Understanding Hoarding Symptoms in Children with Comorbid ADHD

By

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Submitted in partial fulfilment of the requirements for the degree of

Doctor of Psychology (Clinical)

Deakin University

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Abstract

Children and adolescents with attention-deficit/hyperactivity disorder (ADHD) are vulnerable to developing comorbid hoarding disorder with severe negative impacts. Despite this, the comorbidity between hoarding disorder and ADHD in childhood is poorly understood. This thesis is comprised of three studies that together aimed to explore the nature of hoarding symptoms in children and adolescents with comorbid ADHD. A correlated liabilities model was developed that implicated genetic, neurological, and executive functioning components in the comorbidity between hoarding disorder and ADHD. The phenomenology of hoarding symptoms in children with comorbid ADHD was then explored through in-depth interviews with 10 parents of children with ADHD and comorbid clinically significant hoarding symptoms. A core theme of emotional distress emerged in parent reports of the hoarding profiles of children with comorbid ADHD, which appeared to be linked with parental accommodating behaviours, urges to acquire items, and negative impacts on the wider family. The potential role of executive functioning in hoarding symptoms was explored through a pilot study with 37 children aged 8-17 years with ADHD and 26 typically developing controls whose parents completed questionnaires designed to assess their child’s executive functioning, hoarding symptoms, and ADHD symptoms. Results suggested that children with ADHD and clinically significant hoarding symptoms may have poorer executive functioning compared to both children with ADHD without clinically significant hoarding symptoms and typically developing controls. Sixteen of the children with ADHD completed neuropsychological assessments of executive functioning including sustained attention, spatial working memory, planning and inhibition. Performance on these tasks hinted that hoarding symptoms may be associated with poorer spatial working memory, long planning times on simple tasks and short planning times on complex tasks. Combined, these results have important clinical implications for the early detection of hoarding symptoms in children.
and adolescents with comorbid ADHD and for the provision of interventions to prevent the severe symptom progression of hoarding disorder.
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Thesis overview and structure

The vulnerability of children and adolescents with attention-deficit/hyperactivity disorder (ADHD) to developing hoarding disorder is indicated from the reported high rate of comorbidity between these two disorders. Of particular concern, individuals with hoarding disorder and comorbid ADHD experience severe impairments in functioning as a result of hoarding symptoms. Given the early onset and progressive course of hoarding disorder, ADHD could provide a foundation for detecting children who may be on a trajectory to developing hoarding disorder. The overall aim of this thesis was to provide a preliminary examination of the nature of hoarding symptoms in children with comorbid ADHD in order to inform early detection and intervention approaches in this population.

This thesis is in the form of a thesis by publication. It is comprised of three studies that are presented as three papers that are linked thematically around the topic of hoarding in ADHD and structurally through the inclusion of a common introductory chapter, links between the chapters and a summary discussion / conclusion. All chapters of the thesis are in manuscript form, with the text identical to the published paper and those submitted for review. A separate reference list is provided at the end of each chapter.

Following this overview of the thesis structure, Chapter One provides an introduction and overview of background literature as a foundation to the overall aims of the thesis, which are listed at the end of the chapter. A description of hoarding disorder and what is known about its presentation in children is included, as is an overview of the cognitive behavioural model of hoarding disorder. ADHD and, in particular, the comorbidity of ADHD and hoarding in children is described, followed by a summary and rational for the thesis.

The first study is presented in Chapter Two. This study is a critical review of the literature that was undertaken in order to develop a model of the aetiology and maintenance
of comorbid ADHD and hoarding symptoms. Entitled “Hoarding in Attention-Deficit/Hyperactivity Disorder: Understanding the Comorbidity”, it is published in the Journal of Obsessive Compulsive and Related Disorders and is included in journal format as Appendix A. Strong empirical support was found for a correlated liabilities model implicating genetic, neurological, and executive functioning factors in the aetiology and maintenance of hoarding symptoms among individuals with ADHD. This suggests that improving executive functioning may be a necessary addition to interventions for hoarding disorder in individuals with comorbid ADHD.

The findings from this review provide the foundation for the second study (manuscript under review), presented in Chapter Three. Study Two is a qualitative study, in which the phenomenology of hoarding symptoms in children with ADHD and comorbid clinically significant hoarding symptoms as described by caregivers are examined. Semi-structured qualitative interviews were conducted with ten parents of 8-12 year olds with ADHD and clinically significant hoarding symptoms. Six superordinate interconnected themes were identified: emotional distress; parental avoidance and accommodating behaviours; family impacts of hoarding; excessive acquisition and saving; executive functioning; parental insight and intervention.

Given that executive functioning was a factor identified in the correlated liabilities model derived from Study One and was a superordinate theme in Study Two, the aim of the third study (Chapter Four; manuscript under review) was to begin to examine the association between executive functioning and hoarding symptoms in children and adolescents with ADHD. Thirty-seven parents of 8-17 year olds with ADHD and 26 parents of typically developing children who served as a comparison control group completed questionnaires assessing hoarding symptoms, executive functioning, and ADHD symptom severity. A pilot sample of sixteen children from the ADHD group completed neuropsychological assessments.
of executive functioning using the CANTAB, including inhibition, planning, spatial working memory, and sustained attention subtests. Findings suggested poorer global executive functioning in children with ADHD and clinically significant hoarding symptoms, which may be particularly associated with poorer spatial working memory and planning ability. These results provide a preliminary indication of an association between executive functioning and hoarding symptoms in children with ADHD. They need to be interpreted with caution, however, given that it is an underpowered exploratory study.

An overall discussion including a summary of findings, clinical implications, limitations and recommendations for future research is provided as Chapter Five. Although limited by a small sample, the findings from studies two and three provide preliminary support for the correlated liabilities model identified in Study One. In addition to executive functioning difficulties, the findings also indicate the possibility that emotion dysregulation may perpetuate hoarding symptoms in children with ADHD, in particular emotional attachment to items, distress when discarding items, and distress when requests to acquire items are refused by parents. This may have important clinical implications for early detection and intervention approaches for hoarding symptoms among children with ADHD. Specifically, it suggests that the presence of ADHD and executive functioning difficulties may provide a platform for detecting children who may be vulnerable to developing hoarding disorder. Moreover, it provides preliminary support for a combined treatment approach that targets executive functioning difficulties and the cognitive behavioural cycle of hoarding symptoms in children with comorbid ADHD.
Chapter One

Introduction

According to Frost, Steketee, and Tolin (2011), 27% of individuals with hoarding disorder have comorbid attention-deficit/hyperactivity disorder (ADHD). Approximately 8.9% of individuals with childhood ADHD proceed to develop lifetime hoarding symptoms, compared to only 2.7% of those without comorbid ADHD (Fullana et al., 2013). This high rate of comorbidity, together with the negative health, safety, and quality of life impacts that are associated with hoarding disorder (Hall, Tolin, Frost, & Steketee, 2013), point to an urgent need for increased understanding of this comorbidity. Improved understanding of the nature of hoarding symptoms in individuals with comorbid ADHD could inform early detection and intervention approaches that may prevent the severe symptom progression that is commonly evident in hoarding disorder (e.g., Ayers, Saxena, Golshan, & Wetherell, 2010).

While there have been suggestions that common executive functioning deficits may explain the comorbidity between ADHD and hoarding disorder (Fullana et al., 2013; Hall et al., 2013), this claim is yet to be explored in childhood, where hoarding symptoms typically commence (Tolin, Meunier, Frost, & Steketee, 2010). A brief overview of hoarding disorder and ADHD is presented in this introductory chapter as background to the aims of the thesis, including the critical review provided in Chapter Two.

Understanding Hoarding Disorder

Although hoarding has been recognised by clinicians as a disorder for a number of years, Hoarding Disorder has only been officially recognised in the most recent fifth edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5; American Psychiatric Association; [APA], 2013). Traditionally considered as a dimension of OCD (Rasmussen & Eisen, 1989), researchers have more recently identified hoarding as an independent symptom dimension compared to the symptoms of OCD (Pinto et al., 2008) and conceptualised it as a
distinct disorder. In subsequent studies in which the comorbidity between OCD and hoarding disorder has been examined, the overlap between the two disorders occurs in only 18% of those with hoarding disorder (Frost et al., 2011). While the findings from studies on hoarding in OCD have informed research and clinical practice, these studies are limited in their applicability to the majority of individuals with hoarding disorder as they do not have comorbid OCD. Given that hoarding appears to be an independent factor, much of the research has been based on conceptualisation of compulsive hoarding, defined by Frost and Hartl (1996) as (1) the acquisition and difficulty in discarding possessions of limited or no value; (2) cluttered living spaces; and (3) significant distress or impaired functioning as a result of the hoarding symptoms.

Drawing from the definition originally provided by Frost and Hartl (1996), hoarding disorder is defined in the *DSM-5* as a difficulty discarding due to urges to save possessions or due to the occurrence of distress when discarding; accumulation and cluttering of living areas; and significant distress or impairment caused by the symptoms (APA, 2013; see Appendix B). Major reasons for saving objects provided by people who hoard include emotional attachment to items, fear of losing important information and an inflated anticipation of the level of utility of objects (Frost & Hartl, 1996; Kellet, Greenhalg, Beail, & Ridgway, 2010). Although excessive acquisition is not a necessary diagnostic criterion in the *DSM-5*, it is an additional specifier (APA, 2013), on the basis that it may occur in as many as 85-95% of individuals with hoarding disorder (Frost, Tolin, Steketee, Fitch, & Selbo-Bruns, 2009).

**Epidemiology and Impact of Hoarding Disorder**

Although hoarding disorder has been identified in 2.3% of adults (Iervolino et al., 2009) and 2% of adolescents (Ivanov et al., 2013), research into hoarding disorder is still limited. As suggested by the diagnostic criterion of significant impairment in functioning,
these individuals are likely to experience severe and disruptive consequences arising from hoarding disorder. In an analysis of hoarding-related complaints to local health departments in Massachusetts USA, Frost, Steketee, and Williams (2000) reported that 88% of complaints involved individuals living in unsanitary conditions, and 67% of cases had alleged fire hazards. According to a report from the Melbourne Fire Brigade (n.d.), as many as 24% of all preventable fire deaths in the State of Victoria, Australia between 1999 and 2009 were due to hoarding disorder in individuals aged 50 years and over, and Melbourne Fire Brigade responded to 102 hoarding-related incidents between 2012 and 2014 (Worcester Polytechnic Institute, 2014). Severe medical concerns are also more likely in those with hoarding disorder compared to controls, including a higher risk of arthritis, diabetes, obesity, and stroke (Tolin, Frost, Steketee, Gray, & Fitch, 2008). In addition, the effects of hoarding disorder extend to occupational functioning, with Saxena et al. (2011) reporting that in the USA only 15% of 34 adults with hoarding disorder and comorbid OCD were employed at the time of their study.

Beyond these core functioning deficits, individuals with hoarding disorder have been shown to have a reduced quality of life in multiple domains of functioning, including involvement in activities, safety, and life satisfaction (e.g., Saxena et al., 2011; Vorstenbosch et al., 2012). The finding that a poorer quality of life in young adults with hoarding disorder is predicted by hoarding symptoms in childhood (Palermo et al., 2011) highlights the importance of identifying early predictors of hoarding disorder in childhood and adolescence. Greater understanding of the disorder in the early years may inform efforts to prevent the significant quality of life impairments experienced in adulthood. In addition to the effects on the individual, hoarding disorder has a significant impact on society, including utilisation of public health, eviction and fire services, as well as the economic effects arising from high rates of work impairment and unemployment (Tolin et al., 2008). Prevention and early
intervention in hoarding disorder thus has the potential to not only benefit those directly at risk, but also the entire community.

**The Course of Hoarding Disorder**

Hoarding disorder typically has an onset in childhood and early adolescence. In patients with OCD and hoarding disorder, the average age of onset has been reported as 11 years old (Fontenelle, Mendlowicz, Soares, & Versiani, 2004). Both Grisham, Frost, Steketee, Kim, and Hood (2006) and Seedat and Stein (2002) established that the average age of onset of hoarding symptoms was 13 years old, with some cases having symptoms starting as young as two years old. Despite these findings, the focus of research on hoarding disorder has been almost exclusively on adult populations. Hoarding disorder in childhood appears to show a similar clinical picture to that in adulthood (Storch et al., 2007). However, saving and collecting behaviour is more developmentally appropriate in childhood and as such, problematic hoarding symptoms are identified only when these behaviours have an increased severity and cause distress (Storch et al., 2007). Moreover, children may have fewer opportunities for hoarding behaviours due to parental control over their environment and their possessions, thus restricting clutter to the child’s bedroom (Storch et al., 2011).

Hoarding disorder appears to have a progressive course. In a study undertaken by Ayers et al., (2010), symptom severity was shown to be at its lowest in childhood, followed by adolescence, and with a gradual increase in severity until older age. A slight decrease was apparent during 71-80 years of life, followed by a further increase. This study was strengthened by the use of mean severity ratings of any hoarding symptom rather than total hoarding scores to account for the tendency for children to have fewer opportunities to clutter their house when living with parents.

An examination of risk factors for hoarding disorder in childhood is therefore critical to allow prevention efforts, early assessment and treatment of hoarding symptoms as soon as
possible after onset, in order to prevent the development of more severe hoarding symptoms in adulthood. The association between ADHD and hoarding disorder has been previously examined by assessing individuals with comorbid ADHD and hoarding disorder. Given the lower symptom severity in childhood, however, it would appear beneficial for the focus of research to be on the association between ADHD and hoarding symptoms, including those that do not meet clinical diagnostic levels for hoarding disorder.

**Cognitive Behavioural Model of Hoarding Disorder**

The cognitive-behavioural model of hoarding disorder (Frost & Hartl, 1996) proposes potential maintenance factors for hoarding symptoms and may inform understanding of early features associated with hoarding disorder in children with ADHD. This model posits that four components are influential in the development and maintenance of hoarding disorder (Frost & Hartl, 1996): information processing deficits, problems in forming emotional attachments, behavioural avoidance, and inaccurate beliefs regarding the nature and importance of possessions (see Figure 1).

![A cognitive-behavioural model of hoarding disorder by Frost and Hartl (1996).](image-url)
Information processing deficits include difficulties with decision-making, categorisation and organisation, memory, and attention (Frost & Hartl, 1996). It has been consistently demonstrated that these information processing deficits are characteristic of individuals with hoarding disorder (Fitch & Cougle, 2013; Grisham, Norberg, Williams, Certoma, & Kadib, 2010; Hartl et al., 2004; Lawrence et al., 2006).

The second component of the model of excessive emotional attachment to possessions outlined in the cognitive-behavioural model of hoarding disorder has also been found to be a predictor of hoarding symptoms (Wheaton, Fabricant, Berman, & Abramowitz, 2012). As many as 89.7% of individuals with hoarding disorder have difficulty discarding possessions due to their having an excessive sentimental attachment to them (Nordsletten et al., 2013). Emotional attachment to objects is one of many emotion regulation difficulties that are associated with hoarding disorder. Emotion regulation is a multi-faceted concept encompassing the awareness, understanding, and acceptance of emotions, as well as the ability to modulate impulsive behaviours in the presence of negative affect, all in the context of goal-directed behaviour (Gratz & Roemer, 2004). For example, individuals with hoarding disorder have been shown to be more likely to have lower distress tolerance compared to a matched comparison (Timpano, Buckner, Richey, Murphy, & Schmidt, 2009). They are also more likely to have increased anxiety sensitivity, which describes a fear of anxiety (Medley, Capron, Korte, & Schmidt, 2013). In a recent study using structural equation modelling with 241 undergraduates undertaken by Shaw, Llabre, and Timpano (2015), these factors appeared to interact to predict hoarding, with high levels of both affect intolerance and saving beliefs significantly associated with greater hoarding symptom severity. Understanding of these factors was extended through a mediation model developed by Phung, Moulding, Taylor and Nedeljkovic (2015). In a sample of 150 non-clinical adults, Phung et al. (2015) demonstrated
that anxiety sensitivity and negative impulsivity predicted hoarding symptoms, mediated via cognitions about the emotional attachment to possessions.

The cognitive behavioural model also identifies a link between behavioural avoidance and hoarding disorder, whereby individuals with hoarding disorder are likely to avoid discarding, sorting, and decision making (Frost & Hartl, 1996). Although support for the relationship between behavioural avoidance and hoarding disorder has been demonstrated (Wheaton, Abramowitz, Franklin, Berman, & Fabricant, 2011; Wheaton et al., 2012), Fernández de la Cruz et al. (2013) identified that this is likely to be representative of psychopathology in general, and not uniquely associated with hoarding disorder.

The concept of inaccurate beliefs about possessions has also received support (Kellet et al., 2010), with the reasons provided for saving items including anticipating a high level of usefulness of objects and fear of losing important information. Individuals with hoarding disorder have been shown to be more likely to believe in the necessity of perfection and maintaining control over possessions compared to controls (Abramowitz, Lackey, & Wheaton, 2009; Steketee, Frost, & Kyrios, 2003). Few other aetiological and maintenance influences on hoarding disorder have been identified in the literature beyond the cognitive-behavioural model. Given that the direct aetiological causes of hoarding disorder remain unknown, it is important to identify early risk factors and hoarding disorder profiles that can aid in screening individuals who may be at risk for the development of hoarding disorder.

**Current Treatment Approaches for Hoarding Disorder**

Despite having a progressive and persistent course, there is evidence that the symptoms of hoarding disorder in adults can be decreased with appropriate treatment. In particular, Tolin, Frost, & Steketee (2007) reported a reduction in hoarding symptoms in a preliminary evaluation with 14 adults of a cognitive behavioural therapy (CBT) program that was specifically modified for the treatment of hoarding disorder. The approach incorporated
specific techniques to expose individuals to avoided situations, restructure their hoarding-related beliefs, and train them in skills of decision-making, organisation, and problem solving (Tolin et al.). A meta-analysis of 12 treatment studies of CBT for hoarding disorder identified that hoarding disorder symptom severity and functional impairment decreased across studies (Tolin, Frost, Steketee, & Muroff, 2015). The application of a group CBT treatment approach online has also demonstrated effectiveness in reducing clutter and hoarding symptoms in 106 adults with self-identified hoarding problems (Muroff, Steketee, Himle, & Frost, 2010). Among 41 adults who completed a 12-week group CBT intervention, 34% were calculated to show clinically significant reductions in hoarding symptoms (Moulding, Nedeljkovic, Kyrios, Osborne, & Mogan, 2016). It would be beneficial to replicate these results in individuals with a formal diagnosis of hoarding disorder. Alternative treatments have also shown success with adults. For example, participation in a structured facilitated support group was shown to result in decreased hoarding-related beliefs in 22 adults with clinically significant hoarding symptoms compared to 21 waitlist controls (Frost, Ruby, & Shuer, 2012).

Preliminary evidence suggests that hoarding symptoms may be reduced through treatment of both emotion regulation difficulties and cognitive problems, such as information-processing difficulties. In a preliminary case study with a 72 year old, Cermele, Melendez-Pallitto, and Pandina (2001), reported a reduction of hoarding symptoms by treating the cognitive behavioural components of Frost and Hartl’s (1996) model, including both the emotion regulation difficulties and cognitive problems, such as information-processing deficits. Subsequently, a combined treatment approach including cognitive therapy targeting executive functioning and exposure therapy has showed support in 11 older adults with hoarding disorder (Ayers et al., 2014). In a pilot randomised controlled trial, DiMauro, Genova, Tolin, and Kurtz (2014) assessed the effects of cognitive remediation compared to relaxation treatment in 17 adults with hoarding disorder. Participants receiving
the behavioural intervention designed to improve cognitive functioning showed greater improvements in attention abilities compared to the relaxation control group, but no significant differences in hoarding symptom severity. These examples show preliminary support for addressing both emotional and cognitive factors in hoarding disorder, such as emotion regulation and executive functioning.

Emerging evidence supports the use of CBT in treatment of childhood hoarding symptoms. A case study with an 11-year-old boy with hoarding disorder and OCD found reductions in items saved and anxiety about discarding after 11 sessions of CBT (Gallo, Wilson, & Comer, 2013). Similarly, a case study of a nine-year-old girl with hoarding disorder and OCD demonstrated reductions in hoarding symptoms following the implementation of CBT strategies, including psychoeducation, reducing parental accommodating behaviours, and exposure tasks (Ale, Arnold, Whiteside, & Storch, 2014). However a specific treatment for paediatric hoarding disorder is yet to be developed or assessed. It is therefore unclear whether treatment with children with ADHD and comorbid hoarding symptoms should target both cognitive and emotional functioning equally, or whether one aspect, such as cognitive difficulties, should be the primary target of early interventions.

**Overview of ADHD**

ADHD is a common neurodevelopmental disorder in children and adolescents involving symptoms of inattention in tasks and activities, or hyperactivity and impulsivity, such as fidgeting, talking excessively or difficulty waiting their turn (APA, 2013). The Australian prevalence rates of ADHD in 6-17 year olds range from 7.5-11.2% using *DSM-IV* diagnostic criteria (Graetz, Sawyer, Hazell, Arney, & Baghurst, 2001; Sawyer et al., 2001). No Australian prevalence data is available to date utilising the *DSM-5* criteria; however, it is likely that the current prevalence rates are even higher than those evident from *DSM-IV*.
criteria due to criteria changes such as increasing the required age of onset from before seven to before twelve years old and the reduction in the number of criteria needed for individuals older than 17 years (Batstra & Frances, 2012). This illustrates that ADHD is a prominent psychological disorder in children and adolescents in Australia, as in other parts of the world. It is therefore important to examine risk factors for poorer outcomes for children with this disorder and the potential for addressing these in order to minimise such impacts. It has recently been proposed that adult ADHD may be not be a childhood-onset neurodevelopmental disorder (Moffitt et al., 2015). Although a contentious claim, it highlights that ADHD symptomatology may be dependent on developmental stage. Given this, there is a need for research into the relationship between ADHD and hoarding disorder in childhood and adolescence, where ADHD prevalence and presentation may differ to adulthood.

**Comorbidity of ADHD and Hoarding Symptoms in Childhood**

Despite the prevalence and early onset of both hoarding symptoms and ADHD, there is a dearth of research exploring this comorbidity in childhood. Preliminary studies have however indicated that core ADHD symptoms may predict hoarding symptoms in childhood. In particular, inattention may be a strong predictor of hoarding across the lifespan (Hacker et al., 2012; Tolin & Villavicencio, 2011), while hyperactivity/impulsivity may be associated with hoarding in childhood (Hacker et al., 2012) but not adulthood (Anholt et al., 2010; Tolin & Villavicencio, 2011). When examining these relationships among 99 youth with ADHD, Hacker et al. identified that inattentive symptoms significantly predicted the hoarding symptoms of clutter, difficulty discarding, and overall hoarding in children, while hyperactivity/impulsivity was a significant predictor of acquisition and hoarding-related distress. Children with hoarding disorder are therefore likely to acquire items more quickly and impulsively than are those without hoarding disorder (Preston, Muroff, & Wengrovitz,
Despite these explorations of hoarding in childhood, the nature of hoarding symptoms in children with ADHD remains unexplored.

**Summary and Rationale**

There is a critical need to understand and thereby potentially reduce the impact of hoarding symptoms in individuals with ADHD, due to the high level of functional impairment arising from hoarding disorder and the likelihood of compounding negative impact. Despite the high rates of co-occurrence between ADHD and hoarding disorder, little is known about the nature of this comorbidity. A diagnosis of ADHD may provide a foundation for detecting children and adolescents who may be on a trajectory to developing hoarding disorder. Given the young age of onset of hoarding disorder followed by a progressive course and severe impairments, understanding childhood hoarding symptoms in individuals who may be particularly vulnerable is essential. Improving understanding of hoarding symptoms in children with ADHD could inform early detection and intervention approaches that may prevent the significant impacts of hoarding disorder on the individuals, their families, and the wider society. The primary aim of this thesis is to provide a preliminary examination of the nature of hoarding symptoms in children and adolescents with comorbid ADHD. This is examined through three studies.

**Study One Aim**

The aim of Study One is to provide a comprehensive review and evaluation of the existing literature in order to develop a model that may be used to understand the comorbidity between ADHD and hoarding disorder. Various comorbidity models are explored with an evaluation of the empirical evidence supporting these models.

**Study Two Aim**

The aim of Study Two is to explore the phenomenology of hoarding symptoms in children with comorbid ADHD. Given the comorbidity models explored in Study One, it is
necessary to clarify the nature of hoarding symptoms in children with ADHD in order to inform early detection and intervention approaches.

**Study Three Aim**

The aim of the third study is to examine the proposed association between hoarding symptoms and executive functioning in children and adolescents with ADHD. Although several researchers have predicted this association in adults, it is yet to be examined in children and adolescents. It was predicted that 8-17 year olds with ADHD and clinically significant hoarding symptoms would have significantly poorer executive functioning on questionnaire and neuropsychological assessments compared to children with ADHD without hoarding.
References (Chapter One)


Chapter Two

Study One. Hoarding in Attention-Deficit/Hyperactivity Disorder: Understanding the Comorbidity

Abstract

Hoarding disorder has a frequent co-occurrence with attention-deficit/hyperactivity disorder (ADHD). An accurate understanding of the comorbidity between hoarding disorder and ADHD remains unclear but is essential to inform appropriate assessment, prevention and treatment approaches. This paper will provide a review of potential comorbidity models and aetiological mechanisms implicated in both disorders in order to inform understanding of the nature of the comorbidity between hoarding disorder and ADHD. A correlated liabilities model is identified that implicates genetic, neurological, and executive functioning factors in the development and maintenance of hoarding symptoms in individuals with ADHD.

# AUTHORSHIP STATEMENT

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Led the conception of the manuscript, independently conducted the literature search, drafted the manuscript as primary author and undertook the incorporation of revisions suggested by the co-authors.

I declare that the above is an accurate description of my contribution to this paper, and the contributions of other authors are as described below. Signature and date: [Signature and date]

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ii. that there are no other authors according to these criteria,
iii. that the description in Section 4 of my contribution(s) to this publication is accurate,
iv. that the data on which these findings are based are stored as set out in Section 7 below.

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<td>Richard Moulding</td>
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<td>Linda Byrne</td>
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<td>19/01/2016</td>
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Introduction

Although ADHD and hoarding disorder commonly co-occur, relatively little is known about the aetiology of this comorbidity. Hoarding disorder is a new addition to the *Diagnostic and Statistical Manual of Mental Disorders, 5th Edition* (*DSM-5*; American Psychiatric Association, 2013) and is defined as the difficulty discarding items due to urges to save possessions or distress when discarding; accumulation of items and cluttering of living spaces; and significant distress or impairment caused by these symptoms. Prior to its addition in the *DSM-5*, various terms were used to define hoarding disorder, such as compulsive hoarding (Frost & Hartl, 1996). However, the present paper will use the term ‘hoarding disorder’ to reflect the current *DSM-5* designation but will note any important differences in individual studies’ definitions of hoarding disorder.

Among the 2.3% of adults with hoarding disorder (Iervolino et al., 2009), approximately 28% have comorbid ADHD (Frost, Steketee, & Tolin, 2011). This population appears to experience greater deficits in functioning than those with hoarding disorder alone (Hall, Tolin, Frost, & Steketee, 2013), including more difficulties in activities of daily living, increased stress, and higher levels of domestic squalor (Hall et al., 2013). These deficits occur in addition to the hoarding-related impairments of compromised safety (Frost, Steketee, & Williams, 2000), reduced quality of life (Palermo et al., 2011), increased employment and housing difficulties (Saxena et al., 2011), and severe medical issues, including higher risk of arthritis, diabetes, obesity, and stroke compared to individuals without hoarding disorder (Tolin, Frost, Steketee, Gray, & Fitch, 2008).

The severe potential consequences of comorbid hoarding disorder in individuals with ADHD and the high association between the two disorders point to the importance of developing a greater understanding of the features involved in the co-occurrence of these disorders, particularly the causal and maintenance factors. It has recently been suggested that
shared executive functioning deficits may explain the comorbidity between ADHD and hoarding disorder (Fullana et al., 2013; Hall et al., 2013). However, to date, no study has investigated this possibility in a sample of individuals with ADHD with comorbid hoarding disorder. The potential role of executive functioning as a mechanism linking ADHD and hoarding disorder therefore remains unknown.

The present review aims to synthesise the current literature to develop a model that may be used to understand the comorbidity between hoarding disorder and ADHD. To do this, several potential comorbidity models will be explored. The correlated liabilities model will be evaluated by providing a critical analysis of possible evidence of a shared genetic, neurological, and executive functioning aetiology of hoarding disorder among adults with ADHD. Such an analysis may inform understanding of the mechanisms of hoarding symptoms in this population and may highlight clinical implications, including prevention and treatment approaches.

