without MHPs with an associated small to moderate effect size. Again, the finding was no longer significant after correcting for multiple comparisons and the effect size confidence interval was wide.

Severity of Symptoms and Difficulties in Functioning in the Target Child

These findings are presented in Table 2. Target children with at least one sibling with MHPs had higher problem SDQ scores and lower prosocial scores than target children with no siblings with MHPs. The former had greater odds of having more than one treatment episode, having missed school due to MHPs, and of having schoolwork, sleep, and daily routine difficulties. However, none of these findings were statistically significant and all effect sizes and odds ratios were minimal to small. The largest effect sizes were found for SDQ conduct problems, having multiple treatment episodes, and difficulties in daily routine functioning. No significant effect was found for length of current treatment.

Table 1

Age Characteristics of Siblings with and without MHPs

	Siblings with MHPs (*n*=56)	Siblings without MHPs (*n*=108)			
	M (*SD*)	*M* (*SD*)	*t* (df)	*p*	*d*[95% CI]
Sibling age	12.45 (6.7)	15.90 (8.37)	-2.67 (162)	0.008	0.44 [0.11, 0.76]
Difference in age from target child (years)	5.11 (4.03)	6.87 (5.47)	-2.34 (143.1)	0.021	0.35 [0.02, 0.67]
	%(*n*)	%(*n*)	χ^2(df)	*p*	OR[95% CI]
Younger sibling	54.5% (30)	37.1% (39)	4.46 (1)	0.035	2.03 [1.05, 3.94]
Male sibling	58.9% (33)	62.0% (67)	0.15 (1)	0.699	0.88 [0.45, 1.70]
Gender composition					
All male (M)	32.1% (18)	38.9% (42)	0.74 (2)	0.786	MvF=0.77 [0.30, 1.99]
All female (F)	17.86% (10)	16.7% (18)			MvMx=0.73 [0.36, 1.51]
Mixed (Mx)	50.0% (28)	44.4% (48)			FvMx=0.95 [0.39, 2.35]

Table 2

Severity of Symptoms and Difficulties in Functioning in Target Children with and without Siblings with Mental Health Problems

	Target child with sibling with MHPs (*n*=85)	Target child with no siblings with MHPs (*n*=94)			
	M (*SD*)	*M* (*SD*)	*t* (df)	*p*	*d* [95% CI]
Length of time with CAMHS (months)	11.04 (17.00)	11.20 (15.39)	-0.07 (176)	0.945	0.01[-0.28, 0.30]
SDQ emotion subscale	5.35 (2.41)	5.27 (2.64)	0.230 (177)	0.819	0.03[-0.26, 0.32]
SDQ conduct subscale	4.18 (2.84)	3.69 (2.83)	1.14 (177)	0.254	0.19[-0.12, 0.47]
SDQ hyper subscale	6.40 (2.76)	6.23 (2.89)	0.392 (177)	0.695	0.06 [-0.23, 0.35]
SDQ peer subscale	3.73 (2.55)	3.69 (2.50)	0.100 (177)	0.920	0.02 [-0.28, 0.31]
SDQ total score	19.73 (7.98)	18.94 (8.02)	0.663 (177)	0.509	0.10 [-0.20, 0.39]
SDQ prosocial subscale	6.80 (2.20)	6.89 (2.35)	-0.275 (177)	-0.784	0.04 [-0.25, 0.33]

Table 2 (Cont.)

Severity of Symptoms and Difficulties in Functioning in Target Children with and without Siblings with Mental Health Problems

	Target child with sibling with MHPs (*n*=85)	Target child with no siblings with MHPs (*n*=94)			
	% (*n*)	% (*n*)	χ^2 (df)	*p*	OR [95% CI]
More than one treatment episode	29.4% (25)	21.3% (20)	1.57 (1)	0.210	1.54 [0.78, 3.04]
Missed school due to MHPs	69.4% (59)	60.2% (56)	1.64 (1)	0.200	1.50 [0.81, 2.79]
Schoolwork difficulties	28.8% (21)	25.0% (19)	0.27 (1)	0.604	1.21 [0.59, 2.50]
Sleep difficulties	12.3% (9)	11.3% (9)	0.04 (1)	0.836	1.11 [0.42, 2.97]
Daily routine difficulties	8.4% (7)	6.2% (5)	0.31 (1)	0.578	1.40 [0.43, 4.61]

Discussion

Summary of Findings

This study aimed to explore the role of several variables as potential predictors of sibling MHPs, as identified by caregivers. First, it was found that the gender of the sibling and the gender composition of the target child-sibling pair were not significantly associated with MHPs in siblings. The findings in the existing literature have been varied in this regard. Some studies have found significant gender effects for specific types of MHPs and others have found no gender effect (Ma, Roberts et al., 2014a). These have typically matched the general population patterns regarding gender differences in MHPs. For example, attention deficit hyperactivity disorder (ADHD) is more prevalent in males than females while depression is more prevalent in females than males (Merikangas et al., 2010). It may be that because this study examined the risk of any MHP in siblings, including internalising and externalising disorders, a specific gender effect was not found.

Second, age variables were significantly or marginally significantly associated with MHPs in siblings. Siblings with MHPs were significantly younger in age than siblings without MHPs. This is consistent with general population patterns (Sawyer et al., 2000). Siblings with MHPs had greater odds of being younger than the target child. Similar birth order findings have been reported in general population studies where younger siblings had more difficulties in psychosocial functioning than older siblings (Fagan & Najman, 2003). This finding is also consistent with normative sibling theories, such as birth order theory, that hold that birth order in families can significantly impact on the mental health and psychological outcomes of children (Miller, Anderson, & Keala, 2004).

While these two findings are consistent with general population trends, they are also consistent with developmental psychopathology frameworks and impact of illness frameworks specifically relating to families of children with MHPs. Developmental psychopathology frameworks focus on risk and protective factors (e.g. genetic, shared family environment, and non-shared factors) that influence the development of psychopathology in children (Parritz & Troy, 2011). Impact of illness frameworks, such as the disability-stress-coping model, hold that illness impacts the mental health and wellbeing of the 'affected' individual and this relationship is moderated by several factors, including those related to the severity of the illness (Wallander & Varni, 1992). Such models have been extended to include the impact on family members, including siblings (e.g. Taylor, Fuggle, & Charman, 2001).

One of the most prominent theories in sibling research, sibling deviance training, incorporates both developmental psychopathology factors, such as deviant peer associations, and impact of illness factors in that having a brother or sister with MHPs impacts on the mental health and wellbeing of siblings. The theory suggests that younger siblings of children with MHPs are more likely to develop MHPs themselves due to the older target child modelling deviant behaviour, reinforcing deviant behaviours and attitudes, increasing exposure to deviant activities and deviant peer associations, and colluding with the sibling to violate rules and authority figures (Bullock & Dishion, 2002; Feinberg, Solmeyer, & McHale, 2012). Based on this theory, we would expect that younger siblings would be at greater risk of MHPs than older siblings, as was found in this study. Sibling deviance training theory also suggests that siblings that are closer in age may be more susceptible to MHPs as they spend more time with the target child, have closer sibling relationships, and have more commonalities (e.g. similar peer groups), which may increase the likelihood of the

above pathways and activities (Criss & Shaw, 2005). As found in this exploratory study, siblings with MHPs were closer in age to the target child than those without MHPs. Thus far, sibling deviance training has been applied to only cases of externalising behaviour problems and substance use disorders (Feinberg et al., 2012). However, the findings from this study suggest that similar pathways may also be involved in siblings of children with a range of MHPs.

Last, the severity of the target child's MHPs, as assessed by functional measures and psychosocial measures, was not significantly associated with the risk of having siblings with MHPs. The findings from past research have been inconsistent and varied. Some studies have found severity predicts particular types of MHPs in siblings but not others (Ma, Roberts et al., 2014a). Other studies report no significant role for the severity of the target child's MHPs (Barnett & Hunter, 2012). It is unclear what may account for divergent findings.

It may be that it is related to the specific diagnosis in the target child and in their sibling. Some developmental psychopathology theorists and researchers argue that particular types of MHPs have distinct aetiological mechanisms, such as particular genetic markers or neurobiological markers (Chen et al., 2008). To test these hypotheses, they recruit target children with a particular diagnosis, such as ADHD, and divide these children into groups based on the severity of a particular symptom or set of symptoms, such as conduct problems or emotional lability (Sobanski et al., 2010). Their siblings are then assessed for the same diagnosis and set of symptoms. If siblings of children with ADHD and conduct problems have significantly greater risk of conduct problems compared to siblings of children with ADHD only, this would suggest that ADHD and conduct problems may be a distinct subtype with distinct aetiologies, such as particular genetic markers or family environment variables (Christiansen et al.,

2008). If this is the case, and there is evidence in support of this, then we would not expect to find a link between the severity of the target child's symptoms and MHPs in siblings when examining across a broad range of MHPs. That is, having a brother or sister with MHPs is linked to a disorder-specific risk for siblings and we would not expect to find a relationship between the target child MHPs and MHPs in siblings when combining target children with a range of MHPs and assessing siblings for 'any MHPs'. This is confirmed by the findings in this study.

Impact of illness frameworks offer an alternative explanation for the link, or lack thereof, between the severity of the target child's symptoms and MHPs in siblings. Based on the disability-stress-coping model, if the individual has more severe difficulties, this would increase the negative impact on the lives of siblings and likely result in poorer mental health and wellbeing (Wallander & Varni, 1992). For example, caregiver burden has been found to increase when the 'affected' individual has more severe behavioural problems (Pinquart & Sörensen, 2003). Although the findings in this study were not significant, they were in the direction predicted by these theories. That is, target children with siblings with MHPs had more severe psychosocial difficulties compared to those without siblings with MHPs. Furthermore, impact of illness theories include a discussion of the role of the 'affected' individuals' functional status. Greater functional limitations in the individual are hypothesised to lead to more psychosocial stressors and poorer mental health and wellbeing in family members (Wallander & Varni, 1992). In this study, functional status variables, such as missed school and difficulties with daily routines, were also linked to MHPs in siblings in the direction predicted by impact of illness frameworks.

Thus, the link between the severity of the target child's MHP and MHPs in siblings is an exceedingly complex one. As yet, the existing literature is unable to shed

light on this issue in sufficient detail. As highlighted in the preceding discussion, the findings in this study are consistent with multiple theorised aetiological pathways. Thus, it is difficult to unravel the causal issues underlying the link, or lack thereof, between the target child's MHPs and the mental health of siblings.

Theoretical and clinical implications

The findings of this study have several important implications for theoretical frameworks and clinical practice. In regards to clinical practice, this is the first known study that has examined the role of these variables in predicting MHPs in siblings. Past research has focused on examining predictors of the mental health of siblings as a continuum (e.g. Barnett & Hunter, 2012; Fagan & Najman, 2003) and has not been able to identify risk factors for siblings that are most likely to display clinically significant MHPs and require clinical attention and treatment. The findings suggest that clinicians should pay particular attention to younger aged siblings, siblings who are younger than the target child, and those closer in the age to the target child. The findings also point to the need for clinicians to assess the mental health of siblings regardless of the severity of the target child's MHPs.

In regards to theoretical frameworks, we have argued elsewhere that there has been little integration of different theories when interpreting the findings from sibling research (Ma, Roberts, Winefield, & Furber, in press). For example, research on psychopathology in siblings of children with MHPs has tended to focus on developmental psychopathology frameworks. They have not typically considered their findings from the perspectives of other theories that are relevant to the mental health of siblings, particularly impact of illness theories. This study has examined a broad range of variables that include factors highlighted across both theories. For example, birth order is consistent with normative sibling theories and sibling deviance training related

to developmental psychopathology and impact of illness frameworks. We have attempted to consider the findings of this study from the perspectives of multiple theories to provide a more comprehensive consideration of the implications of these findings. In doing so, we have highlighted the relevancy of multiple theories in sibling research and the importance for researchers to consider their findings within the context of a more integrative theoretical perspective.

However, the challenge with providing a more inclusive and integrated discussion is that it is difficult to conclusively support one pathway over another. The majority of factors that influence the mental health of siblings are implicated in multiple frameworks using different aetiological pathways. Future research designed to test the relative contributions of the suggested pathways is needed. Such research may illuminate which pathways significantly contribute to the mental health of siblings and highlight targets for prevention and treatment programs.

Limitations

There is an important methodological limitation that should be considered when interpreting the findings of this study. Siblings with MHPs and those without MHPs were based on caregiver perceptions only. Several issues arise out of this methodology. Firstly, reliance on caregiver reports of MHPs can lead to less accurate estimations of the prevalence of mental health disorders in comparison to the use of standardised measures, such as behaviour rating scales or diagnostic interviews (Aboraya, Rankin, France, El-Missiry, & John, 2006). However, this method has been found to be highly reliable when assessing ‘any mental disorder’ (Hardt & Franke, 2007), as was the case in this study. Second, reliance on caregiver reports may overlook siblings that are experiencing significant difficulties that caregivers are unaware of. Thus, it is unclear if relative age data can be useful in identifying these siblings. Third, the significance of

relative age data may not be related to true differences in these variables between siblings with MHPs and those without MHPs. It may be an artefact of caregiver reports. It may be that caregivers pay particular attention to younger siblings and those closer in age to the target child. For example, they may be concerned that the younger sibling may begin to mimic the behaviours of their older sibling and are therefore more sensitive and attuned to the behaviours of younger siblings. It may also be that older siblings have moved out of home and are no longer easily monitored by caregivers. Given these limitations, this study is best thought of as a preliminary exploration of the role of these factors. Future research should re-examine these variables using more standardised and objective methods of determining the presence of MHPs in siblings.

Future research

As previously described, additional research on relative age variables is needed. It would be beneficial to test the predictive value of these variables. This study has only examined if birth order and age difference can differentiate siblings with and without MHPs. It would be useful to conduct longitudinal studies to explore if these factors can predict the development of MHPs in siblings and the need for future treatment. If so, preventative programs or family-inclusive treatment protocols could be implemented for these siblings when the target child first presents for treatment. Future research should also explore what underlying mechanisms that could explain these findings. For example, sibling deviance training should be explored in a sample of siblings of children with internalising disorders to extend the findings on siblings of children with externalising behaviour problems and substance use disorders. Such research could provide valuable insight into the development of MHPs in children and provide guidance in providing effective treatment and support for siblings of children with MHPs.

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References

Aboraya, A., Rankin, E., France, C., El-Missiry, A., & John, C. (2006). The reliability of psychiatric diagnosis revisited: The clinician's guide to improve the reliability of psychiatric diagnosis. *Psychiatric Edgmont, 3,* 41-50.

Barnett, R.A., & Hunter, M. (2012). Adjustment of siblings of children with mental health problems: Behaviour, self-concept, quality of life, and family functioning. *Journal of Child and Family Studies, 21,* 262-272. doi: 10.1007/s10826-011-9471-2

Bird, H.R., Gould, M.S., & Staghezza, B.M. (1993). Patterns of diagnostic comorbidity in a community sample of children aged 9 through 17 years. *Journal of the American Academy of Child & Adolescent Psychiatry, 32,* 361-368. doi: 10.1097/00004583-199303000-00018

Bullock, B.M., & Dishion, T.J. (2002). Sibling collusion and problem behavior in early adolescence: Toward a process model for family mutuality. *Journal of Abnormal Child Psychology, 30,* 143-153.

Chen, W., Zhou, K., Sham, P., Franke, B., Kuntsi, J., Campbell, D…Asherson, P. (2008). DSM-IV combined type ADHD shows familial association with sibling trait scores: A sampling strategy for QTL linkage. *American Journal of Medical Genetics(B), 147,* 1450-1460. doi:10.1002/ajmg.b.30672

Christiansen, J., Chen, W., Oades, R.D., Asherson, P., Taylor, E.A., Lasky-Su, J.,…Faraone, S.V. (2008). Co-transmission of conduct problems with attention-deficit/hyperactivity disorder: Familial evidence for a distinct disorder. *Journal of Neural Transmission, 115,* 163-175. doi:10.1007/s00702-007-0837-y

Cohen, J. (1988). *Statistical power analysis for the behavioral sciences* (2nd ed.). New Jersey: Erlbaum.

Criss, M.M. & Shaw, D.S. (2005). Sibling relationships as contexts for delinquency training in low-income families. *Journal of Family Psychology, 19,* 592-600.

Fagan, A.A., & Najman, J.M. (2003). Association between early childhood aggression and internalizing behavior for sibling pairs. *Journal of the American Academy of Child & Adolescent Psychiatric, 42,* 1093-1100. doi:10.1097/01.CHI.0000070240.24125.EA

Feinberg, M.E., Solmeyer, A.R., & McHale, S. (2012). The third rail of family systems: Sibling relationships, mental and behavioral health, and preventive intervention in childhood and adolescence. *Clinical Child & Family Psychology Review, 15*, 43-57.

Furber, G., Segal, L., Leach, M., & Cocks, J. (2013). Mapping scores from the Strengths and Difficulties Questionnaire (SDQ) to preference-based utility values. *Quality of Life Research, 23,* 403-411. doi: 10.1007/s11136-013-0494-6

Goodman, R. (2001). Psychometric properties of the strengths and difficulties questionnaire. *Journal of the American Academy of Child & Adolescent Psychiatry, 40*, 1337-1345.

Goodman, R. (2007). The strengths and difficulties questionnaire: A research note. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 38*, 581-586.

Goodman, A., & Goodman, R. (2009). Strengths and difficulties questionnaire as a dimensional measure of child mental health. *Journal of the American Academy of Child & Adolescent Psychiatry, 48*, 400-403.

Hardt, J., & Franke, P. (2007). Validity, reliability, and objectivity of the family history method in psychiatry: A meta analysis. *European Psychiatry, 22,* 49-58.

Hawes, D.J., & Dadds, M.R. (2004). Australian data and psychometric properties of the Strengths and Difficulties Questionnaire. *Australian and New Zealand Journal of Psychiatry*, *38*, 644-651.

Logan, D.E., & King, C.A. (2001). Parental facilitation of adolescent mental health service utilization: A conceptual and empirical review. *Clinical Psychology: Science and Practice, 8,* 319-333. doi: 10.1093/clipsy.8.3.319

Ma, N., Furber, G., Roberts, R., & Winefield, H. (2014). *Caregiver perceptions of mental health problems in siblings of children with mental health problems*. Manuscript submitted for publication.

Ma, N., Roberts, R., Winefield, H., & Furber, G. (2014a). *A dimensional approach to the mental health of siblings of children with mental health problems: A 20-year systematic review.* Manuscript submitted for publication.

Ma, N., Roberts, R., Winefield, H., & Furber, G. (2014b). The prevalence of psychopathology in siblings of children with mental health problems: A 20-year systematic review. *Child Psychiatry and Human Development.* Advance online publication. doi: 10.1007/s10578-014-0459-1

Ma, N., Roberts, R., Winefield, H., & Furber, G. (in press). The utility of qualitative metasynthesis: Advancing knowledge on the wellbeing and needs of siblings of children with mental health problems. *Qualitative Psychology.*

Merikangas, K.R., He, J.P., Burstein, M., Swanson, S.A., Avenevoli, S., Cui, L.,…Swendsen, J. (2010). Lifetime prevalence of mental disorders in U.S. adolescents: Results from the National Comorbidity Survey Replication-Adolescent Supplement (NCS-A). *Journal of the American Academy of Child and Adolescent Psychiatry, 49,* 980-989. doi:10.1016/jaac.2010.05.017

Miller, R.B., Anderson, S., Keals, D.K. (2004). Is Bowen theory valid? A review of basic research. *Journal of Marital and Family Therapy, 30,* 453-466. doi: 10.1111/j.1752-0606.2004.tb01255.x

Parritz, R.H., & Troy, M.F. (2011). *Disorders of childhood: Development and psychopathology* (2nd ed.). USA: Wadsworth, Cengage Learning.

Pinquart, M. & Sörensen, S. (2003). Associations of stressors and uplifts of caregiving with caregiver burden and depressive mood: A meta-analysis. *Journal of Gerontology Series B, Psychological Sciences and Social Sciences, 58,* 112-128. doi:10.1093/geronb/58.2.P112

Ratcliffe, J., Stevens, K., Flynn, T., Brazier, J., & Sawyer, M. (2012a). An assessment of the construct validity of the CHU-9D in the Australian adolescent general population. *Quality of Life Research, 21*, 717-725. doi: 10.1007/s11136-011-9971-y

Ratcliffe, J., Stevens, K., Flynn, T., Brazier, J., & Sawyer, M. (2012b). Whose values in health? An emprivial comparison in the application of adolescent and adult values for the CHU-9D and AQOL-6D in the Australian adolescent general population. *Value in Health, 15,* 730-736. doi: 10.1016/j.jval.2012.04.005

Sawyer, M.G., Arney, F.M., Baghurst, P.S., Clark, J.J., Graetz, B.W., Kosky, R.J.,…Zubrick, S.R. *The mental health of young people in Australia.* Australia: Mental Health and Special Programs Branch, Commonwealth Department of Health and Aged Care.