**Overlap between ADHD and hoarding disorder**

There is substantial evidence for an association between ADHD and hoarding disorder. For example, in an assessment of comorbidity in adults with hoarding disorder, 27.8% were found to have comorbid inattentive ADHD, while 13.7% had hyperactive ADHD (Frost et al., 2011). Furthermore, Sheppard et al. (2010) found that 21.9% of adults with hoarding disorder had comorbid ADHD. In contrast, it appears from a meta-analysis of prevalence rates of ADHD using DSM-IV diagnostic criteria that ADHD only affects approximately 5% of the general adult population (Wilcutt, 2012). A further study using retrospective data indicated that a higher proportion of individuals with childhood ADHD later developed hoarding symptoms in comparison to those without childhood ADHD (Fullana et al., 2013). Other studies also suggest an association between the specific symptoms of ADHD and core hoarding symptoms. In particular, inattention appears to be a strong predictor of hoarding
symptoms across the lifespan (Hacker et al., 2012; Tolin & Villavicencio, 2011), while hyperactivity/impulsivity appears to be associated with hoarding symptoms in childhood (Fitch & Cougle, 2013; Hacker et al., 2012).

Despite developing evidence of a frequent co-occurrence between ADHD and hoarding disorder, an accurate understanding of the mechanisms explaining this comorbidity remains unclear. Neale and Kendler (1995) propose several general comorbidity models that may be used to explain the co-existence of hoarding disorder and ADHD. As used previously by Schmitt and Weidinger (2014) in a study of dermatological comorbidities, Table 1 summarises these models in relation to existing knowledge regarding the association between hoarding disorder and ADHD. It is crucial that research is undertaken to establish a valid model of comorbidity that enables accurate diagnosis of comorbidity, as well as differential diagnosis between ADHD and hoarding disorder (Achenbach, 1995). Moreover, research is needed to gain an understanding of the aetiological mechanisms of ADHD and hoarding disorder in order to inform treatment decisions such as identifying specific treatment targets and determining whether the two disorders should be treated sequentially or in parallel (Achenbach, 1995).

Table 1


<table>
<thead>
<tr>
<th>Model</th>
<th>Description</th>
<th>Evidence for ADHD and Hoarding Disorder</th>
</tr>
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<tbody>
<tr>
<td>Chance</td>
<td>The two disorders have separate liabilities and the comorbidity is due to chance alone.</td>
<td>Model rejected: Prevalence of ADHD in those with hoarding disorder is approximately 28% (Frost et al., 2011), which substantially exceeds the expected prevalence of 5% based on community</td>
</tr>
<tr>
<td>Sampling Bias</td>
<td>Individuals with comorbid conditions are more likely to participate in research than those with either disorder alone, particularly in clinical settings.</td>
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<td>----------------</td>
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</tr>
<tr>
<td>Population Stratification</td>
<td>The two disorders have separate risk factors, but each of the risk factors is likely to co-occur in a specific population, such as twin populations.</td>
<td></td>
</tr>
<tr>
<td>Alternate Forms</td>
<td>The two disorders share a single liability that manifests in either disorder based on chance or environmental risk factors that differ across individuals.</td>
<td></td>
</tr>
<tr>
<td>Random Multiformity</td>
<td>Having ADHD increases the probability of having hoarding disorder, despite not having elevated liability for hoarding disorder (or the opposite pattern).</td>
<td></td>
</tr>
<tr>
<td>Extreme Multiformity</td>
<td>Like random multiformity, the two disorders have separate liabilities but having hoarding disorder only occurs at the extreme end of ADHD symptoms (or the opposite).</td>
<td></td>
</tr>
<tr>
<td>Three Independent</td>
<td>The comorbid disorder is distinct from either disorder populations (Wilcutt, 2012).</td>
<td></td>
</tr>
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*Model rejected:* Previous studies have used community samples recruited through general advertisements that are unlikely to carry the sampling bias typically linked to clinical samples (e.g., Frost et al., 2011).

*Model rejected:* Frost et al. (2011) advertised widely in the community and had no twin participants thereby suggesting that population stratification effects were unlikely, yet identifying a high comorbidity.

*Model not rejected:* Preliminary evidence suggests that hoarding disorder and ADHD may share some genetic and neurological aetiological mechanisms. Further understanding of these factors is needed.

*Model not rejected:* Though it remains untested, it is plausible that having ADHD can generate hoarding symptoms, thus some cases of hoarding may be epiphenomena of ADHD, unrelated to liability for hoarding disorder.

*Model rejected:* Several authors have reported that hoarding disorder is associated with mild and sub-clinical ADHD (e.g., Hacker et al., 2012).

*Model not rejected:* Further research is needed to determine if the nature of
Disorders occurring alone. hoarding and ADHD symptoms in those with the comorbid condition represents a disorder that is distinct from ADHD or hoarding disorder alone.

Correlated Liabilities The biological and/or environmental risk factors for both disorders are correlated. Model not rejected: Hoarding disorder and ADHD have shown similar patterns of neural abnormalities in the frontal lobe. Moreover researchers have recently suggested that executive functioning deficits may present a shared risk factor.

Causal Models One disorder is a liability of the other disorder or both disorders cause each other. Model not rejected: It is unknown and thus remains a possibility that ADHD and hoarding disorder cause each other.

On the basis of Neale and Kendler’s (1995) framework and the current understanding of ADHD and hoarding disorder, the comorbidity between the two disorders may be explained by several alternate models, including alternate forms, random multiformity, three independent disorders, correlated liabilities, and the causal models. The alternate forms model would apply if both ADHD and hoarding disorder result from a single underlying liability. Given that no single cause has been identified for either disorder, this model cannot yet be rejected. However, our current understanding of hoarding disorder and ADHD suggests that several different aetiological factors may contribute to the occurrence of each disorder. It is therefore unlikely that the comorbid condition results from a single cause. The aetiological factors will therefore be discussed as they contribute to the correlated liabilities model. The random multiformity model is also plausible, whereby the comorbid condition represents an epiphenomenon of hoarding disorder in those with ADHD.

According to the three independent disorders model, individuals with ADHD plus hoarding disorder may have a disorder qualitatively and aetiologically distinct from ADHD and hoarding disorders. There would therefore be two possibilities for the comorbid condition:
those with ADHD and hoarding disorder who are above the threshold for the liability of each disorder; and those with the distinct disorder of ADHD plus hoarding disorder. Given that the precise aetiology of hoarding disorder and ADHD is unknown, it remains a possibility that some cases of the comorbid condition could be an independent disorder that arises from a third aetiological mechanism. Very little is known about the nature of hoarding symptoms in those with ADHD, and further qualitative explorations and genetic studies of the comorbid condition are essential to explore these models.

Alternatively, the co-occurrence of ADHD and hoarding symptoms may be conceptualised using the Research Domain Criteria, which focuses on dimensional aspects of behavioural and neural circuits in the development and classification of mental disorders (National Institute of Mental Health, 2008). For example, ADHD and hoarding symptoms could be conceptualised as interrelated constructs, or behavioural dimensions, of the broader domain of Cognitive Systems, that co-vary with different degrees of damage to shared neural circuits, genetic, and behavioural factors (Cuthbert & Kozak, 2013; Morris & Cuthbert, 2012). Although these models cannot be rejected, the existing literature on these concepts is not yet established enough to warrant inclusion in the present review. It is also possible that hoarding disorder is directly caused by ADHD in a subgroup of those with hoarding disorder. Although this is also unable to be rejected as a possibility, reciprocal causation seems unlikely given the current indications that the development of hoarding symptoms begin after the onset of ADHD.

The inability to reject the alternate forms, random multiformity, causal and three independent disorder models as explanations for the comorbidity between ADHD and hoarding disorder highlights the need for further empirical investigation of these models. However, recent interest from researchers in the correlated liabilities model suggests its potential in explaining the comorbidity between ADHD and hoarding disorder. On the basis
of the current interest in shared aetiological and maintenance factors in ADHD and hoarding disorder (e.g., Fullana et al., 2013; Sheppard et al., 2010), the aim of the present review is to evaluate existing evidence for a correlated liabilities model, including genetic factors, neurological influences, and executive functioning difficulties. This model suggests that certain factors may predict which individuals with ADHD are likely to develop hoarding symptoms. Risk factors include a family history of hoarding disorder, neurological damage to or abnormal functioning in the prefrontal areas of the brain, and impaired executive functioning. While other articles have focussed on related disorders such as the comorbidity between OCD and ADHD (Brem, Grünblatt, Drechsler, Riederer, & Walitza, 2014), to date none has considered the overlap between ADHD and hoarding disorder, which is essential given that hoarding disorder is neurobiologically distinct from OCD (An et al., 2009; Saxena, 2008), and appears to share some distinct areas of neurological dysfunction with ADHD which differs from OCD (Bush et al., 1999; Saxena et al., 2004).

Aetiological mechanisms implicated in the ADHD-hoarding disorder link

**Genetic factors**

The contribution of genetic components and neural dysfunction associated with ADHD and hoarding disorder has led to a consensus that the aetiology of both disorders includes substantial biological factors. For example, in a comprehensive study of 5022 pairs of twins, Iervolino et al. (2009) reported that genetic factors explained approximately 50% of the variance in hoarding disorder. Samuels, Bienvenu III, et al. (2007) examined hoarding symptoms in 132 relatives of individuals with OCD and hoarding disorder and 259 relatives of individuals with OCD without hoarding disorder. Their results showed that 49% of the relatives of those with hoarding disorder had hoarding symptoms themselves, compared to 33% of the relatives of individuals with OCD without hoarding disorder. Moreover, in a genetic study of 219 families with hoarding disorder, Samuels, Shugart, et al. (2007) established a
significant linkage to hoarding disorder on chromosome 14. Several genes have been associated with hoarding disorder, including BDNF (Timpano, Schmidt, Wheaton, Wendland, & Murphy, 2011), SLC1A1 (Wendland et al., 2009), COMT (Lochner et al., 2005), and neurotrophic tyrosine kinase receptor type 3 (Alonso et al., 2008).

The aetiology of ADHD indicates some overlap with the genetic liability of hoarding disorder. As with hoarding disorder, ADHD has a high heritability, with a meta-analysis of 79 twin and adoption studies establishing that the heritability of ADHD was between 71% and 73% (Nikolas & Burt, 2010). The BDNF gene has also been associated with ADHD and may indicate a shared genetic vulnerability between hoarding disorder and ADHD, although the results are inconsistent with further replications needed (Forero, Arboleda, Vasquez, & Arboleda, 2009). However, the literature on the genetics of ADHD suggests that, like most psychiatric disorders, the cause of ADHD is not attributable to a single gene, but is more likely to be the result of multiple small effects on many common gene variants (Forero et al., 2009).

**Neurological factors**

Shared neural dysfunctions provide further evidence for a biological link between ADHD and hoarding disorder; specifically, dysfunction in the prefrontal cortex, anterior cingulate gyrus, and amygdala. Greater activation in the ventral prefrontal regions and left amygdala is related to hoarding-specific anxiety (An et al., 2009; Mataix-Cols et al., 2003). For example, An et al. (2009) reported that individuals with hoarding disorder displayed greater activation in the bilateral anterior ventromedial prefrontal cortex when asked to imagine discarding specific objects, which was associated with higher anxiety levels, compared to those without hoarding disorder. Likewise, adolescents with ADHD show greater activity in the amygdala when performing a task of subliminal presentation of fearful faces compared to their typically developing counterparts (Posner et al., 2011). The existing
The literature also highlights hypoactivity in the anterior cingulate gyrus common to both disorders. For example, individuals with hoarding disorder have lower metabolism in the dorsal anterior cingulate gyrus compared to controls (Saxena et al., 2004), and hypoactivity in the anterior cingulate cortex associated with decision making about objects (Tolin et al., 2012). Likewise, reduced activation in the anterior cingulate gyrus is associated with ADHD (Smith, Taylor, Brammer, Halari, & Rubia, 2008) in addition to reduced grey matter volume in this region (Amico et al., 2011; Bledsoe et al., 2013).

The prefrontal cortex has also been associated with both disorders. However, opposing patterns of dysfunction are apparent, with some studies reporting a greater activation in the anterior ventromedial prefrontal cortex and ventral prefrontal cortex in individuals with hoarding disorder (An et al., 2009) and a global reduction in activity in the prefrontal cortex in those with ADHD (Smith et al., 2008; Wilson et al., 2013). For example, hoarding disorder has been associated with greater activation in the prefrontal cortex, particularly in the orbitofrontal cortex (Mataix-Cols et al., 2004; Tolin et al., 2009). In contrast, a meta-analysis of 55 fMRI studies reported that adults with ADHD have hypoactivation in the frontoparietal system (Cortese et al., 2012), while Dickstein, Bannon, Castellanos, and Milham (2006) also found similar patterns of hypoactivity within the fronto-striatal circuit in individuals with ADHD.

Beyond these functional deficits, ADHD and hoarding disorder have each been associated with further structural abnormalities in the prefrontal cortex. In an examination of 86 case studies of brain damage undertaken by Anderson, Damasio, and Damasio (2005), hoarding disorder was associated with damage to the mesial frontal region of the brain. Several other case studies have implicated structural damage to the prefrontal cortex in hoarding disorder (Hahm et al., 2001; Volle et al., 2002). Likewise, a systematic review of diffusion tensor imaging studies in ADHD found alterations in white matter in the fronto-
striatal–cerebellar neurocircuitry in patients with ADHD (van Ewijk, Heslenfeld, Zwiers, Buitelaar, & Oosterlaan, 2012). Individuals with ADHD also have a thinner left medial prefrontal cortex (mPFC; Shaw, Lerch, Greenstein, Sharp, & Clasen, 2006) and delayed cortical maturation in this area (Shaw et al., 2007).

Combined, the current literature highlights a shared vulnerability toward functional and or structural abnormalities in several frontal networks in both ADHD and hoarding disorder. As Castellanos, Sonuga-Barke, Milham, and Tannock (2006) discuss, in their recent paper on ADHD, there is considerable intra-individual variability in ADHD, and the same is true in HD (Preston, Muroff, & Wengrovtiz, 2009) while both may be characterised by dysfunction in the circuits driving cognition, motivation and motor behaviour, both internal and external stimuli continue to operate and modify these circuits. The heterogeneity of these disorders means divergence in some areas. For example, the nature of the prefrontal activity dysfunctions in ADHD and hoarding disorder appears to be in conflict, but these differences may have arisen due to a combination of influences specific to one disorder or the other. Further, it is possible that abnormally low and abnormally high activity both result in similar behavioural and cognitive deficits. For example, both high activation of the prefrontal cortex (Ehlis, Bähne, Jacob, Herrmann, & Fallgatter, 2008), and low prefrontal activity (Tsujimoto et al., 2013) are associated with executive function deficits in working memory. Despite this opposite pattern of dysfunction, empirical evidence provides support for a shared neurological factor between ADHD and hoarding disorder, whereby both disorders are particularly vulnerable to functional and structural dysfunction in the prefrontal areas. Clarification is needed, however, on the prefrontal abnormalities in individuals with ADHD with comorbid hoarding disorder to determine whether they reflect the ADHD or the hoarding disorder pattern of dysfunction. Moreover, future research on the neural activity in
hoarding disorder should consider screening for ADHD to ensure an accurate representation of the hoarding disorder pattern of dysfunction is found.

**The role of executive functions**

Executive functions can be broadly described as higher-order capacities of self-regulation that are instrumental to independent, future-oriented intentional behaviours (Barkley, 2001). These include numerous actions such as planning, inhibition, flexibility, organisation, working memory, updating, and mental set-shifting, among others (Ardila, 2008; Barkley, 2001; Chan et al., 2008; Miyake et al., 2000). Deficits in these functions are typically associated with abnormal activation in, or damage to, the frontal lobe (Alvarez & Emory, 2006), an area linking ADHD and hoarding disorder as outlined above, including the prefrontal cortex (Brozoski, Brown, Rosvold, & Goldman, 1979) and the anterior cingulate gyrus (Smith et al., 2006; Smith et al., 2008).

Like ADHD and hoarding disorder, executive functions are influenced by genetic factors, as indicated by gene mapping and a high heritability. For example, genetic factors are reported to explain between 29% and 68% of the variance in executive functions in typically developing adults (Lee et al., 2012; Lessov-Schlaggar et al., 2007; Swan & Carmelli, 2002). Individuals with ADHD similarly show high heritability of executive functioning, at 77% (Coolidge, Thede, & Young, 2000). Comprehensive genetic studies have identified that the DRD4 gene is implicated in both executive functioning (Barnes et al., 2011; Kebir et al., 2009) and ADHD (Gizer, Ficks, & Waldman, 2009), which may indicate that individuals with ADHD have a genetic vulnerability to executive functioning deficits. Though the attention deficits inherent in many individuals with ADHD may contribute to hoarding symptoms, it is likely that additional executive functioning impairments may better explain which 9% of individuals with ADHD will develop hoarding disorder (Fullana et al., 2013). A review of the literature has identified that the executive functions of working memory,
inhibition, and planning have received the strongest support in the hoarding disorder profile and will therefore be discussed in further detail.

**Review method**

In this review, the findings regarding the executive functioning of adults with hoarding disorder are evaluated. Studies were identified through searches in PubMed and EBSCO PsycInfo performed in October 2014, using the search terms executive function or neuropsychological or working memory or inhibition or planning and hoarding. The results were limited to peer-reviewed articles, English language publications, and human studies. The most pertinent articles from the above search were selected, as well as two articles from their reference lists, based on their clinical relevance to the present paper.

**Empirical evidence in hoarding disorder**

**Working memory**

Working memory has frequently been examined in the context of hoarding disorder, with findings indicating that spatial working memory difficulties are particularly salient in hoarding disorder. The visuospatial sketchpad is a distinct sub-system of working memory that is responsible for the brief storage of visual information, and creating and manipulating mental images (Baddeley & Hitch, 1974). Researchers have consistently reported that the contrasting verbal memory system is not associated with hoarding disorder (Grisham et al., 2007; Hartl et al., 2004; McMillan et al., 2013; & Tolin et al., 2011). This discrepancy between visual and verbal memory is not entirely unexpected given that discarding items involves the visual assessment of locations and the manipulation of this visual information to plan to discard items. Grisham et al. (2007) examined the differences in neuropsychological functions between 30 adults with hoarding disorder and 60 controls. Participants with hoarding disorder had significantly poorer visual memory span compared to controls, suggesting decreased spatial attention and spatial working memory abilities in individuals
with hoarding disorder (Grisham et al., 2007). Likewise, Hartl et al. (2004) found that individuals with hoarding disorder showed significantly poorer recall on the Rey–Osterrieth Complex Figure Test (RCFT) after a 20 min delay compared to controls, demonstrating poorer spatial memory encoding. Despite these findings, participants in these samples all had a diagnosis of OCD, which may have inflated their results due to co-occurring OCD symptoms. However, research has indicated that individuals with OCD with hoarding disorder have an executive functioning profile similar to those with hoarding disorder without OCD (Morein-Zamir et al., 2014); therefore these sampling methods are unlikely to significantly impact results.

In contrast, when comparing a sample of individuals with hoarding disorder to test norms, McMillan et al. (2013) unexpectedly found that individuals with hoarding disorder performed significantly better on the spatial span backward. It remains unclear whether their results represent a better developed spatial working memory system in individuals who hoard or are a reflection of methodological issues. Specifically, the authors suggest that individuals with hoarding disorder may have an improved visuospatial sketchpad due to navigating more cluttered living spaces. In contrast, the results may be attributable to methodological issues such as the lack of a non-clinical control group or learning by participants, with no counterbalancing in the study meaning that spatial span backwards was the fourth memory span test administered to each participant (McMillan et al., 2013). There is therefore a need for clarification regarding the relationship between visual working memory and hoarding disorder.

Planning

Beyond working memory, individuals with hoarding disorder have exhibited weaknesses in planning tasks compared to controls, which is likely to be reflected in their difficulties in planning to discard and acquire items. According to McMillan et al. (2013),
individuals with hoarding disorder had significantly more perseveration errors on the Wisconsin Card Sorting Test (WCST) than age-matched population norms, suggesting poorer cognitive flexibility, which can affect planning. Similar results were reported by Grisham, Norberg, Williams, Certoma, and Kadib (2010) in a sample of individuals with hoarding disorder, using the Cambridge Neuropsychological Test Automated Battery (CANTAB) Stockings of Cambridge computerised assessment, which required participants to mentally plan a series of moves before commencing them. Participants with hoarding disorder solved fewer problems within the minimum number of moves than both the non-clinical controls and the mood or anxiety disorders group (Grisham et al., 2010).

Individuals with hoarding disorder have also exhibited difficulties in the ability to plan a series of actions when completing the RCFT, including drawing disjointed elements of the figure when copying (Hartl et al., 2004) and scoring significantly lower on an organisational score (Tolin et al., 2011). Hoarding symptoms are also correlated with significantly lower planning scores on the RCFT during the copying dimension of this task (Pinto et al., 2011). Pinto et al. (2011) examined these associations in a sample of individuals with OCD, which could inflate their results due to co-occurring OCD symptoms. However, Tolin et al. (2011) unexpectedly found that participants with hoarding disorder had no significant differences in the number of moves taken to complete the Tower of London task when compared to controls. The medium effect size in the findings by Tolin et al. (2011) leads the authors to suggest that significant results may be found with a larger sample size. However, Hartl et al. (2004) identified significant results in a hoarding disorder sample size of 22 compared to the 27 participants with hoarding disorder in the study by Tolin et al. (2011). It is therefore unclear whether the inconsistent results by Tolin et al. (2011) are due to the different neuropsychological tasks used or if they demonstrate an inconsistent pattern of planning abilities among those with hoarding disorder.
Planning difficulties may affect the acquiring and discarding dimensions of hoarding disorder, with individuals less likely to plan the specific items they will obtain (Nordsletten, Fernández De La Cruz, Billotti, & Mataix-Cols, 2013), and more likely to thus accumulate more items than needed or wanted. This may result in a further inability to discard these acquired items, whereby individuals with hoarding disorder struggle to mentally plan and execute the set of actions required to discard items. It would be of interest for further research to examine which symptom dimensions were associated with planning, to provide more accurate treatment targets. Despite spending less time planning items to acquire, individuals with hoarding disorder are likely to show the reverse pattern when discarding items, sometimes planning the steps needed to discard items in great detail. For example, evidence from case studies found that some individuals with hoarding disorder are highly perfectionistic in their planning (Ayers, Bratiotis, Saxena, & Wetherell, 2012). These results are supported by consistent findings of an association between high levels of perfectionism and hoarding symptoms (Frost & Gross, 1993; Frost et al., 2013; Martinelli et al., 2014; Timpano et al., 2011). Though initially this process may appear helpful, it can reflect an inability to prioritise appropriately, paradoxically increasing the difficulty of the plan (Ayers et al., 2012), and is therefore likely to affect treatment outcomes (Muroff, Steketee, Frost, & Tolin, 2013). It also highlights the tendency to focus on specific details, with a difficulty in attending to the more global picture, which is also demonstrated in the studies examining completion of the RCFT (Hartl et al., 2004).

**Inhibition**

A third factor of executive functioning, inhibition, has also been associated with hoarding disorder. Inhibition involves deliberately suppressing a dominant, automatic, or unwanted response (Mirsky, 1996). Specifically, individuals with hoarding disorder have shown significantly higher commission errors on tasks of sustained attention compared to
nonclinical controls (Grisham et al., 2007; Rasmussen et al., 2013) and an anxiety group (Grisham et al., 2007). These findings suggest that inhibition difficulties in individuals with hoarding symptoms are beyond those seen in individuals with heightened anxiety. This is consistent with several studies demonstrating increased impulsivity in individuals with hoarding disorder compared to controls (Fitch & Cougle, 2013; Hall et al., 2013; Tolin & Villavicencio, 2011), which may arise from poor inhibition. Older adults with hoarding disorder have similar difficulties of inhibition, with increased non-perseveration errors on the WCST compared to controls (Ayers et al., 2013).

However, several studies have not found significant between-group differences in inhibition tasks, including the CANTAB Affective Go/No-Go task (AGN; Grisham et al., 2010) and the Stop Signal Reaction Time Task (SSRTT; Blom et al., 2011). These contrasting results may be indicative of the different neuropsychological tasks used to assess inhibition. Although the CANTAB AGN task assesses response inhibition, Grisham et al. (2010) used mean latency to correct response and total omissions as outcome measures, which are likely to be more indicative of set-shifting skills, a function that may be unimpaired in individuals with hoarding disorder (e.g., Lawrence et al., 2006; Tolin et al., 2011). In contrast, the previous findings of significant between-group differences have assessed inhibition using commission errors, which were not reported in the study by Grisham et al. (2010). The results by Blom et al. (2011) may suggest that previous findings of poorer inhibition in individuals with hoarding disorder do not transfer to difficulties in motor inhibition.

**Empirical evidence of shared executive functioning difficulties in ADHD**

The existing literature indicates that the reviewed executive functioning difficulties in hoarding disorder are shared difficulties with ADHD. Like adults with hoarding disorder, individuals with ADHD have also demonstrated spatial working memory difficulties
compared to controls. These include significantly more between search error scores on tasks of spatial working memory (Alderson et al., 2013; Clark et al., 2007; Dowson et al., 2004; McLean et al., 2004), and significantly higher strategy scores (Dowson et al., 2004; McLean et al., 2004), suggesting a deficient use of a consistent search sequence to locate items. The consistent finding of planning difficulties in adults with hoarding disorder is also the case in individuals with ADHD. Compared to controls, adults with ADHD exhibited significantly lower percentage of correct first choices on a Tower of London task (McLean et al., 2004), significantly shorter planning times (Bramham et al., 2009; Young et al., 2007), no significant increases in planning times on more complex tasks (Young et al., 2007), and create less elaborate plans (Fuermaier et al., 2013). In relation to hoarding symptoms, adults with ADHD may therefore spend inadequate time planning to discard items, which may then lead to increased clutter of items.

Response inhibition difficulties are a final executive function that is common to both hoarding disorder and ADHD and several authors have argued that inhibitory control is the foundational core deficit in ADHD (e.g., Boonstra et al., 2010; Pazvantaoğlu et al., 2012). Like individuals with hoarding disorder, adults with ADHD have demonstrated significantly more commission errors on the Continuous Performance Test (Epstein et al., 2001; Murphy et al., 2001), significantly slower responding on the go/no-go task (McLean et al., 2004), have less accurate responses, and more variable reactions than controls (Bozorgpour, Klorman, & Gift, 2013). This may specifically help to explain the link between ADHD and hoarding disorder, whereby adults with ADHD have difficulties inhibiting intentional actions that result in increased hoarding symptoms such as acquiring unneeded items. Combined, these studies illustrate that adults with ADHD may be particularly vulnerable to executive functioning difficulties that are shared with hoarding disorder, which may partly explain the association between the two disorders.
Summary

This review of empirical findings has shown support for the correlated liabilities model in explaining the comorbidity between ADHD and hoarding disorder. The key executive functions of spatial working memory, planning, and inhibition have been specifically implicated in the hoarding disorder profile. A review of these functions in ADHD has found that adults with ADHD may be particularly vulnerable to difficulties in each of these areas. Despite these findings, and the suggestions that executive functioning deficits may explain the co-occurrence of these two disorders, very few studies have evaluated the potential of a shared vulnerability model in individuals with ADHD and comorbid hoarding disorder. Moreover, the discussed genetic and neural influences on each of hoarding disorder, ADHD, and executive functioning may provide further explanation for the association between ADHD and hoarding disorder (see Figure 2). For example, executive functioning may provide a marker for genetic vulnerability toward hoarding symptoms in individuals with ADHD.

Figure 2.

*Executive functioning difficulties as a mechanism of association between ADHD and hoarding disorder.*
Clinical implications

The findings of this review provide support for a shared vulnerability between hoarding disorder and ADHD, including executive functioning difficulties, genetic vulnerability, and neurological impairments. This has several implications for current treatment approaches. Firstly, hoarding disorder has an early age of onset (Tolin, Meunier, Frost, & Steketee, 2010) with a severe symptom progression (Ayers, Saxena, Golshan, & Wetherell, 2010) that results in particularly disruptive consequences for those with comorbid ADHD. Executive functioning difficulties could therefore provide a target for early screening to identify those individuals who may be on a trajectory to develop hoarding disorder in already at-risk individuals with ADHD. This could result in early intervention to prevent the severe symptom progression and associated impaired functioning in later life. However, further research is needed to determine if the discussed weakness in executive functioning in individuals with hoarding disorder compared to controls is due to objective deficits in these areas.

Secondly, if executive functioning difficulties contribute to the maintenance of hoarding symptoms, treatment implications are suggested. There is currently some evidence that hoarding disorder can be successfully treated using cognitive behavioural therapy (CBT) incorporating specific treatment techniques to expose individuals to avoided situations, restructure their hoarding-related beliefs, and train them in skills of decision making, organisation, and problem solving (Muroff et al., 2010; Tolin et al., 2007). However, the current review has indicated that specific executive functioning weaknesses may continue to maintain hoarding symptoms, particularly in those with comorbid ADHD. As such, a combination treatment of hoarding symptoms and executive functioning deficits is indicated. Several studies have shown effective treatment of executive dysfunction in individuals with
ADHD. These include the use of computerised spatial working memory training programs (Gray et al., 2012; Klingberg et al., 2005), behavioural interventions such as response cost and token approaches that show improvements in inhibition (Siniatchkin et al., 2012), and CBT techniques focusing on self-instruction that improve each of the discussed executive functioning deficits (Miranda, Presentación, Siegenthaler, & Jara, 2011).