Sobanski, E., Banaschewski, T., Asherson, P., Buitelaar, J., Chen, W., Franke, B., . . . Faraone, S. V. (2010). Emotional lability in children and adolescents with attention deficit/hyperactivity disorder (ADHD): Clinical correlates and familial

prevalence. *Journal of Child Psychology & Psychiatry, 51*, 915-923. doi:10.1111/j.1469-7610.2010.02217x

Stevens, K. (2009). Developing a descriptive system for a new preference-based measure of health-related quality of life for children. *Quality of Life Research, 18,* 1105-1113. doi: 10.1007/s11136-009-9524-9

Stevens, K. (2011). Assessing the performance of a new generic measure of health-related quality of life for children and refining it for use in health state valuation. *Applied Health Economics and Health Policy, 9,* 157-169. doi: 10.2165/11587350-000000000-00000

Taylor, V., Fuggle, P., & Charman, T. (2001). Well sibling psychological adjustment to chronic physical disorder in a sibling: How important is maternal awareness of their illness attitudes and perceptions? *Journal of Child Psychology and Psychiatry, 42,* 953-962. doi: 10.1111/1469-7610.00791

Verhulst, F.C., & van der Ende, J. (1997). Factors associated with child mental health service use in the community. *Journal of the American Academy of Child & Adolescent Psychiatry, 36,* 901-909. doi: 10.1097/00004583-199707000-00011

Wallander, J.L., & Varni, J.W. (1992). Adjustment in children with chronic physical disorders: Programmatic research on a disability-stress-coping model. In A.M. LaGreca, L. Siegal, J.L. Wallander, & C.E., Walker (Eds.). *Stress and coping in child health.* New York, USA: Guildford Press.

Yeh, M., & Weisz, J.R. (2001). Why are we here at the clinic? Parent-child (dis)agreement on referral problems at outpatient treatment entry. *Journal of Consulting and Clinical Psychology, 69,* 1018-1025. doi: 10.1037/0022-006X.69.6.1018

Chapter 10. Conclusion

10.1 Overview of Chapter

The current chapter discusses the findings and implications of the body of work presented in this thesis. The findings from the individual papers are integrated to form more substantive conclusions and implications than those described in the individual papers. The chapter is structured by how this body of work relates to the main aims of the thesis. To reiterate, the main aims of this thesis were to a) clarify the current state of evidence, b) assess the methodologies used, c) formulate theoretical and clinical implications beyond that found in primary studies, d) highlight what is not yet known in this field and recommendations for future research, and e) conduct a primary research study that addresses some of the gaps in the literature and advances our knowledge of the mental health and wellbeing of siblings of children with MHPs. As highlighted in Chapters 7, 8, and 9, the primary research study was able to address gaps in the literature and contribute to our understanding of siblings. Thus, in the current chapter, the findings of the primary research study are discussed within the context of the first aim of the thesis as it forms part of the current state of evidence on the mental health and wellbeing of siblings.

10.2 Current State of Evidence

Broadly, the results of this thesis have shown that siblings of children with MHPs are at increased risk of psychopathology, difficulties in areas of psychosocial functioning, and more negative and less positive family relationships compared to those without siblings with MHPs. In addition, siblings of children with MHPs experience a range of daily challenges associated with having a brother or sister with MHPs. Thus,

siblings of children with MHPs experience a range of difficulties across all areas relating to mental health and wellbeing. While the evidence suggests this, it should be noted that due to methodological limitations of the literature and due to a paucity of high quality studies, at this time, this conclusion is somewhat tentative. This is particularly the case when examining statistical significance. For example, sample sizes were typically small and thus the studies had limited power to detect statistically significant differences between target and control siblings. The following discussions then outline the main findings in more detail with a focus on trends in the data, what the majority of studies have found, and what higher quality studies show. I do, however, recognise that the findings are not rigorously conclusive, particularly in regards to the quantitative works.

10.2.1 Mental health.

The body of work presented in Chapters 3, 4, 8, and 9 showed that siblings of children with MHPs had higher rates of psychopathology and emotional and behavioural difficulties and poorer psychosocial functioning than control siblings. Consistently across these works, siblings of children with MHPs were at risk of a range of psychopathologies and were at risk of poorer functioning across a range of functioning domains. The magnitude of the difference between siblings of children with MHPs and control siblings was considerable. Based on the higher quality studies from the systematic review and the primary research study, siblings of children with MHPs were two to four times more likely to have MHPs than control siblings. Psychosocial functioning typically differed between these two groups to a moderate to large degree. However, confidence intervals for these effect sizes were, in the majority of cases, considerably wide. Thus, we cannot conclude with confidence that these effect size

estimates are precise and that future studies will report similar effect sizes. The width of effect size confidence intervals is largely dependent upon sample size (Higgins & Green, 2011). The majority of studies in the systematic review had small sample sizes (n<100), which may explain why the effect size confidence intervals were wide. However, wide effect size confidence intervals were also noted in studies with relatively large sample sizes (n>100). This suggests that the width of the confidence interval may be explained by variation in outcome scores across participants (Higgins & Green, 2011). That is, there is considerable variation across siblings in terms of psychological outcomes. Thus, while the majority of studies show that, overall as a group, siblings of children with MHPs have more mental health difficulties than control siblings, it is important to bear in mind that it is likely that there is considerable individual variation in mental health across siblings.

In addition to a broad risk of mental health difficulties, two trends relating to specific areas of risk emerged. First, there was a trend for siblings to be at risk of similar psychopathologies to the target child. This was particularly the case when examining the more general psychopathology categories of externalising and internalising childhood disorders. However, siblings were not necessarily at greater risk of similar psychopathologies when compared to the risk of other forms of psychopathology. That is, siblings of children with depression were at risk of depression but also had equally high risk of other disorders, such as anxiety disorders. Further, this trend may have been an artefact of a focus on concordance in the sibling literature. Second, there was a trend for siblings of children with MHPs to have more difficulties in functioning in particular areas. Regardless of the diagnosis of the target child, target siblings had greater impairments in delinquent and social behaviour problems, somatic complaints, and anxious/depressed behaviours.

In regards to moderators of the mental health of siblings of children with MHPs, the existing literature focused primarily on three moderator variables: Target child and sibling age and gender, parental psychopathology and mental health, and the type or severity of the target child's MHPs. Firstly, target child age and gender did not significantly moderate the mental health of their siblings. In contrast, the age and gender of the sibling was significantly related to their risk of MHPs and/or impaired psychosocial functioning. This is not surprising given that in the general population, age and gender similarly play a role in the risk of MHPs (Parritz & Troy, 2011). The relationship between gender and risk varied depending on the type of outcome measured (e.g. type of psychopathology) and these variations typically matched general population patterns. For example, female siblings of children with MHPs had higher scores on internalising problem scales compared to male siblings (see Chapter 4). Similarly, in the general population, females are at greater risk of internalising disorders than males (Merikangas et al., 2010). When the rate of any MHP was assessed in siblings (Chapter 9), no gender effect was found. In regards to age, siblings younger in age were found to be at greater risk of MHPs and/or impaired functioning. This also matches general population trends (Sawyer et al., 2000).

Two studies examined the role of the age and/or gender composition of the target child-sibling dyad in moderating the mental health of siblings. Szatmari, Offord, and Boyle (1993) found that gender composition (i.e. all male, all female, and mixed gender composition) significantly moderated the risk of MHPs in the target child and sibling (Szatmari et al., 1993). The age composition did not. However, age composition was only examined by comparing dyads where target children and siblings were both 4-11 years old, both 12-16 years old, or each child fell in a different age bracket. Other forms of age compositions were not examined, such as difference in age.

In contrast, the primary research study presented in Chapter 9, found that birth order and age difference were related to the presence of MHPs in siblings. Siblings with MHPs were twice as likely to be the younger sibling than those without MHPs. The former were also closer in age to the target child, approximately within 5 years of age, than those without MHPs. Age-spacing had a small to moderate effect in differentiating siblings with and without MHPs. Also in contrast to Szatmari and colleagues' (1993) study, no significant effect for gender composition was found in the primary research study. Szatmari and colleagues (1993) examined rates of MHPs that tend to differ significantly according to gender (i.e. attention deficit, conduct, and emotional problems; Merikangas et al., 2010). This may explain the divergent findings across their study and those presented in this thesis.

Second, findings on the role of parental psychopathology and mental health varied considerably across studies. As discussed in Chapters 3 and 4, the findings were inconsistent and/or varied according to the type of psychopathology assessed in the parents and the type of psychological outcome measured in the sibling. For example, one study reported a significant association between parental affective disorders and sibling depression while another study found no significant relationship. Too few studies examined the role of parental psychopathology and, as a result, no conclusions about the role of parental psychopathology can be made at this time as it directly relates to the mental health of siblings of children with MHPs. In research with children in general, parental psychopathology has been consistently shown to impact on child mental health (Parritz & Troy, 2011). However, the question remains if this applies to siblings of children with MHPs to a significant degree. It may be that having a brother or sister with MHPs is significantly related to increased sibling risk regardless of the status of the parents.

Third, findings on the role of the type and severity of the target child's MHPs in moderating sibling mental health varied considerably. Similarly to parental mental health, the findings were inconsistent and/or varied according to the psychological outcome measured in the sibling and the comparison group used. For example, siblings of children with ADHD were more likely to have ADHD compared to siblings of children with affective disorders but not in comparison to siblings of children with CD or those of children with anxiety disorders. Very few studies explored the role of the type and severity of the target child's MHPs. As a result, specific combinations of target child MHP, sibling outcome, and comparison group used were typically explored by only one study. Thus, no conclusions about the role of the type and severity of the target child's MHPs can be formed. When diagnosis specific variations were not considered, as was the case in the primary research in this thesis, no significant effect for the severity of symptoms or functional difficulties was found.

10.2.2 Family functioning.

The body of work presented in Chapter 5 and 6 examined the functioning of families of children with MHPs with a focus on the impact on siblings. Although the qualitative work presented in Chapter 6 did not exclusively focus on family functioning, the most salient aspects of sibling experiences related directly to home life and family functioning.

Siblings of children with MHPs have more negative (e.g. conflict) and less positive (e.g. supportive) relationships with the target child and with their parents compared to control siblings. Effect sizes were typically in the small to moderate range. However, the findings varied in terms of statistical significance and effect size confidence intervals were wide. This was likely due to methodological limitations in the

existing literature. The majority of studies had small sample sizes (n<100) resulting in insufficient power to detect statistically significant differences and resulting in limited precision in effect size estimates. However, studies with large sample sizes also reported effect sizes with wide confidence intervals. Thus, it is likely that there is considerable variation in the quality of family relationships across siblings.

The qualitative metasynthesis showed that having a brother or sister with MHPs has a significant, predominantly negative impact on the lives of siblings. This impact was seen across all areas of siblings' lives including relationships, self or personal wellbeing, and daily routines, and was seen across numerous settings including home and school. The qualitative metasynthesis also highlighted several experiences relating to having a brother or sister with MHPs that may contribute to the quality of family relationships. For example, violence, conflict, and aggression created a home life environment of chaos and conflict, and had a significant negative impact on the quality of the target child-sibling relationship. Furthermore, parents' inability to effectively manage and prevent violence directed towards the sibling likely had a significant negative impact on the parent-sibling relationship. 'Being another parent' creates a different family dynamic wherein siblings have similar responsibilities and burdens to parents, yet no authority in family decision-making and no recognition. This would likely cause tension in target child-sibling relationships promoting feelings of resentment and creating a disharmony between the extremes of having to exert control over the target child and manage their behaviours, while at the same time having a complete lack of control over decision making and being a victim of the target child's behaviours. Experiences such as these likely contribute to the impaired family relationship quality found in quantitative studies.

However, the qualitative study does not provide the entire picture. The qualitative literature in this field focuses primarily on impact of illness pathways and primarily on the experiences of siblings. Parents and target children have unique family experiences that also impact on family relationship quality for siblings. For example, target children might view their sibling as insensitive, lacking empathy, and unable to truly understand the difficulties they are experiencing. This may contribute to greater distance in the target child-sibling relationship and may result in less positive and supportive target-child sibling relationships. To form a complete and comprehensive understanding of family functioning as it pertains to siblings of children with MHPs, it is necessary to integrate information from the perspectives of all family members. Furthermore, other pathways besides those related to the impact of illness may contribute to the quality of family relationships. For example, a substantial proportion of siblings of children with MHPs display significant mental health difficulties and this is likely to impact on the quality of sibling and parent-sibling relationships.

Findings from the quantitative systematic review suggest that the type of MHP in the target child may also moderate the quality of family relationships. Two trends emerged from examinations across the entire body of literature. First, sibling relationships in families of children with CD were more positive than control siblings. This was not found in any other families of children with MHPs. As discussed in more detail in Chapter 5, sibling deviance training research has hypothesised that siblings and target children with CD may have closer sibling relationships due to an underlying deviance training family dynamic. For example, if siblings and target children have a close sibling relationship this facilitates increased association with deviant peers, collusion to undermine authorities, and encourages mutual reinforcement of deviant behaviours (Bullock & Dishion, 2002). No qualitative studies focused on families of

children with CD and no studies directly compared sibling relationships in families of children with CD compared to those of children with other MHPs. Thus, due to limited research, it is unclear if this is a reliable finding and, if so, what may lead to more positive sibling relationships in these families.

The second trend was that siblings of children with anxiety disorders may have the least risk of impairments in family functioning compared to those of children with other MHPs. Sibling relationships in families of children with anxiety disorders displayed the least impairment and the presence of anxiety disorders in the target child was actually associated with reduced odds of sibling relationship problems. The qualitative research suggests that siblings of children with anxiety disorders may experience mostly practical challenges, such as delays due to the target child's compulsive behaviours, rather than emotional challenges, such as interpersonal conflict. The former may result in less impact on the mental health and wellbeing of siblings and on the quality of family relationships than the latter.

In light of these findings, small trends found in the quantitative data on the mental health of siblings could be given greater consideration. Siblings of children with anxiety disorders, with the exception of obsessive-compulsive disorder (OCD), did not have elevated rates of psychopathology. This was the only group of siblings that did not display a broad risk of psychopathology above that found in the general population. Thus, it may be that siblings of children with anxiety disorders have the least risk of impairment across a range of mental health and wellbeing domains. It may be that having a brother or sister with an anxiety disorder has less impact on the lives of siblings and is associated with risk factors that do not increase the risk of impaired functioning in areas other than anxiety. For example, parents of children with anxiety disorders have been found to display more over-involvement in child activities than

control parents (Hudson & Rapee, 2002). Parental over-involvement has been primarily linked to the development of anxiety disorders in children over other types of MHPs (Parritz & Troy, 2011). Furthermore, behavioural inhibition, a temperamental construct with a high genetic contribution, has been linked primarily to the development of anxiety disorders (Svihra & Katzman, 2004). It has been suggested that behavioural inhibition is linked to protective factors against other forms of psychopathology, such as substance use (Simons, Dvorak, & Lau-Barraco, 2009). Thus, siblings of children with anxiety disorders may be more likely to have high behavioural inhibition and this may result in lower risk of other forms of MHPs and impairments in functioning. More research in this area is needed to test the reliability and consistency of this trend and to explore possible underlying mechanisms for this reduced risk.

10.2.3 Coping strategies and seeking treatment.

As discussed above, the evidence strongly suggests that siblings of children with MHPs are at risk of a range of impairments in mental health and wellbeing and experience a range of challenges in their every day lives. The body of work presented in Chapters 6 and 8 provide insight into how siblings cope with these challenges and what, if any, support or treatment are provided for siblings that are displaying MHPs. As discussed in Chapter 6, the primary coping strategies used by siblings are avoidance, accommodation, and normalisation. Though no studies with siblings of children with MHPs examined the effectiveness and consequences of these coping strategies, research with adult siblings suggests that these strategies are unhealthy and may be detrimental to siblings, particularly in terms of their relationship with the target child. Only one qualitative study made mention of siblings seeking help from social supports or

professional services and no known studies have explored if siblings receive any form of support.

In the primary research study presented in Chapter 8, I attempted to address this gap and ascertain how many siblings were receiving some form of support and what types of support were used. The vast majority of siblings who were identified by caregivers as experiencing significant emotional and/or behavioural difficulties had, were on the waitlist for, or were currently receiving some form of mental health treatment, most commonly through psychological services.

10.2.4 Summary of key findings.

In sum, in addressing our first aim, the current state of knowledge on siblings of children with MHPs strongly suggests an increased broad risk of impairments across a range of mental health and wellbeing domains compared to those of children without MHPs. The findings also show that siblings experience a range of difficulties, particularly in their daily lives and in family relationships. These difficulties may not necessarily manifest in clinical presentations but nonetheless can have a significant impact on the wellbeing of siblings. Thus, this thesis has highlighted that all areas of siblings' lives are impacted and a holistic, multi-dimensional and multi-system approach is needed when working with these siblings. Few trends revealing specific patterns of risk could be identified. Further, the current state of knowledge on moderators and underlying processes is severely lacking. Thus, while advancements have been made over the past 20 years in our understanding of the mental health and wellbeing of siblings, significant gaps and questions remain.

10.3 Methodologies Used in Sibling Research

One of the biggest contributors to our gaps in knowledge on the mental health and wellbeing of siblings of children with MHPs is methodological issues. In fact, one of the key findings of this thesis is that significant methodological flaws are found across the literature. This section addresses the second aim of my thesis and comments on my assessment of the methodologies used across the sibling literature. First, the methodological limitations found in the literature will be discussed. Second, guidelines for future research with siblings of children with MHPs are outlined. This section addresses methodological issues; topics for future research are discussed in later sections.

10.3.1 Methodological limitations in sibling research.

Five main limitations were found in the research with siblings of children with MHPs that recurred across each of the areas of functioning examined in this thesis. Methodological limitations specific to the areas of functioning were noted in the individual chapters (see Chapters 3, 4, and 5). Only those methodological limitations found across the literature and different areas of functioning are discussed here.

First, the vast majority of studies had small sample sizes with $n < 100$. Thus, the reliability and generalizability of the findings from these studies are limited. As previously discussed, small sample sizes likely impacted on the consistency of findings across studies in terms of statistically significant differences between target siblings and controls. When focusing on the higher quality studies with larger sample sizes, statistically significant differences were often reported. Small sample sizes also impact on the reliability of the effect size estimates and contributed to wide confidence intervals.

Second, there was a lot of variation across studies in how the mental health and wellbeing of siblings were assessed. Three primary methods were used: a) recruitment of all available siblings of a particular target child, b) recruitment of one of the target child's sibling, most commonly the sibling closest in age to the target child, and c) assessing the proportion of target children with siblings with the psychological outcome. Although these methods and their accuracy and/or reliability have not been examined, there are a priori reasons that suggest these different methods would produce different results. The first method is likely to be the most accurate and reliable method as it allows assessments of the entire sample of siblings for each family. Although this would still be subject to sampling error as it only recruits a portion of the entire population of siblings of children with MHPs, it would likely result in a smaller error margin than the other two methods. The second method, in contrast, increases sampling error by recruiting only a subset of siblings within each family. In fact, the majority of studies that recruited only one sibling did not do so in a random fashion, which would increase the reliability and generalizability of the findings, but chose the sibling closest in age to the target child. As shown in Chapter 9, siblings with MHPs were closer in age to the target child than siblings without MHPs. Thus, studies that use this method may overestimate the prevalence of mental health problems in siblings of children with MHPs. The third method is likely to result in the least accurate findings. It is likely to lead to overestimations of sibling risk as it effectively selects any sibling with mental health difficulties. By treating the target child's siblings as one unit, it may also lead to underestimations of sibling risk when the target child has multiple siblings with psychiatric diagnoses or mental health difficulties. Although, in my opinion, this method is not an accurate method of assessing sibling mental health and wellbeing, it has been frequently used across the literature as an indication of risk of

psychopathology for siblings (see Chapter 3) and therefore should be considered in this discussion of sibling research methodology.

Third, there has been considerable variation across the literature in the types of MHPs in the target child. Some studies have focused on individual disorders, such as ADHD, others have focused on specific disorder combinations, such as depression and anxiety, and others have examined siblings of children with a broad range of MHPs. Because of this variation it has been difficult to identify consistent findings and trends in the data, particularly in regards to moderators. Furthermore, some disorders have been extensively researched while others have been neglected. The majority of the high quality research has been conducted with siblings of children with ADHD. Because genetic and shared family environment risk factors have been the focus in ADHD research, impact of illness factors are largely ignored in this literature and there has been little integration of the numerous theories that are relevant to sibling mental health.

Fourth, approximately half of the quantitative studies included some form of control group. The remainder relied on standardised norms or general population prevalence rates. As shown in Chapter 4, reliance on general population data or norms can result in inaccurate conclusions. Based on standardised norms, one might conclude that siblings of children with MHPs do not have significant difficulties in psychosocial functioning. However, comparisons with control siblings show that siblings of children with MHPs experience significantly more mental health difficulties and impairments in psychosocial functioning.

Last, research with siblings of children with MHPs has not been driven by the findings of past research. As argued in Chapter 1, without a systematic review in this field, there has been little guidance for researchers on methodology and on questions

that have been answered and those that remain. Because of this, there has been a lack of meaningful advancement in this field. Later studies tend to repeat methodological limitations and continue to explore moderators that have been consistently shown to have little or no relationship to the mental health of siblings (e.g. target child age and gender). Furthermore, there is little evidence that later studies build upon and advance the scientific contributions made by past studies. For example, each of the qualitative studies included in the metasynthesis presented in Chapter 6 explored similar concepts using similar methodologies with similar populations. As a result, the same findings were repeated across 10 studies over a 15-year period with little to no additional advancements in knowledge made by later studies. While replication and consistency is a cornerstone of scientific knowledge, every study should endeavour to make advancements beyond past research. Thus far, there has been little evidence of this in the sibling literature.

10.3.2 Methodological guidelines for sibling research.

Based on the observations described above, the following is a list of guidelines for future researchers. It should be noted that there are additional guidelines relating to specific areas of functioning, as discussed in the individual chapters. It should also be noted that I recognise that there are often practical limitations to conducting research and it may not be possible to meet all of these guidelines. The following is a list of what I believe to be the more essential guidelines that researchers should endeavour to meet when exploring the mental health and wellbeing of siblings of children with MHPs.

A) Obtain a sample size that is associated with sufficient power to detect differences.

⇒ The following sample size suggestions are based on power calculations from Cohen's (1992) article and the effect sizes most consistently found in the existing literature. Based on a moderate to large effect size found for differences in mental health outcomes between target and control siblings, the most conservative estimations (d=0.50) suggest a sample of 128 or more siblings and controls combined should yield sufficient power (i.e. 0.80). Based on a small to moderate effect size (d≈0.35) found for differences in family relationship quality outcomes, the sample should approximate 260 siblings and controls combined. Statistically significant findings on moderators often yielded a similar effect size to that found for family relationship quality, thus a similar sample size is recommended when exploring moderator variables.

B) Assess all available target siblings.

⇒ If this cannot be achieved, assess one *randomly* selected sibling.

C) Obtain data on the mental health of siblings of children with a range of MHPs.

⇒ As previously argued, given the complexities of childhood diagnoses, it may be more clinically useful to focus on siblings of children with a range of MHPs rather than those of children with a specific disorder. Research with siblings of children with a specific disorder however, is still valuable, particularly in regards to theory development and exploring type of target child MHPs as predictors of sibling mental health.

⇒ If a specific combination of disorder in the target child is of primary interest, include sibling data divided by the individual diagnoses in the target child. For example, if siblings of children with depression and anxiety are of interest,

include segregated data on siblings of children with depression and siblings of children with anxiety.

D) Recruit a matched control group of siblings of children without MHPs.

E) Aim to make significant advancements and contributions beyond past research.

⇒ Consult past research and the systematic reviews presented in this thesis to clarify the current state of evidence and aim to build upon these findings and address literature gaps.

⇒ Use theoretical frameworks, including impact of illness frameworks, as well as the findings of past research to guide explorations of moderators of the mental health and wellbeing of siblings of children with MHPs.

10.4 Theoretical Implications

The following section addresses the third aim of this thesis: To formulate theoretical implications beyond that found in primary studies. As previously noted, due to a paucity of research, no conclusive patterns could be identified regarding moderators of the mental health and wellbeing of siblings. Thus, the current state of evidence cannot directly support or refute existing theories that are relevant to the mental health and wellbeing of siblings of children with MHPs. However, several trends regarding theoretical frameworks were noted across the literature and warrant further consideration.

First, two theoretical frameworks, developmental psychopathology and impact of illness frameworks (see Chapter 1), have been favoured by researchers in this area and although each has received support, there has been little integration and mutual consideration of these frameworks across the literature. I noted that across the existing literature, researchers typically relied on one of these to inform their research and guide

their explorations of moderators. Most commonly, studies on psychopathology and psychosocial functioning in target siblings appealed to developmental psychopathology frameworks while family relationship and qualitative works on the experiences of siblings focused on impact of illness frameworks. Yet, preliminary findings on moderators provide support for both frameworks.

For example, parental psychopathology is a risk factor that features prominently in developmental psychopathology frameworks (Parritz & Troy, 2011). The presence of parental psychopathology also likely plays a role in impact of illness frameworks. For example, the degree to which siblings' lives were impacted by the target child's behaviour, particularly in terms of violence towards siblings, was directly related to how parents managed these behaviours. Given that parental psychopathology significantly affects parenting styles and parental efficacy, parents with mental health difficulties are less likely to effectively manage the target child's behaviour. Thus, assuming that parental psychopathology significantly moderates the relationship between the mental health of the target child and the target sibling, it could have effects through both developmental psychopathology pathways (e.g. harsh and inconsistent parenting) and those specific to impact of illness pathways (e.g. impaired management of the target child's behavioural problems as it impacts on the siblings). It is important that researchers consider both of these frameworks when conducting research with siblings of children with MHPs and when drawing implications from their findings. To date, there has been little evidence of integration and of due consideration given to both theoretical frameworks.

Second, several theories that are relevant to the mental health and wellbeing of siblings of children with MHPs have been overlooked. Normative sibling theories, for example, highlight several variables that should be examined as moderators of the

mental health and wellbeing of siblings. But little research has appealed to these theories and explored the variables highlighted. For example, Brody's (1998) family experiences and sibling relationships model (section 1.4.1.1) highlights parental management of sibling conflict as a key factor in determining the quality of sibling relationships. While several variables from his model have been explored, such as parental differential treatment, no known quantitative research has explored the role of parental management of sibling conflict on the quality of sibling relationships for siblings of children with MHPs. Similarly, birth order theory (Bowen, 1978) has been overlooked (section 1.4.1.3). The primary research study presented in this thesis describes the first known research to explore birth order and age-spacing as predictors of the presence of clinically significant MHPs in siblings of children with MHPs.

Third, there are theories that may be relevant to the mental health and wellbeing of siblings of children with MHPs that have been ignored entirely. The social model of disability (section 1.4.3), for example, highlights the role of prejudice, discrimination, and stigma in determining the extent to which illness can impact on the lives of family members (Dowling & Dolan, 2001). These factors may play a role in the lives of siblings of children with MHPs. For example, siblings may have difficulty forming close, trusting relationships with others. They may not want to confide in others and disclose personal details about their family and home life out of fear of being judged negatively based on having a brother or sister with MHPs. Siblings may not seek social support for the same reasons, resulting in isolation and reliance on less healthy coping strategies. To date, no known research has explored the role of stigma and prejudice in moderating the mental health and wellbeing of siblings of children with MHPs.

In sum, although there are numerous existing theories that can inform research with siblings of children with MHPs, there has been little evidence to date that

researchers have integrated the perspectives of multiple theories or have given due consideration to multiple theories. The primary research study I conducted presents an attempt to integrate the perspectives of multiple theories. Asides from this, the group of researchers and theorists that have achieved the greatest degree of integration, thus far, are those that research sibling deviance training (see section 1.4.1; Bullock & Dishion, 2002; Feinberg et al., 2013). This theory and research incorporates the notion that having a brother or sister with MHPs can impact on your mental health via modelling and reinforcement of dysfunctional behaviours, through facilitating associations with like-minded peers, through increasing exposure to risky behaviours and environments, and through sibling collusion to undermine authorities. It also incorporates factors that feature in developmental psychopathology frameworks, such as the role of deviant peer associations and exposure to illicit substances (Parritz & Troy, 2011). In addition, it incorporates other theories, such as birth order theory, in the sense that most researchers and theorists believe that older siblings are more likely to influence younger siblings and thus focus on the impact of MHPs in the older sibling on the mental health of their younger sibling (Brotman et al., 2004; Bullock & Dishion, 2002).

There are limitations to this theory and research however. First, sibling deviance training does not incorporate other key factors that are likely to play a role in these pathways. For example, it does not consider genetic risk factors that both children would share and would increase their propensity to engage in similar behaviours. This theory also does not consider neurobiological factors shared by siblings and target children that can increase the risk of certain behaviours, such as substance abuse (Simons et al., 2009). Second, sibling deviance training has typically only been applied to target children with conduct disorder, delinquent behaviour problems, and substance use problems (Brotman Gouley, O'Neil, & Klein, 2004; Bullock & Dishion, 2002;

Wasserman, Miller, Pinner, & Jaramillo, 1996). Yet similar pathways may also apply to target children with other disorders and their siblings. For example, modelling of avoidant strategies and accommodation around anxiety related behaviours may facilitate the development and/or maintenance of anxiety symptoms in target children with anxiety disorders and their siblings (Barrett, Rasmussen, & Healy, 2001). Nevertheless, sibling deviance training theory and research based on this theory presents an example of how multiple theoretical frameworks can be integrated to inform our understanding of and research on the mental health and wellbeing of siblings of children with MHPs.

It is hoped that this thesis will draw attention to the relevancy of multiple theories and encourage future researchers to adopt a holistic and inclusive perspective when considering the mental health and wellbeing of siblings of children with MHPs rather than the one-theory view observed thus far.

10.5 Implications for Clinical Practice and Policy

This section addresses the third aim of this thesis and discusses the clinical implications formed from this body of work that extends the findings from the individual papers. First, broad clinical implications are outlined followed by a discussion of prevention, treatment, and/or support programs for siblings of children with MHPs.

10.5.1 Broad clinical implications.

The findings of the body of work presented in this thesis have several broad clinical implications for parents and mental health clinicians working with children and families. First, siblings of children with MHPs are at risk for difficulties in all mental health and wellbeing domains. This points to the need for parents and clinicians to

monitor and assess the functioning of siblings across multiple domains to ensure they receive the appropriate support. It should be noted that the findings from the primary research study suggest that once parents or caregivers are aware that target siblings are displaying MHPs, they actively seek mental health treatment for these siblings. This presumes a level of mental health literacy in parents. As they are the primary gatekeepers between children and mental health services, it is important that clinicians strive to improve the mental health literacy of parents and guide them towards particular behaviours to attend to in siblings. However, as research has shown, a substantial proportion of individuals with MHPs do not receive mental health treatment (Bristow & Patten, 2002). Thus, it is highly recommended that mental health literacy of both children and parents be promoted using universal strategies that can reach the general population as well.

Second, while facilitating mental health treatment for siblings who are displaying MHPs is recommended, it is also important to create and/or implement some form of prevention or support programs for those that may not be displaying clinical levels of impairment. As discussed in greater detail in Chapter 8, siblings displaying MHPs may account for an up to 38% increase in workload for already understaffed and underfunded mental health services. Introducing sibling prevention programs when the target child first presents for mental health treatment may reduce the incidence of MHPs in siblings and the burden on mental health services. Cost-benefit analyses should be incorporated into any research in this area. In addition, prevention or support programs will likely have significant benefits for family functioning and the long-term mental health of both the target child and their siblings if the incidence of MHPs in siblings can be reduced. Alternatively, some form of family inclusive treatment could be implemented. Research has shown that the inclusion of siblings in the treatment of

the target child has significant benefits for all family members (Castorina & Negri, 2011; Rowe, 1992).

Third, mental health clinic policies should recognise the increased risk of impaired functioning for siblings of children with MHPs. Most mental health services for children and adolescents do not have policies or practice guidelines specifically relating to siblings (e.g. New South Wales Ministry of Health, 2011). As a result, there is likely to be significant variation across clinicians and services in the extent to which the mental health of siblings is assessed. There is also likely to be variations in how clinicians manage siblings that are displaying MHPs. Enacting a clinic policy that recognises siblings and outlines assessment, practice, and treatment guidelines for siblings would ensure that siblings are acknowledged and reduce the number of siblings that may not receive appropriate treatment. Admittedly, it is difficult to develop such policies as there are no known best practice guidelines for managing siblings of children with MHPs. Additional research with this population is greatly needed. As a starting point, I recommend that mental health clinic policies include a recommendation or requirement that the clinician enquire about the mental health and functioning of siblings and highlight warning signs of MHPs that parents should be aware of.

Last, a necessary step underpinning each of the aforementioned implications for clinical practice is awareness. Clinicians and mental health clinics need to be aware that there is a substantial risk of poorer mental health and wellbeing in siblings of children with MHPs before any of the above can occur. Large-scale dissemination of research is difficult, particularly when mental health clinicians work in several different areas including government mental health clinics, private practice, and hospitals. I recommend that information on the mental health and wellbeing of siblings be disseminated to universities and programs responsible for training psychologists and

other allied health professionals (e.g. social workers), disseminated as a professional development resource for registered allied health professionals, and disseminated to general practitioners as they represent a primary gatekeeper for mental health services. It is essential that efforts be made to increase awareness of this at-need, high-risk population to facilitate appropriate support and treatment for siblings of children with MHPs.

10.5.2 Implications for treatment, prevention, and/or support programs.

While it is difficult to formulate specific practice guidelines for working with siblings of children with MHPs given the paucity of empirical literature, there are several suggestions that can be formed based on the body of work presented in this thesis. Broadly, as highlighted in the theoretical implications, all of the theories discussed in Chapter 1 have received some support in the sibling literature. Thus, sibling treatment, prevention, and/or support programs should include components that manage the processes highlighted in these theories.

First, there is a large body of literature exploring prevention programs for childhood MHPs based on developmental psychopathology frameworks (e.g. see Barrett, Farrell, Ollendick, & Dadds, 2006; Greenberg, Domitrovich, & Bumbarger, 1999; Stice, Shaw, Bohon, Marti, & Rohde, 2009). These typically include components that address parenting skills and that enhance the coping skills of children and adolescents. These programs have also been included in programs designed for siblings of children with MHPs with significant benefits on parenting practices and the psychosocial functioning of siblings (Brotman, Gouley, Chesir-Teran, Dennis, Klein, & Shrout, 2005).

Second, prevention programs based on normative sibling theories have also been developed. Feinberg and colleagues (2013) tested a program based directly on their model of the pathways from sibling relationships to child adjustment. The Siblings are Special or SIBS program was designed to improve the sibling relationship and prevent the development of child adjustment programs through targeting the sibling relationship pathways to adjustment problems. Three key strategies were used: Improving the quality of the sibling relationship, improving the child's individual interpersonal skills, and addressing parental behaviours that relate to sibling relationships (e.g. management of sibling conflict). This program was then tested using a randomised controlled trial with 174 families. Significant benefits were found including improvements in sibling fair play, sibling positivity, lower levels of internalising problems, and increases in self-control and social competence. However, there was little effect on parenting practices and on child externalising problems. Although this program was developed as a universal prevention program, it may provide insight into program components that are effective in improving sibling relationships in particular. Because this is a universal program, not specifically designed for use with families of children with MHPs, and because the benefits have primarily been on positive constructs, the program components may be particularly useful as additions to a problem-based or treatment-based program that focuses on reducing problematic or maladaptive child and parent behaviours.

Last, programs based on impact of illness frameworks have received the least attention with regard to siblings of children with MHPs. Only pilot studies have been conducted thus far and there is significant variation across these programs. Some programs are primarily support networks that focus on providing social support for siblings and psychoeducation on MHPs (Griffiths & Sin, 2013). Other programs focus

primarily on giving siblings a voice and facilitating an open family discussion on the difficulties that siblings face (Bamberg, Toumbourou, & Marks, 2008). Last, some siblings programs focus on enhancing the coping skills of siblings (Pitman & Matthey, 2004). Based on preliminary findings, these programs have significant benefits for siblings including increased social support and reduced isolation, improvements in family functioning, increases in mental health knowledge, increases in ability to use coping skills, and reductions in stress.

While there are numerous programs that address the range of factors that impact on the mental health and wellbeing of siblings of children with MHPs, there are several key experiences relating to the impact of illness that have not been included in any of these programs. These were particularly highlighted in the qualitative metasynthesis in Chapter 6. These included increased caregiving responsibilities without reward or recognition from parents, parental favouritism, and unhealthy strategies used to cope with the target child's behaviour. As shown in Chapter 6, appealing to research with siblings of children with special needs and adult siblings of individuals with MHPs may provide insight into strategies that could be used to address these difficulties faced by siblings. First, regarding increased caregiving responsibilities, siblings of children with special needs report being included in family decision-making processes (Beilby, Byrnes, & Young, 2012) while siblings of children with MHPs report having no input in decision-making. Inclusion in decision-making processes may give siblings a greater sense of control and collaboration and reduce feelings of burden and resentment. Siblings of children with special needs may also have greater opportunities for respite. For example, almost half of all sibling programs identified in a recent systematic review of interventions for siblings of children with cancer included a focus on recreation and respite (Prchal & Landolt, 2009). In contrast, only sibling support networks include a

recreation aspect and these events happen very infrequently. In fact, siblings were often described as being the source of respite for their parents (see Chapter 6). Thus, it is recommended that any sibling treatment, prevention, or support programs include discussions with parents on the importance of arranging respite for siblings, on acknowledging and rewarding siblings for caregiving tasks, and including siblings in decision-making processes that have a significant impact on their lives.

Second, parental favouritism is another challenge that siblings emphasise. It is important that sibling programs emphasise the need to allow for quality time for parents and siblings. Treatment programs for children with MHPs recommend allowing 15-20 minutes of daily parent-child interaction and activities for each child in the family (Greene, 1998). This time should be spent only with the parent and sibling and should be free from distractions. It may also be useful to remind parents to acknowledge and validate the difficulties that the sibling faces to ensure that their feelings and needs are cared for and not just those of the target child.

Third, the coping strategies used by siblings of children with MHPs – avoidance and accommodation, in particular – have been described by adult siblings as useful at the time but unhealthy and detrimental in the long run. As described in more detail in Chapter 6, adult siblings have found that separating the illness from the target child was a more useful and adaptive way for them to cope. That is, recognising that the target child and their MHPs are separate and defining the illness as the problem rather than viewing the target child as the problem (Kinsella, Anderson, & Anderson, 1996). This strategy can help siblings relate in a healthier way to the target child and may reduce negative emotions towards the target child by depersonalising their behaviour (Dimitropoulos et al., 2009). Some of the above programs have included coping skills training but have not included a discussion of the type of strategies used specifically to

manage the target child's behaviour, particularly in terms of perceptions of their motivations and personalising their behaviour. It is essential that sibling programs include a component that facilitates the development of this strategy, as well as other coping strategies, to replace the less helpful ones that siblings may be using. In order to achieve this, it is also important to include a psychoeducation component that informs siblings on the cause of MHPs and the controllability of related behaviour problems.

In sum, based on the collective works in this thesis, I recommend that sibling programs include components from normative sibling theories, developmental psychopathology pathways, and impact of illness pathways, particularly those discussed above. No such program exists, to the best of my knowledge, however there are several existing programs that include some of these theories and pathways that can be adapted and extended. The development of these programs must be accompanied by evaluations of effectiveness, feasibility, participant uptake, and cost-effectiveness. Furthermore, research on moderators of the mental health and wellbeing of siblings of children with MHPs needs to significantly advance alongside development and evaluations of sibling programs to ensure that such programs are focusing on addressing pathways that have the greatest impact on the mental health and wellbeing of these siblings.

10.6 Recommendations for Future Research

This section addresses the final aim of this thesis: To highlight what is not yet known in this field and highlight recommendations for future research. Recommendations for future research have been covered in great detail in the individual chapters, thus the following discussion will outline the broad areas that future research could explore and what I believe to be the priorities for future research.

First, there is a need for high quality research that assesses the mental health and wellbeing of siblings of children with MHPs. The majority of studies in this field are methodologically flawed. In fact, the highest quality studies found in the systematic review were conducted almost 20 years ago and focused on ADHD only. I have provided guidelines for future sibling research that will hopefully guide future researchers to produce more robust research such that the status of the mental health and wellbeing of siblings of children with MHPs can be more conclusively shown.

Second, longitudinal research that can explore temporal and causal links between the target child's MHP and their sibling is needed. There is an implicit assumption when reading sibling research that there is a causal link, or at least a temporal link, between the mental health of the target child and their siblings. This is yet to be proven. Determining if these links exist will allow more meaningful and directed explorations of sibling-specific pathways in the development of MHPs. Existing longitudinal studies, to date, have not incorporated siblings into their methodology and data collection. The Longitudinal Study of Australian Children, for example, focuses on following the development of one child in the family and obtains very little data on siblings (e.g. age and gender, sibling relationship quality as reported by parents; Sanson et al., 2002). Although these studies are designed to explore an extensive range of determinants of child health and wellbeing, they have not considered the numerous sibling-specific pathways that have been shown to predict health and wellbeing outcomes (Sanson et al., 2002). These existing longitudinal study structures could be expanded to include assessments of sibling mental health and may provide a vehicle by which causal and temporal links can be explored.