Developing evidence in hoarding disorder provides support for a combined treatment approach. For example, a recent pilot study with older adults with hoarding disorder examined a novel treatment of behavioural therapy combined with cognitive rehabilitation, including six sessions targeting executive functioning, such as problem solving, organisational skills, prospective memory, and cognitive flexibility (Ayers et al., 2014). The preliminary findings of a high reduction in hoarding symptom severity following this treatment suggest that combining treatment of hoarding symptoms with executive functioning training may result in improved outcomes for individuals with hoarding disorder (Ayers et al., 2014). In a case analysis of four adults with hoarding disorder who did not have comorbid ADHD, Rodriguez et al. (2013) reported that methylphenidate extended release reduced inattention in all cases and showed a modest reduction in hoarding symptoms in two of the four subjects. Given the prefrontal hyperactivity in hoarding disorder, the positive result with stimulant medication treatment may be somewhat unexpected. However, this highlights the ongoing need for further clarification of the neural activity and medication responses in individuals with hoarding disorder, particularly in those with comorbid ADHD. Further research is required to determine if stimulant medications frequently used in ADHD treatment are effective in treating hoarding symptoms. The long-term prevention of severe hoarding symptoms in individuals with ADHD and comorbid hoarding disorder may thus require a combination treatment of hoarding symptoms as well as interventions that specifically target the executive functions identified in the hoarding disorder profile.
Suggestions for future research

The research reviewed has focused on the influence of genetic factors, neurological impairments, and executive functioning weaknesses on hoarding disorder and ADHD alone; however the nature of these factors in individuals with ADHD and comorbid hoarding disorder remains unknown. It may be helpful for future research to determine the extent to which individuals with comorbid ADHD and hoarding disorder have similar or different genetic compositions, neurological, and executive functioning profiles compared to individuals with either disorder alone. For example, do individuals with comorbid ADHD and hoarding disorder have hyper- or hypo-activity in the prefrontal regions? Likewise, it appears that ADHD and hoarding disorder share common hoarding symptoms that may correspond with opposing planning difficulties. Do the planning impairments in those with comorbid ADHD and hoarding disorder reflect the hoarding disorder profile of complex and perfectionistic plans, or do they follow the ADHD pattern of reduced time spent planning complex tasks?

Clarifying these profiles in those with comorbid ADHD and hoarding disorder could inform treatment approaches. For example, individuals with comorbid ADHD and hoarding disorder may require training to allocate more time on complex plans to discard items, rather than reducing the perfectionistic planning inherent in hoarding disorder. Future attempts to develop and assess a treatment model of hoarding disorder that integrates traditional CBT approaches with interventions that promote executive functioning may reduce the negative impact of hoarding disorder among those with ADHD. In order to improve screening for hoarding disorder and identify appropriate treatment targets, future researchers might also test the correlated liabilities model presented in this review to determine if it should be extended to include other elements common to both disorders, such as emotional regulation. Finally, the available research on executive functioning in the context of hoarding disorder has almost
exclusively focused on adult populations. Given that both executive functioning and hoarding disorder are highly dependent on developmental stage (Ayers et al., 2010; De Luca et al., 2003), there is a need to explore these relationships in children and adolescents.
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Chapter Three

Study Two. Phenomenology of Hoarding in Children with Comorbid ADHD: The Perception of Parents

In study one, it was suggested that a correlated liabilities model implicating genetic, neurological, and executive functioning factors may account for the aetiology and maintenance of comorbid ADHD and hoarding symptoms. It was also evident from the empirical evidence, however, that the random multiformality model (Neale & Kendler, 1995) cannot be rejected, which suggests that hoarding symptoms may be epiphenomena to ADHD. It is thus possible that ADHD may produce behaviours such as clutter and difficulty discarding items that appear to similar to hoarding symptoms, but actually result from core ADHD symptomatology. Indeed it appears from initial research that the core ADHD symptoms of inattention and hyperactivity/impulsivity predict hoarding symptoms in childhood (Hacker et al., 2012; Tolin & Villavicencio, 2011). Moreover, Storch et al. (2011) suggested that children with ADHD and comorbid hoarding symptoms may not have a typical hoarding disorder profile of emotional attachment to objects and significant distress when discarding items. It therefore remains unclear whether hoarding behaviours in children with ADHD are accompanied by the distress and urges to save that are necessary for a diagnosis of hoarding disorder (APA, 2013). Although the multiformality model remains untested with regard to the comorbidity between ADHD and hoarding disorder, it serves to highlight the clinical need to better understand the nature of clinically significant hoarding symptoms in children with comorbid ADHD. This is particularly important in childhood, as hoarding symptoms frequently have an onset in childhood, show a progressive course, and are associated with chronic negative consequences in adulthood (see Burton, Arnold, & Soreni, 2015).
Improved understanding of the phenomenology of hoarding symptoms in children with comorbid ADHD is essential to informing early detection and intervention approaches. For example, if the concerns held by parents regarding hoarding symptoms in children with comorbid ADHD are related to core ADHD symptomatology or associated executive dysfunction, these may be appropriate treatment targets. In contrast, if hoarding symptoms among those children with comorbid ADHD appear similar to a typical hoarding disorder profile and include emotional factors, cognitive behaviour therapy for hoarding disorder may be the recommended treatment approach (e.g., Tolin, Frost, & Steketee, 2007). The phenomenology of hoarding in children with comorbid ADHD are explored from the perspective of parents in this next paper.
AUTHORSHIP STATEMENT

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<td>Fiona Lynch</td>
<td>School of Psychology</td>
<td>Understanding hoarding symptoms in children with comorbid ADHD</td>
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Led the conception of the manuscript, independently conducted the literature search, designed the methodology, conducted the data analysis, interpreted the data, drafted the manuscript as primary author and undertook the incorporation of revisions suggested by the co-authors.

I declare that the above is an accurate description of my contribution to this paper, and the contributions of other authors are as described below.

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Abstract

Individuals with ADHD and comorbid hoarding disorder are vulnerable to severe consequences from hoarding symptoms. Despite this, and the early onset of hoarding disorder, the nature of early hoarding symptoms in children with comorbid ADHD is unknown. We therefore explored the phenomenology of hoarding symptoms among children with ADHD and comorbid clinically significant hoarding symptoms through parental perceptions. Ten parents of 8-12 year olds with ADHD and clinically significant hoarding symptoms completed in-depth semi-structured interviews. The data was analysed using Interpretative Phenomenological Analysis. Six superordinate interconnected themes were identified: emotional distress; parental avoidance and accommodating behaviours; family impacts of hoarding; excessive acquisition and saving; executive functioning; parental insight and intervention. In contrast to previous suggestions, these findings highlight that emotional distress may be core to the hoarding disorder profile of children with ADHD and appeared to be linked to the remaining themes. This has important implications for health practitioners who may consider conceptualising, assessing, and treating hoarding symptoms in children with comorbid ADHD using a cognitive behavioural model of hoarding disorder.


Phenomenology of hoarding in children with comorbid attention-deficit/hyperactivity disorder (ADHD): Perceptions of Parents.
Introduction

Hoarding disorder is a debilitating condition defined in the *Diagnostic and Statistical Manual of Mental Disorders Fifth Edition (DSM-5; American Psychiatric Association [APA], 2013)* as a difficulty in discarding items regardless of their actual value due to urges to save possessions, distress when discarding, and with the resulting clutter causing significant impairments in functioning. Hoarding disorder has an onset in childhood (Tolin, Meunier, Frost, & Steketee, 2010) and descriptions of childhood hoarding disorder suggest a similar picture to that of adulthood (Storch et al., 2011). Notable exceptions to this are that clutter among children with hoarding disorder may be restricted to the child’s bedroom (Storch et al., 2011) and that acquisition of items may be less prominent in children (Plimpton et al., 2009). Difficulty discarding items therefore seems to be the key indicator of childhood hoarding disorder (Plimpton et al., 2009). Further understanding of hoarding disorder in childhood is needed, however, in order to inform efforts from health practitioners to prevent its chronic and impairing consequences (Burton, Arnold, & Soreni, 2015).

Historically, hoarding disorder was considered to be a variant of obsessive compulsive disorder (OCD; Rasmussen & Eisen, 1989). However, it has been demonstrated that only 18% of those with hoarding disorder meet criteria for OCD (Frost, Steketee, & Tolin, 2011), and hoarding is now conceptualised as a distinct disorder by researchers, as well as in the *DSM-5* (APA, 2013; Pinto et al., 2008). In contrast, over 27% of individuals with hoarding disorder have comorbid attention-deficit/hyperactivity disorder (ADHD; Frost et al., 2011). In a study of 155 individuals aged 4-82 years, Sheppard et al. (2010) found that 21.9% of individuals with childhood-onset hoarding OCD had comorbid ADHD. Although this study is benefited by a large sample with a broad age range, its focus on individuals diagnosed with OCD limits its generalisability to individuals with ADHD and hoarding symptoms without OCD. This shortcoming is addressed in a study by Frost et al. (2011) who,
in using *DSM-5* diagnostic criteria for hoarding disorder, established that 27.8% of
individuals with hoarding disorder had comorbid inattentive ADHD, while 13.7% met the
criteria for hyperactive ADHD. In an epidemiological study of 2963 individuals with ADHD
who had hoarding disorder across nine European countries, Fullana et al. (2013) reported that
8.9% of individuals with childhood ADHD had lifetime hoarding symptoms, compared to
only 2.7% of those without ADHD. Although the rates of hoarding disorder in individuals
with ADHD are lower than reported rates of ADHD in those with hoarding disorder, ADHD
in childhood may provide a foundation for detecting children vulnerable to hoarding
symptoms. These studies consistently demonstrate that there is an association between
ADHD and hoarding symptoms. Of concern, individuals with comorbid hoarding disorder
and ADHD are vulnerable to increased difficulties in activities of daily living, higher levels
of domestic squalor, and increased overall stress (Hall, Tolin, Frost, & Steketee, 2013).

It has been suggested that the core ADHD symptoms and the executive functioning
deficits common among those with ADHD may help to explain the high comorbidity of
hoarding symptoms (Fullana et al., 2013; Hacker et al., 2012; Hall et al., 2013). This may
indicate support for the multiformity model of comorbidity (Neale & Kendler, 1995), which
suggests that hoarding symptoms may be an epiphenomena of ADHD, rather than arising
from a separate aetiological mechanism (for discussion, see Lynch, McGillivray, Moulding,
& Byrne, 2015). However, this model is yet to be systematically evaluated in the comorbidity
between ADHD and hoarding disorder. Accordingly, children with ADHD may have
symptoms of clutter and difficulty discarding items that are phenotypically similar to
hoarding symptoms, but lack the distress and the urges to save items that are necessary for a
diagnosis (APA, 2013). This contention is supported by the clinical experience of Storch et al.
(2011) who noted that children with ADHD and comorbid hoarding symptoms typically do
not have a strong emotional connection with their possessions and do not display significant
levels of distress when items are discarded. If this is accurate, executive functioning retraining (e.g., Ayers et al., 2014) may prove to be a more effective treatment approach for individuals with ADHD and hoarding symptoms compared to cognitive behavioural therapy (CBT), which is currently favoured when treating hoarding disorder (Muroff, Steketee, Himle, & Frost, 2010; Tolin, Frost, & Steketee, 2007). Despite the early onset of both hoarding symptoms (Tolin, Meunier, et al., 2010) and ADHD (APA, 2013), as well as the high rates of this comorbidity in childhood (Shepperd et al., 2010), the phenomenology of this comorbidity in childhood remains a relatively underexplored area.

There is an urgent need to enhance understanding of the nature of hoarding symptoms in children with comorbid ADHD in order to inform and to promote identification and early intervention with these children. In particular, this may shed light on the extent to which hoarding symptoms in children with ADHD are akin to hoarding disorder, or lacks the attachment to possessions as suggested by Storch et al. (2011). The aim of the present study was to describe the phenomenology of hoarding symptoms among children with ADHD and comorbid clinically significant hoarding symptoms. Specifically, what is the experience of hoarding symptoms in children with ADHD and clinically significant hoarding? Given that poor insight is typical of children with hoarding symptoms (Storch et al., 2007; Tolin, Fitch, Frost, & Steketee, 2010), we instead utilized parents as the key informants of the phenomenology of hoarding symptoms in their children within the context of the family environment.

Method

Participants

A purposive sample was selected of 10 parents (9 female, 1 male) of 8 to 12 year olds with ADHD and clinically significant hoarding symptoms. The selection of a homogenous sample is recommended for phenomenological research (Smith & Osborn, 2008) in order to
optimally understand the lived experience of individuals with a particular phenomenon (Creswell, 2013). Participants were recruited from a larger quantitative project exploring executive functioning and hoarding in children, which had a sample size of 22 parents at the time of recruitment for the current study. Parents whose children scored above 40 on the Children’s Saving Inventory ($M=59$, $SD=10.29$), a level considered reflective of clinically significant hoarding symptoms (Frost & Hristova, 2011), were invited to participate in the present study. All children had received a diagnosis of ADHD by their paediatrician. Those with a diagnosis of an intellectual disability were excluded. Of the 11 participants who were eligible to participate, 10 consented to take part in the present study. The remaining participant could no longer be contacted due to change of contact details. The children of parents interviewed included 8 males and 2 females and were aged between 8 to 12 years old ($M=9$ years 3 months, $SD=1$ year 3 months). All children were taking psychotropic medication, including methylphenidate, clonidine, and fluoxetine. Of the ten children, five had comorbid diagnoses including two with oppositional defiant disorder (ODD), one child with generalised anxiety disorder, one with autism spectrum disorder (Asperger’s), and one with comorbid anxiety and ODD.

**Materials**

**Demographics Questionnaire.** Participants completed a demographics questionnaire, which assessed age, sex, medications, and psychological disorders.

**Children’s Saving Inventory.** The Children’s Saving Inventory (CSI; Storch et al., 2011) was used to assess the presence of hoarding symptoms. The CSI is based on the Savings Inventory-Revised (Frost, Steketee, & Grisham, 2004) and is a 23-item parent-rated questionnaire that examines hoarding symptoms across four subscales: Difficulty Discarding; Clutter; Acquisition; Distress/Impairment. Parents rated each question on a 5-point scale of the frequency and severity of hoarding symptoms in the previous week, from 0 (**none/not at**
all/never) to 4 (almost all/extreme/ very often). For example, parents were asked “How often does your child decide to keep things he/she does not need and has little space for?” or “How much of your home is difficult to walk through because of your child’s clutter?”. The CSI produces a total score that can range from 0 to 92, with higher scores indicating more hoarding symptoms. A total score above 40 is considered within the clinical range of likely hoarding problems on the Saving Inventory Revised (Pertusa et al., 2010) and was used to identify children who are likely to display clinically significant levels of hoarding symptoms in the present study. The CSI has strong internal consistency ($r = .84-.96$), test-retest reliability ($r = .85-.92$), and inter-item correlations and has been validated in 8-17 year olds, with evidence of convergent, discriminant, and construct validity (Storch et al., 2011).

**Semi-structured Interview.** Participants completed an in-depth semi-structured telephone interview. Participants were asked five core questions detailed below. Probes were used to gain more detailed information where necessary, as recommended by Smith and Osborn (2008). The duration of the interviews ranged from 10 minutes to 27 minutes.

1. Tell me about the items (name of child) keeps.
2. Tell me everything about discarding (name of child)’s possessions from the moment you or he/she decide to get rid of the item(s), through to the outcome.
3. Tell me about (name of child)’s experience of acquiring new items.
4. Describe your concerns about (name of child)’s clutter, difficulty discarding, or acquiring items.
5. Tell me about the development of (name of child)’s clutter, difficulty discarding items, and acquiring items, from the moment you first noticed these behaviours, to now, and any changes over time.
Procedure

Participants completed the demographic questionnaire and CSI as part of a larger quantitative study. Parents whose child scored above 40 on the CSI and who had consented to be contacted for future research received an invitation to participate in the present study and an explanatory statement detailing the study. Participants were contacted by telephone within two weeks of mailing the explanatory statement to determine their interest in taking part in the present study and to arrange a time for the telephone interview (see Appendix C). Verbal consent was obtained prior to commencing the telephone interview. The data was collected through in-depth semi-structured telephone interviews conducted by the first author. Telephone interviews were chosen due to the nation-wide locations of participants. Each interview was recorded with consent from the participant. Ethics approval was granted by the host university’s human research ethics committee (see Appendix D).

Data Analysis

All interviews were de-identified and transcribed verbatim before being analysed using interpretative phenomenological analysis (IPA; Smith, 1996) methodology as detailed by Smith and Osborn (2008). The aim of IPA is to explore the participant’s experience and perceptions of the phenomenon through dynamic interactions with the researcher (Smith, 1996). As such IPA was chosen as it is best able to answer the research question of the experience of hoarding symptoms in children with ADHD and clinically significant hoarding. Each transcript was read several times by the first author and initial annotations were made in the margin. These initial responses were transformed into concise phrases, which captured the emerging themes of the text. Connections between emerging themes were then analysed to develop clustering of superordinate themes. This methodology was repeated for each transcript. Individual participant themes were used to develop a final list of superordinate themes. IPA acknowledges that the researcher plays an active and dynamic role in the
collection and interpretation of the data, where the data presented is the best representation possible of the individual’s experience considering the researcher’s subjective interpretations (Smith & Osborn, 2008). To increase trustworthiness of the interpretations, several standards of conduct for qualitative research were employed (Creswell, 2013; Yardley, 2000), including: transparency, such as detailing participant accounts through the use of quotations to support themes; peer review to provide the researcher with a critique of the methods, meanings, and interpretations made; triangulation of data, through the use of a structured self-report questionnaire and telephone interview. A small sample size is recommended for IPA in order to allow detailed interpretative accounts of the cases examined (Smith & Osborn, 2008).

**Results**

Six superordinate themes emerged from the IPA: (i) emotional distress; (ii) parental avoidance and accommodating behaviours; (iii) family impacts of hoarding; (iv) excessive acquisition and saving; (v) executive functioning; (vi) parental insight and intervention. In addition to these themes, several participants identified a common pattern of the course of hoarding for this sample. Most participants reported an early onset of hoarding symptoms, ranging from three to seven years of age. Parents described a large variety of items acquired with no common themes, including straws, used scratchy tickets, pieces of paper, little car parts, boxes, rocks, small toys, bottle caps, empty containers, wrapping and tags, jewellery, and clothing items. The majority of parents first noticed their child’s distress when discarding items, but then retrospectively identified earlier problematic acquiring behaviours that had gone unnoticed. A progressive worsening of symptom severity was noted, with parents identifying that their child acquired more items and had increasing difficulty with discarding as they age. Seven participants identified a family history of undiagnosed hoarding symptoms in multiple family members, including four mothers with hoarding symptoms, four fathers and four extended-family members. The first five participants were asked if they had noticed
any changes in hoarding behaviours concurrent with pharmacological treatment of ADHD. This question was removed for subsequent participants due to its leading nature. Of the initial participants, however, two had noticed no changes with ADHD treatment, one reported that hoarding symptoms had worsened since commencing treatment which was attributed to developmental changes, one reported reduced urges to acquire, and the other reported their child was “slightly” more organised with items when taking medication.

**Theme one: Emotional distress**

All parents reported that their child had strong emotional distress when hoarding items. Nine of the ten participants reported that their child exhibited high levels of emotional distress when discarding, ranging from crying to physical aggression in some children. When describing throwing an item away, one mother stated “I would get tears and tantrums and meltdowns when I wanted to throw it away”. For many children, this emotional reaction extended beyond the time spent discarding and was not avoided by discarding the items without the child present. For example, one mother noted:

> Generally I do it when she’s at school. Sometimes she’ll notice if I’ve been in her room and cleaned it up. She’s like ‘where’s this, and where’s this?’, that sort of thing. Yeah so she just gets quite aggressive and things like that.

These experiences indicate that for some children, the emotional reactions when discarding were beyond emotion dysregulation and represent a more meaningful, sentimental attachment to objects. One mother stated “I’m always surprised how attached he can be to inanimate objects”. Another mother described her child’s experience of this emotional attachment when discarding items:

> He gets a really different look on his face. And it’s like a very maternal look. And it’s as if he’s patting a really cute baby rabbit. He goes all doey eyed…He looks like he’s very attached emotionally to it and that taking it away would break his heart.
Theme two: Parental avoidance and accommodating behaviours

Parental avoidance and accommodating behaviours emerged as a superordinate theme present among all parents. Most parents identified that they discarded items when their child was at school. For example, one mother stated:

Other times I’ll sneakily throw it out. Other times I will just go and put it in her room; that little bit of rubbish and then wait until I decide I’m going to clean her room up and then I’ll sneakily throw it out. So she thinks that I’ve put it in her room; she doesn’t go looking for it.

Parents appeared to avoid exposing themselves and their child to the distress associated with discarding. One mother explained this avoidance as follows:

I don’t tend to involve him with the decision making about discarding things because he gets worked up and starts to get agitated about wanting to keep everything. So I kind of end up being sneaky about it and sneak things away gradually.

Parents also noted they accommodated hoarding symptoms, particularly acquisition of items. Many parents identified that this accommodating was due to difficulty saying ‘no’ to their child, and appeared linked with avoiding the emotional distress associated with refusing acquiring. One parent stated “But then on the times that I’m not really sure and ok, maybe it’s five dollars, then I let him get it…I’m quite weak like that. So is my husband, we don’t do ‘no’ very well”. One mother noted that accommodating acquisition may be maintained by associated increased gratitude towards parents: “Then he’s really happy and I’m the best mum in the world”. High levels of emotional reactivity by children often resulted in perceived sense of a loss of parental control over the situation and over hoarding symptoms, which contributed to parents accommodating these behaviours. One parent stated that her child “won’t let me throw them away”, while another stated “you can’t stop him (acquiring items)…and I think he knows that”. One mother noted:
Once he gets it in his head he really wants something, he really, he’s able, I don’t know how, but he’s over the years managed to make it a very difficult life for me if I don’t get the thing that he wants.

Descriptions by several parents also identified that parents with low tolerance of hoarding symptoms were likely to resist their child’s urge to acquire, whereas parents with high levels of tolerance of hoarding symptoms may inadvertently accommodate these behaviours. One mother noted:

I’m probably quite accepting of things so probably for a long, like the last few years I don’t even bother having a discussion about it because I just think: ‘well he likes keeping empty boxes, or the bits and pieces’ and I just let it accrue.

Despite attempts by some parents to resist their child’s desires to acquire items, third parties, particularly grandparents, often accommodated hoarding symptoms. For example, one mother stated “I also found out my mum…sends a lot of stuff home with him too. And he told me that grandma told him not to tell mummy”.

Theme three: Family impacts of hoarding

Eight participants reported that the hoarding symptoms had a significant negative impact on their child’s functioning, on parents and siblings, and on family dynamics including the parent-child relationship. The emotional impact of hoarding symptoms on parents was particularly salient, where parents expressed feeling “embarrassed”, “upset”, “traumatised”, “grumpy”, “frustrated”, and “angry”. For example, when explaining a situation in which she resisted her son’s desires to acquire, one mother stated “I’m still traumatised by the whole incident for a week because, you know, I feel completely embarrassed by everyone staring at us and the behaviour and everything”.

The emotional impact on parents appeared to be linked to the child’s emotional distress, with more intense reactions by children resulting in increased parental distress. One
parent also noted the reciprocal nature of this relationship, where the parent’s emotional state could also affect the child’s emotional reactions, stating “I think it just depends on the mood that she’s in and the mood that I’m in as to how she copes with it (discarding items)”.

According to a number of interviews, disagreement about the management of hoarding symptoms caused conflict in the relationship between parents. For example:

My husband is even worse at saying ‘no’ than I am, but he comes home and he complains to me about how he can’t take (name of child) anywhere because (name of child) always wants to buy stuff. And then I’m like ‘well it’s not my fault’, so I think then there might be some couple strain over the issue.

For many families, the lack of organisation and subsequent clutter impacted the entire family environment. One parent noted that the clutter of items was “invading the whole house environment…and so even in his own room I’m the one that’s imposed the organisation” and another stated she was concerned of the risk that “people are going to fall over them”. Hoarding symptoms also impacted on the individual child’s functioning, leading to difficulty finding, using, and organising items, particularly in the school context. For example, one mother stated “He gets to a point where he covers every single surface and nothing’s in any particular order and he struggles to use it”. In addition to the impact on the individual parents and child, many parents identified that the hoarding symptoms placed a strain on the parent-child relationship. One parent described her interaction with her son when she tried to discard an item: “You get abused, he starts yelling at you and he’ll take it and he’ll go hide it”.

A subtheme of secrecy in the parent-child relationship emerged from many interviews. Several parents identified that during the past few years their child had become “a bit more secretive” about items and would retrieve items from bins after they had been discarded. These behaviours also highlight a strong sentimental attachment to items that are considered
of high value to the child, which distinguishes these hoarding symptoms from that of impulsive acquisition. A major impact on the parent-child relationship appeared to be the parental difficulty understanding their child’s reasoning for saving or acquiring items. One mother noted that she was particularly confused that her child “likes collecting what I think is rubbish”. Another parented stated “That’s when she tries to explain that she needs it, why she needs it…Really basic reasons and reasons that aren’t valid for me, for her to be able to keep it”.

**Theme four: Excessive acquisition and saving**

Nine of the ten participating parents described that there were a large variety of items acquired and saved, with indiscriminate acquisition. The majority of items were small, and were acquired frequently from a range of sources including schools, friends or family, doctor’s clinics, and restaurants. Some children had an urge to acquire items that appeared driven by their impulsivity and rapid emotional attachment to objects. For example, one mother stated:

So what I’m saying is, and I think it’s because of the ADHD, in that moment he feels the need to have that thing right now. Like ‘I desperately need this, my life cannot continue’. I can see him thinking this you know; ‘my life cannot go forward unless I have this thing’. Right? And he’s crazy for it.

Some children appeared to consider items as unique pieces requiring saving, despite their functional similarity to other objects. For example, one mother stated “Like he got upset about a straw the other day. He wanted to keep it…because it was a black straw and ‘there’s not many black straws’”. Parents were particularly concerned by the acquisition and saving of items that they considered rubbish, which appeared to distinguish their child’s collecting behaviour from that of other children. When comparing her child to his peers, one mother noted that “Whereas (child’s name) doesn’t perceive a box that he’s gotten a present from,
you know, he’s unwrapped it; they would see that as rubbish and he, that may as well be a gift as well”.

Once acquired, parents noted that their children gave many different reasons for saving items. The most common reason parents reported was that their child states they need the item or are still using it. Some children appeared to receive comfort from having the items and knowing that the items are there. Several parents also noted that their child saved items due to fear of losing important information. One mother stated:

He often says ‘oh it’s got the proper name on it’, um like the toy item. Or ‘it’s got more details on it that I need’ or it might have a website link or an app link or something like that. He says he needs it, all that information he has to keep.

Once acquired, parents identified that their children frequently forgot about the items unless they were requested to discard the item. For example, one parent noted:

I think he just forgets about things a lot of the time. He has to have it in the moment, and then forgets, and just forgets why he got it or forgets why he had it, and can’t process the ‘well I’ll just throw it out’ Right? ‘It’s served its purpose’.

**Theme five: Executive functioning**

Seven of the ten parents discussed the influence of their child’s difficulties with executive functioning on hoarding symptoms, including the areas of organisation, memory, inattention, and impulsivity. Parents described their children as having difficulties with several organisation skills, such as categorising and clustering items. Organisational difficulties appeared to have a significant impact on the level of clutter, and was frequently highlighted as a factor that parents were concerned about. When discussing her child’s items, one mother noted that “They just end up in a mess really. Not really organised. I’m looking at her room, her room is so messy at the moment I can’t even get in to look”. Several parents
reported that they were concerned that their child’s difficulty with organisation would impact their hoarding symptoms and functioning in the future. One mother stated:

I guess I’m just more concerned about his organisational skills for the future. You know, like he’s going to go to high school eventually or get a job eventually. He needs to be able to manage things and let things go, as we all do.

In contrast, poor memory recall of items appeared to reduce clutter by allowing parents to discard items when their child had forgotten about them. One mother contrasted this to her child’s typically developing sibling who had “a memory like an elephant”, which lead to her avoiding throwing his items out. As one mother stated:

If I throw that away without him being there, he mightn’t ever think about it again for another, you know, one day when someone mentions something he might remember it and then go looking for it.

In addition to organisation and memory, several parents noted that their child had difficulty sustaining their attention on the task of discarding items and that they were distracted by items around the home, leading to increased acquisition and clutter. Parents also highlighted that increased impulsivity appeared to be linked with excessive acquisition:

I mean at that moment he feels like that is something that he, it’s just an impulsive, like because he’s got ADHD he’s impulsive about his emotions. So at that very moment, that is the most important thing in the world.

**Theme six: Parental insight and intervention**

Several parents identified that they had difficulty defining as atypical the hoarding behaviours children had when they were young, due to the lack of available normative comparisons. This often resulted in parents not noticing the severity of hoarding symptoms until children were older, particularly among first-born children:
It probably wasn’t until the younger children have gotten older and then I see the difference. That’s generally how things work with (child’s name). That for us, we don’t see him as unusual until we see what the other two do or don’t do and then we realise the difference.

The high levels of emotional distress displayed by their child was often the first indicator to parents that their child’s behaviours may be atypical. For example, when describing her child’s difficulty discarding items at approximately four years old, one mother stated that “I would get tears and tantrums and meltdowns when I wanted to throw it away…and this is when I started realising that I needed a strategy to help manage this stuff”. This difficulty understanding the severity of their child’s hoarding symptoms may have resulted in reduced implementation of intervention strategies. Five of the ten parents identified that they had employed several strategies to reduce the impact of hoarding symptoms, including imposing organisation on their child’s environment, placing boundaries on where items can be kept, and educating their child on the reasons for discarding. For example, one mother stated “So I try and explain to her that as she’s getting older, she doesn’t need to keep these baby toys and that there’s other little kids and babies out there that do need stuff”. For some families, this process is time consuming, which is exacerbated by the ADHD symptoms which meant that repeated explanations were required in order to increase the child’s understanding. As one parent noted:

I spend probably a few weeks preparing him when I know that I have like a day off or you know, something…so for the last few I remind him every day: ‘oh (child’s name), we’re going to get into your room soon, we’re going to tidy it up’. And I do this every day for probably about two or three weeks so that he knows that that’s what’s going to be happening.
Parents who engaged in these strategies noticed that it reduced the level of their child’s distress when discarding. Two parents also noted long-term reduction in hoarding symptoms through parental early intervention. For example, one mother stated:

I do want to say though that I’m glad it’s being looked at and recognised because I noticed it so early and I think if I hadn’t intervened early with strategies, I could be dealing with a much bigger problem…I think early intervention is key.

Discussion

The purpose of this study was to describe the phenomenology of hoarding symptoms among children with ADHD and clinically significant hoarding symptoms through the perceptions of their parents. The emergent themes identified that the children with ADHD and comorbid hoarding symptoms experienced significant distress when discarding or attempting to acquire items that appeared to be particularly associated with parental accommodating behaviours, family impacts of hoarding symptoms, perceived sense of loss of parental control over hoarding symptoms, and was likely to provide an indicator to parents that behaviours may be atypical.

Parents also described high levels of sentimental attachment to inanimate objects by their children, mirroring findings that in 89.7% of adults with hoarding disorder, emotional attachment to items results in significant distress when discarding possessions (Nordsletten, Fernández De La Cruz, Billotti, & Mataix-Cols, 2013). Although emotion regulation difficulties are prominent in children with ADHD and could contribute to the emotional distress (Seymour, Chronis-Tuscano, Halldorsdottir, Stupica, & Owens, 2012), the reported pattern of strong emotional attachment to objects that results in increased distress when discarding appears to be more consistent with hoarding disorder (e.g., Nordsletten et al., 2013). These findings contrast statements by Storch et al. (2011) that emotional attachment and distress when discarding are not often observed in children with ADHD and hoarding
symptoms. Combined with the reports that children with ADHD had strong urges to acquire and save items, these accounts seem to describe a typical presentation of hoarding disorder beyond the accumulation of items and clutter that may be expected from the core ADHD symptoms of impulsivity and inattention. In particular, parents reported their child with ADHD and clinically significant hoarding symptoms provided reasons for saving items that are typical of a hoarding disorder profile, including the anticipated utility of objects, fear of losing important information, and emotional attachment to objects (Frost & Hartl, 1996; Kellett, Greenhalgh, Beail, & Ridgway, 2010).

Indeed, the emergent themes more generally support the application of Frost and Hartl’s (1996) cognitive behavioural model of hoarding disorder to hoarding symptoms in children with ADHD. The parent informants identified each of the four components influencing hoarding symptom development and maintenance that are highlighted in this model, including: emotional attachment to possessions; overvaluation of the importance of possessions; information processing deficits; and behavioural avoidance. In addition to the findings on emotional attachment and over-valuation of possessions, the influence of information processing deficits, which we referred to as executive functioning difficulties, was also supported by parent report. This finding adds weight to the emerging evidence of an association between executive functioning and hoarding symptoms in children with ADHD (e.g., Fullana et al., 2013; Lynch et al., 2015). For example, previous studies have identified a significant association between hoarding disorder and difficulties with organisation (Tolin et al., 2007), memory (see Woody, Kellman-McFarlane, & Welsted, 2014), and impulsivity (Hall et al., 2013). Interestingly, while memory difficulties in adults may contribute to a fear of losing and discarding items (Hartl et al., 2004), our study suggests that it appears to aid parental discarding of their child’s items, thereby reducing clutter symptoms in childhood.
Behavioural avoidance is the final component of Frost and Hartl’s (1996) model that is present in adults with hoarding disorder. This involves avoidance of the negative emotions associated with discarding items, resulting in increased hoarding (Timpano, Shaw, Cougle, & Fitch, 2014; Wheaton, Abramowitz, Franklin, Berman, & Fabricant, 2011). This may extend to parental avoidance through accommodating hoarding symptoms in children with ADHD (Drury, Ajmi, Fernandez de la Cruz, Nordsletten, & Mataix-Cols, 2014; Wilbram Kellett, & Beail, 2008). In the paediatric OCD and anxiety literature, parental accommodation has been shown to occur through parental behaviour change in order to help a child avoid negative emotional states (Lebowitz et al., 2013). The present findings also support the identified association between parental accommodation and high levels of hoarding symptom severity and emotional distress in children (Lebowitz et al., 2013). Although some parents noted that they accommodate hoarding symptoms in order to avoid their child’s distress, others reported a long-term reduction in symptom severity/distress when they resisted their child’s urges to save and acquire. The potential role of parental accommodation in the development and maintenance of clinical symptoms such as hoarding thus requires further exploration (Lebowitz et al., 2013). However, parents may also be motivated to accommodate their child’s behaviour in order to avoid their own distress arising from efforts to prevent their child’s hoarding symptoms and the emotional sequelae that arise from this. Alternatively, prior research has suggested that parents may reduce hoarding symptoms by discarding their child’s items or by refusing to allow a child’s acquisition of items, which typically results in increased externalising symptoms in children (Storch et al., 2007; Storch et al., 2011).

In addition to enhancing knowledge about the nature of hoarding symptoms in children with comorbid ADHD, the themes identified in this study increase understanding of the family impact of childhood hoarding symptoms. Previous studies have identified that the social-emotional impacts of hoarding symptoms on relatives may be equivalent to the impact
experienced on the individuals with hoarding disorder themselves (Drury et al., 2014). Specifically, relatives of individuals with hoarding disorder show significant impairments due to emotional problems, poor emotional wellbeing, and increased physical pain (Drury et al., 2014). This finding is receiving greater clinical and research recognition, and has prompted the development of a Family Impact Scale for Hoarding, which assesses the family accommodation and burden that may be specific to hoarding disorder (Nordsletten et al., 2014). While previous studies have focused on adult populations, the present account describes similar emotional and functional impairments experienced by parents of children with hoarding symptoms. The emotional impact for parents appeared to be influenced by their difficulty understanding hoarding symptoms, which is consistent with two prior qualitative studies of the experiences of family members of individuals with hoarding disorder (Sampson, 2013; Wilbram et al., 2008). For example, parents reported a conflict between their perception of the value of an item and their child’s strong urge to save it. Previous studies have also acknowledged the difficulty experienced by parents in defining problematic hoarding symptoms in the absence of developmentally appropriate normative comparisons (Frank et al., 2014). Finally, the reports from parents that hoarding symptoms in their children reduced after the introduction of intervention strategies is encouraging and provides preliminary support for the development of an early intervention approach to hoarding symptoms in children with ADHD.

Although the present study has informed understanding of hoarding symptoms in children with ADHD and has added significant weight to a conceptualisation of hoarding disorder according to the cognitive behavioural model, there are several limitations to acknowledge. While the use of informant reports may provide more accurate details of hoarding symptoms (Tolin, Meunier, et al., 2010), it limits the ability to explore the specific cognitive and emotional patterns that may have contributed to the described symptoms.
Moreover nine of the ten parents were female, which may have added gender effects to answers provided. A second limitation is that the study used a purposive sample of those who had identified high levels of hoarding symptoms on parental-report measures. This may have increased the likelihood that the parents endorsed emotional and functional impacts of their child’s hoarding symptoms. However, this also allowed the researchers to explore the nature of hoarding symptoms in children with the comorbid condition. This paper therefore sought not to confirm that these symptoms were present, but to describe the experience of these symptoms in children with ADHD and comorbid hoarding symptoms.

Third, the study also utilized a limited sample of informants for 10 children with ADHD, however, the sample size is consistent with other studies in hoarding disorder using the in-depth qualitative approach of IPA (e.g., Wilbram et al., 2008). Finally, the children in the present sample had not received a formal diagnosis of hoarding disorder. Although this was advantageous in that it allowed exploration of early hoarding symptoms prior to clinical intervention, the interpretations made relate to sub-clinical hoarding symptoms and not hoarding disorder per se. However, the findings are consistent with the increasing emphasis in the literature on understanding the dimensional nature of symptoms, compared to the previous focus on distinct categorical disorders (National Institute of Mental Health, 2008). This approach also allowed us to examine the clinical picture of those with high hoarding symptoms who have been diagnosed with ADHD (i.e., reflecting our core question of the nature of hoarding symptoms in ADHD). In contrast, relying on those with an existing hoarding clinical diagnosis may have begged the question (i.e., a clinical interview means that the phenomenology of their hoarding disorder is identifiable in the first instance, and thus the interviews would have been less illuminating). While it is expected that the current results may be transferable to other contexts (Yardley, 2000), further quantitative studies are needed to examine the generalizability of the emerging themes to the wider population.
Although this study provides some useful preliminary findings about the association between these two disorders, the limitations of the design are acknowledged. In particular, the hypotheses that hoarding behaviour equates to hoarding disorder and an epiphenomena of ADHD may not necessarily be mutually exclusive. A more rigorous design and larger and more diverse sample is recommended to test the hypothesis that these are indeed distinct disorders.

**Conclusions and Clinical Implications**

This study explored the phenomenology of hoarding symptoms in children with comorbid ADHD. Parents’ descriptions of their child’s behaviours included high levels of emotional distress associated with hoarding symptoms, which appeared to be linked to parental accommodating behaviours, impaired child and family functioning, and early identification and intervention of hoarding symptoms. Parental and clinician awareness of the potential emotional distress when discarding or attempting to acquire items may therefore improve identification of early hoarding symptoms in children with comorbid ADHD. In contrast to previous suggestions that emotional factors were not prevalent in the hoarding profile of children with ADHD, these findings suggest that hoarding symptoms in children with ADHD may be best understood using the Frost and Hartl (1996) cognitive behavioural model of hoarding disorder. This has important clinical implications for health practitioners, who may consider intervention approaches for hoarding symptoms among children with comorbid ADHD that include treatment specifically targeting the unhelpful cognitive-behavioural cycle of hoarding disorder (e.g., Tolin, Frost, & Steketee, 2007). It is possible that reducing the high levels of emotional reactions in children with ADHD and hoarding symptoms may subsequently decrease parental accommodating behaviours, thereby further reducing hoarding symptoms. Further research is needed to determine if early identification
and intervention for hoarding symptoms results in reduced symptom progression, particularly in children with ADHD who may already be vulnerable to hoarding disorder.

References (Chapter Three)


Chapter Four

Study Three. Executive Functioning and Hoarding in Children with Comorbid Attention-Deficit/Hyperactivity Disorder

It was apparent from Study Two that some children with ADHD show a typical hoarding disorder profile, resulting in distress and negative impacts for both the individual and their family members. The determination of the signs and correlates of hoarding symptoms in childhood is therefore critical in order to inform early assessment and treatment approaches. This could assist in the prevention of the severe symptom progression of hoarding disorder in children with ADHD who may be particularly vulnerable to heightened levels of functional impairment arising from hoarding disorder. As reported in Study One, there is substantial evidence in support of the claim that hoarding disorder and ADHD may share executive functioning difficulties and specifically identified weaknesses in inhibition, planning, spatial working memory, and sustained attention in the hoarding disorder profile. Further support for the potential association between executive functioning and hoarding symptoms in ADHD is evidenced by the superordinate theme of executive functioning identified in Study Two. Moreover, it has been suggested by researchers in this area that executive functioning deficits may inform understanding of the comorbidity between ADHD and hoarding disorder (Fullana et al., 2013; Hall, Tolin, Frost, & Steketee, 2013). This hypothesis, however, is yet to be tested in children. Approximately 50% of children with ADHD have executive function deficits (Lambek et al., 2011), yet less than 10% have clinically significant hoarding (Fullana et al., 2013). It is therefore unclear if children with ADHD and comorbid hoarding symptoms have an executive functioning profile that is distinct from those with ADHD alone, where executive functioning may already be impaired. The contention regarding the potential association between executive functioning and hoarding in children and adolescents with ADHD is further explored in Study Three.
# AUTHORSHIP STATEMENT

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<tr>
<td>Fiona Lynch</td>
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<td>Understanding hoarding symptoms in children with comorbid ADHD</td>
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Led the conception of the manuscript, independently conducted the literature search, designed the methodology, conducted the data analysis, interpreted the data, drafted the manuscript as primary author and undertook the incorporation of revisions suggested by the co-authors.

I declare that the above is an accurate description of my contribution to this paper, and the contributions of other authors are as described below.  
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Abstract

Theoretical models implicate executive functioning in the comorbidity between hoarding symptoms and ADHD. The purpose of the current study was to examine whether executive functioning in children with ADHD is associated with hoarding symptom severity. Parents of children with ADHD (n = 37) and typically developing controls (n = 26) completed questionnaires assessing hoarding, executive functioning, and ADHD symptom severity. We piloted key neuropsychological tests on a small number of sixteen children with ADHD including inhibition, planning, spatial working memory, and sustained attention. Parents of children with ADHD and clinically significant hoarding reported more executive functioning difficulties compared to children with ADHD without hoarding and typically developing controls. The results hint towards an association between hoarding symptom severity and performance on neuropsychological assessments of planning and spatial working memory abilities. These preliminary findings suggest that hoarding symptoms in children with ADHD may be associated with specific difficulties in executive functioning.

Introduction

Hoarding disorder is associated with considerable functional impairment, including severe medical issues (Tolin, Frost, Steketee, Gray, & Fitch, 2008), compromised safety (Frost, Steketee, & Williams, 2000), reduced quality of life (Palermo et al., 2011), and employment and housing difficulties (Saxena et al., 2011). First included in the 5th edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5; American Psychiatric Association [APA], 2013), hoarding disorder is characterized by compulsive acquisition and difficulty discarding possessions, leading to the accumulation of clutter in living spaces, resulting in significant distress or impairment (APA, 2013; Frost & Hartl, 1996). Hoarding disorder typically has an onset in childhood or early adolescence followed by increased symptom progression over the lifetime (Ayers, Saxena, Golshan, & Wetherell, 2010; Tolin, Meunier, Frost, & Steketee, 2010). However, hoarding research has predominantly focused on adult populations.

On the basis of data reported by Frost, Steketee, and Tolin (2011) in a sample of 217 adults meeting criteria for hoarding disorder, there may be as many as 27% of adults who meet criteria for hoarding disorder with comorbid attention-deficit/hyperactivity disorder (ADHD). In a prevalence study of 155 individuals with OCD, Sheppard et al. (2010) reported that 35.5% of participants with ADHD and OCD had comorbid hoarding disorder. Despite the severe consequences of hoarding in those with ADHD (Hall et al., 2013), the nature of the association between the two disorders remains unclear. Emerging evidence suggests that executive functioning deficits may be associated with the development of hoarding symptoms in those with ADHD (Fullana et al., 2013; Hall et al., 2013; Lynch, McGillivray, Moulding, & Byrne, 2015). Executive functions are higher-order capacities that regulate intentional behaviours, including working memory, planning, inhibition, and sustained attention (Barkley, 2001; Miyake et al., 2000). Both individuals with ADHD and hoarding disorder
show impairments in the same neural substrates that underlie executive functioning, although
the direction of activity levels is sometimes opposing (see Lynch et al., 2015). Specifically,
executive functioning is particularly dependent on the frontal lobes (Alvarez & Emory, 2006),
areas which are abnormally underactive in ADHD (Wilson et al., 2013) and overactive in
hoarding disorder (An et al., 2009). Children and adolescents with ADHD show executive
functioning deficits compared to controls, including demonstrating poorer spatial working
memory (Cockcroft, 2011; Toplak, Bucciarelli, & Jain, 2009; Vance, Ferrin, Winther, &
Gomez, 2013), planning (Toplak et al., 2009), inhibition (Miller, Ho, & Hinshaw, 2012;
Toplak et al., 2009), and sustained attention (Günther, Konrad, De Brito, Herpertz-Dahlmann,
& Vloet, 2011; Miller et al., 2012).

Executive dysfunction similar to that apparent in children and adolescents with
ADHD has also been shown in individuals with hoarding disorder. For example,
neuropsychological assessments performed on adults with hoarding disorder have shown that
they have significantly poorer spatial working memory compared to adults without hoarding
disorder, including poorer spatial span (Grisham, Brown, Savage, Steketee, & Barlow, 2007;
Miller et al., 2012) and poorer spatial memory encoding (Hartl et al., 2004). Difficulty in
spatial working memory is expected to be associated with difficulty discarding items, given
the visual processing of information required to discard. Individuals with hoarding disorder
have also shown difficulties in planning compared with controls, with more perseveration
errors on the Wisconsin Card Sorting Test (McMillan, Rees, & Pestell, 2013), fewer
problems solved on the CANTAB Stockings of Cambridge task (Grisham, Norberg, Williams,
Certoma, & Kadib, 2010), and difficulties displayed when copying the Rey-Osterrieth
Complex Figure Test (Hartl et al., 2004; Pinto et al., 2011; Tolin, Villavicencio, Umbach, &
Kurtz, 2011). However, the planning difficulties in individuals with hoarding disorder appear
to be contrary to those in ADHD with unknown effects on those with the comorbid condition.
Specifically, individuals with ADHD spend reduced time planning tasks (Bramham et al., 2009; Fuermaier et al., 2013) while the high levels of perfectionism typical of hoarding disorder (e.g., Martinelli et al., 2014; Tolin et al., 2011) may be associated with longer perfectionistic planning times. It is therefore possible that individuals with ADHD may have enhanced difficulty planning to discard or acquire items, thus increasing their vulnerability to hoarding symptoms.

Adults with hoarding disorder display more commission errors on sustained performance tasks compared to controls, suggesting difficulties in inhibition (Grisham et al., 2007; Rasmussen, Brown, Steketee, & Barlow, 2013). Inhibition involves suppressing an automatic response to a stimulus (Mirsky, 1996), and may be particularly associated with difficulties resisting urges to acquire items in hoarding disorder. However, there are some inconsistencies in results, with Grisham et al. (2010) and Blom et al. (2011) failing to find significant differences in inhibition between individuals with hoarding disorder and controls. Further research is therefore needed to clarify the nature of the relationship between inhibition and hoarding symptoms, particularly in childhood where inhibition abilities may differ depending on developmental stage (Rezazadeh, Wilding, & Cornish, 2011). Finally, a number of studies have found consistently that individuals with hoarding disorder are particularly impaired in their sustained attention abilities, including on continuous performance tasks (Grisham et al., 2007; Tolin & Villavicencio, 2011), the Wisconsin Card Sorting Test (McMillan et al., 2013), and behavioural reports of sustained attention in both adults (Hall et al., 2013; Tolin & Villavicencio, 2011) and children (Hacker et al., 2012).

Discarding items requires planning, sustained attention and decision making, which may be impaired in some children with ADHD, therefore increasing their hoarding symptoms. These results highlight the importance of exploring executive functioning across both performance tasks and questionnaires, as performance tasks provide an important assessment of
impairment, but may not approximate more complex multi-step tasks required in a child’s naturalistic environment (Chan, Shum, Touloupoulou, & Chen, 2008). In comparison, questionnaires may provide a more ecologically valid indication of a child’s everyday functioning in their natural environment (Gioia, Kenworthy, & Isquith, 2010).

It therefore appears from the literature that individuals with ADHD and executive functioning impairments may be at risk of comorbid clinically significant hoarding symptoms. However, several studies have also demonstrated an association between hoarding disorder and core ADHD symptoms of inattention and hyperactivity/impulsivity (e.g., Hacker et al., 2012; Tolin & Villavicencio, 2011). It is therefore unclear whether the potential association between executive functioning and hoarding symptoms in individuals with ADHD is specific to executive functioning difficulties, or is dependent on ADHD symptom severity. No study to date has examined the potential association between executive functioning difficulties and hoarding symptoms in children and adolescents with ADHD. Exploring this may improve earlier detection of hoarding symptoms among children with ADHD in order to prevent the severe hoarding symptom progression in children vulnerable to developing hoarding disorder. It is proposed that shared neural deficits may predispose individuals to executive functioning difficulties which may be associated with both ADHD and hoarding symptoms.

The aim of the present paper was to begin to examine the association between hoarding symptoms and executive functioning, examining these constructs in a sample of 8-17 year olds with ADHD and typically developing controls. It was predicted that children with ADHD and clinically significant hoarding symptoms would demonstrate poorer executive functioning compared to both children with ADHD without hoarding and typically developing controls. It was also predicted that neuropsychological assessments of inhibition, planning, spatial working memory, and sustained attention would be significantly associated with hoarding symptoms in children with ADHD.
Method

Participants

The participants were 37 children with ADHD and 26 typically developing controls, all recruited through advertisements in the community. All participants in the ADHD group had received a diagnosis of ADHD according to the DSM (APA, 2013 or its predecessor). ADHD diagnosis was confirmed by viewing a copy of the diagnostic report where available, or details of the practitioner who provided the diagnosis. Participants were excluded if they had a diagnosed intellectual disability. Parents of thirteen children with Autism Spectrum Disorder (ASD) initially completed the study with modified instructions to rate hoarding items that were not due to restricted interests typical of ASD, in accordance with the DSM-5 (APA, 2013). However, children with comorbid ASD scored significantly higher on the Children’s Saving Inventory and Behavior Rating Inventory of Executive Function compared to the remaining ADHD sample, suggesting that they may not have been part of the intended population, and they were thus excluded from analyses. Children with ADHD were aged 8 to 17 years (M age = 10.98, SD = 2.37), with 29 males and 8 females. According to the parental informants, 24 of the children with ADHD had comorbid conditions, including oppositional defiant disorder, anxiety disorders, obsessive-compulsive disorder, and dyslexia. Participants with ADHD were classified into two groups: those with clinically significant hoarding or those without clinically significant hoarding according to the clinical cut-off score of 40 on the Children’s Saving Inventory. Seventeen children with ADHD who were located in the state where the research was conducted were invited to take part in a pilot of neuropsychological assessments. Of those children, 16 completed the neuropsychological assessments, including eight who were classified into the ADHD with hoarding and eight were classified as ADHD without hoarding.
Children in the typically developing control group were excluded if parents reported a
developmental disorder or children scored above the cut-off for clinically significant ADHD
symptoms on the Vanderbilt ADHD Diagnostic Parent Rating Scale (n = 11 excluded).
Children in the final control group were aged 8 to 17 years (mean age = 11.92, SD = 2.46),
including 13 males and 13 females.

Materials

Demographics. A demographic questionnaire assessed children’s age, medications,
psychiatric diagnoses, and parental education (see Appendix E for questionnaires).

ADHD. The Vanderbilt ADHD Diagnostic Parent Rating Scale (VADPRS; Wolraich,
Hannah, Baumgaertel, & Fuerer, 1998) was completed by parents to assess the severity of the
child’s ADHD symptoms. Parents completed the 47 item scale rating the severity of each
behaviour on a four-point scale ranging from Never (0) to Very Often (3). The VADPRS was
developed from the teacher’s version of the scale and includes 18 items assessing all
diagnostic criteria of ADHD according to DSM-IV including Inattention and
Hyperactivity/Impulsivity subscales, which are still applicable to DSM-5 (APA, 2013). An
additional three subscales assess comorbid conditions, including eight items assessing
oppositional defiant disorder symptoms, 14 items assessing conduct disorder symptoms, and
seven items that screen for anxiety and depression. The VADPRS has shown adequate
reliability and validity (Wolraich et al., 2003) and the comorbidity scales have shown
adequate reliability and validity in screening for comorbid disorders (Becker, Langerg,
Vaughn, & Epstein, 2012). Reliability of subscales in the current sample was good, ranging
from α= .83 to .96.

Hoarding. The Children’s Saving Inventory (CSI; Storch et al., 2011) was completed
by one parent of each participant to assess hoarding symptoms. The CSI is based on the
Savings Inventory Revised (SI-R; Frost, Steketee, & Grisham, 2004) and requires parents to
rate 23 items on the extent to which their child has displayed the behaviours over the previous week on a 5-point scale, from None/not at all/never (0) to Almost all/extreme/very often (4). The CSI contains four subscales: Discarding; Clutter; Acquisition; Distress/Impairment. Twenty scores combine to produce a Total Hoarding score used to assess overall hoarding symptoms, ranging from 0 to 80, with higher scores indicating greater hoarding severity. A clinical cut-off for hoarding disorder of 40 was used to categorise children with ADHD into “with hoarding” and “without hoarding” groups. A total score of 40 on the SI-R has been established as a clinical cut-off score (Frost & Hristova, 2011; Pertusa et al., 2010) and is the total score that would be obtained if an individual answered “moderate” or “2” on each of the 20 items of the CSI contributing to the total score, which is consistent with prior studies (e.g., Hacker et al., 2012). The CSI has adequate psychometrics in 8-17 year olds, including convergent ($r = .53-.69$), discriminant ($r = .12-.23$), and construct validity, strong internal consistency ($r = .84-.96$), test-retest reliability ($r = .85-.92$), and inter-item correlations (Storch et al., 2011). Reliability in the present sample was adequate, $\alpha = .82$ to .93. The CSI is yet to be confirmed as a valid measure of hoarding in this cohort, however it is the most comprehensive instrument available to assess hoarding symptoms in children.

**Executive functioning questionnaire.** The Behavior Rating Inventory of Executive Function Parent Version (BRIEF-P; Gioia, Isquith, Guy, & Kenworthy, 2000) was used as a parental indication of executive functioning in children. Parents completed 72 items of the BRIEF-P, with Items 73 through to 86 excluded as these items do not contribute to subscale or total scores. Parents were asked to rate how often their child has displayed the behaviour of each item over the past six months: Never (1), Sometimes (2), Often (3). Higher scores indicate poorer executive functioning. The BRIEF-P contains three subscales that contribute to a Behavior Rating Index, including Inhibit, Shift, and Emotional Control. Five subscales contribute to the Metacognitive Index, including Initiate, Working Memory, Plan/Organize,
Organization of Materials, and Monitor. A total Global Executive Composite (GEC) score is derived from the sum of the Behavior Rating Index (BRI) and Metacognitive Index (MCI). Raw scores are converted to T-Scores based on age and gender. Missing values were assigned a score of 1, consistent with scoring directions (Gioia et al., 2000) - no cases had more than two missing values on a single subscale. Reliability in the current sample was good, ranging from $\alpha = .88$ to .94.

Neuropsychological assessments. Four tasks from the Cambridge Neuropsychological Test Automated Battery (CANTAB; Cambridge Cognition, 2006) were administered to assess executive functioning components of inhibition, planning, spatial working memory, and sustained attention. The CANTAB has been validated in samples aged from 4 to 92 (Cambridge Cognition, 2006). The Stop Signal Task (SST) was used to assess inhibition and requires participants to respond to a pictured arrow pointing left or right with the corresponding press pad button. When the arrow is accompanied by a short sound, participants are required to inhibit their response for that arrow. The measures of performance on this task were the proportion of successful stops.

Planning abilities were assessed using the Stockings of Cambridge (SOC), a computerised adaptation of the Tower of London task. Participants were presented with a display of coloured balls in the top half of the screen and were required to copy the display in the bottom half. This task includes problems that are solved in a minimum of two moves, three moves, four moves and five moves, and it controls for motor speed. Outcome measures for this task included the number of problems solved in minimum moves, mean initial thinking time, and mean subsequent thinking time.

The Rapid Visual Information Processing (RVP) requires participants to detect a target sequence of three numbers amongst digits from 2 to 9 displayed in a pseudo-random order at the rate of 100 digits per minute. The RVP assesses sustained attention across a four
minute period. Performance was measured by RVP A’ (A prime) which measures sensitivity of detecting the target ranging from 0.00 to 1.00, with higher scores indicating better performance.

Finally, participants completed the Spatial Working Memory task (SWM) during which they selected coloured boxes to find a blue ‘token’ hidden beneath one, in a series of three box, four box, six box, and eight box designs. Once a blue token appears in a box, it will not repeat that box again. Performance was indicated by total errors; between errors where a box in which a token has previously been found is revisited; within search errors where a box which has been found to be empty is revisited within the same search.

The Wechsler Abbreviated Scale of Intelligence – Second Edition (WASI-II; Wechsler, 2011) was used to assess cognitive ability. The Full Scale Intelligent Quotient – two subscales (FSIQ-2) was used to control for the potential influence of cognitive ability on the analyses. Children completed the Vocabulary subscale, which requires participants to define items presented visually and orally, and a Matrix Reasoning subscale, which requires participants to choose the correct response option to complete the missing square from matrices in order to complete a pattern. Each of these subscale raw scores was converted to a standard subscale score that was combined to produce a FSIQ-2 raw score, which was converted to a T-score according to age. All analyses used the FSIQ-2 T-score. The WASI-II FSIQ-2 shows adequate validity and reliability, at $r=.93$ on average for children and adolescents (Wechsler, 2011).