Third, research on moderators of the mental health and wellbeing of siblings of children with MHPs is greatly needed. As suggested by effect size confidence intervals,

there is considerable variation in psychological outcomes across siblings of children with MHPs. This variation is an important obstacle to the formation of a more conclusive evidence base on the mental health and wellbeing of siblings of children with MHPs, particularly in regards to conclusions around effect sizes. Future developments in the identification of moderators of sibling mental health that can explain this variation will allow researchers to control for sources of variation and produce more conclusive, reliable, and precise findings. The difficulty with research on moderators in this field is that there is an extremely large number of variables that could contribute to the mental health and wellbeing siblings, as highlighted in the theories discussed in Chapter 1. A more systematic or structured approach to researching moderators, than has been used in sibling research thus far, is needed. The existing sibling research has examined moderators that are implicated in multiple theories and may impact on the mental health of siblings through several pathways. For example, as previously discussed, parental psychopathology can moderate the mental health of siblings through genetic risk factors, family environment risk factors, and impact of illness pathways. As a result, the existing research on moderators has provided little insight into the pathways to the development of MHPs and impaired wellbeing for siblings and into how clinicians and parents can reduce the risk of mental health difficulties for siblings of children with MHPs. I recommend following a similar logic and methodology to that used in twin studies. That is, comparisons should be made across different groups of siblings of children with MHPs that differ in some important aspect (e.g. genetic overlap). For example, comparisons could be made between siblings that currently live in the same household as the target child and those that have reached adulthood and have moved out of home. Such research could highlight the relative contribution of impact of illness factors and pathways (i.e. impact of direct,

daily contact with the target child in the home environment), because genetic influences and childhood rearing environments are shared across these groups of siblings and are, to some extent, controlled for. I recommend that this research focus on factors that are implicated in sibling-specific pathways to the development of MHPs and impaired psychosocial functioning, such as birth order. Insight into these pathways is the unique contribution that research with siblings can provide and can contribute to research in other fields, such as developmental psychopathology and family systems research.

Last, research on sibling treatment, prevention, and support programs for siblings of children with MHPs is needed. There have only been a handful of studies that explore these programs and this research has been limited to the pilot study phase. Considering the findings of this thesis, it is essential that this is a focus of research. Siblings of children with MHPs are experiencing significant difficulties in all areas of their lives but we have few strategies and existing programs to support them.

10.7 Limitations

Similarly to the recommendations for future research, the limitations of the work presented in this thesis have been covered in detail in the individual chapters. Thus, this section will focus on discussing the limitations of the research process used in the thesis as a whole.

First, the inclusion/exclusion criteria for the systematic review were formulated early on. In hindsight, it might have been more useful to include only studies that included a matched control group. This may have justified the use of meta-analytic strategies, albeit based on only a small sample of studies. Because this was the first systematic review in the area, it was considered more appropriate to integrate all of the relevant data rather than screen out studies for methodological reasons or quality.

Alternatively, I could have conducted statistical comparisons of sibling scores and general population data that would have similarly allowed for more effect size data that may have justified meta-analyses. However, in my opinion, this would have only compounded the methodological flaws in the existing literature. I acknowledge that other researchers may have differing views and may have chosen to analyse the available data in a different fashion.

Second, it may have been useful to include a search of the unpublished grey literature. Given the paucity of literature, locating additional data within the grey literature could have been beneficial and strengthened and extended the conclusions in this thesis. However, given that the initial search yielded over 6000 records, including this search strategy would have resulted in an unmanageable dataset within the time span of this thesis.

Third, during the data collection phase of the primary research study, it became apparent that the original questions were not yielding the data we intended to obtain. It would have been useful to pilot the questions with a small sample of participants. Unfortunately, this was not done and almost half of the allotted sample had already been surveyed by the time the need was realised. Furthermore, it would have been useful to conduct surveys in multiple clinical settings. Given that the rate of treatment utilisation for siblings with MHPs is likely to be impacted by individual clinicians and clinic practices, it would have been highly beneficial to obtain data from multiple clinics. However, recruitment was conducted at a clinic within one of the largest child and adolescent mental health services in Australia. It is also not likely that multisite recruitment could have occurred within the constraints of this thesis project and because of the large dataset that resulted from the systematic review. I highly recommend that future research replicate this research in other clinical settings.

Last, the focus of the vast majority of this thesis was on psychological difficulties in siblings of children with MHPs and examining the proportion of these siblings experiencing clinically significant difficulties. This was largely a result of the focus of the existing literature. Siblings who are not presenting with clinically significant MHPs, but who may nonetheless experience a range of challenges and difficulties, are overlooked. This subset of siblings is no less important and in need of support, as suggested in the qualitative literature. Furthermore, explorations of moderators focused on those that increase the risk of MHPs and impairments in functioning and family relationships. This overlooks protective factors that are of equal importance and value. I would have liked to give equal attention to these siblings and to factors that serve to protect the mental health and wellbeing of siblings in the face of significant risk factors and challenges.

10.8 Overall Significance and Contribution to Knowledge

In sum, despite limitations, the body of work presented in this thesis has significantly contributed to our understanding of siblings of children with MHPs with important implications for clinical practice, research, and theoretical frameworks.

Firstly, I conducted the first systematic review on the mental health and wellbeing of siblings of children with MHPs. In using a holistic approach that addressed multiple aspects and perspectives on mental health and wellbeing, several trends across the entire body of literature on these siblings were noted leading to important contributions to knowledge. Methodological limitations were found across the literature and I have recommended several clear guidelines for future research to address these limitations. Limited integration of relevant and applicable theories was noted. I have highlighted the importance of each of these theories and have

recommended that future research use an integrative approach to further advance knowledge on siblings of children with MHPs. Recommendations for support, prevention, and/or treatment programs for siblings of children with MHPs were outlined that draw on the entire body of literature and all of the theories relevant to the wellbeing of siblings. This is the first known attempt to provide a comprehensive, inclusive, and integrated approach to supporting the needs of siblings of children with MHPs.

Secondly, I conducted a primary research study in collaboration with CAMHS. Importantly, this research has highlighted the significant and direct impact that these siblings can have on clinical practice and mental health service provision. This relationship has been overlooked in research and may underlie the limited recognition of and support provided for siblings of children with MHPs. Furthermore, this research study explored potential predictors of MHPs in siblings, a key gap in the existing literature particularly in terms of birth order theory and age difference.

Thus, the body of work presented in this thesis has systematically and comprehensively reviewed the past literature, contributed to the current research base in a manner that extends past research, and has provided clear and direct guidelines for future research.

10.9 Final Thoughts: Siblings of Children with Mental Health Problems – the Unseen Amongst the Already Overlooked

To conclude this thesis, there is an important point about research with siblings that I would like to emphasise. Siblings, as a group, are too commonly unrecognised in our considerations of the role of family in determining our mental health and wellbeing. However, I found that siblings of children with MHPs are further overlooked in our

consideration of siblings. Extensive research has been conducted with twin siblings. There are large-scale projects and considerable funding resources devoted to these siblings. This is certainly understandable given the advancements that can be made in genetics and family environment research. Non-twin siblings receive much less attention despite the fact that they represent the majority proportion of types of siblings in the general population and are also exposed to risk factors for the development of MHPs (see Chapter 3). Siblings of children with special needs have similarly received considerable attention in terms of research and funding. While there is no doubt that this population is in need of research and attention, siblings of children with MHPs experience similar difficulties and, perhaps, are at even greater risk of poor mental health and wellbeing. For example, effect sizes for differences between psychological outcomes for siblings of children with special needs and control siblings are typically in the small range (see Chapter 1) while those for siblings of children with MHPs are in the moderate to large range. Yet our efforts to understand and support siblings of children with MHPs are far behind that of siblings of children with special needs. I would hope in the future, with increasing awareness, that siblings of children with MHPs can receive similar opportunities for understanding and support. I hope that this thesis can contribute to this goal and shine a light on these siblings that are too often forgotten.

References

Achenbach, T.M. (1991). *Manual for the Child Behaviour Checklist/4-18 and 1991 profile.* Vermont, USA: University of Vermont, Department of Psychiatry.

Achenbach, T.M., & Rescorla, L.A. (2001). *Manual for the ASEBA School-Age forms & profiles.* Vermont, USA: University of Vermont, Research Center for Children, Youth, & Families.

Alderfer, M.A., Long, K.A., Lown, E.A., Marsland, A.L., Ostrowski, N.L., Hock, J.M., & Ewing, L.J. (2010). Psychosocial adjustment of siblings of children with cancer: A systematic review. *Psycho-Oncology, 18,* 789-805. doi: 10.1002/pon.1638

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). Virginia, USA: American Psychiatric Publishing.

Angermeyer, M.C., & Dietrich, S. (2006). Public beliefs about and attitudes towards people with mental illness: A review of population studies. *Acta Pscyhiatrica Scandinavica, 113,* 163-179. doi: 10.1111/j.1600-0447.2005.00699.x

Austin, J.K. (1993). Families with chronically ill children. In C.S. Fawcett (Ed.). *Family psychiatric nursing.* Missouri, USA: C.V. Mosby Co.

Australian Bureau of Statistics. (2008). *National Survey of Mental Health and Wellbeing: Summary of results, 2007.* Canberra, Australia: Australian Bureau of Statistics.

Australian Institute of Health and Welfare. (2009). *Measuring the social and emotional wellbeing of Aboriginal and Torres Strait Islander peoples.* Canberra, Australia: Australian Institute of Health and Welfare.

Bamberg, J.H., Toumbourou, J.W., & Marks, R. (2008). Including the siblings of youth substance abusers in a parent-focused intervention: A pilot test of the Best Plus Program. *Journal of Psychoactive Drugs, 40,* 281-291. doi: 10.1080/02791072.2008.10400643

Barrett, P.M., Farrell, L.J., Ollendick, T.H., & Dadds, M. (2006). Long-term outcomes of an Australian universal prevention trial of anxiety and depression symptoms in children and youth: An evaluation of the friends program. *Journal of Clinical Child and Adolescent Psychology, 35,* 403-411. doi: 10.1207/s15374424jccp3503_5

Beilby, J.M., Byrnes, M.L., & Young, K.N. (2012). The experiences of living with a sibling who stutters: A preliminary study. *Journal of Fluency Disorders, 37,* 135-148. doi: 10.1016/jfludis.2012.02.002

Bernsen, R.M.D., de Jongste, J.C., & van der Wouden, J.C. (2003). Birth order and sibship size as independent risk factors for asthma, allergy, and eczema. *Pediatric Allergy and Immunology, 14,* 464-469. doi: 10.1046/j.0905-6157.2003.00108.x

Bird, H.R., Gould, M.S., & Staghezza, B.M. (1993). Patterns of diagnostic comorbidity in a community sample of children aged 9 through 17 years. *Journal of the American Academy of Child & Adolescent Psychiatry, 32,* 361-368. doi: 10.1097/00004583-199303000-00018

Bowen, M. (1978). *Family therapy in clinical practice.* New York, USA: Jason Aronson.

Breslau, N. (1982). Siblings of disabled children: Birth order and age-spacing effects. *Journal of Abnormal Child Psychology, 10,* 85-95. doi: 10.1007/BF00915953

Bristow, K., & Patten, S. (2002). Treatment-seeking rates and associated mediating factors among individuals with depression. *Canadian Journal of Psychiatry, 47,* 660-665.

Brody, G.H. (1998). Sibling relationship quality: Its causes and consequences. *Annual Review Psychology, 49,* 1-24. doi:10.1146/annurev.psych.49.1.1

Bronfenbrenner, U. (1979). *The ecology of human development.* Massachusetts, USA: Harvard University Press.

Brotman, L.M., Gouley, K.K., Chesir-Teran, D., Dennis, T., Klein, R.G., & Shrout, P. (2005). Prevention for pre-schoolers at high risk for conduct problems: Immediate outcomes on parenting practices and child social competence. *Journal of Clinical Child and Adolescent Psychology, 34,* 724-734. doi: 10.1207/s15374424jccp3404_14

Brotman, L.M., Gouley, K.K., O'Neal, C., & Klein, R.G. (2004). Preschool-aged siblings of adjudicated youths: Multiple risk factors for conduct problems. *Early Education and Development, 15,* 387-406.

Buist, K.L. (2010). Sibling relationship quality and adolescent delinquency: A latent growth curve approach. *Journal of Family Psychology, 24,* 400-410. doi:10.1037/a0020351

Bullock, B.M., & Dishion, T.J. (2002). Sibling collusion and problem behaviour in early adolescence: Toward a process model for family mutuality. *Journal of Abnormal Child Psychology, 30,* 143-153. doi: 10.1023/A:1014753232153

Cardino, A.G., Marshall, E.J., Cold, B., Macdonald, A.M., Ribchester, T.R., Davies, N.J.,…Murray, R.M. (1999). Heritability estimates for psychotic disorders: The Maudsley twin psychosis series. *Archives of General Psychiatry, 56,* 162-168. doi:10.1001/archpsyc.56.2.162

Castorina, L.L., & Negri, L.M. (2011). The inclusion of siblings in social skills training groups for boys with Asperger syndrome. *Journal of Autism & Developmental Disorders, 41,* 73-81. doi: 10.1007/s10803-010-1023.x

Chan, R.C.K., Xu, T., Heinrichs, R.W., Yu, Y., & Gong, Q.Y. (2010). Neurological soft signs in non-psychotic first-degree relatives of patients with schizophrenia: A systematic review and meta-analysis. *Neuroscience & Biobehavioral Reviews, 34,* 889-896. doi: 10.1016/j.neubiorev.2009.11.012

Cicirelli, V.G. (1982). Sibling influence throughout the lifespan. In M.E. Lamb & B. Sutton-Smith (Eds). *Sibling relationships: Their nature and significance across the lifespan*. New York: Lawreunce Erlbaum Associates, Inc.

Cohen, J. (1988). *Statistical power analysis for the behavioral sciences* (2nd ed.). New Jersey, USA: Erlbaum.

Cohen, J. (1992). A power primer. *Psychological Bulletin, 112*, 155-159. doi:10.1037/0033-2909.11.1.155

Dauz-Williams, P., Piamjariyakui, U., Graff, J.C., & Stanton, A. (2010). Developmental disabilities: Effects on well siblings. *Issues in Comprehensive Pediatric Nursing, 33,* 39-55. doi: 10.3109/01460860903486515

Davey Smith, G., & Egger, M. (2001). Going beyond the grand mean: Subgroup analysis in meta-analysis of randomised trials. In: M. Egger, G. Davey Smith, & D.G. Altman (Eds.). *Systematic reviews in health care: Meta-Analysis in context* (2nd ed.). London: BMJ.

Department of Health, Government of South Australia. (2010). *South Australia's Mental Health and Wellbeing Policy, 2010-2015.* South Australia, Australia: Department of Health, Government of South Australia.

DiLalla, L.F., Millineaux, P.Y., & Elam, K.K. (2008). Twins. In: M.M. Haith & J.B. Benson (Eds.). *Encyclopedia of infant and early childhood development.* San Diego, USA: Academic Press.

Dowling, M., & Dolan, L. (2001). Families with children with disabilities – Inequalities and the social model. *Disability & Society, 16,* 21-35. doi: 10.1080/713662027

Dunn, J. (1983). Sibling relationships in early childhood. *Child Development, 54,* 787-811. http://www.jstor.org/stable/1129886

Dunn, J., Slomkowski, C., Beardsall, L., & Rende, R. (1994). Adjustment in middle childhood and early adolescence: Links with earlier and contemporary sibling relationships. *Journal of Child Psychology and Psychiatry, 35,* 491-504. doi: 10.1111/j.1469-7610.1994.tb01736.x

Eckstein, D. (2000). Empirical studies indicating significant birth-order related personality differences. *Journal of Individual Psychology, 56,* 481-494.

Ehringer, M.A., Rhee, S.H., Young, S., Corley, R., & Hewitt, J.K. (2006). Genetic and environmental contributions to common psychopathologies of childhood and adolescence: A study of twins and their siblings. *Journal of Abnormal Child Psychology, 34,* 1-17. doi:10.1007/s10802-005-9000-0

Erel, O., & Burman, B. (1995). Interrelatedness of marital relations and parent-child relations: A meta-analytic review. *Psychological Bulletin, 118,* 108-132. doi: 10.1037/0033-2909.118.1.108

Fagan, A.A., & Najman, J.M. (2003). Association between early childhood aggression and internalizing behavior for sibling pairs. *Journal of the American Academy of Child & Adolescent Psychiatric, 42,* 1093-1100. doi:10.1097/01.CHI.0000070240.24125.EA

Farrell, L., Barrett, P., & Piacentini, J. (2006). Obsessive-compulsive disorder across the developmental trajectory: Clinical correlates in children, adolescents, and adults. *Behaviour Change, 23,* 103-120.

Farrell, P. (1994). *Children with emotional and behavioural difficulties: Strategies for assessment and intervention.* London: Falmer Press.

Feinberg, M.E., Solmeyer, A.R., & McHale, S.M. (2012). The third rail of family systems: Sibling relationships, mental and behavioural health, and preventive intervention in childhood and adolescence. *Clinical Child and Family Psychology Review, 15,* 43-57. doi: 10.1007/s10567-011-0104-5

Feinberg, M.E., Solmeyer, A.R., Hostetler, M.L., Sakuma, K.L., Jones, D., & McHale, S.M. (2013). Siblings are special: Initial test of a new approach for preventing youth behaviour problems. *Journal of Adolescent Health, 53,* 166-173. doi: 10.1016/j.jadohealth.2012.10.004

Furber, G., Segal, L., Leach, M., & Cocks, J. (2013). Mapping scores from the Strengths and Difficulties Questionnaire (SDQ) to preference-based utility values. *Quality of Life Research, 23,* 403-411. doi: 10.1007/s11136-013-0494-6

Garley, D., Gallop, R., Johnston, N., & Pipitone, J. (1997). Children of the mentally ill: A qualitative focus group approach. *Journal of Psychiatric Mental Health Nursing, 4,* 97-103. doi: 10.1046/j.1365-2850.1997.00036.x

Gates, L., Lineberger, M.R., Crockett, J., & Hubbard, J. (1988). Birth order and its relationship to depression, anxiety, and self-concept test scores in children. *The Journal of Genetic Psychology: Research and Theory on Human Development, 149,* 29-34. doi: 10.1080/00221325.1988.10532136

Green, H., McGinnity, A., Meltzer, H., Ford, T., & Goodman, R. (2005). *Mental health of children and young people in Great Britain, 2004*. Office for National Statistics, Department of Health and Scottish Executive.

Green, M.T., Clopton, J.R., & Pope, A.W. (1996). Understanding gender differences in referral of children to mental health services. *Journal of Emotional and Behavioral Disorders, 4,* 182-190. doi: 10.1177/106342669600400305

Greenberg, M.T., Domitrovich, C., & Bumbarger, B. (1999). *Preventing mental disorders in school-age children: A review of the effectiveness of prevention programs.* Prevention Research Center for the Promotion of Human Development, College of Health and Human Development, Pennsylvania State University.

Greene, R.W. (1998). *The explosive child.* New York, USA: HarperCollins Publishers.

Griffiths, A.J.F., Miller, J.H., Suzuki, D.T., Lewontin, R.C., & Gelbart, W.M. (2000). *An introduction to genetic analysis* (7th ed.). New York: W.H. Freeman.

Griffiths, C., & Sin, J. (2013). Rethinking siblings and mental illness. *The Psychologist, 26,* 808-810.

Harrison, P.J., & Weinberger, D.R. (2005). Schizophrenia genes, gene expression, and neuropathology: On the matter of their convergence. *Molecular Psychiatry, 10,* 40-68. doi:10.1038/sj.mp.4001558

Helzer, J.D., Chmura Kraemer, H., Krueger, R.F., Wittchen, H.U., Sirovatka, P.J., Regier, D.A. (Eds.). (2008). *Dimensional approaches in diagnostic classification: Refining the research agenda for DSM-V.* Arlington, V.A.; American Psychiatric Publishing.

Hettema, J.M., Neale, M.C., & Kendler, K.S. (2001a). A review and meta-analysis of the genetic epidemiology of anxiety disorders. *American Journal of Psychiatry, 158,* 1568-1578. doi: 10.1176/appi.ajp.158.10.1568

Hettema, J.M., Prescott, C.A., & Kendler, K.S. (2001b). A population-based twin study of generalised anxiety disorder in men and women. *Journal of Nervous & Mental Disease, 189,* 413-420.