Procedure

Parents responded to advertisements in the community by proceeding directly to the online questionnaire or contacting the researchers. After completing several screening questions to ensure their child met the inclusion criteria, parents completed the questionnaire online or a mailed copy according to their preference (see Appendix F to H). Parents of
children with ADHD returned a copy of their child’s diagnostic report where available. All participants with ADHD in the state in which the research was conducted were invited to take part in the neuropsychological assessments, which were completed within one month of parents completing the questionnaires. Sixteen participants elected to complete the neuropsychological assessment session consisting of the WASI-II and CANTAB tasks. The entire battery took approximately 60-90 minutes to complete, with the WASI-II administered first followed by the CANTAB tasks in the order presented above. The WASI-II was administered in a room set up according to manual directions. The CANTAB was on a touchpad placed on the table directly in front of the participant. Breaks were taken as needed during the testing period, but the majority of participants completed the protocol without breaks.

Results

Statistical Analyses

All data was analysed using SPSS Statistics Version 22.0. Missing values for the VADPRS and CSI were missing completely at random according to Little’s test, and less than 5% of responses were missing on each item. Missing values were therefore replaced using expectation maximisation methods as recommended by Tabachnick and Fidel (2013). Two participants with ADHD did not complete the VADPRS and were excluded pairwise from the relevant analyses. Inspection of z scores indicated no univariate outliers with a z score $\geq 3.29$ (Tabachnick & Fidel, 2013). The BRIEF Global Executive Composite was not normally distributed for the ADHD samples with a significant kurtosis ratio $>3.29$ and significant Shapiro-Wilk tests. However, ANOVAs are considered robust to non-normality with samples above 20 degrees of freedom for error and where the smallest group comprises over 20% of the sample (Field, 2009; Tabachnick & Fidel, 2013). Several subscales did not show homogeneity of variance, including BRIEF Plan/Organise, BRIEF Organisation,
BRIEF Metacognition Index, BRIEF Global Executive Composite, and VADPRS Inattention. A more stringent alpha value of $p < .01$ was set for these subscales to protect the Type I error rate (Tabachnick & Fidel, 2013). ANCOVA was used to test the group differences in executive functioning when controlling for ADHD symptoms. Homogeneity of regression slopes and equality of variance were met with a non-significant interaction term.

**Sample Characteristics**

Table 2 shows that there were no significant between group differences in age, gender, or parental level of education. As expected, the control group had significantly fewer participants with comorbid disorders and who were taking medication compared to the ADHD groups, which did not differ significantly from each other. The group with ADHD without hoarding did not differ significantly from typically developing controls on any of the CSI subscales or on total scores.
Table 2

Sample characteristics

<table>
<thead>
<tr>
<th>Variable</th>
<th>ADHD with hoarding</th>
<th>ADHD without hoarding</th>
<th>Typically developing controls</th>
<th>( F/\chi^2 )</th>
</tr>
</thead>
<tbody>
<tr>
<td>( M ) (SD)</td>
<td>( n = 22 )</td>
<td>( n = 15 )</td>
<td>( n = 26 )</td>
<td></td>
</tr>
<tr>
<td>Age</td>
<td>11.33 (2.50)</td>
<td>10.46 (2.13)</td>
<td>11.92 (2.46)</td>
<td>1.76</td>
</tr>
<tr>
<td>% Male</td>
<td>81.8</td>
<td>73.3</td>
<td>50</td>
<td>5.82</td>
</tr>
<tr>
<td>Parental Education (%)</td>
<td></td>
<td></td>
<td></td>
<td>8.78</td>
</tr>
<tr>
<td>Did not finish secondary school</td>
<td>4.5</td>
<td>0</td>
<td>3.8</td>
<td></td>
</tr>
<tr>
<td>Year 12 or equivalent</td>
<td>13.6</td>
<td>6.7</td>
<td>20.0</td>
<td></td>
</tr>
<tr>
<td>Certificate level</td>
<td>9.1</td>
<td>13.3</td>
<td>4.0</td>
<td></td>
</tr>
<tr>
<td>Diploma/Advanced Diploma</td>
<td>18.2</td>
<td>26.7</td>
<td>16.0</td>
<td></td>
</tr>
<tr>
<td>Graduate Diploma/Certificate</td>
<td>9.1</td>
<td>0</td>
<td>16.0</td>
<td></td>
</tr>
<tr>
<td>Bachelor Degree</td>
<td>22.7</td>
<td>26.7</td>
<td>12.0</td>
<td></td>
</tr>
<tr>
<td>Postgraduate Degree</td>
<td>22.7</td>
<td>40</td>
<td>28.0</td>
<td></td>
</tr>
<tr>
<td>Taking Medication (%)</td>
<td>77</td>
<td>86.7</td>
<td>0</td>
<td>40.56**</td>
</tr>
<tr>
<td>Comorbid Disorder (%)</td>
<td>50</td>
<td>33.3</td>
<td>7.7</td>
<td>10.67**</td>
</tr>
<tr>
<td>ODD</td>
<td>36</td>
<td>13</td>
<td>0</td>
<td></td>
</tr>
<tr>
<td>Anxiety disorder</td>
<td>18</td>
<td>27</td>
<td>7.7</td>
<td></td>
</tr>
<tr>
<td>Learning disorder</td>
<td>4.5</td>
<td>0</td>
<td>0</td>
<td></td>
</tr>
<tr>
<td>CSI Total</td>
<td>51.64 (7.86)</td>
<td>28.40 (10.93)</td>
<td>25.58 (13.41)</td>
<td>36.35**</td>
</tr>
<tr>
<td>CSI Acquisition</td>
<td>13.32 (4.27)</td>
<td>7.73 (3.62)</td>
<td>7.73 (4.15)</td>
<td>13.45**</td>
</tr>
<tr>
<td>CSI Clutter</td>
<td>17.95 (3.40)</td>
<td>8.33 (3.69)</td>
<td>9.08 (5.51)</td>
<td>36.75**</td>
</tr>
<tr>
<td>CSI Discarding</td>
<td>16.14 (4.07)</td>
<td>9.73 (3.88)</td>
<td>7.23 (3.57)</td>
<td>33.26**</td>
</tr>
<tr>
<td>CSI Distress/Impairment</td>
<td>9.64 (2.34)</td>
<td>5.07 (2.19)</td>
<td>4.04 (2.79)</td>
<td>31.87**</td>
</tr>
</tbody>
</table>

Note. ODD: Oppositional Defiant Disorder. CSI: Children’s Saving Inventory.

**\( p < .01 \).

Parental Report Questionnaires

The three groups were compared on parental report questionnaires using a one-way ANOVA (see Table 3). There was a significant main effect for all VADPRS, and BRIEF
subscales except BRIEF Organisation, due to a more stringent alpha value being required for this subscale. Post hoc testing using the Tukey HSD test identified that children with ADHD and hoarding had significantly higher scores on the VADPRS Combined and Inattention scales compared to both other groups, with large effect sizes ($\eta^2 = .78$ and $.75$ respectively). However, only the controls differed from the ADHD groups on the VADPRS Hyperactivity/Impulsivity subscale ($\eta^2 = .72$). Children with ADHD with hoarding had significantly higher scores than the ADHD without hoarding group on the VADPRS ODD subscale, while both groups differed significantly from controls on the VADPRS ODD, CD, and Anxiety/Depression comorbidity scales.

The ADHD with hoarding group reported significantly poorer executive functioning compared to both the ADHD without hoarding and typically developing control groups on BRIEF GEC and BRIEF BRI with large effect sizes ($\eta^2 = .57$ and $.64$ respectively). The ADHD without hoarding group had significantly higher scores compared to controls on BRIEF GEC, BRIEF BRI, and BRIEF. Exploration of BRIEF subscales indicated that children with ADHD with hoarding scored significantly higher than the other two groups on Initiate, Working Memory, Plan/Organise, and Monitor subscales.

A one-way ANCOVA was conducted to evaluate the differences in executive functioning between groups when controlling for ADHD symptoms. After adjusting for VADPRS ADHD Combined symptoms, there was no significant difference between groups on the BRIEF GEC, $F(2, 57) = .177, p = .18, \eta^2_{\text{partial}} = .06$. In order to determine which ADHD symptoms were affecting these group differences, further analyses were conducted that found a significant difference between groups on BRIEF GEC remained when adjusting for VADPRS Hyperactivity/Impulsivity $F(2, 57) = 5.34, p < .01, \eta^2_{\text{partial}} = .16$, but not VADPRS Inattention $F(2, 57) = 1.19, p = .31, \eta^2_{\text{partial}} = .04$. 
<table>
<thead>
<tr>
<th>Measure</th>
<th>ADHD with hoarding M (SD)</th>
<th>ADHD without hoarding M (SD)</th>
<th>Typically developing controls M (SD)</th>
<th>F</th>
<th>d (ADHD vs. ADHD)</th>
<th>d (ADHD with hoarding vs. TD controls)</th>
</tr>
</thead>
<tbody>
<tr>
<td>VADPRS: Combined</td>
<td>44.81 (7.14)</td>
<td>35.50 (10.33)</td>
<td>9.92 (8.73)</td>
<td>102.31**</td>
<td>1.05</td>
<td>4.38</td>
</tr>
<tr>
<td>VADPRS: Inattention</td>
<td>24.19 (2.70)</td>
<td>18.07 (5.77)</td>
<td>6.23 (5.38)</td>
<td>86.91**</td>
<td>1.36</td>
<td>4.22</td>
</tr>
<tr>
<td>VADPRS: Hyperactivity/Impulsivity</td>
<td>20.62 (5.54)</td>
<td>17.43 (5.30)</td>
<td>3.69 (4.49)</td>
<td>73.18**</td>
<td>.59</td>
<td>3.36</td>
</tr>
<tr>
<td>VADPRS: ODD</td>
<td>16.62 (4.60)</td>
<td>12.14 (5.28)</td>
<td>5.12 (5.30)</td>
<td>30.72**</td>
<td>.91</td>
<td>2.32</td>
</tr>
<tr>
<td>VADPRS: CD</td>
<td>5.57 (5.29)</td>
<td>4.00 (3.06)</td>
<td>.88 (1.93)</td>
<td>9.99**</td>
<td>.36</td>
<td>1.18</td>
</tr>
<tr>
<td>VADPRS: Anxiety/Depression</td>
<td>10.00 (5.23)</td>
<td>9.50 (4.97)</td>
<td>4.31 (4.91)</td>
<td>8.91**</td>
<td>.10</td>
<td>1.12</td>
</tr>
<tr>
<td>BRIEF: GEC</td>
<td>80.55 (5.97)</td>
<td>69.20 (9.68)</td>
<td>54.19 (12.90)</td>
<td>40.39**</td>
<td>1.41</td>
<td>2.62</td>
</tr>
<tr>
<td>BRIEF: BRI</td>
<td>80.09 (9.12)</td>
<td>70.45 (10.59)</td>
<td>49.92 (10.91)</td>
<td>54.17**</td>
<td>.97</td>
<td>3.00</td>
</tr>
<tr>
<td>BRIEF: Inhibit</td>
<td>79.41 (10.39)</td>
<td>71.67 (10.88)</td>
<td>48.77 (9.70)</td>
<td>57.79**</td>
<td>.73</td>
<td>3.05</td>
</tr>
<tr>
<td>BRIEF: Shift</td>
<td>75.32 (10.17)</td>
<td>66.67 (12.80)</td>
<td>48.81 (10.66)</td>
<td>35.94**</td>
<td>.75</td>
<td>2.55</td>
</tr>
<tr>
<td>BRIEF: Emotional Control</td>
<td>72.59 (10.57)</td>
<td>64.60 (11.87)</td>
<td>51.96 (12.27)</td>
<td>19.25**</td>
<td>.71</td>
<td>1.80</td>
</tr>
<tr>
<td>BRIEF: MCI</td>
<td>77.27 (5.72)</td>
<td>69.93 (6.62)</td>
<td>58.96 (12.93)</td>
<td>22.29**</td>
<td>1.19</td>
<td>1.83</td>
</tr>
<tr>
<td>BRIEF: Initiate</td>
<td>72.68 (8.36)</td>
<td>63.07 (12.49)</td>
<td>53.15 (11.56)</td>
<td>19.54**</td>
<td>.90</td>
<td>1.94</td>
</tr>
<tr>
<td>BRIEF: Working Memory</td>
<td>78.00 (5.30)</td>
<td>67.73 (7.57)</td>
<td>51.27 (9.70)</td>
<td>69.85**</td>
<td>1.57</td>
<td>3.42</td>
</tr>
<tr>
<td>BRIEF: Plan/Organise</td>
<td>76.09 (7.18)</td>
<td>63.60 (11.07)</td>
<td>53.62 (14.76)</td>
<td>21.91**</td>
<td>1.34</td>
<td>1.94</td>
</tr>
<tr>
<td>BRIEF: Organization</td>
<td>65.50 (6.30)</td>
<td>61.13 (8.48)</td>
<td>57.15 (12.33)</td>
<td>4.42*</td>
<td>.58</td>
<td>.85</td>
</tr>
<tr>
<td>BRIEF: Monitor</td>
<td>73.27 (5.29)</td>
<td>62.07 (13.34)</td>
<td>47.23 (10.29)</td>
<td>42.86**</td>
<td>1.10</td>
<td>3.18</td>
</tr>
</tbody>
</table>

*Note.* VADPRS: Vanderbilt ADHD Diagnostic Parent Rating Scale; ODD: Oppositional Defiant Disorder; CD: Conduct Disorder; BRIEF: Behavior Rating Inventory of Executive Function.
Functioning; GEC: Global Executive Functioning; BRI: Behavior Regulation Index; MCI: Metacognitive Index.

*p < .05. **p < .01.

**Neuropsychological Assessments**

Of the 16 participants who completed the neuropsychological assessments, one participant did not complete the Stop Signal Task due participant’s request to cease the task, which appeared to be due to anxiety. Analyses were completed with this participant included and excluded on the completed tests with no effect on the significance of results. As such, they were only excluded pairwise for the SST. There were no significant differences in FSIQ-2 scores between children with ADHD and clinically significant hoarding ($M = 94.63; SD = 15.79$) and children with ADHD without hoarding ($M = 104.50; SD = 16.03$). A chi-square test found no significant difference in parental education level between groups, $\chi^2(4, N = 16) = 4.00, p = .41$. Significant Shapiro-Wilk’s values in each group indicated that scores were not normally distributed on a number of variables. Given the large number of non-normal distributions and the small sample sizes, to be conservative, the nonparametric method of Mann-Whitney U test was conducted to evaluate differences in neuropsychological performance between those with clinically significant hoarding and without (See Table 4). There was a significant group difference on the SOC Subsequent Thinking Time 3 moves with a large effect size and SWM Within Errors. No significant differences between groups emerged on other neuropsychological assessment outcomes.

In order to evaluate the association between neuropsychological performance and specific hoarding domains, ADHD samples were combined (n = 16) in order to conduct a Spearman’s Rho correlations (Table 5). SOC Subsequent Thinking Time on the three move problem was significantly positively correlated with CSI Total and CSI Distress/Impairment while CSI Clutter and CSI Acquisition showed a non-significant correlation with moderate effect size. SOC Initial Thinking Time 5 moves was also positively correlated with CSI
Distress/Impairment and moderate effect sizes were observed for non-significant correlations with CSI Total and CSI Discarding. SOC Subsequent Thinking Time 5 moves showed a significant negative association with CSI Acquisition, and moderate non-significant effect sizes with CSI Total and CSI Clutter. CSI Discarding was significantly positively associated with SWM Total Errors and SWM Between Errors. The likelihood of increased Type I error due to multiple comparisons is acknowledged. However, post-hoc procedures for correcting for multiple comparisons were not employed as they were likely to further reduced power in an already underpowered study (Tabachnick & Fidel, 2013).
Table 4

Comparison of neuropsychological performance between children with ADHD and clinically significant hoarding and children with ADHD without hoarding

<table>
<thead>
<tr>
<th>Variable</th>
<th>ADHD with hoarding (Median)</th>
<th>ADHD without hoarding (Median)</th>
<th>IQR</th>
<th>U</th>
<th>p</th>
<th>r</th>
</tr>
</thead>
<tbody>
<tr>
<td>SST Proportion Successful Stops</td>
<td>.42&lt;sup&gt;a&lt;/sup&gt;</td>
<td>.44</td>
<td>.20</td>
<td>22.00</td>
<td>.54</td>
<td>.18</td>
</tr>
<tr>
<td>SOC Problems Solved in Minimum Moves</td>
<td>7.00</td>
<td>7.00</td>
<td>2.75</td>
<td>32.00</td>
<td>1.00</td>
<td>.00</td>
</tr>
<tr>
<td>SOC Initial Thinking Time</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>3 moves</td>
<td>5325.75</td>
<td>2338.75</td>
<td>4449.88</td>
<td>19.50</td>
<td>.20</td>
<td>.33</td>
</tr>
<tr>
<td>4 moves</td>
<td>3516.88</td>
<td>3887.13</td>
<td>3955.19</td>
<td>30.00</td>
<td>.88</td>
<td>.05</td>
</tr>
<tr>
<td>5 moves</td>
<td>2887.25</td>
<td>4077.38</td>
<td>4357</td>
<td>24.00</td>
<td>.44</td>
<td>.21</td>
</tr>
<tr>
<td>SOC Subsequent Thinking Time</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>3 moves</td>
<td>2693.20</td>
<td>.00</td>
<td>3020.10</td>
<td>6.50</td>
<td>.01</td>
<td>.69</td>
</tr>
<tr>
<td>4 moves</td>
<td>3543.70</td>
<td>5714.45</td>
<td>6680.09</td>
<td>26.00</td>
<td>.57</td>
<td>.16</td>
</tr>
<tr>
<td>5 moves</td>
<td>1657.02</td>
<td>3002.27</td>
<td>2926.60</td>
<td>20.50</td>
<td>.23</td>
<td>.30</td>
</tr>
<tr>
<td>SWM Total Errors</td>
<td>52.50</td>
<td>43.00</td>
<td>23.50</td>
<td>20.00</td>
<td>.23</td>
<td>.32</td>
</tr>
<tr>
<td>SWM Between Errors</td>
<td>52.00</td>
<td>43.00</td>
<td>23.50</td>
<td>21.50</td>
<td>.28</td>
<td>.28</td>
</tr>
<tr>
<td>SWM Within Errors</td>
<td>2.50</td>
<td>.00</td>
<td>3.00</td>
<td>11.50</td>
<td>.03</td>
<td>.56</td>
</tr>
<tr>
<td>RVP A’</td>
<td>.94</td>
<td>.91</td>
<td>.09</td>
<td>27.00</td>
<td>.65</td>
<td>.13</td>
</tr>
</tbody>
</table>

Note. WASI FSIQ-2: Wechsler Abbreviated Scale of Intelligence Full Scale Intelligence Quotient - two subscales; SST: Stop Signal Task; SOC: Stockings of Cambridge; SWM: Spatial Working Memory; RVP: Rapid Visual Processing A Prime.

<sup>a</sup>n = 7 for SST ADHD with hoarding.
## Table 5

*Correlation coefficients between CSI subscales and neuropsychological measures.*

<table>
<thead>
<tr>
<th>Variable</th>
<th>CSI Total</th>
<th>CSI Clutter</th>
<th>CSI Acquisition</th>
<th>CSI Discarding</th>
<th>CSI Distress/Impairment</th>
</tr>
</thead>
<tbody>
<tr>
<td>SST Proportion of Successful Stops&lt;sup&gt;a&lt;/sup&gt;</td>
<td>-.04</td>
<td>-.09</td>
<td>-.12</td>
<td>.12</td>
<td>.17</td>
</tr>
<tr>
<td>SOC Problems Solved in Minimum Moves</td>
<td>.06</td>
<td>.37</td>
<td>.33</td>
<td>.04</td>
<td>-.03</td>
</tr>
<tr>
<td>SOC Initial Thinking Time</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>3 moves</td>
<td>.11</td>
<td>.10</td>
<td>.11</td>
<td>.10</td>
<td>.11</td>
</tr>
<tr>
<td>4 moves</td>
<td>-.14</td>
<td>-.22</td>
<td>-.12</td>
<td>-.15</td>
<td>.12</td>
</tr>
<tr>
<td>5 moves</td>
<td>.46</td>
<td>.16</td>
<td>.35</td>
<td>.46</td>
<td>.53*</td>
</tr>
<tr>
<td>SOC Subsequent Thinking Time</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>3 moves</td>
<td>.61*</td>
<td>.45</td>
<td>.45</td>
<td>.33</td>
<td>.52*</td>
</tr>
<tr>
<td>4 moves</td>
<td>.21</td>
<td>.23</td>
<td>.28</td>
<td>.06</td>
<td>.15</td>
</tr>
<tr>
<td>5 moves</td>
<td>-.40</td>
<td>-.46</td>
<td>-.50*</td>
<td>-.17</td>
<td>-.23</td>
</tr>
<tr>
<td>SWM Total Errors</td>
<td>.36</td>
<td>.10</td>
<td>.27</td>
<td>.54*</td>
<td>.14</td>
</tr>
<tr>
<td>SWM Between Errors</td>
<td>.33</td>
<td>.07</td>
<td>.24</td>
<td>.55*</td>
<td>.11</td>
</tr>
<tr>
<td>SWM Within Errors</td>
<td>.34</td>
<td>.06</td>
<td>.16</td>
<td>.33</td>
<td>.21</td>
</tr>
<tr>
<td>RVP A’</td>
<td>.19</td>
<td>.05</td>
<td>.08</td>
<td>.04</td>
<td>.31</td>
</tr>
</tbody>
</table>

*Note.* SST: Stop Signal Task; SOC: Stockings of Cambridge; SWM: Spatial Working Memory; RVP: Rapid Visual Processing A Prime.

<sup>a</sup> n = 15 for SST.

<sup>*</sup> p < .05.
Discussion

To our knowledge, this is the first study to have examined the association between executive functioning and hoarding symptoms among children and adolescents with ADHD. Before discussing specific findings with hypotheses, it is important to acknowledge that this is an underpowered exploratory study, the results of which need to be interpreted with caution given the possibility of Type I error. Nonetheless, the results of subjective parent ratings lend support to the hypothesis that poorer executive functioning may be associated with higher levels of hoarding symptoms in children and adolescents with ADHD. Executive functioning differentiated between the three groups, with those with ADHD and clinically significant hoarding symptoms displaying poorer overall executive functioning compared to children with ADHD without hoarding and typically developing controls. Poorer performance on neuropsychological tasks of spatial working memory and planning appeared to be associated with hoarding symptom dimensions of difficulty discarding, distress/impairment, and increased acquisition. This may provide preliminary support to previous suggestions in the literature that poor executive functioning may help to explain the presence of hoarding symptoms in individuals with ADHD (Fullana et al., 2013; Hall et al., 2013).

Consistent with the hypotheses, children with ADHD and clinically significant hoarding appeared to show poorer global executive functioning across the majority of behaviour and metacognitive executive functions when compared to children without hoarding. Consistent with prior findings (Hacker et al., 2012), children with ADHD and clinically significant hoarding had greater ADHD symptom severity compared to those with ADHD without clinically significant hoarding. This appeared to be specific to ADHD inattention symptoms, while ADHD hyperactivity/impulsivity did not differentiate between the two ADHD groups. Although this parallels the adult literature (Anholt et al., 2010; Tolin
& Villavicencio, 2011), it is in contrast to previous childhood studies that have identified a significant association between hyperactivity/impulsivity and hoarding (Fitch & Cougle, 2013; Hacker et al., 2012). The present study extended these results in its finding that parental reports of their child’s executive functioning did not differentiate between the three groups after controlling for overall ADHD symptom severity. It is unlikely that this represents a response bias by parents (Furnham, 1986) or shared method variance, as only inattention and not hyperactivity/impulsivity seem to negate the differences. It is possible that given that ADHD inattention and global executive functioning were highly correlated, both may have been assessing similar constructs. Alternatively, severity models suggest that greater ADHD symptom severity is associated with greater deficits in neuropsychological performance (e.g., Nikolas & Nigg, 2013). Therefore it may be that the association between executive functioning and hoarding symptoms in children with ADHD is moderated by ADHD inattention symptom severity. Further exploration of this possibility is needed with a larger sample size.

The questionnaire findings were partially supported by significant between group differences on performance on neuropsychological tasks of planning and spatial working memory. Children with ADHD and hoarding spent a longer time planning subsequent moves on the SOC 3 moves task compared to those without hoarding, which may support the hoarding disorder profile of longer planning times and may indicate perfectionism while planning (e.g., Ayers, Bratiotis, Saxena, & Wetherell, 2012; Timpano, Keough, Traeger, & Schmidt, 2011). Moreover the significant association between hoarding related distress/impairment and longer initial and subsequent planning times is similar to the hoarding disorder profile of increased distress when planning to discard items (Nordsletten et al., 2013). This finding occurs in the context of previous neuroimaging studies that have identified that anxiety about discarding items in individuals with hoarding disorder is
associated with greater activity in the ventral prefrontal regions and amygdala (An et al., 2009; Mataix-Cols et al., 2003).

Previous findings have identified that individuals with ADHD do not significantly increase planning times on more complex tasks (Young, Morris, Toone, & Tyson, 2007). The current results hint that when tackling more complex tasks, children with ADHD and hoarding may reduce planning time while children with ADHD without hoarding may increase planning time. An analysis of the significance of this trend, however, was not possible due to the small sample size. This divergent pattern of planning suggests that multi-step complex tasks such as discarding several items, or advanced planning to acquire items may be poorly planned in children with ADHD, whereas they may spend more time planning simple task such as discarding one item leading to increased distress. It is therefore possible that, in contrast to a typical ADHD profile of short planning time and less elaborate plans (e.g., Bramham et al., 2009; Fuermaier et al., 2013), children with ADHD and clinically significant hoarding symptoms may have a hoarding disorder-like profile of spending a longer time planning simple tasks. If this was the case, it would be likely to increase distress in these children.

Moreover, the low planning times demonstrated on complex tasks was significantly associated with increased acquisition in children with ADHD, with moderate effect sizes evident with respect to clutter and total hoarding symptoms. This is consistent with the pattern whereby individuals with hoarding disorder are less likely than comparison individuals to plan the items they will acquire (Nordsletten et al., 2013), thus resulting in accumulation of items and potential clutter. Although limited by sample size, overall, these findings are consistent with prior studies identifying planning difficulties in people with hoarding disorder (Grisham et al., 2010; Pinto et al., 2011).
Errors in spatial working memory were associated with difficulty discarding items in children with ADHD. This supports previous results in adults with hoarding disorder who displayed poorer visual memory span (Grisham et al., 2007) and poorer spatial memory encoding compared to controls (Hartl et al., 2004). Hartl et al. (2004) suggest that these spatial working memory difficulties may increase hoarding related anxiety and clutter, as individuals prone to poor visual memory may avoid organising items in out-of-view storage. Among children with ADHD, spatial working memory difficulties may therefore be a precursor to hoarding-related anxiety and difficulty discarding items. Contrary to predictions, the hoarding group did not differ significantly from the comparison group on measures of sustained attention or inhibition. This was unexpected, given previous findings of poorer sustained attention in adults with hoarding disorder (Grisham et al., 2007; Tolin & Villavicencio, 2011) and the greater scores on VADPRS Inattention in children with ADHD and hoarding in the present study. It is possible that medications may have influenced sustained attention performance (e.g., Bédard et al., 2015), with the majority of the ADHD sample taking medications. The RVP also provides a very structured assessment of sustained attention that may not be ecologically valid compared to the multi-step procedure required in discarding possessions (see Chan et al., 2008). Moreover, the discrepancy between neuropsychological performance on the sustained attention task and parental ratings of the child’s attention may demonstrate a performance deficit rather than a skill deficit, where children were able to demonstrate higher performance than usual in controlled settings with an examiner present (Draeger, Prior, & Sanson, 1986). The current study found no significant correlations between the CANTAB and BRIEF. This may support the suggestion that they could be measuring different constructs and highlights the importance of considering both forms of measurement (e.g., Chan et al., 2008).
Consistent with the questionnaire findings, children with clinically significant hoarding did not display poorer inhibition compared to those without hoarding. Although this may be due to small sample sizes, it parallels the non-significant findings in adults by Grisham et al. (2010) and Blom et al. (2011). However, this is in contrast to other findings that adults with hoarding disorder exhibit more commission errors on sustained performance tasks compared to controls (Grisham et al., 2007; Rasmussen et al., 2013). This discrepancy may be understood in the context of continuous performance tasks also assessing sustained attention and that they thus may be more susceptible to ‘task impurity’ of assessing multiple components of executive functioning, potentially contributing to a measurement error (Burgess, 1997). Moreover, the SST does not assess emotionally salient aspects of inhibition that may be related to hoarding symptoms, such as resisting the urge to acquire items.

These initial results have several potential clinical implications. First, the results show preliminary support for what is known as the correlated liabilities model for the comorbidity between ADHD and hoarding symptoms (Lynch et al., 2015; Neale & Kendler, 1995), with a shared underlying neuropsychological dysfunction. Specifically, the executive functioning profile of children with ADHD and clinically significant hoarding symptoms appeared to be worse than that for those with ADHD alone. This may point toward a shared vulnerability toward dysfunction in the frontal networks in both disorders (e.g., An et al., 2009; Wilson et al., 2013) associated with impaired executive functioning (Alvarez & Emory, 2006). Second, planning among children with ADHD and hoarding symptoms may be similar to a typical hoarding disorder profile, and may be particularly associated with high levels of distress when discarding items, potentially due to perfectionistic planning. If substantiated, this may have important treatment implications, as individuals with ADHD and significant hoarding symptoms could benefit from executive functioning training in order to reduce excessive planning times for simple tasks, in combination with increasing distress tolerance (see Phung,
Moulding, Taylor, & Nedeljkovic, 2015; Timpano, Buckner, Richey, Murphy, & Schmidt, 2009; Timpano et al., 2011). Finally, the present study has implicated planning, spatial working memory and inattention as potential treatment targets for early intervention for hoarding symptoms in children and adolescents with ADHD.

Although this study has provided an initial examination of the association between hoarding symptoms and executive functioning using both performance and questionnaire assessments, the findings must be considered in the context of several limitations. First, as previously noted, this preliminary investigation had a small sample which likely resulted in an underpowered study. For example, while the ANOVA for the BRIEF GEC between groups had adequate power at .93 (Faul, Erdfelder, Lang, & Buchner, 2007), the ANCOVA controlling for VADPRS Combined had a very low power at .19. It is therefore possible that further significant differences may have been found with a larger sample.

Second, children in this study were categorised as having clinically significant hoarding symptoms - however, this is not equivalent to a diagnosis of hoarding disorder. It is also acknowledged that the cut-off used in the present study is yet to receive validation in children. Moreover the CSI has several limitations as it is not a diagnostic instrument and is yet to be validated in this cohort. Although the CSI has been shown to significantly correlate with anxiety/depression scales (Storch et al., 2011), the present study found no significant difference between anxiety/depression scores of children with ADHD and hoarding compared to those with ADHD without hoarding. Further research is needed to explore executive functioning in children with ADHD and a diagnosed hoarding disorder in order to better understand this comorbidity. The high rate of comorbid ODD may also have influenced some results such as the potential for higher scores on the BRIEF.

Third, it is possible that the questionnaire findings represent a shared method variance due to parental ratings on all outcomes. This limitation is acknowledged and the
neuropsychological findings may provide a more objective account of specific executive functioning differences. Moreover, it is possible that the comorbid group has a vulnerability to increased executive functioning difficulties associated with higher levels of psychopathology in general, and may not be specifically associated with the hoarding symptom/ADHD comorbidity. Finally, the typically developing control group were not administered the neuropsychological assessments, thereby preventing the comparison between the neuropsychological functioning of those with ADHD without hoarding and typically developing controls.

In conclusion, this pilot study has provided preliminary evidence that children and adolescents with ADHD and clinically significant hoarding symptoms may exhibit poorer executive functioning, particularly in planning and spatial working memory abilities. The existing executive functioning difficulties in children with ADHD may therefore be associated with specific hoarding symptomatology, including difficulty discarding items, increased acquisition, and distress or impairment. This improves our understanding of the neuropsychological functioning of children with ADHD and comorbid hoarding symptoms, and it provides initial support for early intervention to target improved executive functioning in children with ADHD who may be vulnerable to developing hoarding disorder.
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Chapter Five
Discussion

Overview of Thesis Findings

The nature of hoarding symptoms in children and adolescents with comorbid ADHD was explored in the three studies comprising this thesis. A shared vulnerability model to hoarding disorder was proposed, and preliminary support for this model was identified. Specifically, children with ADHD and clinically significant hoarding symptoms displayed behaviours that appear to parallel a typical hoarding disorder profile and were associated with poorer executive functioning compared to comparison children.

In Study One, potential comorbidity models were explored and applied to ADHD and hoarding disorder in order to improve our understanding of the nature of this comorbidity. Substantial evidence supporting a correlated liabilities model was identified and it was proposed that ADHD and hoarding disorder may have a shared genetic, executive functioning, and neurological vulnerability. This is consistent with the empirical evidence from hoarding disorder research which suggests that specific neuropsychological and neurophysiological deficits are associated with hoarding symptoms (see Grisham & Baldwin, 2015). Although the two disorders appear to share difficulties in the same executive functions and neurological networks, individuals with ADHD and hoarding disorder appear to have the opposite pattern of planning difficulties and prefrontal activity. Specifically, a review of the evidence indicated that individuals with hoarding disorder may have hyper-activity in prefrontal areas, compared to the hypo-activity typical of ADHD. Moreover, the hoarding disorder profile includes longer planning times with perfectionistic development of plans, whereas individuals with ADHD typically display short planning times on complex tasks. This has important implications for clinical practice and for research, where the opposing patterns of planning and prefrontal activity in hoarding disorder and ADHD have not been previously
considered. Despite the strengths of the proposed correlated liabilities model, existing evidence does not allow rejection of other comorbidity models. It therefore remains possible that hoarding symptoms may be epiphenomena of core ADHD symptomatology; thus appearing phenotypically similar to hoarding disorder but potentially lacking the distress and urges to save features that are typical of hoarding disorder.

To expand the findings of Study One, the phenomenology of hoarding behaviours in children with ADHD and comorbid clinically significant hoarding symptoms was explored in Study Two from the perspective of parents. According to parents, children with ADHD and clinically significant hoarding symptoms displayed behaviours and reactions that mirrored a typical hoarding disorder profile and including distress when discarding items, emotional attachment to objects, urges to save items, and a range of parental accommodating behaviours. Emotional distress was a core theme that appeared to be related to the negative impact of hoarding on the academic, emotional, social, and environmental functioning of the child, parents, and sometimes siblings. The negative impact of hoarding symptoms on functioning and wellbeing in children with comorbid ADHD signalled a clear need for further exploration of this comorbidity.

One of the six superordinate themes identified in Study Two was executive functioning, with parents reporting that organisation, memory, attention, and impulsivity appeared to be associated with hoarding symptoms. The correlated liabilities model proposed in Study One and the results of Study Two therefore both pointed toward a need to examine the association between executive functioning and hoarding symptoms in children and adolescents with ADHD.

Study Three used both ecologically valid parental questionnaires and neuropsychological assessments were used in Study Three to explore differences in executive functioning between children with ADHD and clinically significant levels of hoarding.
symptoms, children with ADHD without clinically significant hoarding symptoms, and typically developing controls. Executive functioning appeared to differentiate between the three groups. According to parental report, the children with ADHD and clinically significant hoarding symptoms exhibit poorer executive functioning than the other two groups. The selection of the neuropsychological assessments conducted in Study Three was informed by the findings derived from Study One that planning, spatial working memory, inhibition, and sustained attention are core executive functioning difficulties in the hoarding disorder profile. Although limited by sample size, the results from these assessments hinted that poorer spatial working memory may be associated with increased difficulty discarding items in children and adolescents with ADHD. A divergent pattern of planning was identified, which suggested that longer planning times on simple tasks may be associated with hoarding related distress and impairment, whereas shorter planning times on complex tasks may be associated with increased acquisition. These findings expand previous suggestions that ADHD and hoarding disorder may be associated through executive functioning (e.g., Fullana et al., 2013; Hall et al., 2013) and provide preliminary support for the presence of these relationships in children and adolescents. Further exploration in studies with larger samples and increased statistical power is clearly required.

**Clinical Implications**

The findings of this thesis have potential clinical implications for understanding the maintenance, early detection, and treatment of hoarding symptoms in children and adolescents with comorbid ADHD. Preliminary support is provided for the correlated liabilities model developed in Study One, with findings also suggesting that the additional components of Frost and Hartl’s (1996) cognitive behavioural model of hoarding disorder may be necessary to best understand hoarding symptoms in children with ADHD. For example, children with ADHD and comorbid hoarding symptoms appeared to be vulnerable
to poorer executive functioning than comparison children. However, the presence of high emotional reactions and urges to save suggest the relevance in this population of the additional core maintenance factors of the Frost and Hartl model, including inaccurate beliefs about the nature of possessions and emotional attachment problems. As a novel finding, there was also an indication that the final behavioural avoidance component of Frost and Hartl’s model may be applied to parental avoidance and parental accommodation of hoarding symptoms in children with comorbid ADHD. Rather than solely focusing on the individual child, hoarding disorder in children with ADHD may therefore be best conceptualised in the context of the family.

Combined, these results suggest that hoarding disorder in children with comorbid ADHD likely mirrors a genuine hoarding disorder profile, with hoarding symptoms maintained by unhelpful cognitive-behavioural cycles and executive functioning difficulties. This conceptualisation has implications for understanding the comorbidity between hoarding disorder and ADHD, as it suggests that executive functioning, neural and genetic factors are insufficient for a full understanding of the comorbidity between these disorders. In order to include the additional cognitive-behavioural components that were identified, the correlated liabilities model proposed in Study One could be extended to include emotion dysregulation. A range of difficulties with emotion regulation have previously been associated with hoarding disorder, including avoidance of negative emotions (Wheaton, Fabricant, Berman, & Abramowitz, 2012), negative impulsivity (Timpano et al., 2013), distress intolerance (Timpano, Keough, Traeger, & Schmidt, 2011), and anxiety sensitivity (Medley, Capron, Korte, & Schmidt, 2013). Of these factors, anxiety sensitivity and negative impulsivity have been shown to predict hoarding symptoms, mediated via cognitions about the emotional attachment to possessions (Phung, Moulding, Taylor, & Nedeljkovic, 2015). Likewise, individuals with ADHD appear susceptible to emotion regulation difficulties in comparison
to controls, including extremes and changes in emotions (Anastopoulos et al., 2011; Sjöwall, Roth, Lindgvist, & Thorell, 2013), poorer emotion regulation when completing frustration tasks (Walcott & Landau, 2004), and impaired emotion regulation according to the Difficulty in Emotion Regulation Scale (Seymour, Chronis-Tuscano, Halldorsdottir, Stupica, & Owens, 2012). Moreover, emotion regulation is likely to be associated with the neurological and executive functioning components already identified in the correlated liabilities model. For example, the amygdala is proposed to be implicated in the regulation of emotional responses (Aznar & Klein, 2013; Mitchell, 2011) and is overactive in hoarding disorder (An et al., 2009) and ADHD (Posner et al., 2011).

Although emotion regulation has historically been viewed as a distinct entity from executive functioning, this dichotomy is being eroded with ‘hot’ executive functions referring to those requiring affective responses (Zelazo & Müller, 2002). This association is evident in emerging empirical research supporting the association between emotion regulation and the executive function of decision making (Martin & Delgado, 2011; Panno & Marco, 2013; Zelazo & Carlson, 2012). Perhaps a more comprehensive correlated liabilities model would therefore include an association between executive functioning, neural vulnerability, emotion regulation and genetic factors in comorbid ADHD and hoarding symptoms.

Together, these findings inform early detection and intervention approaches. It is suggested that ADHD may provide a platform to identify children who may be at-risk of developing hoarding disorder. Among these children, executive functioning difficulties may indicate a genetic or neural vulnerability towards hoarding symptoms. As such, executive functioning difficulties may be used as a factor for early detection of a vulnerability to hoarding disorder among children with ADHD and elevated hoarding symptoms prior to a formal diagnosis. This is an important development, as despite its early onset, hoarding disorder frequently remains undiagnosed until adulthood when its symptoms have progressed...
and functioning has deteriorated. Earlier diagnosis of hoarding disorder could also result in
early intervention to prevent the progression to more severe symptom. The preliminary
findings of this thesis suggest that early intervention of hoarding symptoms in children with
ADHD likely needs to include a combined approach of cognitive-behaviour therapy (CBT)
and treatment targeting the executive functioning difficulties. Specifically, the unhelpful
cognitive-behaviour cycle of hoarding symptoms could be treated using CBT for hoarding
disorder to target emotional attachment issues and urges to save (e.g., Muroff, Steketee,

Training in executive functioning, specifically targeting spatial working memory and
planning difficulties, may augment therapy outcomes in children with ADHD and comorbid
hoarding symptoms. There is evidence to support executive functioning treatment in children
with ADHD (e.g., Gray et al., 2012; Miranda, Presentación, Siegenthaler, & Jara, 2011) and
emerging treatment trials in hoarding disorder include cognitive skills training (e.g., Muroff
et al., 2010; Tolin et al., 2007). However, when considering interventions for planning skills
in children with ADHD and comorbid hoarding symptoms, treatment may require reducing
planning times for simple tasks as opposed to increasing planning detail, as might be
expected of typical ADHD treatment (e.g., Sibley et al., 2015). In addition to CBT and
executive functioning training, a more complete intervention for hoarding symptoms in
children with ADHD may be achieved through also targeting perpetuating parental factors.
Although emerging evidence supports an association between family accommodation and
hoarding disorder (Lebowitz et al., 2013), no study to date has investigated incorporating
family education and intervention into hoarding disorder treatment for children and
adolescents.
Limitations

The findings of this thesis must be considered in the context of several limitations. Firstly, the participants sampled had not received a clinical diagnosis of hoarding disorder. Although this allowed preliminary exploration of the phenomenology and maintenance of early hoarding symptoms in children with ADHD, it did not directly examine the comorbidity between the two disorders at a diagnostic level. Nonetheless, the current exploration of hoarding symptoms is consistent with the Research Domain Criteria proposed by the National Institute of Mental Health (2008) and which focuses on the dimensional aspects of behavioural and neural circuits rather than distinct categorical disorders.

Second, the small sample size employed in Study Three may have reduced statistical power and increased the likelihood of a null hypothesis being rejected (Ellis, 2010). This particularly applies to the unexpected absence of poorer executive functioning when controlling for ADHD symptoms in children with clinically significant hoarding and ADHD. Finally, it was unfortunately beyond the scope of this preliminary investigation to examine the additional genetic and neurological factors proposed in the correlated liabilities model in Study One.

There are several additional limitations to hoarding disorder research in childhood that may have affected the findings of this research. Although hoarding disorder is proposed to have an early age of onset, few studies have investigated hoarding symptoms in childhood. While the present thesis addressed this gap by specifically exploring hoarding symptoms in children and adolescents, conclusions are limited given that a thorough understanding of hoarding disorder in childhood has not yet been developed. It is unclear, for example, whether the descriptions of hoarding symptoms provided in Study Two are similar to those of children with hoarding disorder without ADHD.
A further limitation arises from the use of neuropsychological assessments in hoarding disorder research. Several researchers have unexpectedly found non-significant results when examining neuropsychological performance in hoarding disorder (e.g., Grisham, Norberg, Williams, Certoma, & Kadib, 2010; McMillan, Rees, & Pestell, 2013). It is difficult to conclude that these cognitive functions are not related to hoarding symptoms. Although neuropsychological tests provide a standardised assessment that allows normative comparisons to be made and performance deficits to be identified, they are typically very structured, time-limited tasks performed under the observation of an examiner (Harvey, 2012). As such, it is possible that they do not provide a representation of more complex multi-step tasks present in the naturalistic environment (Chan, Shum, Toulopoulou, & Chen, 2008). This is particularly relevant for children with ADHD, who may be able to sustain attention and prevent hyperactivity whilst under examination during short performance tasks but not with respect to the demands of general daily living (Chan et al., 2008). Moreover, performance tests of executive functioning provide information on the presence of impairment but not impact on daily functioning (Chan et al., 2008). In hoarding disorder research, executive functioning difficulties may therefore go undetected in neuropsychological assessments, but may nonetheless be specifically impaired in emotionally laden tasks such as discarding or acquiring items (Grisham et al., 2010). This is likely a further limitation of the current thesis which did not assess these emotionally laden functions through the inclusion of ‘hot’ executive functioning tasks, such as decision making or goal setting (Zelazo & Müller, 2002).

**Conclusion and Future Directions**

The overall aim of this thesis was to explore the nature of hoarding symptoms in children and adolescents with comorbid ADHD. It was concluded that a correlated liabilities model may be useful in understanding the comorbidity between hoarding symptoms and
ADHD, including shared executive functioning difficulties, emotion dysregulation, neurological and genetic vulnerability. This parallels the cognitive-behavioural model of hoarding disorder (Frost & Hartl, 1996), which may be applied to children with ADHD in order to understand the maintenance of hoarding symptoms, including information processing difficulties, emotional attachment to objects, urges to save, and parental accommodation. If confirmed, these findings may inform treatment approaches in suggesting the value of inclusion of CBT and executive functioning training components in interventions for children with ADHD and comorbid hoarding symptoms.

This preliminary investigation has highlighted a need for future research across many important areas. Firstly, the correlated liabilities model and executive functioning findings suggest a potential shared vulnerability involving neurological dysfunction that requires further exploration. Future research is needed to further investigate the neurological profile of individuals with ADHD and comorbid hoarding disorder, particularly the prefrontal network to determine if it shows the hoarding disorder profile of hyper-activity or the ADHD profile of hypo-activity. This suggestion is offered in the context of the current findings indicating that the planning profile of children with ADHD and comorbid hoarding symptoms during simple tasks showed a similar pattern to that of hoarding disorder. A second area for future research is an exploration of the potential role of emotion regulation in the comorbidity of hoarding disorder and ADHD. While Study Two showed initial support for this additional component, further quantitative studies are required to determine if this factor is significantly associated with hoarding symptoms among children with ADHD.

Thirdly, future research and clinical practice could attempt to develop and assess an intervention approach for hoarding symptoms in childhood that combines traditional CBT skills with training in executive functioning. This would be particularly relevant for children with ADHD who may be at particular risk of severe functional consequences arising from
hoarding symptoms. Moreover, the potential role of parental accommodation in maintaining hoarding symptoms requires further investigation to determine if it is an appropriate treatment target. Finally, given the early onset of these symptoms, there is a need to address the identified dearth of research into early detection and early intervention for hoarding symptoms in childhood and adolescence. The findings from this thesis that hoarding symptoms are present from a young age, with significant deleterious impact on functioning and wellbeing on children and their families, point to a clear need for research with a focus on hoarding symptoms in children, particularly those with comorbid ADHD.
References (Chapter Five)


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Hoarding in attention deficit hyperactivity disorder: Understanding the comorbidity

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Hoarding disorder has a frequent co-occurrence with attention-deficit/hyperactivity disorder (ADHD). An accurate understanding of the comorbidity between hoarding disorder and ADHD remains unclear but is essential to inform appropriate assessment, prevention and treatment approaches. This paper will provide a review of potential comorbidity models and aetiological mechanisms implicated in both disorders in order to inform understanding of the nature of the comorbidity between hoarding disorder and ADHD. A correlated liabilities model is identified that implicates genetic, neurological, and executive functioning factors in the development and maintenance of hoarding symptoms in individuals with ADHD.

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1. Introduction

Although ADHD and hoarding disorder commonly co-occur, relatively little is known about the aetiology of this comorbidity.

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Hoarding disorder is a new addition to the Diagnostic and Statistical Manual of Mental Disorders, 5th Edition (DSM-5; American Psychiatric Association, 2013) and is defined as the difficulty discarding items due to urges to save possessions or distress when discarding; accumulation of items and cluttering of living spaces; and significant distress or impairment caused by these symptoms. Prior to its addition in the DSM-5, various terms were used to define hoarding disorder, such as compulsive hoarding (Frost & Hartl, 1996). However, the present paper will use the term ‘hoarding disorder’ to reflect the
current DSM-5 designation but will note any important differences in individual studies’ definitions of hoarding disorder.

Among the 2.3% of adults with hoarding disorder (Iervolino et al., 2009), approximately 28% have comorbid ADHD (Frost, Steketee, & Tolin, 2011). This population appears to experience greater deficits in functioning than those with hoarding disorder alone (Hall, Tolin, Frost, & Steketee, 2013), including more difficulties in activities of daily living, increased stress, and higher levels of domestic squalor (Hall et al., 2013). These deficits occur in addition to the hoarding-related impairments of compromised safety (Frost, Steketee, & Williams, 2000), reduced quality of life (Palermo et al., 2011), increased employment and housing difficulties (Saxena et al., 2011), and severe medical issues, including higher risk of arthritis, diabetes, obesity, and stroke compared to individuals without hoarding disorder (Tolin, Frost, Steketee, Gray, & Fitch, 2008).

The severe potential consequences of comorbid hoarding disorder in individuals with ADHD and the high association between the two disorders point to the importance of developing a greater understanding of the features involved in the co-occurrence of these disorders, particularly the causal and maintenance factors. It has recently been suggested that shared executive functioning deficits may explain the comorbidity between ADHD and hoarding disorder (Fullana et al., 2013; Hall et al., 2013). However, to date, no study has investigated this possibility in a sample of individuals with ADHD with comorbid hoarding disorder. The potential role of executive functioning as a mechanism linking ADHD and hoarding disorder therefore remains unknown.

The present review aims to synthesise the current literature to develop a model that may be used to understand the comorbidity between hoarding disorder and ADHD. To do this, several potential comorbidity models will be explored. The correlated liabilities model will be evaluated by providing a critical analysis of possible evidence of a shared genetic, neurological, and executive functioning aetiology of hoarding disorder among adults with ADHD. Such an analysis may inform understanding of the mechanisms of hoarding symptoms in this population and may highlight clinical implications, including prevention and treatment approaches.

2. Overlap between ADHD and hoarding disorder

There is substantial evidence for an association between ADHD and hoarding disorder. For example, in an assessment of comorbidity in adults with hoarding disorder, 27.8% were found to have comorbid inattentive ADHD, while 13.7% had hyperactive ADHD (Frost et al., 2011). Furthermore, Sheppard et al. (2010) found that 21.9% of adults with hoarding disorder had comorbid ADHD. In contrast, it appears from a meta-analysis of prevalence rates of ADHD using DSM-IV diagnostic criteria that ADHD only affects approximately 5% of the general adult population (Wilcutt, 2012). A further study using retrospective data indicated that a higher proportion of individuals with childhood ADHD later developed hoarding symptoms in comparison to those without childhood ADHD (Fullana et al., 2013). Other studies also suggest an association between the specific symptoms of ADHD and core hoarding symptoms. In particular, inattention appears to be a strong predictor of hoarding symptoms across the lifespan (Hacker et al., 2012; Tolin & Villavicencio, 2011), while hyperactivity/impulsivity appears to be associated with hoarding symptoms in childhood (Fitch & Cougle, 2013; Hacker et al., 2012).

Despite developing evidence of a frequent co-occurrence between ADHD and hoarding disorder, an accurate understanding of the mechanisms explaining this comorbidity remains unclear. Neale and Kendler (1995) propose several general comorbidity models that may be used to explain the co-existence of hoarding disorder and ADHD. As used previously by Schmitt and Weidinger (2014) in a study of dermatological comorbidities, Table 1 summarises these models in relation to existing knowledge regarding the association between hoarding disorder and ADHD. It is crucial that research is undertaken to establish a valid model of comorbidity that enables accurate diagnosis of comorbidity, as well as differential diagnosis between ADHD and hoarding disorder (Achenbach, 1995). Moreover, research is needed to gain an understanding of the aetiological mechanisms of ADHD and hoarding disorder in order to inform treatment decisions such as identifying specific treatment targets and determining whether the two disorders should be treated sequentially or in parallel (Achenbach, 1995).

On the basis of Neale and Kendler’s (1995) framework and the current understanding of ADHD and hoarding disorder, the comorbidity between the two disorders may be explained by several alternate models, including alternate forms, random multifactor, three independent disorders, correlated liabilities, and the causal models. The alternate forms model would apply if both ADHD and hoarding disorder result from a single underlying liability. Given that no single cause has been identified for either disorder, this model cannot yet be rejected. However, our current understanding of hoarding disorder and ADHD suggests that several different aetiological factors may contribute to the occurrence of each disorder. It is therefore unlikely that the comorbid condition results from a single cause. The aetiological factors will therefore be discussed as they contribute to the correlated liabilities model. The random multifactor model is also plausible, whereby the comorbid condition represents an epiphenomenon of hoarding disorder in those with ADHD.

According to the three independent disorders model, individuals with ADHD plus hoarding disorder may have a disorder qualitatively and aetologically distinct from ADHD and hoarding disorders. There would therefore be two possibilities for the comorbid condition: those with ADHD and hoarding disorder who are above the threshold for the liability of each disorder; and those with the distinct disorder of ADHD plus hoarding disorder. Given that the precise aetiology of hoarding disorder and ADHD is unknown, it remains a possibility that some cases of the comorbid condition could be an independent disorder that arises from a third aetiological mechanism. Very little is known about the nature of hoarding symptoms in those with ADHD, and further qualitative explorations and genetic studies of the comorbid condition are essential to explore these models.

Alternatively, the co-occurrence of ADHD and hoarding symptoms may be conceptualised using the Research Domain Criteria, which focuses on dimensional aspects of behavioural and neural circuits in the development and classification of mental disorders (National Institute of Mental Health, 2008). For example, ADHD and hoarding symptoms could be conceptualised as interrelated constructs, or behavioural dimensions, of the broader domain of Cognitive Systems, that co-vary with different degrees of damage to shared neural circuits, genetic, and behavioural factors (Cuthbert & Kozak, 2013; Morris & Cuthbert, 2012). Although these models cannot be rejected, the existing literature on these concepts is not yet established enough to warrant inclusion in the present review. It is also possible that hoarding disorder is directly caused by ADHD in a subgroup of those with hoarding disorder. Although this is also unable to be rejected as a possibility, reciprocal causation seems unlikely given the current indications that the development of hoarding symptoms begin after the onset of ADHD.

The inability to reject the alternate forms, random multifactor, causal and three independent disorder models as explanations for the comorbidity between ADHD and hoarding disorder highlights the need for further empirical investigation of these models. However, recent interest from researchers in the correlated liabilities model suggests its potential in explaining the comorbidity between ADHD and hoarding disorder. On the basis of the current interest in
Table 1

<table>
<thead>
<tr>
<th>Model</th>
<th>Description</th>
<th>Evidence for ADHD and hoarding disorder</th>
</tr>
</thead>
<tbody>
<tr>
<td>Chance</td>
<td>The two disorders have separate liabilities and the comorbidity is due to chance alone.</td>
<td>Model rejected: Prevalence of ADHD in those with hoarding disorder is approximately 28% (Frost et al., 2011), which substantially exceeds the expected prevalence of 5% based on community populations (Wilcutt, 2012).</td>
</tr>
<tr>
<td>Sampling bias</td>
<td>Individuals with comorbid conditions are more likely to participate in research than those with either disorder alone, particularly in clinical settings.</td>
<td>Model rejected: Previous studies have used community samples recruited through general advertisements that are unlikely to carry the sampling bias typically linked to clinical samples (e.g., Frost et al., 2011).</td>
</tr>
<tr>
<td>Population stratification</td>
<td>The two disorders have separate risk factors, but each of the risk factors is likely to co-occur in a specific population, such as twin populations.</td>
<td>Model rejected: Frost et al. (2011) advertised widely in the community and had no twin participants thereby suggesting that population stratification effects were unlikely, yet identifying a high comorbidity.</td>
</tr>
<tr>
<td>Alternate forms</td>
<td>The two disorders share a single liability that manifests in either disorder based on chance or environmental risk factors that differ across individuals.</td>
<td>Model not rejected: Preliminary evidence suggests that hoarding disorder and ADHD may share some genetic and neurological aetiological mechanisms. Further understanding of these factors is needed.</td>
</tr>
<tr>
<td>Random multiformity</td>
<td>Having ADHD increases the probability of having hoarding disorder, despite not having elevated liability for hoarding disorder (or the opposite pattern).</td>
<td>Model not rejected: Though it remains untested, it is plausible that having ADHD can generate hoarding symptoms; thus some cases of hoarding may be epiphenomena of ADHD, unrelated to liability for hoarding disorder.</td>
</tr>
<tr>
<td>Extreme multiformity</td>
<td>Like random multiformity, the two disorders have separate liabilities but having hoarding disorder only occurs at the extreme end of ADHD symptoms (or the opposite).</td>
<td>Model rejected: Several authors have reported that hoarding disorder is associated with mild and sub-clinical ADHD (e.g., Hacker et al., 2012).</td>
</tr>
<tr>
<td>Three independent disorders</td>
<td>The comorbid disorder is distinct from either disorder occurring alone.</td>
<td>Model not rejected: Further research is needed to determine if the nature of hoarding and ADHD symptoms in those with the comorbid condition represents a disorder that is distinct from ADHD or hoarding disorder alone.</td>
</tr>
<tr>
<td>Correlated liabilities</td>
<td>The biological and/or environmental risk factors for both disorders are correlated.</td>
<td>Model not rejected: Hoarding disorder and ADHD have shown similar patterns of...</td>
</tr>
</tbody>
</table>
shared aetiological and maintenance factors in ADHD and hoarding disorder (e.g., Fullana et al., 2013; Sheppard et al., 2010), the aim of the present review is to evaluate existing evidence for a correlated liabilities model, including genetic factors, neurological influences, and executive functioning difficulties. This model suggests that certain factors may predict which individuals with ADHD are likely to develop hoarding symptoms. Risk factors include a family history of hoarding disorder, neurological damage to or abnormal functioning in the prefrontal areas of the brain, and impaired executive functioning. While other articles have focussed on related disorders such as the comorbidity between OCD and ADHD (Brem, Grünblatt, Drechsler, Riederer, & Walitza, 2014), to date none has considered the overlap between ADHD and hoarding disorder, which is essential given that hoarding disorder is neurobiologically distinct from OCD (An et al., 2009; Saxena, 2008), and appears to share some distinct areas of neurological dysfunction with ADHD which differs from OCD (Bush et al., 1999; Saxena et al., 2004).