Higgins, J. P. T., & Green, S. (Eds.). (2011). *Cochrane handbook for systematic reviews of interventions* (Version 5.1.0): The Cochrane Collaboration.

Hoffman, L. (1981). *Foundations of family therapy: A conceptual framework for systems change.* New York, USA: Basic Books.

Hudson, J.L., & Rapee, R.M. (2002). Parent-child interactions in clinically anxious children and their siblings. *Journal of Clinical Child and Adolescent Psychology, 31,* 548-555. doi: 10.1207/153744202320802214

Kempton, T., Armistead, L., Wierson, M., & Forehand, R. (1991). Presence of a sibling as a potential buffer following parental divorce: An examination of young adolescents. *Journal of Clinical Child Psychology, 20,* 434-438. doi:10.1207/s15374424jccp2004_12

Kendler, K.S. (2001). Twin studies of psychiatric illness: An update. *Archives of General Psychiatry, 58,* 1005-1014. doi:10.1001/archpsyc.58.11.1005

Kendler, K.S., & Eaves, L.J. (1986). Models for the joint effect of genotype and environment on liability to psychiatric illness. *American Journal of Psychiatry, 143,* 279-289.

Kendler, K.S., Davis, C.G., & Kessler, R.C. (1997). The familial aggregation of common psychiatric and substance use disorders in the National Comorbidity

Survey: A family history study. *The British Journal of Psychiatry, 170,* 541-548. doi: 10.1192/bjp.170.6.541

Kessler, R.C., McLaughlin, K.A., Greif Green, J., Gruber, M.J., Sampson, N.A., Zaslavsky, A.M.,…Williams, D.R. (2010). Childhood adversities and adult psychopathology in the WHO World Mental Health Surveys. *The British Journal of Psychiatry, 197,* 378-385. doi:10.1192/bip.bp.110.080499

Kinsella, K.B., Anderson, R.A. & Anderson, W.T. (1996). Coping skills, strengths, and needs as perceived by adult offspring and siblings of people with mental illness: A retrospective study. *Psychiatric Rehabilitation Journal, 20,* 24-32. doi: 10.1037/h0095388

Kristensen, P., & Bjerkedal, T. (2007). Explaining the relation between birth order and intelligence. *Science, 316,* 1717. doi: 10.1126/science.1141493

Lombroso, P.J., Pauls, D.L., & Leckman, J.F. (1994). Genetic mechanisms in childhood psychiatric disorders. *Journal of the American Academy of Child and Adolescent Psychiatry, 39,* 921-938.

Manuel, J.C. (2001). Risk and resistance factors in the adaptation in mothers of children with juvenile rheumatoid arthritis. *Journal of Pediatric Psychology, 26,* 237-246. doi: 10.1093/jpepsy/26.4.237

McLaughlin, K.A., Gadermann, A.M., Hwang, I., Sampson, N.A., Al-Hamzawi, A., Andrade, L.H.,…Kessler, R.C. (2012). Parent psychopathology and offspring mental disorders: Results from the WHO World Mental Health Surveys. *The British Journal of Psychiatry, 200,* 290-299. doi: 10.1192/bjp.bp.111.101253

Merikangas, K.R., He, J.P., Burstein, M., Swanson, S.A., Avenevoli, S., Cui, L.,…Swendsen, J. (2010). Lifetime prevalence of mental disorders in U.S. adolescents: Results from the National Comorbidity Survey Replication-

Adolescent Supplement (NCS-A). *Journal of the American Academy of Child and Adolescent Psychiatry, 49,* 980-989. doi:10.1016/jaac.2010.05.017

Milevsky, A. (2005). Compensatory patterns of sibling support in emerging adulthood: Variations in loneliness, self-esteem, depression, and life satisfaction. *Journal of Social and Personal Relationships, 22,* 743-755. doi:10.1177/0265407505056447

Milevsky, A., & Levitt, M.J. (2005). Sibling support in early adolescence: Buffering and compensation across relationships. *European Journal of Developmental Psychology, 2,* 299-320. doi:10.1080/1740562054000048

Miller, R.B., Anderson, S., Keals, D.K. (2004). Is Bowen theory valid? A review of basic research. *Journal of Marital and Family Therapy, 30,* 453-466. doi: 10.1111/j.1752-0606.2004.tb01255.x

New South Wales Ministry of Health [NSW]. (2011). *NSW Child and Adolescent Mental Health Services (CAMHS) competency framework.* NSW, Australia: NSW Ministry of Health.

Noland, V., Liller, K., McDermott, R., Coulter, M., & Seraphine, A.E. (2004). Is adolescent sibling violence a precursor to college dating violence? *American Journal of Health Behavior, 28,* S13-S23.

Parritz, R.H., & Troy, M.F. (2011). *Disorders of childhood: Development and psychopathology* (2nd ed.). USA: Wadsworth, Cengage Learning.

Patterson, G.R. (1986). Siblings: Fellow travellers in the coercive family process. In R.J. Blanchard & D.C. Blanchard (Eds.). *Advances in the Study of Aggression*. New York: USA: Academic Press.

Petticrew, M., & Roberts, H. (2006). *Systematic reviews in the social sciences: A practical guide*. UK: Blackwell Publishing.

Pike, A., & Plomin, R. (1996). Importance of nonshared environmental factors for childhood and adolescent psychopathology. *Journal of the American Academy of Child and Adolescent Psychiatry, 35,* 560-570.

Prchal, A., & Landolt, M.A. (2009). Psychological interventions with siblings of pediatric cancer patients: A systematic review. *Psycho-Oncology, 18,* 1241-1251. doi: 10.1002/pon.1565

Ravens-Sieberer, U., Wille, N., Erhart, M., Bettge, S., Wittchen, J., Rothenberger, A.,…Dopfner, M. (2008). Prevalence of mental health problems among children and adolescents in Germany: Results of the BELLA study within the National Health Interview and Examination Survey. *European Child and Adolescent Psychiatry, 17,* 22-23. doi:10.1007/s00787-008-1003-2

Rosenzweig, J.M., Brennan, E.M., & Ogilvie, A.M. (2002). Work-family fit: Voices of parents of children with emotional and behavioural disorders. *Social Work, 47,* 415-424. doi: 10.1093/sw/47.4.415

Rowe, J. (1992). In support of sibling inclusion: A literature review. *Journal of Child & Adolescent Psychiatric Mental Health Nursing, 5,* 27-33. doi: 10.1111/j.1744-6171.1992.tb00125.x

Rutter, M. (2011). Research review: Child psychiatric diagnosis and classification: Concepts, findings, challenges, and potential. *Journal of Child Psychology and Psychiatry, 52,* 647-660. doi:10.1111/j.1469-7610.2011.02367.x

Rutter, M., & Redshaw, J. (1991). Annotation: Growing up as a twin: Twin-singleton differences in psychological development. *Journal of Child Psychology and Psychiatry, 32,* 885-895. doi:10.1111/j.1469-7610.1991.tb01916.x

Sandelowski, M. & Barroso, J. (2007). *Handbook for synthesising qualitative research.* New York: Springer Publishing Company, Inc.

Sanson, A., Nicholson, J., Ungerer, J., Zubrick, S., Wilson, K., Ainley, J.,…Wake, M. (2002). *LSAC discussion paper No. 1: Introducing the Longitudinal Study of Australian Children.* Melbourne, Australia: Australian Institute of Family Studies.

Sawyer, M.G., Arney, F.M., Baghurst, P.S., Clark, J.J., Graetz, B.W., Kosky, R.J.,…Zubrick, S.R. *The mental health of young people in Australia.* Australia: Mental Health and Special Programs Branch, Commonwealth Department of Health and Aged Care, Australia.

Sharpe, D., & Rossiter, L. (2002). Siblings of children with a chronic illness: A meta-analysis. *Journal of Pediatric Psychology, 27,* 699-710. doi: 10.1093/jpepsy/27.8.699

Simons, J.S., Dvorak, R.D., & Lau-Barraco, C. (2009). Behavioural inhibition and activation systems: Differences in substance use expectancy organization and activation in memory. *Psychology of Addictive Behaviours, 23,* 315-328. doi: 10.1037/a0015834

Sitskoom, M.M., Aleman, A., Ebisch, S.J.H., Appels, M.C.M., & Kahn, R.S. (2004). Cognitive deficits in relatives of patients with schizophrenia: A meta-analysis. *Schizophrenia Research, 71,* 285-295. doi: 10.1016/j.schres.2004.03.007

Smith, M. J., Barch, D. M., Wolf, T. J., Mamah, D., & Csernansky, J. G. (2008). Elevated rates of substance use disorders in non-psychotic siblings of individuals with schizophrenia. *Schizophrenia Research, 106*, 294-299. doi:10.1016/j.schres.2008.07.015

Stalker, K., & Connors, C. (2004). Children's perceptions of their disabled siblings: 'she's different but it's normal for us'. *Children & Society, 18,* 218-230. doi: 10.1002/chi.794

Stice, E., Shaw, H., Bohon, C., Marti, C.N., & Rohde, P. (2009). A meta-analytic review of depression prevention programs for children and adolescents: Factors that predict magnitude of intervention effects. *Journal of Consulting and Clinical Psychology, 77,* 486-503. doi: 10.1037/a0015168

Sullivan, G.M., & Feinn, R. (2012). Using effect size – or why the p value is not enough. *Journal of Graduate Medical Education, 4,* 279-282. doi: 10.4300/JGME-D-12-00156.1

Sulloway, F.J. (1995). Birth order and evolutionary psychology: A meta-analytic overview. *Psychological Inquiry, 6,* 75-80. doi: 10.1207/s15327965pli0601_15

Summers, C.R., White, K.R., & Summer, M. (1994). Siblings of children with a disability: A review and analysis of the empirical literature. *Journal of Social Behavior & Personality, 9,* 169-184.

Svihra, M., & Katzman, M.A. (2004). Behavioural inhibition: A predictor of anxiety. *Paediatric Child Health, 9,* 547-550.

Szatmari, P., Boyle, M.H., & Offord, D.R. (1993). Familial aggregation of emotional and behavioural problems of childhood in the general population. *American Journal of Psychiatry, 150,* 1398-1403.

Tancredy, C.M., & Fraley, R.C. (2006). The nature of adult twin relationships: An attachment-theoretical perspective. *Journal of Personality and Social Psychology, 90,* 78-93. doi:10.1037/0022-3514.90.1.78

Thorpe, K., Golding, J., MacGillivray, I., & Greenwood, R. (1991). Comparison of prevalence of depression in mothers of twins and mothers of singletons. *BMJ, 302,* 875-878.

Tienari, P., Wynne, L.C., Sorri, A., Lahti, I., Läksy, K., Moring, J., …Wahlberg, K.E. (2004). Genotype-environment interaction in schizophrenia-spectrum disorder:

Long term follow-up study of Finnish adoptees. *The British Journal of Psychiatry, 184,* 216-222. doi:10.1192/bjp.184.3.216

United Nations. (1989). *Convention on the rights of the child.* Retrieved 8 Nov 2012 from http://treaties.un.org

Valentine, J.C., Pigott, T.D., & Rothstein, H.R. (2010). How many studies do you need? A primer on statistical power for meta-analysis. *Journal of Educational and Behavioral Statistics, 35,* 215-247. doi: 10.3102/1076998609346961

Verhulst, F.C., & van der Ende, J. (1997). Factors associated with child mental health service use in the community. *Journal of the American Academy of Child & Adolescent Psychiatry, 36,* 901-909. doi: 10.1097/00004583-199707000-00011

Vermaes, I.P., van Susante, A.M., van Bakel, H.J. (2012). Psychological functioning of siblings in families of children with chronic health conditions: A meta-analysis. *Journal of Pediatric Psychology, 37,* 166-184. doi: 10.1093/pepsy/jsr081

Volling, B.L., McElwain, N.L., & Miller, A.L. (2002). Emotion regulation in context: The jealousy complex between young siblings and its relations with child and family characteristics. *Child Development, 73,* 581-600. doi: 10.1111/1467-8624.00425

Waldinger, R.J., Vaillant, G.E., & Orav, J. (2007). Childhood sibling relationships as a predictor of major depression in adulthood: A 30-year prospective study. *American Journal of Psychiatry, 164,* 949-954. doi: 10.1176/appi.ajp.164.6.949

Wallander, J.L., & Varni, J.W. (1992). Adjustment in children with chronic physical disorders: Programmatic research on a disability-stress-coping model. In A.M. LaGreca, L. Siegal, J.L. Wallander, & C.E., Walker (Eds.). *Stress and coping in child health.* New York, USA: Guildford Press.

Wallander, J.L., Varni, J.W., Babani, L.V., Banis, H.T., Wilcox, K.T. (1989). Family resources as resistance factors for psychological maladjustment in chronically ill and handicapped children. *Journal of Pediatric Psychology, 14,* 157-173.

Wasserman, G., Miller, L.S., Pinner, E., & Jaramillo, B. (1996). Parenting predictors of early conduct problems in urban, high-risk boys. *Journal of the American Academy of Child & Adolescent Psychiatry, 35,* 1227-1236. doi: 10.1098/00004583-199609000-00020

Yeh, M., & Weisz, J.R. (2001). Why are we here at the clinic? Parent-child (dis)agreement on referral problems at outpatient treatment entry. *Journal of Consulting and Clinical Psychology, 69,* 1018-1025. doi: 10.1037/0022-006X.69.6.1018

Appendix A

Search Keyword List and Search Strategy

Sibling	Child	Mental health problem			Exclusions
• Brother*	• Adolescen*	• Acute stress disorder*	• Emotional adjustment	• Psychiatric difficult*	• Aspergers
• First-degree relative*	• Child*	• Addiction*	• Emotional difficult*	• Psychiatric disorder*	• Autis*
• Sibling relation*	• Young	• Adjustment disorder*	• Emotional disturb*	• Psychiatric illness*	• Brain injur*
• Sibling*	• Youth*	• Affective disorder*	• Emotional problem*	• Psychiatric problem*	• Cancer
• Sister*		• Aggressive*	• Gambling	• Psychiatric symptom*	• Cystic fibrosis
		• Anorexia nervosa	• Gender identity disorder*	• Psychological diagnos*	• Deaf
		• Anxi*	• Generalised anxiety disorder*	• Psychological difficult*	• Developmental disab*
		• Anxiety disorder*	• Hyperactiv*	• Psychological disorder*	• Developmental disorder*
		• Attention deficit*	• Impulse control disorder*	• Psychological illness	• Epilep*
		• Behav*	• Major depressi*	• Psychological problem*	• Intellectual disab*
		• Behav* Difficult*	• Mania	• Psychological symptom*	• Mental retard*
		• Behav* problem*	• Mental disorder*	• Psychopatholog*	• Physical disorder*
		• Behaviour disorder*	• Mental health	• Psychosis	• 1990-July 2011
		• Bipolar disorder*	• Mental illness*	• Risk taking	• English
		• Bulimia	• Mentally ill*	• Schizo*	
		• Conduct disorder*		• Seasonal affective disorder*	

Appendix A (Cont.)

Search Keyword List and Search Strategy

Sibling	Child	Mental health problem			Exclusions
		• Delinquen*	• Neurosis	• Self mutilation	• Peer-review journal articles
		• Depress*	• Obsession*	• Separation anxiety	• Exclude dissertations
		• Depressive disorder*	• Obsessive compulsive disorder*	• Social adjustment	
		• Dissociative disorder*		• Social behaviour	
		• Dissociative identity disorder*	• Panic disorder*	• Social difficult*	
			• Personality disorder*	• Social problem*	
		• Drug abuse	• Phobia*	• Social skills	
		• Eating disorder*	• Posttraumatic Stress Disorder*	• Social*	
		• Emotion*		• Substance-related disorder*	
		• Emotional	• Psychiatric	• Thought disturb*	
			• Psychiatric diagnos*	• Wellbeing	

Note. The terms used differed slightly across electronic databases due to differences in search language and syntax across the databases. *Wildcard syntax (includes articles including the term with any letters or words following the asterisk).

Appendix B

Data Extraction: Prevalence of Psychopathology

Reference	Sibling diagnosis measured		Sibling prevalence	Control prevalence	OR [95% CI]
Siblings of children with ADHD					
Biederman et al. (1990a)	**ADHD**	Pre-baseline morbidity risk	20.8%	5.6%	(-)**
Faraone et al. (1996)		Baseline Lifetime	18.0%	5.0%	4.62 [1.87, 11.42]**
Faraone et al. (2000)		4-year follow-up	26.0%	10.0%	3.24 [1.69, 6.21]**
		Baseline	29.4%	13.7%	1.87 [1.02, 3.42]*
Mikami & Pfiffner (2008)		Lifetime	14.3%	0.0%	5.02 [0.28, 90.05]
Hassan et al. (2011)		Lifetime	11.25%	1.5%[3]	
Hebrani & Behdani (2007)		Lifetime	21.3%	5.29%[4]	
Kollins et al. (2009)		1-year	44.1%	0.41%[1]	
Christiansen et al. (2008)		Lifetime	29.45%	5.29%[4]	
Biederman et al. (1990b)	**CD/ODD**	Pre-baseline CD/ODD	25.8%	3.0%	11.13 [1.30, 95.25]*
Faraone et al. (1996)		Baseline: CD	7.0%	3.0%	2.52 [0.80, 7.93]
		ODD	18.0%	9.0%	2.42 [1.17, 5.00]*
		4-year follow-up: CD	11.0%	4.0%	2.72 [1.05, 7.06]*
		ODD	30.0%	12.0%	3.11 [1.70, 5.70]**
Faraone et al. (2000)		Baseline: CD	7.0%	3.8%	1.89 [0.63, 5.70]
		ODD	16.8%	9.2%	2.00 [0.96, 4.18]
Christiansen et al. (2008)		1-year: CD/ODD	2.7%	0.13-0.94%[1]	
Sobanski et al. (2010)		1-year: ODD	15.48%	0.94%[1]	

Appendix B (Cont.)

Data Extraction: Prevalence of Psychopathology

Reference	Sibling diagnosis measured		Sibling prevalence	Control prevalence	OR [95% CI]
Siblings of children with ADHD					
Faraone et al. (1996)	**SUD**	Baseline: Any SUD	9.0%	11.0%	0.77 [0.36, 1.67]
		Alcohol abuse	3.0%	5.0%	0.73 [0.23, 2.32]
		Alcohol dependence	5.0%	5.0%	0.84 [0.30, 2.38]
		Drug abuse	1.0%	2.0%	0.49 [0.08, 2.97]
		Drug dependence	4.0%	4.0%	1.04 [0.32, 3.35]
		4-year follow-up: Any SUD			
		Alcohol abuse	17.0%	16.0%	1.03 [0.57, 1.89]
		Alcohol dependence	11.0%	14.0%	1.68 [0.76, 3.75]
		Drug abuse	7.0%	7.0%	1.02 [0.43, 2.43]
		Drug dependence	5.0%	4.0%	1.55 [0.51, 4.74]
		4-year follow-up:	5.0%	5.0%	1.09 [0.40, 3.01]
Milberger et al. (1997)		Psychoactive SUD	17.0%	16.0%	1.06 [0.56, 2.03]
Biederman et al. (1991)	**Affective Disorder**	Pre-baseline: Any AFF	7.1%	0.0%	5.88 [0.33, 104.5]
		DEP	6.2%	0.0%	5.14 [0.29, 92.26]
		Bipolar Disorder	0.9%	0.0%	0.97 [0.04, 24.42]
		Dysthymia	0.9%	0.0%	0.97 [0.04, 24.42]
Faraone et al. (1996)		Baseline: DEP	10.0%	5.0%	2.01 [0.81, 4.97]
		Bipolar Disorder	3.0%	2.0%	2.27 [0.45, 11.42]
		Dysthymia	1.0%	3.0%	0.36 [0.07, 2.02]
		4-year follow-up: DEP	17.0%	8.0%	2.26 [1.11, 4.62]*
		Bipolar Disorder	9.0%	4.0%	2.69 [0.95, 7.59]
		Dysthymia	4.0%	6.0%	0.62 [0.21, 1.83]
Hassan et al. (2011)		DEP	0.5%	0.9%[3]	
		Bipolar disorder	0.0%	0.9%[3]	

Appendix B (Cont.)