3. Aetiological mechanisms implicated in the ADHD-hoarding disorder link

3.1. Genetic factors

The contribution of genetic components and neural dysfunction associated with ADHD and hoarding disorder has led to a consensus that the aetiology of both disorders includes substantial biological factors. For example, in a comprehensive study of 5022 pairs of twins, Iervolino et al. (2009) reported that genetic factors explained approximately 50% of the variance in hoarding disorder. Samuels, Bienvenu III, et al. (2007) examined hoarding symptoms in 132 relatives of individuals with OCD and hoarding disorder and 259 relatives of individuals with OCD without hoarding disorder. Their results showed that 45% of the relatives of those with hoarding disorder had hoarding symptoms themselves, compared to 33% of the relatives of individuals with OCD without hoarding disorder. Moreover, in a genetic study of 219 families with hoarding disorder, Samuels, Shugart, et al. (2007) established a significant linkage to hoarding disorder on chromosome 14. Several genes have been associated with hoarding disorder, including BDNF (Timpano, Schmidt, Wheaton, Wendland, & Murphy, 2011), SLC1A1 (Wendland et al., 2009), COMT (Lochner et al., 2005), and neurotrophic tyrosine kinase receptor type 3 (Alonso et al., 2008).

The aetiology of ADHD indicates some overlap with the genetic liability of hoarding disorder. As with hoarding disorder, ADHD has a high heritability, with a meta-analysis of 79 twin and adoption studies establishing that the heritability of ADHD was between 71% and 73% (Nikolas & Burt, 2010). The BDNF gene has also been associated with ADHD and may indicate a shared genetic vulnerability between hoarding disorder and ADHD, although the results are inconsistent with further replications needed (Forero, Arboleda, Vasquez, & Arboleda, 2009). However, the literature on the genetics of ADHD suggests that, like most psychiatric disorders, the cause of ADHD is not attributable to a single gene, but is more likely to be the result of multiple small effects on many common gene variants (Forero et al., 2009).

3.2. Neurological factors

Shared neural dysfunctions provide further evidence for a biological link between ADHD and hoarding disorder; specifically, dysfunction in the prefrontal cortex, anterior cingulate gyrus, and amygdala. Greater activation in the ventral prefrontal regions and left amygdala is related to hoarding-specific anxiety (An et al., 2009; Mataix-Cols et al., 2003). For example, An et al. (2009) reported that individuals with hoarding disorder displayed greater activation in the bilateral anterior ventromedial prefrontal cortex when asked to imagine discarding specific objects, which was associated with higher anxiety levels, compared to those without hoarding disorder. Likewise, adolescents with ADHD show greater activity in the amygdala when performing a task of subliminal presentation of fearful faces compared to their typically developing counterparts (Posner et al., 2011). The existing literature also highlights hypoactivity in the anterior cingulate gyrus common to both disorders. For example, individuals with hoarding disorder have lower metabolism in the dorsal anterior cingulate gyrus compared to controls (Saxena et al., 2004), and hypoactivity in the anterior cingulate cortex associated with decision making about objects (Tolin et al., 2012). Likewise, reduced activation in the anterior cingulate gyrus is associated with ADHD (Smith, Taylor, Brammer, Halari, & Rubia, 2008) in addition to reduced grey matter volume in this region (Amico, Stauber, Koutsouleris, & Frodl, 2011; Bledsoe, Semrud-Clikeman, & Pliszka, 2013).

The prefrontal cortex has also been associated with both disorders. However, opposing patterns of dysfunction are apparent, with some studies reporting a greater activation in the anterior ventromedial prefrontal cortex and ventral prefrontal cortex in individuals with hoarding disorder (An et al., 2009) and a global reduction in activity in the prefrontal cortex in those with ADHD (Smith et al., 2008; Wilson et al., 2013). For example, hoarding disorder has been associated with greater activation in the prefrontal cortex, particularly in the orbitofrontal cortex (Mataix-Cols et al., 2004; Tolin, Kiehl, Worhunsky, Book, & Maltby, 2009). In contrast, a meta-analysis of 55 fMRI studies reported that adults with ADHD have hypoactivity in the frontoparietal system (Cortese et al., 2012), while Dickstein, Bannon, Castellanos, and Milham (2006) also found similar patterns

Table 1 (continued)

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<thead>
<tr>
<th>Model</th>
<th>Description</th>
<th>Evidence for ADHD and hoarding disorder</th>
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<tbody>
<tr>
<td>Causal models</td>
<td>One disorder is a liability of the other disorder or both disorders cause each other.</td>
<td>neural abnormalities in the frontal lobe. Moreover researchers have recently suggested that executive functioning deficits may present a shared risk factor. Model not rejected: It is unknown and thus remains a possibility that ADHD and hoarding disorder cause each other.</td>
</tr>
</tbody>
</table>
of hypoactivity within the fronto-striatal circuit in individuals with ADHD.

Beyond these functional deficits, ADHD and hoarding disorder have each been associated with further structural abnormalities in the prefrontal cortex. In an examination of 86 case studies of brain damage undertaken by Anderson, Damasio, and Damasio (2005), hoarding disorder was associated with damage to the mesial frontal region of the brain. Several other case studies have implicated structural damage to the prefrontal cortex in hoarding disorder (Hahm, Kang, Cheong, & Na, 2001; Volle, Beato, Levy, & Dubois, 2002). Likewise, a systematic review of diffusion tensor imaging studies in ADHD found alterations in white matter in the fronto-striatal–cerebellar neurocircuitry in patients with ADHD (van Ewijk, Hesenfeld, Zweiers, Buitelaar, & Oosterlaan, 2012). Individuals with ADHD also have a thinner left medial prefrontal cortex (mPFC; Shaw, Lerch, Greenstein, Sharp, & Clasen, 2006) and delayed cortical maturation in this area (Shaw et al., 2007).

Combined, the current literature highlights a shared vulnerability toward functional and or structural abnormalities in several frontal networks in both ADHD and hoarding disorder. As Castellanos, Sonuga-Barke, Milham, and Tannock (2006) discuss, in their recent paper on ADHD, there is considerable intra-individual variability in ADHD, and the same is true in HD (Preston, Muroff, & Wengrovitz, 2009) while both may be characterised by dysfunction in the circuits driving cognition, motivation and motor behaviour, both internal and external stimuli continue to operate and modify these circuits. The heterogeneity of these disorders means divergence in some areas. For example, the nature of the prefrontal activity dysfunctions in ADHD and hoarding disorder appears to be in conflict, but these differences may have arisen due to a combination of influences specific to one disorder or the other. Further, it is possible that abnormally low and abnormally high activity both result in similar behavioural and cognitive deficits. For example, both high activation of the prefrontal cortex (Ehls, Bähne, Jacob, Herrmann, & Fallgatter, 2008), and low prefrontal activity (Tsujimoto et al., 2013) are associated with executive function deficits in working memory. Despite this opposite pattern of dysfunction, empirical evidence provides support for a shared neurological factor between ADHD and hoarding disorder, whereby both disorders are particularly vulnerable to functional and structural dysfunction in the prefrontal areas. Clarification is needed, however, on the prefrontal abnormalities in individuals with ADHD with comorbid hoarding disorder to determine whether they reflect the ADHD or the hoarding disorder pattern of dysfunction. Moreover, future research on the neural activity in hoarding disorder should consider screening for ADHD to ensure an accurate representation of the hoarding disorder pattern of dysfunction is found.

4. The role of executive functions

Executive functions can be broadly described as higher-order capacities of self-regulation that are instrumental to independent, future-oriented intentional behaviours (Barkley, 2001). These include numerous actions such as planning, inhibition, flexibility, organisation, working memory, updating, and mental set-shifting, among others (Ardila, 2008; Barkley, 2001; Chan, Shum, Toulouropoulos, & Chen, 2008; Miyake et al., 2000). Deficits in these functions are typically associated with abnormal activation in, or damage to, the frontal lobe (Alvarez & Emory, 2006), an area linking ADHD and hoarding disorder as outlined above, including the prefrontal cortex (Brozoski, Brown, Rosvold, & Goldman, 1979) and the anterior cingulate gyrus (Smith, Taylor, Brammer, Toone, & Rubia, 2006; Smith et al., 2008).

Like ADHD and hoarding disorder, executive functions are influenced by genetic factors, as indicated by gene mapping and a high heritability. For example, genetic factors are reported to explain between 29% and 68% of the variance in executive functions in typically developing adults (Lee et al., 2012; Lessov-Slaggar, Swan, Reed, Wolf, & Carmelli, 2007; Swan & Carmelli, 2002). Individuals with ADHD similarly show high heritability of executive functioning, at 77% (Coolidge, Thede, & Young, 2000). Comprehensive genetic studies have identified that the DRD4 gene is implicated in both executive functioning (Barnes, Dean, Nandam, O’Connell, & Belgerove, 2011; Kebir, Tabbane, Sengupta, & Joobter, 2009) and ADHD (Gizer, Ficks, & Waldman, 2009), which may indicate that individuals with ADHD have a genetic vulnerability to executive functioning deficits. Though the attention deficits inherent in many individuals with ADHD may contribute to hoarding symptoms, it is likely that additional executive functioning impairments may better explain which 9% of individuals with ADHD will develop hoarding disorder (Fullana et al., 2013). A review of the literature has identified that the executive functions of working memory, inhibition, and planning have received the strongest support in the hoarding disorder profile and will therefore be discussed in further detail.

4.1. Review method

In this review, the findings regarding the executive functioning of adults with hoarding disorder are evaluated. Studies were identified through searches in PubMed and EBSCO PsychInfo performed in October 2014, using the search terms executive function or neuropsychological or working memory or inhibition or planning and hoarding. The results were limited to peer-reviewed articles, English language publications, and human studies. The most pertinent articles from the above search were selected, as well as two articles from their reference lists, based on their clinical relevance to the present paper.

4.2. Empirical evidence in hoarding disorder

4.2.1. Working memory

Working memory has frequently been examined in the context of hoarding disorder, with findings indicating that spatial working memory difficulties are particularly salient in hoarding disorder. The visuospatial sketchpad is a distinct sub-system of working memory that is responsible for the brief storage of visual information, and creating and manipulating mental images (Baddeley & Hitch, 1974). Researchers have consistently reported that the contrasting verbal memory system is not associated with hoarding disorder (Grisham, Brown, Savage, Steketee, & Barlow, 2007; Hartl et al., 2004; McMillan, Rees, & Pessell, 2013; Tolin, Villavicencio, Umbach, & Kurtz, 2011). This discrepancy between visual and verbal memory is not entirely unexpected given that discarding items involves the visual assessment of locations and the manipulation of this visual information to plan to discard items. Grisham et al. (2007) examined the differences in neuropsychological functions between 30 adults with hoarding disorder and 60 controls. Participants with hoarding disorder had significantly poorer visual memory span compared to controls, suggesting decreased spatial attention and spatial working memory abilities in individuals with hoarding disorder (Grisham et al., 2007). Likewise, Hartl et al. (2004) found that individuals with hoarding disorder showed significantly poorer recall on the Rey-Osterrieth Complex Figure Test (RCFT) after a 20 min delay compared to controls, demonstrating poorer spatial memory encoding. Despite these findings, participants in these samples all had a diagnosis of OCD, which may have inflated their results due to co-occurring OCD symptoms. However, research has indicated that individuals with OCD with hoarding disorder have an executive functioning profile similar to those with hoarding disorder without OCD (Morein-Zamir et al., 2008).
2014); therefore these sampling methods are unlikely to significantly impact results.

In contrast, when comparing a sample of individuals with hoarding disorder to test norms, McMillan et al. (2013) unexpectedly found that individuals with hoarding disorder performed significantly better on the spatial span backward. It remains unclear whether their results represent a better developed spatial working memory system in individuals who hoard or are a reflection of methodological issues. Specifically, the authors suggest that individuals with hoarding disorder may have an improved visuospatial sketchpad due to navigating more cluttered living spaces. In contrast, the results may be attributable to methodological issues such as the lack of a non-clinical control group or learning by participants, with no counterbalancing in the study meaning that spatial span backwards was the fourth memory span test administered to each participant (McMillan et al., 2013). There is therefore a need for clarification regarding the relationship between visual working memory and hoarding disorder.

4.2.2. Planning

Beyond working memory, individuals with hoarding disorder have exhibited weaknesses in planning tasks compared to controls, which is likely to be reflected in their difficulties in planning to discard and acquire items. According to McMillan et al. (2013), individuals with hoarding disorder had significantly more perseveration errors on the Wisconsin Card Sorting Test (WCST) than age-matched population norms, suggesting poorer cognitive flexibility, which can affect planning. Similar results were reported by Grisham, Norberg, Williams, Certoma, and Kadib (2010) in a sample of individuals with hoarding disorder, using the Cambridge Neuropsychological Test Automated Battery (CANTAB) Stockings of Cambridge computerised assessment, which required participants to mentally plan a series of moves before commencing them. Participants with hoarding disorder solved fewer problems within the minimum number of moves than both the non-clinical controls and the mood or anxiety disorders group (Grisham et al., 2010).

Individuals with hoarding disorder have also exhibited difficulties in the ability to plan a series of actions when completing the RCFT, including drawing disjointed elements of the figure when copying (Hartl et al., 2004) and scoring significantly lower on an organisational score (Tolin et al., 2011). Hoarding symptoms are also correlated with significantly lower planning scores on the RCFT during the copying dimension of this task (Pinto et al., 2011). Pinto et al. (2011) examined these associations in a sample of individuals with OCD, which could inflate their results due to co-occurring OCD symptoms. However, Tolin et al. (2011) unexpectedly found that participants with hoarding disorder had no significant differences in the number of moves taken to complete the Tower of London task when compared to controls. The medium effect size in the findings by Tolin et al. (2011) leads the authors to suggest that significant results may be found with a larger sample size. However, Hartl et al. (2004) identified significant results in a hoarding disorder sample size of 22 compared to the 27 participants with hoarding disorder in the study by Tolin et al. (2011). It is therefore unclear whether the inconsistent results by Tolin et al. (2011) are due to the different neuropsychological tasks used or if they demonstrate an inconsistent pattern of planning abilities among those with hoarding disorder.

Planning difficulties may affect the acquiring and discarding dimensions of hoarding disorder, with individuals less likely to plan the specific items they will obtain (Nordsletten, Fernández De La Cruz, Bilotti, & Mataix-Cols, 2013), and more likely to thus accumulate more items than needed or wanted. This may result in a further inability to discard these acquired items, whereby individuals with hoarding disorder struggle to mentally plan and execute the set of actions required to discard items. It would be of interest for further research to examine which symptom dimensions were associated with planning, to provide more accurate treatment targets. Despite spending less time planning items to acquire, individuals with hoarding disorder are likely to show the reverse pattern when discarding items, sometimes planning the steps needed to discard items in great detail. For example, evidence from case studies found that some individuals with hoarding disorder are highly perfectionistic in their planning (Ayers, Bratiotis, Saxena, & Wetherell, 2012). These results are supported by consistent findings of an association between high levels of perfectionism and hoarding symptoms (Frost & Gross, 1993; Frost, Rosenfield, Steketee, & Tolin, 2013; Martinelli, Chasson, Wetterneck, Hart, & Björgvinsson, 2014; Timpano, Exner, et al., 2011). Though initially this process may appear helpful, it can reflect an inability to prioritise appropriately, paradoxically increasing the difficulty of the plan (Ayers et al., 2012), and is therefore likely to affect treatment outcomes (Muroff, Steketee, Frost, & Tolin, 2013). It also highlights the tendency to focus on specific details, with a difficulty in attending to the more global picture, which is also demonstrated in the studies examining completion of the RCFT (Hartl et al., 2004).

4.2.3. Inhibition

A third factor of executive functioning, inhibition, has also been associated with hoarding disorder. Inhibition involves deliberately suppressing a dominant, automatic, or unwanted response (Miskry, 1996). Specifically, individuals with hoarding disorder have shown significantly higher commission errors on tasks of sustained attention compared to nonclinical controls (Grisham et al., 2007; Rasmussen, Brown, Steketee, & Barlow, 2013) and an anxiety group (Grisham et al., 2007). These findings suggest that inhibition difficulties in individuals with hoarding symptoms are beyond those seen in individuals with heightened anxiety. This is consistent with several studies demonstrating increased impulsivity in individuals with hoarding disorder compared to controls (Fitch & Cougle, 2013; Hall et al., 2013; Tolin & Villavicencio, 2011), which may arise from poor inhibition. Older adults with hoarding disorder have similar difficulties of inhibition, with increased non-perseveration errors on the WCST compared to controls (Ayers et al., 2013).

However, several studies have not found significant between-group differences in inhibition tasks, including the CANTAB Affective Go/No-Go task (AGN; Grisham et al., 2010) and the Stop Signal Reaction Time Task (SSRT; Blom et al., 2011). These contrasting results may be indicative of the different neuropsychological tasks used to assess inhibition. Although the CANTAB AGN task assesses response inhibition, Grisham et al. (2010) used mean latency to correct response and total omissions as outcome measures, which are likely to be more indicative of set-shifting skills, a function that may be unimpaired in individuals with hoarding disorder (e.g., Lawrence et al., 2006; Tolin et al., 2011). In contrast, the previous findings of significant between-group differences have assessed inhibition using commission errors, which were not reported in the study by Grisham et al. (2010). The results by Blom et al. (2011) may suggest that previous findings of poorer inhibition in individuals with hoarding disorder do not transfer to difficulties in motor inhibition.

4.3. Empirical evidence of shared executive functioning difficulties in ADHD

The existing literature indicates that the reviewed executive functioning difficulties in hoarding disorder are shared difficulties with ADHD. Like adults with hoarding disorder, individuals with ADHD have also demonstrated spatial working memory difficulties compared to controls. These include significantly more between search error scores on tasks of spatial working memory (Alderson,
Hudec, Patros, & Kasper, 2013; Clark et al., 2007; Dowson et al., 2004; McLean et al., 2004), and significantly higher strategy scores (Dowson et al., 2004; McLean et al., 2004), suggesting a deficient use of a consistent search sequence to locate items. The consistent finding of planning difficulties in adults with hoarding disorder is also the case in individuals with ADHD. Compared to controls, adults with ADHD exhibited significantly lower percentage of correct first choices on a Tower of London task (McLean et al., 2004), significantly shorter planning times (Bramham et al., 2009; Young, Morris, Toone, & Tyson, 2007), no significant increases in planning times on more complex tasks (Young et al., 2007), and create less elaborate plans (Fuermaier et al., 2013). In relation to hoarding symptoms, adults with ADHD may therefore spend inadequate time planning to discard items, which may then lead to increased clutter of items.

Response inhibition difficulties are a final executive function that is common to both hoarding disorder and ADHD and several authors have argued that inhibitory control is the foundational core deficit in ADHD (e.g., Boonstra, Kooy, Oosterlaan, Sergeant, & Buitelaar, 2010; Pazvantoglu et al., 2012). Like individuals with hoarding disorder, adults with ADHD have demonstrated significantly more commission errors on the Continuous Performance Test (Epstein, Johnson, Varia, & Conners, 2001; Murphy, Barkley, & Bush, 2001), significantly slower responding on the go/no-go task (McLean et al., 2004), have less accurate responses, and more variable reactions than controls (Bozorgpour, Klorman, & Gift, 2013). This may specifically help to explain the link between ADHD and hoarding disorder, whereby adults with ADHD have difficulties inhibiting intentional actions that result in increased hoarding symptoms such as acquiring unneeded items. Combined, these studies illustrate that adults with ADHD may be particularly vulnerable to executive functioning difficulties that are shared with hoarding disorder, which may partly explain the association between the two disorders.

5. Summary

This review of empirical findings has shown support for the correlated liabilities model in explaining the comorbidity between ADHD and hoarding disorder. The key executive functions of spatial working memory, planning, and inhibition have been specifically implicated in the hoarding disorder profile. A review of these functions in ADHD has found that adults with ADHD may be particularly vulnerable to difficulties in each of these areas. Despite these findings, and the suggestions that executive functioning deficits may explain the co-occurrence of these two disorders, very few studies have evaluated the potential of a shared vulnerability model in individuals with ADHD and comorbid hoarding disorder. Moreover, the discussed genetic and neural influences on each of hoarding disorder, ADHD, and executive functioning may provide further explanation for the association between ADHD and hoarding disorder (see Fig. 1). For example, executive functioning may provide a marker for genetic vulnerability toward hoarding symptoms in individuals with ADHD.

6. Clinical implications

The findings of this review provide support for a shared vulnerability between hoarding disorder and ADHD, including executive functioning difficulties, genetic vulnerability, and neurological impairments. This has several implications for current treatment approaches. Firstly, hoarding disorder has an early age of onset (Tolin, Meunier, Frost, & Steketee, 2010) with a severe symptom progression (Ayers, Saxena, Golshah, & Wetherell, 2010) that results in particularly disruptive consequences for those with comorbid ADHD. Executive functioning difficulties could therefore provide a target for early screening to identify those individuals who may be on a trajectory to develop hoarding disorder in already at-risk individuals with ADHD. This could result in early intervention to prevent the severe symptom progression and associated impaired functioning in later life. However, further research is needed to determine if the discussed weakness in executive functioning in individuals with hoarding disorder compared to controls is due to objective deficits in these areas.

Secondly, if executive functioning difficulties contribute to the maintenance of hoarding symptoms, treatment implications are suggested. There is currently some evidence that hoarding disorder can be successfully treated using cognitive behavioural therapy (CBT) incorporating specific treatment techniques to expose individuals to avoided situations, restructure their hoarding-related beliefs, and train them in skills decision making, organisation, and problem solving (Muroff, Steketee, Himle, & Frost, 2010; Tolin, Frost, & Steketee, 2007). However, the current review has indicated that specific executive functioning weaknesses may continue to maintain hoarding symptoms, particularly in those with comorbid ADHD. As such, a combination treatment of hoarding symptoms and executive functioning deficits is indicated. Several studies have shown effective treatment of executive dysfunction in individuals with ADHD. These include the use of computerised spatial working memory training programs (Gray et al., 2012; Klingberg et al., 2005), behavioural interventions such as response cost and token approaches that show improvements in inhibition (Siniatchkin et al., 2012), and CBT techniques focusing on self-instruction that improve each of the discussed executive functioning deficits (Miranda, Presentación, Siegenthaler, & Jara, 2011).

Developing evidence in hoarding disorder provides support for a combined treatment approach. For example, a recent pilot study with older adults with hoarding disorder examined a novel treatment of behavioural therapy combined with cognitive rehabilitation, including six sessions targeting executive functioning, such as problem solving, organisational skills, prospective memory, and cognitive flexibility (Ayers et al., 2014). The preliminary findings of a high reduction in hoarding symptom severity following this treatment suggest that combining treatment of hoarding symptoms with executive functioning training may result in improved outcomes for individuals with hoarding disorder (Ayers et al., 2014). In a case analysis of four adults with hoarding disorder who did not have comorbid ADHD, Rodriguez et al. (2013) reported that methylphenidate extended release reduced inattention in all cases and showed a modest reduction in hoarding symptoms in two of the four subjects. Given the prefrontal hyperactivity in hoarding disorder, the positive result with stimulant medication treatment may be somewhat unexpected. However, this highlights the ongoing need for further clarification of the neural activity and medication responses in individuals with hoarding disorder, particularly in those with comorbid ADHD. Further research is required to determine if stimulant medications frequently used in ADHD treatment are effective in treating hoarding symptoms. The long-term prevention of severe hoarding symptoms in individuals with ADHD and comorbid hoarding disorder may thus require a
combination treatment of hoarding symptoms as well as interventions that specifically target the executive functions identified in the hoarding disorder profile.

7. Suggestions for future research

The research reviewed has focused on the influence of genetic factors, neurocognitive impairments, and executive functioning weaknesses on hoarding disorder and ADHD alone; however the nature of these factors in individuals with ADHD and comorbid hoarding disorder remains unknown. It may be helpful for future research to determine the extent to which individuals with comorbid ADHD and hoarding disorder are similar or different from unrelated ADHD and hoarding disorder. Given that both executive functioning and hoarding disorder share common hoarding symptoms that may correspond with opposing planning difficulties. Do the planning impairments in those with comorbid ADHD and hoarding disorder reflect the hoarding disorder profile of complex and perfectionistic plans, or do they follow the ADHD pattern of reduced time spent planning complex tasks?

Clarifying these profiles in those with comorbid ADHD and hoarding disorder could inform treatment approaches. For example, individuals with comorbid ADHD and hoarding disorder may require training to allocate more time on complex plans to discard items, rather than reducing the perfectionistic planning inherent in hoarding disorder. Future attempts to develop and assess a treatment model of hoarding disorder that integrates traditional CBT approaches with interventions that promote executive functioning may reduce the negative impact of hoarding disorder among those with ADHD. In order to improve screening for hoarding disorder and identify appropriate treatment targets, future researchers might also test the correlated liabilities model presented in this review to determine if it should be extended to include other elements common to both disorders, such as emotional regulation. Finally, the available research on executive functioning in the context of hoarding disorder has almost exclusively focused on adult populations. Given that both executive functioning and hoarding disorder are highly dependent on developmental stage (Ayers et al., 2010; De Luca et al., 2009), there is a need to explore these relationships in children and adolescents.

References


Appendix B

DSM-5 Diagnostic Criteria for Hoarding Disorder

A. Persistent difficulty discarding or parting with possessions, regardless of their actual value.
B. This difficulty is due to a perceived need to save the items and to distress associated with discarding them.
C. The difficulty discarding possessions results in the accumulation of possessions that congest and clutter active living areas and substantially compromises their intended use. If living areas are uncluttered, it is only because of the interventions of third parties (e.g., family members, cleaners, authorities).
D. The hoarding causes clinically significant distress or impairment in social, occupational, or other important areas of functioning (including maintaining a safe environment for self and others).
E. The hoarding is not attributable to another medical condition (e.g., brain injury, cerebrovascular disease, Prader-Willi syndrome).
F. The hoarding is not better explained by the symptoms of another mental disorder (e.g., obsessions in obsessive-compulsive disorder, decreased energy in major depressive disorder, delusions in schizophrenia or another psychotic disorder, cognitive deficits in major neurocognitive disorder, restricted interests in autism spectrum disorder).

Specify if:

**With excessive acquisition:** If difficulty discarding possessions is accompanied by excessive acquisition of items that are not needed or for which there is no available space.

Specify if:

**With good or fair insight:** The individual recognizes that hoarding-related beliefs and behaviors (pertaining to difficulty discarding items, clutter, or excessive acquisition) are problematic.

**With poor insight:** The individual is mostly convinced that hoarding-related beliefs and behaviors (pertaining to difficulty discarding items, clutter, or excessive acquisition) are not problematic despite evidence to the contrary.

**With absent insight/delusional beliefs:** The individual is completely convinced that hoarding-related beliefs and behaviors (pertaining to difficulty discarding items, clutter, or excessive acquisition) are not problematic despite evidence to the contrary.

Appendix C

Plain Language Statement Qualitative Study

Plain Language Statement and Consent Form

TO: Prospective participants

Plain Language Statement

Date: August 2013

Full Project Title: Hoarding in children and adolescents with ADHD: Cognitive, emotion regulation, and familial factors

Principal Researcher: Professor Jane McGillivray

Student Researcher: Fiona Lynch

Associate Researcher: Dr. Richard Moulding, Dr. Linda Byrne

Background and Purpose

Recent reports have shown that a high proportion of individuals with hoarding disorder also have ADHD. These hoarding behaviours could be preventable if early detection and treatment is available. The present study aims to investigate the nature of hoarding behaviours in children and adolescents with ADHD. Initial research on hoarding behaviours in this population has prompted the need for clarification of the nature of hoarding behaviours in children and adolescents with ADHD to increase our understanding of the family experience of hoarding.

You and your child have been invited to participate in this research because your child is aged between 8 and 17 years and has a diagnosis of ADHD and you have consented to the researchers contacting you for participation in a further study. You have received this information directly from the researchers.

Procedures

If you agree to participate, you will complete a 30 minute telephone interview at a time convenient to you as arranged with the researchers. This interview will be audio recorded for later transcribing purposes to ensure the accuracy of the data.

Possible benefits

By participating in this project, you and your child are contributing to a very valuable area of research. The results of this project will increase our understanding of the nature of the lived experience of hoarding behaviours in families with children and adolescents with ADHD. It
is anticipated that your results will have potential benefits for numerous health professionals, individuals with ADHD, as well as the general community.

**Inconvenience/discomfort**

There are no anticipated risks to yourself or your child.

If you do find that you are worried about any of your responses or find participating in the project distressing, please contact Professor Jane McGillivray as soon as convenient. If any of these questionnaires cause you or your child distress, services such as Lifeline (13 11 14) or Kids Help Line (1800 55 1800) can be contacted.

**Dissemination of the research results**

A summary of the research findings will be available for interested participants at the completion of the study. To receive a copy of these publications, please inform the researchers at the commencement of your interview.

**Reimbursement**

You will not be paid for your participant in this project.

**Privacy and confidentiality**

You can be assured that we respect your privacy, and information obtained in connection with this project will remain confidential. All information will be stored according to University regulations and kept in a locked cabinet within the School of Psychology at Deakin University for a minimum of 5 years after publication. All transcripts will be labelled only with a unique identification number, and audio recordings of the interview will be stored on a password protected computer.

**Participation is voluntary**

Participating in this study is voluntary and you are not obliged to consent to participate. If you do decide to participate, you are free to withdraw yourselves and your results from the study at any time prior to the completion of the project. Your decision to withdraw will not affect your relationship with Deakin University in any way. Should you have any further questions, please contact a member of the research team prior to consenting to participate. If you decide to withdraw from this project, please notify a member of the research team so your details and data can be removed from the project. Should you wish to withdraw, you can email Fiona Lynch at flynch@deakin.edu.au or Professor Jane McGillivray. If you decide to participate in this project after reading the Plain Language Statement, please sign the consent forms and return them with your questionnaires.

**Complaints**

If you have any complaints about any aspect of the project, the way it is being conducted or any questions about your rights as a research participant, then you may contact:

The Manager, Research Integrity, Deakin University, 221 Burwood Highway, Burwood Victoria 3125, Telephone: 9251 7129, research-ethics@deakin.edu.au

Please quote project number 2013-225.
Further information

If you would like to contact the researchers about any aspect of this study, please contact the Chief Investigator:

Professor Jane McGillivray
Deakin University, School of Psychology
Phone: +61 3 9244 6426
Email: jane.mcgillivray@deakin.edu.au
Appendix D

Ethics Approval Letter

Human Research Ethics
Deakin Research Integrity
70 Elgin Road Burwood Victoria
Postal 221 Burwood Highway
Burwood Victoria 3126 Australia
Telephone: 03 9251 7123 Facsimile 03 9244 6581
research-ethics@deakin.edu.au

Memorandum

To: A/Prof Jane McGillivray
    School of Psychology
    cc: Ms Fiona Lynch

From: Deakin University Human Research Ethics Committee (DUHREC)

Date: 03 October, 2013

Subject: 2013-225
Hoarding in children and adolescents with ADHD: Cognitive, emotion regulation, and familial factors
Please quote this project number in all future communications

The application for this project was considered at the DUHREC meeting held on 23/09/2013.

Approval has been given for Ms Fiona Lynch, under the supervision of A/Prof Jane McGillivray, School of Psychology, to undertake this project from 3/10/2013 to 3/10/2017.

The approval given by the Deakin University Human Research Ethics Committee is given only for the project and for the period as stated in the approval. It is your responsibility to contact the Human Research Ethics Unit immediately should any of the following occur:

- Serious or unexpected adverse effects on the participants
- Any proposed changes in the protocol, including extensions of time
- Any events which might affect the continuing ethical acceptability of the project
- The project is discontinued before the expected date of completion
- Modifications are requested by other HRECs.

In addition you will be required to report on the progress of your project at least once every year and at the conclusion of the project. Failure to report as required will result in suspension of your approval to proceed with the project.

DUHREC may need to audit this project as part of the requirements for monitoring set out in the National Statement on Ethical Conduct in Human Research (2007).

Human Research Ethics Unit
research-ethics@deakin.edu.au
Telephone: 03 9251 7123
Appendix E

Online Questionnaire

1. What is your child’s date of birth (D.O.B)? ……/……../……

2. What is your child’s sex?
   (1) Male  (2) Female  (3) Other

3. Does your child have an Intellectual Disability
   (1) No  (2) Yes
   If yes: Thank you for your interest in this study. Unfortunately you are not eligible to take part in the current research project.

4. Please select your country of residence
   If Australia:
   a. Please select your State or Territory
      (1) Victoria  (2) New South Wales  (3) Queensland  (4) Western Australia  (5) South Australia  (6) Tasmania  (7) Australian Capital Territory  (8) Northern Territory

5. Does your child have a diagnosis of ADHD?
   (1) No  (2) Yes
   If yes:
   5a.1. If you have a copy of your child’s diagnosis, please upload it now or
   5a.2. What type of professional provided your child’s diagnosis of ADHD
      (1) Psychologist  (2) Paediatrician  (3) General Practitioner  (4) Psychiatrist  (5) Other, please specify:........................

5b. How old was your child when they received a diagnosis?

If located in Victoria:
5c. You have the option of your child completing a once-off interactive session taking approximately 1 hour. During this session, your child will complete a range of different tasks mainly involving computerised assessments of attention, planning, and memory. Following completion of the study, you will be mailed a summary of your child’s results on the computerised tasks of attention, planning, and memory.
Would you like the researchers to contact you to determine your interest in this taking part in this session?
   (1) Yes  (2) No
If yes, Please complete the following details and our researchers will contact you within two weeks:

5c.1. Your name:

5c.2. Phone Number:

5c.3. Email:

6. Does your child have an Autism Spectrum Disorder?
   (1) No   (2) Yes
   If Yes: Participants receive different instructions for CSI questionnaire

7. Is your child currently taking any medications?
   (1) No   (2) Yes, please specify………………………………………………

8. Does your child have a diagnosed psychological disorder other than ADHD (e.g., an Anxiety Disorder)?
   (1) No   (2) Yes, please specify………………………………………………

9. Please select your sex
   (1) Male   (2) Female
   (3) Other

10. What is your D.O.B.? ……/……/……

11. What is your occupation? ……………………………………………………………………………

12. What is the highest level of education you have completed?
   (1) Did not finish secondary school   (2) Year 12 or equivalent
   (3) Certificate Level   (4) Diploma/Advanced Diploma
   (5) Graduate Diploma/Graduate Certificate   (6) Bachelor Degree
   (7) Postgraduate Degree

13. Do you have a diagnosed psychological disorder?
    (1) No   (2) Yes, please specify………………………………………………
Children’s Saving Inventory

*Instructions:* Below are some items that describe some children and adolescents. Please select the number that best describes your child in the past week. Please report only on your child’s behaviour and things, not those of others in the family.

<table>
<thead>
<tr>
<th>None</th>
<th>A little/Minimal</th>
<th>Some/Moderate</th>
<th>Most/Much</th>
<th>Almost All/Complete</th>
</tr>
</thead>
<tbody>
<tr>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
</tbody>
</table>

1. How much of your child’s room (or where s/he sleeps, plays etc.) is cluttered with possessions? 0 1 2 3 4
2. How much control does your child have over his/her urges to acquire possessions that s/he does not need? 0 1 2 3 4
3. How much time do you spend dealing with your child’s possessions (e.g., organising, discarding, arranging)? 0 1 2 3 4
4. How much control does your child have over his/her urges to save possessions that s/he does not need? 0 1 2 3 4
5. How much of your home is difficult to walk through because of your child’s clutter? 0 1 2 3 4

<table>
<thead>
<tr>
<th>Not at all</th>
<th>Mild</th>
<th>Moderate</th>
<th>Considerable/Severe</th>
<th>Extreme</th>
</tr>
</thead>
<tbody>
<tr>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
</tbody>
</table>

6. To what extent does your child have difficulty throwing things away that s/he does not need? 0 1 2 3 4
7. How distressing does your child find the task of throwing things away? 0 1 2 3 4
8. To what extent does your child get upset when other people touch or move his/her things? 0 1 2 3 4
9. Tow what extent does your child get upset when you (or another adult) remove or throw away items that you do not think your child needs? 0 1 2 3 4
10. How distressed or uncomfortable would your child become if he/she could not acquire something he/she wanted (but didn’t need)? 0 1 2 3 4
11. To what extent does attachment to things interfere with your child’s functioning at school, at home, or with friends? 0 1 2 3 4
12. How strong is your child’s urge to buy or acquire free things for which he/she has no immediate use? 0 1 2 3 4
13. How strong is your child’s urge to save something you know he/she may never use? 0 1 2 3 4
14. How much does your child’s attachment to things interfere with his/her relationships with other children or family members? 0 1 2 3 4
<table>
<thead>
<tr>
<th>Question</th>
<th>Never</th>
<th>Rarely</th>
<th>Sometimes/Occasionally</th>
<th>Frequently/Often</th>
<th>Very Often</th>
</tr>
</thead>
<tbody>
<tr>
<td>15. How often does your child avoid trying to discard possessions</td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>because it is too stressful or time consuming?</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>16. How often does your child feel compelled to acquire something</td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>he/she sees? e.g., when shopping or offered free things?</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>17. How often does your child decide to keep things he/she does not</td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>need and has little space for?</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>18. How frequently does your child’s clutter in the home prevent you</td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>from inviting people to visit?</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>19. How often does your child actually buy (or insist that you buy or</td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>acquire for free) things for which he/she has no immediate use or need?</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>20. How frequently do you avoid taking your child with you when you</td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>go shopping because of his/her acquiring problem?</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>21. To what extent does your child have so many things that his/her</td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>room, play area, etc. are cluttered?</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>22. To what extent does your child’s clutter prevent him/her or other</td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>family members from using part of the home for its intended purpose</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>(e.g., sleeping in his/her bed, using a bathroom sink, using his/her</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>desk, etc.)?</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>23. How often is your child unable to discard a possession you would</td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>like him/her to get rid of?</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
BRIEF: Behavior Rating Inventory of Executive Functioning
Parent Form

Instructions: Below is a list of statements. We would like to know if your child has had problems with these behaviours over the past 6 months. Please answer all the items the best that you can. Please DO NOT SKIP ANY ITEMS. Think about your child as you read each statement and circle your response:

- **N** if the behaviour is **Never** a problem
- **S** if the behaviour is **Sometimes** a problem
- **O** if the behaviour is **Often** a problem

For example, if your child **never** has trouble completing homework on time, you would select **N** for this item:

Has trouble completing homework on time

<table>
<thead>
<tr>
<th></th>
<th>N</th>
<th>S</th>
<th>O</th>
</tr>
</thead>
<tbody>
<tr>
<td>1.</td>
<td>Overreacts to small problems</td>
<td>N</td>
<td>S</td>
</tr>
<tr>
<td>2.</td>
<td>When given three things to do, remembers only the first or last</td>
<td>N</td>
<td>S</td>
</tr>
<tr>
<td>3.</td>
<td>Is not a self-starter</td>
<td>N</td>
<td>S</td>
</tr>
<tr>
<td>4.</td>
<td>Leaves playroom a mess</td>
<td>N</td>
<td>S</td>
</tr>
<tr>
<td>5.</td>
<td>Resists or has trouble accepting a different way to solve a problem with schoolwork, friends, chores, etc.</td>
<td>N</td>
<td>S</td>
</tr>
<tr>
<td>6.</td>
<td>Becomes upset with new situations</td>
<td>N</td>
<td>S</td>
</tr>
<tr>
<td>7.</td>
<td>Has explosive, angry outbursts</td>
<td>N</td>
<td>S</td>
</tr>
<tr>
<td>8.</td>
<td>Tries the same approach to a problem over and over even when it does not work</td>
<td>N</td>
<td>S</td>
</tr>
<tr>
<td>9.</td>
<td>Has a short attention span</td>
<td>N</td>
<td>S</td>
</tr>
<tr>
<td>10.</td>
<td>Needs to be told to begin a task even when willing</td>
<td>N</td>
<td>S</td>
</tr>
<tr>
<td>11.</td>
<td>Does not bring home homework, assignment sheets, materials, etc.</td>
<td>N</td>
<td>S</td>
</tr>
<tr>
<td>12.</td>
<td>Acts upset by a change in plans</td>
<td>N</td>
<td>S</td>
</tr>
<tr>
<td>13.</td>
<td>Is disturbed by change of teacher or class</td>
<td>N</td>
<td>S</td>
</tr>
<tr>
<td>14.</td>
<td>Does not check work for mistakes</td>
<td>N</td>
<td>S</td>
</tr>
<tr>
<td>15.</td>
<td>Has good ideas but cannot get them on paper</td>
<td>N</td>
<td>S</td>
</tr>
<tr>
<td>16.</td>
<td>Has trouble coming up with ideas for what to do in play or free time</td>
<td>N</td>
<td>S</td>
</tr>
<tr>
<td>17.</td>
<td>Has trouble concentrating on chores, schoolwork, etc.</td>
<td>N</td>
<td>S</td>
</tr>
<tr>
<td>18.</td>
<td>Does not connect doing tonight’s homework with grades</td>
<td>N</td>
<td>S</td>
</tr>
<tr>
<td>19.</td>
<td>Is easily distracted by noises, activity, sights etc.</td>
<td>N</td>
<td>S</td>
</tr>
<tr>
<td>20.</td>
<td>Becomes tearful easily</td>
<td>N</td>
<td>S</td>
</tr>
<tr>
<td>21.</td>
<td>Makes careless errors</td>
<td>N</td>
<td>S</td>
</tr>
<tr>
<td>22.</td>
<td>Forgets to hand in homework, even when completed</td>
<td>N</td>
<td>S</td>
</tr>
<tr>
<td>23.</td>
<td>Resists change of routine, foods, places, etc.</td>
<td>N</td>
<td>S</td>
</tr>
<tr>
<td>24.</td>
<td>Has trouble with chores or tasks that have more than one step</td>
<td>N</td>
<td>S</td>
</tr>
<tr>
<td>25.</td>
<td>Has outbursts for little reason</td>
<td>N</td>
<td>S</td>
</tr>
<tr>
<td>26.</td>
<td>Mood changes frequently</td>
<td>N</td>
<td>S</td>
</tr>
<tr>
<td>27.</td>
<td>Needs help from an adult to stay on task</td>
<td>N</td>
<td>S</td>
</tr>
<tr>
<td>28.</td>
<td>Gets caught up in details and misses the big picture</td>
<td>N</td>
<td>S</td>
</tr>
<tr>
<td>29.</td>
<td>Keeps room messy</td>
<td>N</td>
<td>S</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>---</td>
<td>---</td>
<td>---</td>
<td>---</td>
</tr>
<tr>
<td>30. Has trouble getting used to new situations (classes, groups, friends)</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>31. Has poor handwriting</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>32. Forgets what he/she was doing</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>33. When sent to get something, forgets what he/she is supposed to get</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>34. Is unaware of how his/her behaviour affects or bothers others</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>35. Has good ideas but does not get job done (lacks follow-through)</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>36. Becomes overwhelmed by large assignments</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>37. Has trouble finishing tasks (chores, homework)</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>38. Acts wilder or sillier than others in groups (birthday parties, recess)</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>39. Thinks too much about the same topic</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>40. Underestimates time needed to finish tasks</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>41. Interrupts others</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>42. Does not notice when his/her behaviour causes negative reactions</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>43. Gets out of seat at the wrong times</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>44. Gets out of control more than friends</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>45. Reacts more strongly to situations than other children</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>46. Starts assignments or chores at the last minute</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>47. Has trouble getting started on homework or chores</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>48. Has trouble organising activities with friends</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>49. Blurs things out</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>50. Mood is easily influenced by the situation</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>51. Does not plan ahead for school assignments</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>52. Has poor understanding of own strengths and weaknesses</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>53. Written work is poorly organised</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>54. Acts too wild or “out of control”</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>55. Has trouble putting the brakes on his/her actions</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>56. Gets in trouble if not supervised by an adult</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>57. Has trouble remembering things, even for a few minutes</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>58. Has trouble carrying out the actions needed to reach goals (saving money for special item, studying to get a good grade)</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>59. Becomes too silly</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>60. Work is sloppy</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>61. Does not take initiative</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>62. Angry or tearful outbursts are intense but end suddenly</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>63. Does not realise that certain actions bother others</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>64. Small events trigger big reactions</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>65. Talks at the wrong time</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>66. Complains there is nothing to do</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>67. Cannot find things in room or school desk</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>68. Leaves a trail of belongings wherever he/she goes</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>69. Leaves messes that others have to clean up</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>70. Becomes upset too easily</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>71. Lies around the house a lot (“couch potato”)</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
<tr>
<td>72. Has a messy closet</td>
<td>N</td>
<td>S</td>
<td>O</td>
</tr>
</tbody>
</table>
**Vanderbilt ADHD Diagnostic Parent Rating Scale**

*Instructions:* Each rating should be considered in the context of what is appropriate for the age of your child. When completing this form, please think about your child’s behaviours in the past 6 months.

**Behaviour:**

<table>
<thead>
<tr>
<th>Never</th>
<th>Occasionally</th>
<th>Often</th>
<th>Very Often</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Does not pay attention to details or makes careless mistakes, for example homework</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>2. Has difficulty sustaining attention to tasks or activities</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>3. Does not seem to listen when spoken to directly</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>4. Does not follow through on instructions and fails to finish schoolwork (not due to oppositional behaviour or failure to understand)</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>5. Has difficulty organizing tasks and activities</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>6. Avoids, dislikes, or is reluctant to engage in tasks that require sustained mental effort</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>7. Loses things necessary for tasks or activities (school assignments, pencils or books)</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>8. Is easily distracted by extraneous stimuli</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>9. Is forgetful in daily activities</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>10. Fidgets with hands or feet or squirms in seat</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>11. Leaves seat when remaining seated is expected</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>12. Runs about or climbs excessively in situations when remaining seated is expected</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>13. Has difficulty playing or engaging in leisure/play activities quietly</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>14. Is “on the go” or often acts as if “drive by a motor”</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>15. Talks too much</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>16. Blurts out answers before questions have been completed</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>17. Has difficulty waiting his/her turn</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>18. Interrupts or intrudes on others (e.g., butts into conversations or games)</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>19. Argues with adults</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>20. Loses temper</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>21. Actively defies or refuses to comply with adults’ requests or rules</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>22. Deliberately annoys people</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>23. Blames others for his or her mistakes or misbehaviours</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>24. Is touchy or easily annoyed by others</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>25. Is angry or resentful</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>26. Is spiteful and vindictive</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>27. Bullies, threatens, or intimidates others</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>28. Initiates physical fights</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>---</td>
<td>---</td>
<td>---</td>
<td>---</td>
</tr>
<tr>
<td>29. Lies to obtain goods for favours or to avoid obligations (i.e., “cons” others)</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>30. Is truant from school (skips school) without permission</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>31. Is physically cruel to people</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>32. Has stolen items of nontrivial value</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>33. Deliberately destroys others’ property</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>34. Has used a weapon that can cause serious harm (bat, knife, brick, gun)</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>35. Is physically cruel to animals</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>36. Has deliberately set fires to cause damage</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>37. Has broken into someone else’s home, business, or car</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>38. Has stayed out at night without permission</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>39. Has run away from home overnight</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>40. Has forced someone into sexual activity</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>41. Is fearful, anxious, or worried</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>42. Is afraid to try new things for fear of making mistakes</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>43. Feels worthless or inferior</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>44. Blames self for problems, feels guilty</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>45. Feels lonely, unwanted, or unloved: complains that “no one loves him/her”</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>46. Is sad, unhappy, or depressed</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>47. Is self-conscious or easily embarrassed</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
</tbody>
</table>

**Academic & Social Performance:**

<table>
<thead>
<tr>
<th></th>
<th>Excellent</th>
<th>Above Average</th>
<th>Average</th>
<th>Somewhat of a Problem</th>
<th>Problematic</th>
</tr>
</thead>
<tbody>
<tr>
<td>48. Overall school performance</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
</tr>
<tr>
<td>49. Reading</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
</tr>
<tr>
<td>50. Writing</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
</tr>
<tr>
<td>51. Mathematics</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
</tr>
<tr>
<td>52. Relationship with parents</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
</tr>
<tr>
<td>53. Relationship with siblings</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
</tr>
<tr>
<td>54. Relationship with peers</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
</tr>
<tr>
<td>55. Participation in organized activities (eg, teams)</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
</tr>
</tbody>
</table>
Appendix F

Plain Language Statement for Online Questionnaire

PLAIN LANGUAGE STATEMENT AND CONSENT FORM

TO: Prospective participants

Plain Language Statement

Date: February 2015

Full Project Title: Hoarding in children and adolescents with ADHD: Cognitive, emotion regulation, and familial factors

Principal Researcher: Professor Jane McGillivray

Student Researcher: Fiona Lynch

Associate Researcher: Dr Richard Moulding, Dr Linda Byrne

Background and Purpose

Hoarding disorder has an onset in childhood and adolescents, despite often going unnoticed until adulthood. Early detection and treatment should therefore be available at a young age. The present study aims to investigate the specific influences on hoarding behaviours in children and adolescents to identify these early treatment targets when intervention may be most effective. The present study will examine the relationship between attention, planning, memory skills, decision making, emotion regulation, and familial patterns with behaviours of collecting, acquiring and difficulty discarding in children and adolescents who are typically developing as well as those with ADHD.

You are invited to participate in this research if your child is aged between 8 and 17 years, and does not have a diagnosis of an intellectual disability.

Procedures

If you agree to participate, you will be required to complete some questions on behaviours relating to collecting, discarding, attention, planning, and memory of yourself and your child. These will take approximately 40 minutes. If you wish to complete the questionnaires in hardcopy, please contact the researchers to receive a mailed-out copy of the questionnaires.

If your child has ADHD, a formal diagnosis from a professional (GP, Paediatrician, Clinical Psychologist) is a requirement of participation. Where you have a copy of a written report confirming the diagnosis, we would appreciate a copy of that report. If the diagnosis was conveyed verbally, we would appreciate a description of who provided the diagnosis, what type of professional were they, and approximately how old your child was when it was...
received. The questionnaire includes these questions for you to complete. You are not expected to seek or pay for a written report.

**Possible benefits**

By participating in this project, you and your child are contributing to a very valuable area of research. The results of this project will increase our understanding of the interaction between attention, memory, planning, decision making, emotion regulation and hoarding behaviours in children and adolescents. It is anticipated that your results will have potential benefits for numerous health professionals, children with hoarding, individuals with ADHD, individuals at risk of hoarding, as well as the general community.

**Inconvenience/discomfort**

There are no anticipated risks to yourself or your child.

If you do find that you are worried about any of your responses or find participating in the project distressing, please contact Professor Jane McGillivray as soon as convenient. If any of these questionnaires cause you or your child distress, services such as Lifeline (13 11 14) or Kids Help Line (1800 55 1800) can be contacted.

**Dissemination of the research results**

A summary of the research findings will be available for interested participants at the completion of the study. To receive a copy of these publications, please email the researchers at flynch@deakin.edu.au.

**Reimbursement**

You will not be paid for your participant in this project. As a token of our appreciation, Australian participants have the option to enter a draw to win a Coles-Myer voucher. If you wish to enter this draw, please complete your details at the end of the questionnaire. You will be contacted directly by the researchers should you receive this voucher.

**Privacy and confidentiality**

You can be assured that we respect your privacy, and information obtained in connection with this project will remain confidential. All information will be stored according to University regulations and kept in a locked cabinet within the School of Psychology at Deakin University for a minimum of 5 years after publication. Electronic information will be stored on a password protected computer.

**Participation is voluntary**

Participating in this study is voluntary and you are not obliged to consent to participate. If you do decide to participate, you are free to withdraw yourselves and your results from the study at any time prior to the completion of the project. Your decision to withdraw will not affect your relationship with Deakin University in any way. Should you have any further questions, please contact a member of the research team prior to consenting to participate. If you decide to withdraw from this project, please notify a member of the research team so your details and data can be removed from the project. Should you wish to withdraw, you can email Fiona Lynch at flynch@deakin.edu.au or Professor Jane McGillivray. If you decide to
participate in this project after reading the Plain Language Statement, please click the button below and complete the questionnaires.

**Complaints**

If you have any complaints about any aspect of the project, the way it is being conducted or any questions about your rights as a research participant, then you may contact:

The Manager, Research Integrity, Deakin University, 221 Burwood Highway, Burwood Victoria 3125, Telephone: 9251 7129, research-ethics@deakin.edu.au

Please quote project number 2013-225.

**Further information**

If you would like to contact the researchers about any aspect of this study, please contact the Chief Investigator:

Professor Jane McGillivray  
Deakin University, School of Psychology  
Phone: +61 3 9244 6426  
Email: jane.mcgillivray@deakin.edu.au

If you would like to participate in this project, please click the following button to indicate that you have read the plain language statement and agree to give your consent to participate in the study.

[I agree]
Plain Language Statement and Consent Form

TO: Prospective participants

<table>
<thead>
<tr>
<th>Date: August 2013</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Full Project Title:</strong> Hoarding in children and adolescents with ADHD: Cognitive, emotion regulation, and familial factors</td>
</tr>
<tr>
<td><strong>Principal Researcher:</strong> Professor Jane McGillivray</td>
</tr>
<tr>
<td><strong>Student Researcher:</strong> Fiona Lynch</td>
</tr>
<tr>
<td><strong>Associate Researchers:</strong> Dr. Richard Moulding, Dr. Linda Byrne</td>
</tr>
</tbody>
</table>

**Background and Purpose**

Recent reports have shown that a high proportion of individuals with hoarding disorder also have ADHD. These hoarding behaviours could be preventable if early detection and treatment is available. The present study aims to investigate the specific influences on hoarding behaviours in children and adolescents to identify these early treatment targets when intervention may be most effective. The present study will examine the relationships between attention, planning, memory skills, decision making, with behaviours of collecting, acquiring and difficulty discarding in children and adolescents with ADHD.

You and your child have been invited to participate in this research because your child is aged between 8 and 17 years, has a diagnosis of ADHD, and you indicated to the researchers that you were interested in taking part. You have received this information directly from the researchers.

**Procedures**

If you agree to participate your child will complete a range of different tasks mainly involving computerised assessments of attention, planning, and memory, which will take approximately 1 hour. This testing session will be at the Burwood campus of Deakin University unless otherwise arranged with you. It is important that you also explain the purpose of this study to your child, and ask them if they are willing to participate. A separate explanatory statement for your child has also been provided. Following completion of the study, you will be mailed a summary of your child’s results on the computerised tasks of attention, planning, and memory. These will be simple percentiles, and will not identify if your child is particularly strong or weak in any skill.

**Possible benefits**

By participating in this project, you and your child are contributing to a very valuable area of research. The results of this project will increase our understanding of the interaction between
attention, memory, planning, decision making, emotion regulation and hoarding behaviours in children and adolescents with ADHD. It is anticipated that your results will have potential benefits for numerous health professionals, individuals with ADHD, as well as the general community.

Inconvenience/discomfort

There are no anticipated risks to yourself or your child.

If you do find that you are worried about any of your responses or find participating in the project distressing, please contact Professor Jane McGillivray as soon as convenient. If any of these questionnaires cause you or your child distress, services such as Lifeline (13 11 14) or Kids Help Line (1800 55 1800) can be contacted.

Reimbursement

You will not be paid for your participant in this project. As a token of our appreciation, you have the option to enter a draw to win a Coles-Myer voucher. If you wish to enter this draw, please tick the appropriate box on the consent form. You will be contacted directly by the researchers should you receive this voucher.

Privacy and confidentiality

You can be assured that we respect your privacy, and information obtained in connection with this project will remain confidential. All information will be stored according to University regulations and kept in a locked cabinet within the School of Psychology at Deakin University for a minimum of 5 years after publication. Electronic information will be stored on a password protected computer.

Participation is voluntary

Participating in this study is voluntary and both you and your child are not obliged to consent to participate. If you do decide to participate, you are free to withdraw yourselves and your results from the study at any time prior to the completion of the project. Your decision to withdraw will not affect your relationship with Deakin University in any way. Should you have any further questions, please contact a member of the research team prior to consenting to participate. If you decide to withdraw from this project, please notify a member of the research team so your details and data can be removed from the project. Should you wish to withdraw, you can email Fiona Lynch at flynch@deakin.edu.au or Professor Jane McGillivray.

Complaints

If you have any complaints about any aspect of the project, the way it is being conducted or any questions about your rights as a research participant, then you may contact:

The Manager, Research Integrity, Deakin University, 221 Burwood Highway, Burwood Victoria 3125, Telephone: 9251 7129, research-ethics@deakin.edu.au

Please quote project number 2013-225.

Further information

If you would like to contact the researchers about any aspect of this study, please contact the Chief Investigator:

Professor Jane McGillivray
Deakin University, School of Psychology
Phone: +61 3 9244 6426
Email: jane.mcgillivray@deakin.edu.au
Hello,

My name is Fiona and I am a student at Deakin University. I am doing a project for my degree on how children think and behave. I want to know the ways children do activities and how they collect and throw out items in their room. I hope this will help me to understand more about how children choose which items to keep and which items throw out.

I would like you and other children in your neighbourhood to be to be part of my project. If you agree to take part you will do some short activities with me. The activities will involve some fun computer activities like looking for shapes, arrows and numbers, as well as some math and word activities. The activities are easy to complete and you will have time to practice them before you complete them. They will take around 1 hour to complete.

When the project is finished I will write a report and it might be published as articles. If I use your results in the articles, you will not be named or have any details written about you.

This project is voluntary, so you can do it or not, and you can change your mind about it later. You just have to tell me or your parents and we will take you out of the project. You won’t have to explain why.

You can ask questions about the project at any time, including during the activities. If you feel worried about the project, or have any questions, you can talk to me or your parents. At the end of the project, your parents will get a short report on how you did on the tasks compared to most children your age. If you do not want them to get this, please let us know.

Thank you for thinking about helping me to find out more about how children think and behave. If you are happy to take part in the project, tell your parents that you would like to take part.

If you have any questions, please ask me.

Fiona Lynch