Data Extraction: Prevalence of Psychopathology

Reference	Sibling diagnosis measured		Sibling prevalence	Control prevalence	OR [95% CI]
Siblings of children with ADHD					
Geller et al. (2006)	**Affective Disorders**	Morbid risk:			Hazard ratio:
		Bipolar disorder	7.3%	3.0%	1.9 [0.3, 11.3]
		Recurrent DEP	25.3%	12.9%	2.1 [0.6, 8.0]
		Bipolar disorder/Recurrent DEP	34.3%	15.7%	2.2 [0.8, 6.4]
Faraone et al. (1996)	**Anxiety Disorder**	Baseline: Two or more ANX	16.0%	9.0%	1.97 [0.94, 4.14]
		Panic disorder	1.0%	1.0%	0.74 [0.05, 11.94]
		Agoraphobia	3.0%	5.0%	0.73 [0.23, 2.32]
		Overanxious disorder	16.0%	13.0%	1.26 [0.66, 2.42]
		Simple phobia	10.0%	9.0%	1.13 [0.52, 2.43]
		Social phobia	13.0%	4.0%	3.59 [1.32, 9.75]*
		Separation anxiety	13.0%	3.0%	4.52 [1.52, 13.47]*
		GAD	2.0%	2.0%	1.11 [0.18, 6.77]
		OCD	1.0%	0.0%	2.24 [0.09, 55.42]
		4-year follow-up: Two or more ANX	23.0%	10.0%	2.56 [1.35, 4.87]**
		Panic disorder	3.0%	1.0%	2.15 [0.41, 11.25]
		Agoraphobia	12.0%	10.0%	1.24 [0.60, 2.55]
		Overanxious disorder	24.0%	15.0%	1.71 [0.95, 3.03]
		Simple phobia	20.0%	15.0%	1.41 [0.77, 2.57]
		Social phobia	17.0%	8.0%	2.38 [1.14, 4.98]*
		Separation anxiety	17.0%	6.0%	3.35 [1.48, 7.61]**
		GAD	3.0%	3.0%	1.06 [0.28, 4.02]
		OCD	1.0%	3.0%	0.21 [0.02, 1.87]

Appendix B (Cont.)

Data Extraction: Prevalence of Psychopathology

Reference	Sibling diagnosis measured		Sibling prevalence	Control prevalence	OR [95% CI]
Siblings of children with ADHD					
Last et al. (1991)	**Anxiety Disorders**	ANX	8.3%	10.8%	0.78 [0.19, 3.22]
Siblings of children with CD/ODD					
Hovens et al. (1994)		% of target children with sibling with: ADHD	7.0%	7.7%[2]	
		CD/ODD	50.0%	19.6%[5]	
		DEP	7.0%	10.1%[5,6]	
		Other emotional difficulties	21.0%	14.3%[5,6]	
		SUD	28.0%	11.4%[5]	
Hill et al. (2002)		Lifetime alcohol dependence	77.78%	6.4%[5]	
Siblings of children with SUDs					
Hovens et al. (1994)		% of target children with sibling with: ADHD	10.0%	7.7%[2]	
		CD/ODD	36.0%	19.6%[5]	
		SUD	24.0%	11.4%[5]	
		DEP	10.0%	10.1%[5,6]	
		Other emotional difficulties	6.0%	14.3%[5,6]	

Appendix B (Cont.)

Data Extraction: Prevalence of Psychopathology

Reference	Sibling diagnosis measured		Sibling prevalence	Control prevalence	OR [95% CI]
Siblings of children with Delinquency					
Wasserman et al. (1996)		Baseline 1-year: ADHD	21.43%	0.41%[1]	
		ODD	11.90%	1.47%[1]	
		CD	3.17%	0.85%[1]	
		Affective Disorder	2.0%	1.95%[1]	
		Anxiety Disorder	31.0%	1.47%[1]	
Pine et al. (1998)		1-2 year follow-up: ADHD	11.59%	0.41%[1]	
		ODD	8.7%	1.47%[1]	
		CD	2.9%	0.85%[1]	
		Affective Disorder	1.4%	1.95%[1]	
		Simple Phobia	23.19%	0.73%[1]	
		Separation Anxiety	5.8%	0.0%[1]	
		Social Phobia	7.25%	0.24%[1]	
		Agoraphobia	2.9%	0.12%[1]	
Siblings of children with Affective Disorders					
Kelvin et al. (1996)	**ADHD**	Lifetime	13.0%	1.5%[3]	
Kelvin et al. (1996)	**CD/ODD**	1-year: CD	16.0%	1.3%[3]	
		ODD	6.5%	3.0%[3]	
Seguin et al. (2003)	**SUD**	SUD	30.0%	0.0%	6.07 [0.26, 140.7]
Ryan et al. (1992)	**Affective Disorder**	Major affective disorder (DEP/Mania/ Schizoaffective)	24.42%	6.49%	4.65 [1.66, 13.05]**
Kelvin et al. (1996)		1-year: DEP	13.0%	0.9%[3]	
		Dysthymia	6.5%	0.9%[3]	

Appendix B (Cont.)

Data Extraction: Prevalence of Psychopathology

Reference	Sibling diagnosis measured		Sibling prevalence	Control prevalence	OR [95% CI]
Siblings of children with Affective Disorders					
Geller et al. (2006)	**Affective Disorder**	Morbid risk:			Hazard ratio:
		Bipolar disorder	36.7%	3.0%	7.2 [2.1, 24.8]*
		Recurrent DEP	11.8%	12.9%	1.2 [0.4, 3.9]
		Bipolar disorder/Recurrent DEP	48.8%	15.7%	3.5 [1.5, 7.8]*
Seguin et al. (2003)		Mood disorder	60.0%	0.0%	18.78 [0.83, 424.2]
Kelvin et al. (1996)	**Anxiety Disorder**	1-year: Overanxious	6.5%	1.29%[1]	
		Panic disorder	10.0%	0.2%[3]	
		Phobia disorder	6.5%	0.8%[3]	
		Separation anxiety	6.5%	0.4%[3]	
Seguin et al. (2003)		ANX	10.0%	0.0%	2.05 [0.07, 58.66]
		Post traumatic stress	20.0%	0.0%	3.82 [0.16, 94.14]
Siblings of children with Anxiety Disorders					
Lenane et al. (1990)	**ADHD**	Lifetime: ADHD	9.0%	7.7%[2]	
	ODD	ODD	13.0%	12.6%[5]	
	CD	CD	7.0%	6.8%[5]	
Lenane et al. (1990)	**Affective Disorder**	Lifetime: Any AFF	18.0%	14.3%[5,6]	
Dia & Harrington (2006)		Prevalence of professionally diagnosed DEP	4.62%	10.1%[5]	
Farrell et al. (2006)		% of target children with sibling with DEP	3.6%	3.7%[7]	
Last et al. (1991)	**Anxiety Disorder**	ANX	21.7%	10.8%	2.27 [0.89, 5.81]
Reddy et al. (2001)		OCD/Subthreshold OCD	0.0%	0.0%	n/a

Appendix B (Cont.)

Data Extraction: Prevalence of Psychopathology

Reference	Sibling diagnosis measured		Sibling prevalence	Control prevalence	OR [95% CI]
Siblings of children with Anxiety Disorders					
Lenane et al. (1990)	**Anxiety Disorders**	Lifetime: OCD	5.0%	2.5%[8]	
		Separation anxiety	2.0%	7.6%[5]	
		Phobia	2.0%	19.3%[5]	
		Overanxious disorder	0.0%	1.29%[1]	
		Age-corrected: OCD/OCD PD/Subclinical OCD	35.0%	2.5%[8]	
Dia & Harrington (2006)		Separation anxiety	1.5%	7.6%[5]	
		Panic disorder	3.1%	2.3%[5]	
		Generalised anxiety	3.1%	2.2%[5]	
		Simple phobia	1.5%	19.3%[5]	
		Social anxiety	1.5%	9.1%[5]	
		OCD	4.6%	2.5%[8]	
		Post-traumatic stress	0.0%	5.0%[5]	
		Any ANX	12.3%	31.9%[5]	
Do Rosario-Campos et al. (2005)		OCD	14.6%	2.5%[8]	
Farrell et al. (2006)		% of target children with sibling with OCD	5.95%	2.5%[8]	
		ANX	14.29%	13.2%[9]	
Toro et al. (1992)		% of target children with sibling with OCD	5.56%	2.5%[8]	

Appendix B (Cont.)

Data Extraction: Prevalence of Psychopathology

Reference	Sibling diagnosis measured		Sibling prevalence	Control prevalence	OR [95% CI]
Siblings of children with Comorbid Disorders					
Waldman et al. (1998)	**ADHD**	Any ADHD: Low severity	64.0%	40.0%	2.61 [1.36, 5.00]*
		Medium severity	32.0%	18.0%	2.12 [1.07, 4.19]*
		High severity	22.0%	9.0%	2.85 [1.30, 6.21]*
		Combined Type: Low severity	32.0%	18.0%	2.12 [1.07, 4.19]*
		Medium severity	15.0%	6.0%	2.71 [1.08, 6.78]*
		High severity	7.0%	2.0%	3.90 [1.08, 4.05]*
		Inattentive Type: Low severity	22.0%	13.0%	1.89 [0.87, 4.08]
		Medium severity	12.0%	8.0%	1.61 [0.61, 4.26]
		High severity	10.0%	5.0%	2.04 [0.69, 6.03]
		Hyperactive/Impulsive Type: Low severity	10.0%	9.0%	1.09 [0.38, 3.16]
		Medium severity	5.0%	4.0%	1.24 [0.29, 5.38]
		High severity	5.0	2.0%	2.53 [0.56, 11.47]
Sakai et al. (2010)	**CD/ODD**	1-year: CD	38.0%	13.5%	3.92 [2.34, 6.58]**
Waldman et al. (1998)		ODD: Low severity	71.0%	48.0%	2.62 [1.32, 5.20]*
		Medium severity	29.0%	14.0%	2.54 [1.26, 5.13]*
		High severity	17.0%	5.0%	3.89 [1.62, 9.35]**
		CD: Low severity	27.0%	13.0%	2.46 [1.20, 5.07]*
		Medium severity	12.0%	2.0%	6.86 [2.36, 19.92]**
		High severity	7.0%	1.0%	7.38 [1.88, 28.94]**

Appendix B (Cont.)

Data Extraction: Prevalence of Psychopathology

Reference	Sibling diagnosis measured		Sibling prevalence	Control prevalence	OR [95% CI]
Siblings of children with Comorbid Disorders					
Sakai et al. (2010)	**CD/ODD**	1-year: CD	38.0%	13.5%	3.92(2.34-6.58)**
Waldman et al. (1998)		ODD: Low severity	71.0%	48.0%	2.62(1.32-5.20)*
		Medium severity	29.0%	14.0%	2.54(1.26-5.13)*
		High severity	17.0%	5.0%	3.89(1.62-9.35)**
		CD: Low severity	27.0%	13.0%	2.46(1.20-5.07)*
		Medium severity	12.0%	2.0%	6.86(2.36-19.92)**
		High severity	7.0%	1.0%	7.38(1.88-28.94)**
Anderson et al. (2007)		1-year: CD	33.3%	0.13%[1]	
Hopfer et al. (2003)	**SUD**	Risk ratio: Marijuana abuse	20-40%	5-20%	2.65(-)*
		Risk ratio: Marijuana dependence	10-20%	0-10%	2.25(-)*
Sakai et al. (2010)		1-year: Alcohol Dependence	9.5%	6.9%	1.43(0.68-3.02)
		Alcohol Use Disorder	33.1%	18.8%	2.36(1.47-3.80)**
Anderson et al. (2007)		1-year: SUD	10.3%	2.34%[1]	

Note. (-): Not reported/not applicable; ADHD: Attention Deficit Hyperactivity Disorder; SUD: Substance Use Disorder; CD: Conduct Disorder; ODD: Oppositional Defiant Disorder; MHP: Mental health problems; AFF: Affective disorders; DEP: Depressive Disorder/Major Depressive Disorder; ANX: Anxiety disorders; OCD: Obsessive Compulsive Disorder; OR: Odds Ratio; CI: Confidence Interval; Studies contained within the same row drew on the same participant pool.

*$p<.05$; **$p<.01$;

[1]As reported by Lewinsohn, Hops, Roberts, Seeley & Andrews (1993); [2]As reported by Fulton et al. (2005); [3]As reported by Green, McGinnity, Meltzer, Ford, & Goodman (2005); [4]As reported by Polancyzk, de Lima, Horta, Biederman, & Rohde (2007); [5]As reported by Merikangas et al. (2010); [6]As reported by He, personal communication (Jan 13, 2012); [7]As reported by Sawyer et al. (2000); [8]As reported by Karno, Goulding, Sorenson, & Burnam (1988); [9]As reported by Boyd, Kostanski, Gullone, Ollendick, & Shek (2000); [10]As reported by Enzmann, Marshall, Killias, Junger-Tas, Steketee, & Gruszczynska (2010).

Appendix C

Data Extraction: Continuous/Dimensional Mental Health Data

Reference	Target Child Difficulty	Measure	Outcome	Sibling Score *M* (*SD*)	Relationship to control/norm	*d* [95% CI]
			Externalising Problems Scales			
Faraone et al. (1996)	ADHD	CBCL	Baseline: Delinquent	52.5(5.8)	+ve/Normal	*d=0.32* [0.06, 0.59]**
			Aggressive	52.2(5.8)	+ve/Normal	*d=0.25* [-0.03, 0.50]
			4-year followup: Delinquent	54.1(7.4)	+ve/Normal	*d=0.44* [0.16, 0.72]**
			Aggressive	53.9(7.7)	+ve/Normal	*d=0.38* [0.10, 0.66]**
Listug-Lunde et al. (2008)	ADHD		Externalising total	52.5 (12.2)	+ve/Normal	*d=0.59* [0.06, 1.10]*
Pine et al. (1997)	DEL		Baseline: Total externalising	55.3(15.1)	Normal	
			Aggressive behaviour	59.5(10.0)	Normal	
			Delinquent behaviour	58.8(9.7)	Normal	
			15-month followup: Total externalising	57.4(12.0)	Normal	
			Aggressive behaviour	58.9(10.2)	Normal	
			Delinquent behaviour	60.3(9.8)	Borderline	
Dennis & Brotman (2003)	DEL		Aggressive behaviour	54.1(6.2)	Normal	
Brotman et al. (2005b)			Total externalising	51.0(10.2)	Normal	
Hudson & Rapee (2002)	ANX		Total externalising	49.0(7.7)	+ve/Normal	*d=0.35* [-0.11, 0.79]
Dia & Harrington (2006)	ANX		Total externalising	56.1(10.5)	Normal	
Deal & MacLean (1995)	Mixed (MHPs)		Externalising total	46.3(10.3)	+ve/Normal	*d=0.25* [-0.49, 0.95]
			Delinquent	58.6(5.9)	+ve/Normal	*d=1.43* [-0.55, 2.14]
			Aggressive	52.7(4.0)	+ve/Normal	*d=0.40* [-0.35, 1.09]

Appendix C (Cont.)

Data Extraction: Continuous/Dimensional Mental Health Data

Reference	Target Child Difficulty	Measure	Outcome	Sibling Score *M* (*SD*)	Relationship to control/norm	*d* [95% CI]
			Externalising Problems Scales			
Cohen et al. (1996)	Mixed (MHPs)		Total externalising-Parent rated	57.8(11.2)	Normal	
			Teacher rated	56.6(8.7)	Normal	
Copeland et al. (2004)	Mixed (Attention problems and		Total externalising-Parent rated	51.3(12.5)	Normal	
			-Sibling rated	50.1(10.6)	Normal	
Hudziak et al. (2004)	aggression)		Delinquent behaviour	57.2(7.5)	Normal	
			Aggressive behaviour	58.0(8.1)	Normal	
			Symptoms of Externalising Disorders			
Listug-Lunde et al. (2008)	ADHD	Disruptive Behavior Rating Scale	Hyperactive symptoms	1.39(2.42)	+ve	*d=0.65* [0.15, 1.12]**
			Inattentive symptoms	2.03(2.76)	+ve	*d=0.71* [0.21, 1.18]**
Milberger et al. (1996)	ADHD	Kiddie-SADS-E	Symptom scores:			
			ADHD	3.43(4.01)	+ve	*d=0.45* [0.23, 0.72]**
			Conduct disorder	0.81(1.36)	+ve	*d=0.38* [0.13, 0.62]**
Kuntsi et al. (2010)	ADHD	Conners' Rating Scales	ADHD Subscale			
			Parent-rated	54.80(13.62)	+ve/Normal	*d=0.21* [0.07, 0.35]*
			Teacher-rated	56.54(12.41)	+ve/Normal	*d=0.56* [0.42, 0.70]*
Christiansen et al. (2008)			ADHD Combined Type symptom scores:	54.67(7.55)	Normal	
			Parent-rated	55.39(12.36)	Normal	
Sobanski et al. (2010)			Teacher-rated			
			Emotional lability:	54.3(12.8)	Normal	
			Parent-rated	55.5(13.6)	Normal	
			Teacher-rated			

Appendix C (Cont.)

Data Extraction: Continuous/Dimensional Mental Health Data

Reference	Target Child Difficulty	Measure	Outcome	Sibling Score *M* (*SD*)	Relationship to control/norm	*d* [95% CI]
Symptoms of Externalising Disorders						
Waldman et al. (1998)	Mixed (ADHD, ODD, CD)	Emory Diagnostic Rating Scale	Number of symptoms: Hyperactive/impulsive	1.02(1.03)	+ve	*d=0.38* [0.06, 0.69]*
			Inattentive	1.12(1.04)	+ve	*d=0.32* [0.01, 0.63]*
			Oppositional Defiant Disorder	1.09(0.92)	+ve	*d=0.20* [-0.11, 0.52]
			Conduct Disorder	0.30(0.41)	+ve	*d=0.73* [0.41, 1.04]*
Stallings et al. (1997)	Mixed (SUD, DEL)	NIMH Diagnostic Interviews	Mean symptom count: Antisocial	13.44(7.74)	+ve	*d=1.63* [0.90, 2.25]**
			Alcohol problem	9.00(7.39)	+ve	*d=1.26* [0.57, 1.86]**
Faraone et al. (1996)	ADHD	CBCL	Baseline: Attention problems	51.4(3.7)	+ve/Normal	*d=0.23* [-0.04, 0.49]
			4-year followup	54.3(8.5)	+ve/Normal	*d=0.54* [*0.26, 0.82]***
Deal & MacLean (1995)	Mixed (MHPs)		Attention problems	55.0(7.1)	+ve/Normal	*d*=0.38 [-0.08, 1.38]
Copeland et al. (2004)	Mixed (Attention problems and aggression)		Attention problems-Parent rated	56.8 (9.4)	Normal	
			-Sibling rated	55.3 (7.7)	Normal	
Internalising Problems Scales						
Listug-Lunde et al. (2008)	ADHD	Disruptive Behavior Rating Scale	Hyperactive symptoms	1.39(2.42)	+ve	*d=0.65* [0.15, 1.12]**
			Inattentive symptoms	2.03(2.76)	+ve	*d=0.71* [0.21, 1.18]**
Faraone et al. (1996)	ADHD	CBCL	Baseline: Withdrawn	53.0(6.4)	+ve/Normal	*d=0.33* [0.06, 0.59]*
			Somatic complaints	52.8(6.1)	+ve/Normal	*d=0.02* [-0.25, 0.28]
			Anxious/Depressed	53.2(6.1)	+ve/Normal	*d=0.24* [-0.03, 0.51]
			4-year followup: Withdrawn	52.6(5.5)	+ve/Normal	*d=0.37* [0.09, 0.64]*
			Somatic complaints	54.5(6.9)	+ve/Normal	*d=0.40* [0.13, 0.71]**
			Anxious/Depressed	53.7(5.7)	+ve/Normal	*d=0.39* [0.11, 0.67]**

Appendix C (Cont.)

Data Extraction: Continuous/Dimensional Mental Health Data

Reference	Target Child Difficulty	Measure	Outcome	Sibling Score *M* (*SD*)	Relationship to control/norm	*d* [95% CI]
			Internalising Problems Scales			
Listug-Lunde et al. (2008)	ADHD		Total internalising	52.8(11.0)	+ve/Normal	*d=0.72* [0.23, 1.20]*
Pine et al. (1997)	DEL		Baseline total internalising	53.6(14.9)	Normal	
			15-month follow-up	53.8(13.2)		
Hudson & Rapee (2002)	ANX		Total internalising	51.1(10.1)	+ve/Normal	*d=0.29* [-0.17, 0.73]
Dia & Harrington (2006)	ANX		Total internalising	56.5(11.7)	Normal	
Cohen et al. (1996)	Mixed (MHPs)		Total internalising – Parent rated	55.6(10.9)	Normal	
			- Teacher rated	54.6(9.5)		
Deal & MacLean (1995)	Mixed (MHPs)		Internalising total	58.8(8.5)	+ve/Normal	*d=1.46* [0.54, 2.13]**
			Withdrawn	56.9(7.2)	+ve/Normal	*d=0.80* [0.01, 1.49]*
			Somatic complaints	59.0(10.5)	+ve/Normal	*d=0.85* [-0.05, 1.54]*
			Anxious/Depressed	58.7(6.7)	+ve/Normal	*d=1.24* [0.39, 1.94]**
Copeland et al. (2004)	Mixed (Attention problems and aggression)		Total internalising – Parent rated	53.06(11.96)	Normal	
			- Sibling rated	50.24(10.37)	Normal	
Hudziak et al. (2004)			Withdrawn	56.56(7.70)	Normal	
			Somatic complains	57.09(6.52)	Normal	
			Anxious/Depressed	57.08(8.17)	Normal	
			Symptoms of Internalising Disorders			
Listug-Lunde et al. (2008)	ADHD	Children's Depression Inventory	Total depression T-score	46.3(11.3)	+ve/Normal	*d=0.25* [-0.22, 0.72]

Appendix C (Cont.)

Data Extraction: Continuous/Dimensional Mental Health Data

Reference	Target Child Difficulty	Measure	Outcome	Sibling Score *M* (*SD*)	Relationship to controls/norms	*d* [95% CI]
Symptoms of Internalising Disorders						
Barrett et al. (2001)	OCD	Children's Depression Inventory	Total raw depression score	11.0	+ve/Normal	
			Negative mood	2.4	+ve/Normal	
			Anhedonia	4.2	+ve/Normal	
			Ineffectiveness	2.4	+ve/Normal	
			Negative self-esteem	1.0	+ve/Normal	
Hudson & Rapee (2002)	ANX		Total depression T-score	42.4(4.7)	-ve/Normal	*d=0.52* [-0.91, 0.01]
Deal & MacLean (1995)	Mixed (MHPs)		Total raw depression score	6.6(4.7)	+ve/Normal	*d=0.72* [-0.06, 1.42]
Listug-Lunde et al. (2008)	ADHD	Multi-dimensional Anxiety Scale for Children	Total anxiety T-score	52.3(9.7)	+ve/Normal	*d=0.27* [-0.21, 0.74]
Barrett et al. (2001)	OCD		Total anxiety T-score	49.4	+ve/Normal	
			Physical symptoms	11.4	+ve/Normal	
			Harm avoidance	16.8	+ve/Normal	
			Social anxiety	13.8	+ve/Above	
			Separation anxiety	7.4	average	
			Anxiety disorder index	12.2	-ve/Normal	
					+ve/Normal	
Hudson & Rapee (2002)	ANX	What I Think & Feel/Revised	Total anxiety score	10.9(7.5)	+ve/Normal	*d=0.47* [-0.01, 0.91]
Deal & MacLean (1995)	Mixed (MHPs)	Children's Manifest Anxiety Scale	General anxiety	14.4(6.8)	+ve/Above	*d=1.27* [0.42, 1.97]**
			Physiological anxiety	4.4(2.3)	average	
			Worry/Oversensitivity	6.3(2.9)	+ve/Normal	*d=0.99* [0.18, 1.69]*
			Concentration anxiety	4.1(3.2)	+ve/Normal	*d=1.50* [0.61, 2.21]**
					+ve/Normal	*d=0.72* [0.06, 1.41]**

Appendix C (Cont.)

Data Extraction: Continuous/Dimensional Mental Health Data

Reference	Target Child Difficulty	Measure	Outcome	Sibling Score *M* (*SD*)	Relationship to controls/norms	*d* [95% CI]
Symptoms of Internalising Disorders						
Milberger et al. (1996)	ADHD	Kiddie-SADS-E	Symptom scores:			
			Agoraphobia	0.18(0.55)	+ve	*d=0.02* [-0.22, 0.26]
			Major Depressive Disorder	2.10(3.05)	+ve	*d=0.09* [-0.15, 0.34]
			Separation Anxiety	1.15(1.50)	+ve	*d=0.33* [0.08, 0.57]**
			Simple Phobia	1.59(1.89)	+ve	*d=0.24* [0.00, 0.48]
			Social Phobia	1.02(1.74)	+ve	*d=0.24* [-0.01, 0.48]
			Overanxious Disorder	1.60(1.74)	+ve	*d=0.25* [0.01, 0.49]*
			Panic Disorder	0.52(1.66)	+ve	*d=0.07* [-0.18, 0.31]
McDougall et al. (2006)	ADHD	DSM-IV Scale	Number of symptoms: Separation Anxiety:			
			Inattentive type ADHD	0.63	+ve	-
			Combined type ADHD	1.74	+ve	*d=0.67*(-)*
			Generalised Anxiety			
			Inattentive type ADHD	0.79	+ve	-
			Combined type ADHD	3.32	+ve	*d=0.88*(-)**
Waldman et al. (1998)	Mixed (ADHD, ODD, CD)	Emory Diagnostic Rating Scale	Number of symptoms: Depression/Dysthymia	0.28(0.40)	+ve	*d=0.43* [0.11, 0.74]*
Stallings et al. (1997)	Mixed (SUD, DEL)	NIMH Diagnostic Interviews	Mean symptom count: Depression	8.80(5.77)	+ve	*d=0.64* [0.57, 1.86]*

Appendix C (Cont.)

Data Extraction: Continuous/Dimensional Mental Health Data

Reference	Target Child Difficulty	Measure	Outcome	Sibling Score *M* (*SD*)	Relationship to controls/norms	*d* [95% CI]
			Social Problems Scales			
Faraone et al. (1996)	ADHD	SAICA	Baseline: School behaviour problems	1.9(0.9)	+ve	*d=0.23* [0.00, 0.46]*
			Spare-time activities (Reversed)	1.7(0.6)	+ve	*d=0.17* [-0.39, 0.06]
			Spare-time problems	1.5(0.6)	+ve	*d=0.17* [-0.06, 0.39]
			Activities with peers (Reversed)	1.7(0.7)	+ve	*d=0.30* [0.07, 0.53]*
			Problems with peers	1.5(0.7)	+ve	*d=0.15* [-0.08, 0.38]
			Boy-girl relationships (Reversed)	2.2(0.7)	+ve	*d=0.14* [-0.37, 0.09]
			Problems with opposite sex	1.3(0.6)	Equal	*d=0.00* [-0.23, 0.23]
			4-year followup: School behaviour problems	2.0(0.9) 2.0(0.6)	+ve +ve	*d=0.65* [0.41, 0.87]** *d=0.18* [-0.04, 0.40]
			Spare-time activities (Reversed)	1.7(0.7)	+ve	*d=0.46* [0.23, 0.68]*
			Spare-time problems	1.9(0.7)	+ve	*d=0.31* [0.08, 0.53]*
			Activities with peers (Reversed)	1.6(0.8)	+ve	*d=0.30* [0.07, 0.52]*
			Problems with peers	2.4(0.9)	Equal	*d=0.00* [-0.22, 0.22]
			Boy-girl relationships (Reversed)	1.3(0.6)	+ve	*d=0.19* [-0.03, 0.42]
Faraone et al. (1998)			Problems with opposite sex Total Social adjustment score	18.24(4.71)	+ve	*d=0.55* [0.26, 0.83]**
Faraone et al. (1996)	ADHD	CBCL	Baseline:Social problems	52.5(6.3)	+ve/Normal	*d=0.40* [-0.07, 0.46]
			4-year followup: Social problems	53.8(6.9)	+ve/Normal	*d=0.52* [0.25, 0.81]**
Deal & MacLean (1995)	Mixed (MHPs)		Social problems	56.9(8.8)	+ve/Normal	*d=0.95* [0.1, -1.64]**
Hudziak et al. (2004)	Mixed (Attention problems and aggression)		Social problems	56.91(7.56)	Normal	
Barrett et al. (2001)	OCD	Children's Depression Inventory	Interpersonal problems	1.2	+ve/Normal	

Appendix C (Cont.)

Data Extraction: Continuous/Dimensional Mental Health Data

Reference	Target Child Difficulty	Measure	Outcome	Sibling Score *M* (*SD*)	Relationship to controls/norms	*d* [95% CI]
Global Functioning Scales						
Dia & Harrington (2006)	ANX	CBCL	Total behaviour problems	57.7(10.2)	Normal	
Cohen et al. (1996)	Mixed (MHPs)		Total behaviour problems – Parent rated	57.5(11.0)	Normal	
			- Sibling rated	55.7(9.9)	Normal	
Rettew et al. (2004)	Mixed (Attention problems and aggression)		Total behaviour problems	30.5(17.68)	Normal	
Deal & MacLean (1995)	Mixed (MHPs)		Total behaviour problems	53.8(9.1)	+ve/Normal	*d=1.15* [0.31, 1.84]**
Müller et al. (2011)	ADHD	Conners' Rating Scales	Total across all scales:	51.8(-)	Normal	
			Parent-rated	52.9(-)	Normal	
			Teacher rated			
Faraone et al. (1996)	ADHD	Global assessment of functioning	Global Assessment of Functioning Scale			
			Baseline	62.0(12.4)	-ve	*d=0.34* [0.11, 0.57]**
			4-year follow-up	61.3(9.7)	-ve	*d=0.61* [-0.37, 0.83]**
Kelvin et al. (1996)	Mixed (ANX and/or DEP)		Children's Global Assessment Scale –			
			Minimal impairment	51.6%		
			Mild impairment	32.3%		
			Moderate impairment	16.1%		

Note. (-): Not reported; +ve: Sibling scores were higher than control scores; -ve: Sibling scores were lower than control scores; Target child difficulties: ADHD: Attention deficit hyperactivity disorder; ANX: Anxiety disorders; CD: Conduct disorder; DEL: Adjudicated for delinquency; DEP: Depression; MHPs: Range of mental health problems; OCD: Obsessive compulsive disorder SUDs: Substance use disorders
Measures: CBCL; Child Behavior Checklist; SAICA: Social Adjustment Inventory for Children and Adolescents
*p<.05; **p<.01; ***p<.005

Appendix D

Data Extraction: Family Relationships Data

Reference	Target child MHP	Outcome Measured	Sibling Score *M* (*SD*)	Comparison to Control Scores	*d* [95% CI]
		Sibling Relationships			
		Positive Aspects			
Biederman et al. (1993)	ADHD	% of target children with impaired sibling activity (e.g. less play, less talking)	22%	-ve	(-)**
Faraone et al. (1996)	ADHD	Activities at baseline (Reverse scored)	1.8(0.7)	-ve	*d=0.29* [0.06, 0.51]*
		4-year followup	2.1(0.8)	-ve	*d=0.53* [0.30, 0.75]*
Mikami & Pfiffner (2008)	ADHD	Warmth/closeness	(-)	-ve	*(-)*
Daniels & Moos (1990)	CD	Sibling resources (e.g. emotional support)	11.98	+ve	*(-)*
Slomkowski et al. (1997)	Delinquency	Positive qualities (e.g. warmth, admiration)	66.5(13.3)	+ve	*d=0.73* [0.24, 1.24]*
Daniels & Moos (1990)	Affective	Sibling resources	10.4	-ve	*(-)*
Barrett et al. (2001)	OCD	Warmth	3.05	-ve	*(-)*
Lindhout et al. (2003)	ANX	Affection	18.75(6.36)	-ve	*d=0.34* [-0.24, 0.89]
Deal & MacLean (1995)	MHPs	Warmth/Closeness	21.5(5.0)	Equal	*d=0.00* [-0.59, 0.93]
		Intimacy	2.9(0.7)	-ve	*d=0.13* [-0.60, 0.84]
		Similarity (e.g. in interests, personality)	2.8(0.9)	-ve	*d=0.23* [-0.50, 0.93]
		Admiration of sibling	3.5(1.0)	-ve	*d=0.09* [-0.60, 0.80]
		Admiration by sibling	2.9(0.7)	-ve	*d=0.11* [-0.62, 0.82]
Dumas (1996)	Externalising problems	Approval (e.g. of conduct)	0.009(0.010)	-ve	*d=0.43* [-0.33, 1.13]
		Positive affect (e.g. affectionate gestures)	0.156(0.181)	-ve	*d=1.15* [0.32, 1.86]**
		Compliance to sibling's requests and instructions	0.546(0.243)	-ve	*d=0.44* [-0.32, 1.14]

Appendix D (Cont.)

Data Extraction: Family Relationships Data

Reference	Target child MHP	Outcome Measured	Sibling Score *M* (*SD*)	Comparison to Control Scores	*d* [95% CI]
		Sibling Relationships			
Negative Aspects					
Biederman et al. (1993)	ADHD	% of target children with impaired sibling relations (e.g. avoidance)	28%	+ve	*(-)***
Faraone et al. (1996)	ADHD	Sibling relationship problems: Baseline	1.8(0.8)	+ve	*d=0.27* [0.03, 0.49]*
		4-year followup	1.9(0.8)		*d=0.40* [0.17, 0.69]*
Geller et al. (2000)	ADHD	% of target child with poor sibling relations	22.5%	+ve	*(-)*
Mikami & Pfiffner (2008)	ADHD	Conflict (e.g. bullying)	(-)	+ve	*(-)**
Tseng et al. (2011)	ADHD	Problems (e.g. bullying)	1.7(0.6)	+ve	*d=0.32* [-0.02, 0.66]
Daniels & Moos (1990)	CD	Sibling stressors (e.g. criticalness)	11.84	+ve	*(-)*
Schachar & Wachsmuth (1990)	ODD	Problems (e.g. blaming)	38.0	+ve	*(-)**
	CD		32.0	+ve	*(-)*
Tseng et al. (2011)	ODD	Problems (e.g. bullying)	2.1(0.7)	+ve	*d=1.04* [0.48, 1.58]**
Slomkowski et al. (1997)	Delinquency	Negative qualities (e.g. destroying sibling property)	29.9(0.7)	+ve	*d=0.07* [-0.46, 0.54]
		Influence (e.g. sibling going along with 'bad' act)	24.4(6.5)	+ve	*d=1.33* [0.70, 1.79]**
Daniels & Moos (1990)	DEP	Sibling stressors (e.g. criticalness)	12.25	+ve	*(-)*
Geller et al. (2000)	Bipolar disorder	% of target children with poor sibling relations	40.7%	+ve	*(-)***
Geller et al. (2001)	DEP	Adult sibling relationship problems (e.g. avoidance) over past 5 years			
		Target with MHPs in past 12 months	2.7(1.1)	+ve	*d=1.25* [0.74, 1.75]*
		Without MHPs in past 12 months	2.1(1.1)	+ve	*d=0.69* [0.12, 1.26]
		Target with MHPs in past 5 years	2.9(1.1)	+ve	*d=1.12* [0.61, 1.60]**
		Without MHPs in past 5 years	2.4(1.2)	+ve	*d=0.60* [0.00, 1.17]

Appendix D (Cont.)

Data Extraction: Family Relationships Data

Reference	Target child MHP	Outcome Measured	Sibling Score *M* (*SD*)	Comparison to Control Scores	*d* [95% CI]
		Sibling Relationships			
Negative Aspects					
Puig-Antich et al. (1993)	DEP	Poor sibling relations (e.g. fights)	2.3(2.1)	+ve	*d=0.51* [0.09, 0.91]**
Barrett et al. (2001)	OCD	Relative power/dominance	-0.4	Equal	*(-)*
		Conflict	2.8	+ve	*(-)*
Lindhout et al. (2003)	ANX	Hostility	14.4(3.4)	+ve	*d=0.44* [-0.14, 0.99]
Deal & MacLean (1995)	MHPs	Relative power/dominance	-2.0(1.7)	+ve	*d=0.22* [-0.51, 0.93]
		Conflict	9.0(3.2)	+ve	*d=0.50* [-0.25, 1.20]
Dumas (1996)	Externalising problems	Attempts to control siblings	0.08(0.06)	+ve	*d=0.10* [-0.82, 0.63]
		Aggression	0.05(0.05)	+ve	*d=0.11* [-0.62, 0.83]
Stormont-Spurgin & Zentall (1995)	Hyperactivity and/or aggression	% of targets with siblings who retaliate aggressively			
		Targets with aggression	8.3%	Equal	*Difference between groups; (-)**
		Targets with hyperactivity	14.6%	+ve	
		Targets with hyperactivity and aggression	31.3%	+ve	
Tseng et al. (2011)	ADHD+ODD	Problems (e.g. bullying)	2.3(0.72)	+ve	*d=1.37* [1.03, 1.70]**

Appendix D (Cont.)

Data Extraction: Family Relationships Data

Reference	Target child MHP	Outcome Measured	Sibling Score *M* (*SD*)	Comparison to Control Scores	*d* [95% CI]
		Parent-Sibling Relationships			
		Positive Aspects			
Faraone et al. (1996)	ADHD	Activity (e.g. spend time, affectionate, reverse-scored)			
		Mother-sibling			
		Baseline	1.5(0.7)	-ve	*d=0.30* [0.07, 0.53]*
		4-year follow-up	1.7(0.7)	-ve	*d=0.31* [0.01, 0.46]*
		Father-sibling			
		Baseline	1.7(0.9)	-ve	*d=0.23* [-0.08, 0.53]
		4-year followup	1.8(0.8)	Equal	*d=0.00* [-0.22, 0.22]
Dadds et al. (1992)	CD	Mean % of intervals that contained behaviour:			
		Mother-sibling positive (e.g. praise)	50.5%(17.3)	-ve	*d=0.95* [-0.29, 1.59]
		Father-sibling positive	41.4%(23.5)	-ve	*d=0.65* [-0.01, 1.27]
	DEP	Mother-sibling positive	64.2%(21.6)	-ve	*d=0.11* [-0.56, 0.78]
		Father-sibling positive	51.4%(21.8)	-ve	*d=0.22* [-0.46, 0.89]
	CD+DEP	Mother-sibling positive	75.2%(14.5)	+ve	*d=0.60* [-0.18, 1.35]
		Father-sibling positive	51.6%(9.7)	-ve	*d=0.26* [-0.10, 0.50]
		Negative Aspects			
Faraone et al. (1996)	ADHD	Problems with parents (e.g. noncompliance)			
		Baseline	1.4(0.6)	+ve	*d=0.38* [0.15, 0.61]*
		4-year followup	1.6(0.7)	+ve	*d=0.49* [0.26, 0.71]*

Appendix D (Cont.)

Data Extraction: Family Relationships Data

Reference	Target child MHP	Outcome Measured	Sibling Score *M* (*SD*)	Comparison to Control Scores	*d* [95% CI]
		Parent-Sibling Relationships			
		Negative Aspects			
Dadds et al. (1992)	CD	Mean % of intervals that contained:			
		Mother-sibling aversive	19.1%(14.5)	+ve	*d=1.40* [0.69, 2.06]*
		Father-sibling aversive	6.0% (9.3)	+ve	*d=0.36* [-0.27, 0.97]
	DEP	Mother-sibling aversive	14.8%(18.3)	+ve	*d=0.90* [-0.18, 1.59]
		Father-sibling aversive	3.5%(4.9)	+ve	*d=0.10* [-0.58, 0.72]
	CD+DEP	Mother-sibling aversive	11.8%(14.5)	+ve	*d=0.92* [-0.11, 1.68]
		Father-sibling aversive	3.4%(7.6)	+ve	*d=0.07*[-0.68, 0.81]
Hudson & Rapee (2002)	ANX	Intrusive involvement (e.g. unsolicited help)			
		Mother	4.66(1.36)	+ve	*d=0.64* [0.09, 1.21]*
		Father	5.20(1.32)	+ve	*d=0.19* [-0.35, 0.74]
Barrett et al. (2001)	OCD	Rivalry/Partiality[a]	3.5	Favour self +ve	*n/a*
Lindhout et al. (2003)	ANX	Parental differential treatment	6.92(3.64)	+ve	*d=0.67* [0.07, 1.22]*
Deal & MacLean (1995)	MHPs	Parental partiality/favouritism[b]	5.7(1.1)	Favour target child -ve	*(-)**
		Maternal partiality/favouritism[a]	3.0(0.7)	Neither favoured -ve	*(-)*
		Paternal partiality/favouritism[a]	2.7(0.6)	Favour target child -ve	*(-)**

Note. (-): Not reported; [a]Midpoint=3.0; <3.0=Parents favour target child; >3.0=Parents favour self; [b]Midpoint=6.0; <6.0=Parent favour target child; >6.0=Parents favour self; ADHD: Attention deficit hyperactivity disorder; ANX: Anxiety disorders; CD: Conduct disorder; DEP: Depression; MHPs: Range of MHPs; OCD: Obsessive-compulsive disorder; ODD: Oppositional defiant disorder
**p*<.05; ***p*<.01

Appendix E

Data Extraction: Qualitative Data

Reference	Extracted data: Summary data and quotes
Limited mental health knowledge	
Litzelfelner (1995)	***Summary data:*** - Did not seem to have much understanding of their siblings' difficulty; 3/4 did not know target child's diagnosis; Confusion and lack of understanding ***Quotes:*** "My brother, we don't really know what's wrong with him, he's just got a hard time living in reality, he lives in a fantasy world" "Well, I know where he gets it, he gets it from my dad…I know it's genetic or something like that" "My grandfather was manic depressive…and I think Johnny got it from him" "Rob and Jane are both chemical, but I don't know where it came from, it could have come from my mom or my dad's side" "…she has little fits in front of people. I think she got it from my dad" "If he grew up and had kids, wouldn't you kinda wonder? Cause some of this stuff runs in families, so to me it's important to know cause if I grow up and have kids I can say, well this has been in my family"
Garley & Johnson (1994)	***Summary data:*** - Lack of understanding of the illness; Struggled to make sense of the illness experience for themselves, felt inadequate knowledge; made own attributions as to purpose of disease (e.g. attention-seeking, manipulative); Future: Thoughts of dying ***Quotes:*** "I've had like cake and everything…and she won't even *touch* cake. She goes, 'I just don't *feel* like it,' and everyone must feel like something every now and then you know, but she always says she doesn't. Sometimes I'd like to be like that." "That's why she keeps on going – 'cause she gets attention from my parents, the hospital, her friends, and I think she likes all that…If she had no one to go to, then maybe she'd smarten up" "I guess I'm pretty ignorant about the whole disease." "That [meeting with psychiatrist] was just a one family meeting. I never really learned anything about it [the condition]."

Appendix E (Cont.)

Data Extraction: Qualitative Data

Reference	Extracted data: Summary data and quotes
Limited mental health knowledge	
Slowik et al. (2004)	***Summary data:*** - Lack of understanding about nature of illness and amount of responsibility the target child could be expected to take for their behaviour
Deal & MacLean (1995)	***Summary data:*** - No one discussed hospitalised target child with them - Siblings were fearful about the target child's future and their own future
Kendall (1998, 1999)	***Summary data:*** - None said they expected the disruptions to end
Violence, conflict, aggression	
Litzelfelner (1995)	***Summary data:*** - Described negative behaviours of target child which pervaded all conversations in focus group; Violent/aggressive behaviours ***Quotes:*** "My brother, he's thirteen, he gets very violent, he threatens me with knives and one time he hit me in the back with a broom so hard it left three welts on my back" "He has a really bad temper and beats up on my little sister" "…we've had to replace a lot of the doors because of my brother, he knocks them off of the hinges and we have lots of holes in the walls." "Jane used to be violent, she used to scratch and draw blood" "My sister, she's got a temper, she gets in fights with my brother"

Appendix E (Cont.)

Data Extraction: Qualitative Data

Reference	Extracted data: Summary data and quotes
Violence, Conflict, Aggression	
Kendall (1998, 1999)	***Summary data:*** - Never knowing what was coming next-what problem would have to be dealt with – when not if; Violence, aggression, manipulation, control - Parents reported younger siblings mimicking disruptive behaviours, family members, especially siblings, taking revenge or escalating the disruptive event, and ineffective family communication patterns - Siblings feeling victimised by physical violence, verbal aggression, manipulation, control; (e.g. by acts of defiance, self-centredness); Feeling 'dumped on' and displaced in their home, invasion of privacy, always feeling 'on guard'; Felt they received the most abuse because they were more available and accessible than peers, parents or others; 'easy targets' and were abused without impunity because parents with either too exhausted or overwhelmed to intervene – target child confirmed they often got away with things at home that they didn't at school - Believed their parents should do more to control target child's behaviour; Also anxious and afraid because of abuse - Reported parents as often minimising and disbelieving seriousness of aggression; many parents described behaviour as 'normal' sibling rivalry whereas none of the siblings did; target child often minimising abuse/justifying e.g. "they deserved what they got" "that's just what you do when you get angry" ***Quotes:*** "He hits me every day. He just all of a sudden hauls off and hits me all of a sudden. I don't know why. My mom says its part of his hyperactivity. My mom tells me not to worry about it, that it will get better. I don't see how" "For the longest time he had a baseball bat in his room that he would take out and treatment to whack me with if I didn't do what he wanted. Oh yeah, and one time he got a stick and threw it right at me and it went through my glasses and into my eye. A year ago, I fell out of a tree and when I was resting on the couch he jumped on me and jammed his finger in my eye and I had this huge bloody eye from it for a long time" Target child – "Why not hit him? He's right there in my face all the time and he's always bothering me. Nothing ever happen when I do – he yells and tells mom and mom sends us both to our room. Big deal. I can't hit anybody at school even though I want to a lot. I'd get in too much trouble. So when I get home, yeah my brother gets most of it. I don't care. He deserves it for being in my face all the time." Target child – "he deserved it because he was ugly"; "it's just something I do"; "that's just what you do when you get angry"

Appendix E (Cont.)

Data Extraction: Qualitative Data

Reference	Extracted data: Summary data and quotes
Violence, Conflict, Aggression	
Tahhan et al. (2010)	***Summary data:*** - Target child described as being out of control resulting in physical damage to home; Violence towards parents and siblings - Tension in home; Dissatisfaction living in household where their child would erupt without provocation - Parent knew that their child needed intensive mental helath treatment in part because siblings lived in fear that at any moment they could be attacked both verbally, and in many cases, physically by the target child ***Quotes:*** "He would come home and as soon as he would open that door, all hell would break loose. The door would come open with his feet and he would just lose it because he's held it in all day and he's ready to explode." - Sibling "His behaviour impacted the family so enormously…when you get to the point where the younger children are scared of the behaviour of the older one you have to realise something has to give" – Parent
Deal & MacLean (1995)	***Summary data:*** - 80% of siblings reported frequent acts of physical aggression by target child; only 20% of control siblings reported frequent acts of physical aggression; Felt picked on
Barrett et al. (2001)	***Summary data:*** Bullying ***Quotes:*** "He sometimes picks on me, and it makes me angry"
Being Another Parent	
Deal & MacLean (1995)	***Summary data:*** - Internalised expectations; feelings of responsibility for their sibling's disturbance or its potential reoccurrence

Appendix E (Cont.)

Data Extraction: Qualitative Data

Reference	Extracted data: Summary data and quotes
Being Another Parent	
Garley & Johnson (1994)	***Summary data:*** - Assumed roles not typical of siblings; Role conflict; personal responsibility ('mothering qualities'), burdensome; - Felt responsibility to target child's illness and possible reoccurrence, protectiveness depicting target child as fragile and vulnerable; confusion regarding level of responsibility; feelings of impotence; difficulty dealing with demands of family and friends; - Parents expected tolerance, understanding, sensitivity to needs; sometimes messages delivered by others were harsh and insensitive, evoking further feelings of guilt and frustration ***Quotes:*** "Before she was like my equal…Now she's like a little puppy, you know. I feel like I have to carry her around with me, like make sure she's OK" "I don't really want to have to be held down [at school lunch hours] with watching her eat lunch"
Kendall (1998, 1999)	***Summary data:*** - Reported that parents expected them to care and protect target child; including befriend, play with, give medication, tutoring; prevent bad behaviour, provide respite for parents yet rarely get it themselves - Two of the younger (both female) siblings viewed responsibility positively – gave them a 'special' role; rest say responsibility expectations as unreasonable, burdensome; Felt excluded from decision making, yet expected to take responsibilities without complaint, reward, recognition; - Resentful yet still worry about target child's wellbeing - Expectation that they were also expected to be somewhat invisible – not requiring too much help or attention from the parents; felt overlooked and ignored - Parents described these responsibility relationships as what siblings 'just do'; siblings said that when they were having difficulty or needed extra attention, they were often told by their parents that they would be all right and not to worry about it. ***Quotes:*** "I worry about him getting hurt by somebody else. He doesn't listen, and one of these times he's going to get into real trouble, and like if he bad-talks somebody and he doesn't have control of himself and then they say, 'I'm going to punch you,' and he doesn't hear them, the he's going to get his lights knocked out. So I try to make sure he doesn't get into that kind of trouble"

Appendix E (Cont.)

Data Extraction: Qualitative Data

Reference	Extracted data: Summary data and quotes
Being Another Parent	
Rosenzweig et al. (2002)	***Summary data:*** - For families with rigid wok schedules or single-parent families, siblings sometimes provided care for the children with MHPs backed by their parents who responded in emergencies. Extended family members rarely gave care on a regular basis (e.g. aunts, uncles, grandparents) - Several families placed their target child in overnight summer camps for a week or two, so that they could take a vacation with the other children or make up for missed work. ***Quotes:*** "Mostly my teenage daughter takes care of him after school and definitely on the weekends...I have to do this in order to support four kids. I can't afford to pay somebody to watch a special needs kid"
Life Dominated by Target Child	
Garley & Johnson (1994)	***Summary data:*** - Profound and all-encompassing impact; being unable to control their involvement or distance themselves – target child having an all-consuming predominance in sibling's life - Overwhelming sense of imposition and intrusiveness experienced in living each day with the illness; Life controlled by target child ***Quotes:*** "When we're travelling…and we have to stop and eat, it's always the biggest issue. We can't go to McDonald's because there's nothing there my sister would eat…We have to go to a restaurant that has waitresses…or to…a Burger King with a salad bar."
Tahhan et al. (2010)	***Summary data:*** - Unable to plan family activities because of the target child's MHPs
Litzelfelner (1995)	***Summary data:*** - Resentment that the family was dominated by target child and that no-one else's needs were being met
Kendall (1998, 1999)	***Summary data:*** - Family life often centred on target child; Target child demands to be centre of attention; Resentful that family life controlled by target child; child 'ruining' events; didn't look forward to certain events because of this (e.g. birthday parties)

Appendix E (Cont.)

Data Extraction: Qualitative Data

Reference	Extracted data: Summary data and quotes
Life Dominated by Target Child	
Barrett et al. (2001)	***Summary data:*** - Life revolving around IC; Protracted showering habits; delays in daily routines ***Quotes:*** "…takes too long in the shower, and then we have to hurry up if we are going out; N does everything slowly, like getting into the house with the key. I get angry because I have to wait and wait to get in" "She is slow in doing things; day-dreaming and wasting water" "N always locks up the house all the time. She went out when I was playing in the yard, and I was locked out and had to ride around for an hour." "N has really long showers and uses all the hot water. There is no water left when I go in for just 5 minutes. I get annoyed at him" "asks questions over and over, and it is really annoying hearing them" "N has really long showers and uses all the hot water and that is really annoying" "N…turns the taps in the bathroom off so tight that I had to get Dad to help me. I couldn't turn the tap on" "In the morning N…gets ready really late because he has to go and check the bathroom: the toothbrushes in the cup, taps off, towels straight. As a result, we are late for school" "N takes ages to have a shower (half an hour) to dry herself (half an hour), and to get dressed (45 minutes). I am never at school on time, and we have to go to the office and get a late note, and it is very embarrassing" "We are never on time, and I am late for my job at the restaurant (their father's), and this annoys Dad a lot"
Impact on Relationships	
Litzelfelner (1995)	***Summary data:*** - Anger was expressed more frequently by siblings than parents, mainly about target child behaviour perceived as 'manipulative' and by implication, controllable; Stigma of mental illness, leading to alienation within their own peer group - Sadness about loss of previous relationship with target child
Rissanen et al. (2008)	***Summary data:*** - Siblings received less attention at home than target child

Appendix E (Cont.)

Data Extraction: Qualitative Data

Reference	Extracted data: Summary data and quotes
Impact on Relationships	
Garley & Johnson (1994)	***Summary data:*** - Repeated questioning by school mates - Some reported getting closer to family members; Changes in family relationships, in themselves and others; - Feelings of loss, abandonment, anguish; some report becoming more distant to family members; target child as having privileged position in family, which was generally attributed to illness itself; indulged by parents, more attention - Feeling overwhelmed, frustrated, unfairly treated; Family tension ***Quotes:*** "It feels like it's two against one sometimes…It makes me worry about…me and my mom. The relationship between me and her has drifted…Those two are really tight, you know" "One whole grocery load was all these diet crackers…and I don't like crackers in the first place…I had to remind her [mother] 'cause she thought 'I thought it would be healthy for you'. I was like 'No, I'm not going to eat those melba toast things'." "She's just drifted, like she's no longer…in the picture. She just doesn't want to have anything to do with it [their peer group]…She's just pushed herself out, like until she loses weight or gets her perfect body…She doesn't really want to come out. It's like hard for me. I miss my sister. Even though I see her every night, she's not the same. She used to be so hyper, you know. She'd do funny things and now she's just like hunched over…She looks drained." "It's not normal…I used to be living with a normal sister. We'd get into arguments over 'No, don't wear my clothes tonight,' those little things, but now over salt in the water! And I didn't even *touch* the water."
Deal & MacLean (1995)	***Summary data:*** - Few siblings reported receiving more parental attention than the target child compared to control siblings - No one had ever asked them about how they felt
Rosenzweig et al. (2002)	***Summary data:*** - Some parents were able to meet needs of the target child and siblings by devising and keeping to a rigid schedules - Parents made compromises in work aspirations, standards for home care, and the attention paid to other siblings

Appendix E (Cont.)

Data Extraction: Qualitative Data

Reference	Extracted data: Summary data and quotes
Impact on Relationships	
Tahhan et al. (2010)	***Summary data:*** - Relationships between family members were strained because of their child's problems; Parents reported feeling neglectful to their other children ***Quotes:*** "When he's not here it gives you an opportunity to spend time with another child"
Kendall (1998, 1999)	***Summary data:*** - Family relationships: Parents and siblings named target child as most significant problem; target child named 'other people' as their most significant problem - Family conflict, poor peer relationships, difficult relationship with extended family - Siblings wanted parents to understand and acknowledge experience, stop playing 'favourites'; felt powerless, resignation, unworthy of attention, love or care reinforced by parental rejection - Parents felt that they should have done something ore for their children or families including giving more time to siblings ***Quotes:*** "I don't ask for much, I'm just here and that's about it. Go to school, take care of my brother, that's about it" "Just there, not doing anything to cause anyone to notice"
Barrett et al. (2001)	***Summary data:*** - Reported causing some difficulty with peers as target child would follow him to their house; Family conflict ***Quotes:*** "N's taking so long to shower and get dressed really upsets things at home because my cousins, aged 2, 3, and 4 years of age who are living with us, can't use the bathroom'; 'When N is in the shower, she wants the children out of the bathroom before she will get out, and she yells at me, and I have to argue with the kids and argue with N, and there is a big yelling match, and the kids are throwing fits" "he follows me around to my mates' homes. I get really angry because he follows me"

Appendix E (Cont.)

Data Extraction: Qualitative Data

Reference	Extracted data: Summary data and quotes
Impact on Self	
Deal & MacLean (1995)	***Summary data:*** - Siblings believe that although similar to target child in appearance and interests, very dissimilar in personality and conduct (not found in control siblings); Believed they would not act like the target child, denying they would emulate their behaviour; - Feelings of guilt, neglect, fear, and victimised
Garley & Johnson (1994)	***Summary data:*** - Some positives: Personal maturation, increased tolerance and empathy - Wide and diverse range of emotions (e.g. loss), often fraught with contradiction and ambivalence, e.g. loyalty, understanding v anger and resentment; often leading to feelings of guilt and self reproach for their lack of tolerance; Feelings of injustice, hurtfulness, frustration, resentment; neglected; - Altered own self-percept negatively (e.g. body image and eating patterns); Felt as if had no identity of their own; Some unwittingly reinforced this by suppressing own feelings, or actively encouraging friends to acknowledge and preserve the special identity of ill sister ***Quotes:*** "There's a few [friends] who are really supportive…and those are the ones I tend to like more now. I used to like…my friends who like to have fun better…but now…I find me liking the people who really care." "It's like she's a mouse compared to me, like if I stand in the mirror next to her I feel like I'm two of her…I just feel big compared to her…really bulky, and dirty sort of you know, like fat and ughh."
Litzelfelner (1995)	***Summary data:*** - 2/4 siblings reported increased stress; Described feelings of anxiety, particularly in relation to deliberate self-harm; loss and sadness ***Quotes:*** "I get really stressed out sometimes"

Appendix E (Cont.)

Data Extraction: Qualitative Data

Reference	Extracted data: Summary data and quotes
Impact on Self	
Slowik et al. (2004)	***Summary data:*** - Could feel anxiety, guilt, anger and sadness both towards and about the target child; Resentment
Barrett et al. (2001)	***Summary data:*** Impact of target child's MHPs ratings: Younger sibling - 'somewhat of a problem' Young sibling - overall 'somewhat' involved; 'a little bit' distressed Younger sibling - 'a lot of a problem' Older sibling - 'a lot of a problem' Younger sibling - 'very much a problem'
Kendall (1999)	***Summary data:*** - Resentment; anxiety, fear, victimised; Powerless, resignation, unworthy of attention, love or care, loss - Process that began as intense anger (without aggression) about their mistreatment that eventually changed to sadness and resignation; anger resentment, sadness and alienation from the family's preoccupation with the ADHD child; resentment towards parents for tolerating and defending behaviour; received the message that they were not worth 'sticking up' for.

Appendix E (Cont.)

Data Extraction: Qualitative Data

Reference	Extracted data: Summary data and quotes
Avoidance	
Garley & Johnson (1994)	***Summary data:*** Brother avoided parental pressure and responsibility for target child due to remaining largely uninvolved and on the periphery of IC life; Siblings would fantasise about moving out of home and living independently, freedom from the impact of the target child's MHPs
Litzelfelner (1995)	***Summary data:*** Reported little effect as they were not around the target child much ***Quotes:*** "It doesn't really effect my life much because I spend a lot of time at work and school and stuff, but when I'm home it does" "I doesn't really effect me at all, I was always either gone or doing something, it never really put a cramp in my life"
Kendall (1998, 1999)	***Summary data:*** - Much energy spent on coping with disruption; avoidance of the target child when mood was volatile ***Quotes:*** "I just stay out of his way most of the time and try to just go with the flow. If I challenge him on something, or disagree with him, he becomes loud and mean. I know to get out of his path when he gets angry and raises his voice. I try to avoid the situation until he cools down"
Garley & Johnson (1994)	***Summary data:*** - Avoid sister's chronic complaints ***Quotes:*** "Usually I don't like it when she's here [at home], because that's when I know about the problems, and I don't like knowing about it. If I don't know about it…it can't bug me, but if she's home and she's sick, then I know about it and it bugs me."

Appendix E (Cont.)

Data Extraction: Qualitative Data

Reference	Extracted data: Summary data and quotes
Accommodation	
Garley & Johnson (1994)	***Summary data:*** - Accommodates sister's chronic complaints
Kendall (1998, 1999)	***Summary data:*** - Resigned to their situations and learning to avoid and accommodate; Accommodating: sibling conformed to target child's needs and affect ***Quotes:*** "I've learned to check to see how he's feeling before I even say hi when I come home from school. If he looks upset I don't say anything because I know he will yell at me. If he looks bored then I know I better talk with him or he'll yell at me." "You know, it's like, when you get home don't ask him how his day was and have him just start talking and all that stuff. But when he wants you to listen, listen, and that way he won't get so mad. So – when you're listening to him, don't like butt in or anything because that also gets him real mad. I've learned not to talk to him about what's important to me because he won't listen or he'll say its stupid. So I only talk to him about what he wants to talk about and that way he won't get mad at me."
Barrett et al. (2001)	***Summary data:*** - All siblings engaged in some form of accommodation; - Accomodation through participation in OCD behaviour was moderate whereas modification of their functioning was mild - Only one sibling reported feeling distressed from engaging in accommodation – said that the target child became extremely distressed and anxious, grew mildly angry and had a mild increase in time spent to do her rituals, when she did not help her - All parents reported observing sibling distress

Appendix E (Cont.)

Data Extraction: Qualitative Data

Reference	Extracted data: Summary data & Quotes
Normalisation	
Litzelfelner (1995)	***Summary data:*** - Normalised their issues compared to other families; Did not feel like things were any different at their home than anyone else's ***Quotes:*** - "Does your brother or sister get more attention? 'She gets more attention because she's a girl' 'He gets more attention because he's the baby of the family'." - "Do you all ever have to help around the house more? 'Yes, because my brother works at two jobs and he never has to do anything.' 'Yes, but only because my schedule has more free time than my brother's'." "I do lots more because I'm the oldest, when mom stayed in bed a lot I took over the responsibilities" "I don't think my life is any different because of my brother, yes, he puts a lot of stress into it, but I don't think it's really different from other people's" "Everyone has problems with their family every once in a while" "Everyone comes from a dysfunctional family, to blame that on the problems you're having right now is a cop out."
Garley & Johnson (1994)	***Summary data:*** - Normalised sister's chronic complaints

Appendix F. Reprint of paper 1 publication

Ma, N., Roberts, R., Winefield, H. & Furber, G. (2014) The prevalence of psychopathology in siblings of children with mental health problems: A 20-year systematic review.
Child Psychiatry and Human Development, March, pp. 1-21