Asian Journal of Psychiatry 42 (2019) 104–114

Contents lists available at ScienceDirect

Asian Journal of Psychiatry

journal homepage: www.elsevier.com/locate/ajp

A narrative synthesis of possible causes and risk factors of hoarding behaviours

Aditi Hombali[a], Vathsala Sagayadevan[a], Weng Mooi Tan[b], Rebecca Chong[b], Hon Weng Yip[c], Janhavi Vaingankar[a], Siow Ann Chong[a], Mythily Subramaniam[a,*]

[a] Research Division, Institute of Mental Health, Buangkok Green Medical Park, 10 Buangkok View, 539747, Singapore
[b] Agency for Integrated Care, Community Mental Health Division, Singapore
[c] Policy & Planning Division, Municipal Services Office, Singapore

ARTICLE INFO

Keywords:
Hoarding disorder
Review
Causes
Risk factors

ABSTRACT

Background: Hoarding is a disorder characterized by excessive acquisition and persistent difficulty in discarding possessions. The behaviour has adverse emotional, physical, social, financial, and legal outcomes for the person with the disorder and family members, and might pose a significant public health problem. Hoarding has been included as a distinct disorder in the Diagnostic and Statistical Manual of Mental Disorders Fifth edition (DSM-5). The prevalence of hoarding disorder is approximately 2–6% globally. The current state of the evidence does not offer clear understanding of the causes of hoarding behaviours. A systematic review of the extant literature was carried out to determine the possible causal factors of hoarding behaviours.

Methods: This review is conducted in line with PRISMA guidelines. The following electronic databases: Medline through Ovid, EMBASE and PsycINFO were searched for relevant articles published between January 2000 and November 2018. Only articles published in English language were included. Two reviewers independently scrutinized the studies and included them in this review.

Results: Our search strategy returned a total of 396 references. Preliminary findings suggest that individuals with hoarding behaviours may have a genetic susceptibility; abnormal neural activity in the fronto-temporal, parahippocampal gyrus and insular parts of the brain has also been identified. Traumatic life experiences have also been posited to predispose individuals to hoard.

Conclusion: Although the understanding of hoarding disorder hasgrown in recent years, greater efforts are still needed to clarify the etiology and mechanisms of hoarding disorder as these may help in planning of more holistic interventions to treat the problem.

1. Introduction

Hoarding is a disorder that is characterized by a persistent difficulty in discarding or parting with possessions regardless of their actual value. The individual has strong urges to save items and experiences distress discarding them, which results in the accumulation of a high degree of clutter (Timpano et al., 2011). Hoarding symptoms have been linked with increased psychiatric co-morbidity and lower functioning levels which not only have negative repercussions for the individual, but also the family, and the wider community (Timpano et al., 2011; Tolin et al., 2008a,b). The accumulation of trash, food, and/or animals by the person with hoarding can create serious threats to personal safety and pose fire and health risks. These issues can affect the person with hoarding, their family and extend to their neighbours and beyond.

Epidemiological studies have found the prevalence of hoarding symptoms to range between 2 and 6% in community samples (Timpano et al., 2011; Iervolino et al., 2009; Nordsletten et al., 2013a; Samuels et al., 2008) and a weighted prevalence of 1.5% using the Diagnostic and Statistical Manual of Mental Disorders – Fifth edition (DSM-5) (American Psychiatric Association, 2013) criteria for Hoarding Disorder (HD) (Nordsletten et al., 2013a; Albert et al., 2015). Hoarding was previously classified as a symptom criterion for obsessive-compulsive disorder (OCD) and obsessive-compulsive personality disorder (OCPD) (Pertusa et al., 2010; Pertusa et al., 2008; Frost et al., 2011), however, hoarding now represents a clinically distinct entity (Albert et al., 2015; Mataix-Cols et al., 2010; Mataix-Cols and Pertusa, 2012; Saxena et al., 2007). Several studies have also identified hoarding behaviours in the absence of other symptoms of OCD (Frost et al., 2000; Samuels et al.,

* Corresponding author.
E-mail address: mythily@imh.com.sg (M. Subramaniam).

https://doi.org/10.1016/j.ajp.2019.04.001
Received 20 July 2018; Received in revised form 7 March 2019; Accepted 7 April 2019

2002a, b; Pertusa et al., 2012). Frost et al (Frost et al., 2011) for instance, found that only 18% of individuals with pathological hoarding have a co-morbid OCD diagnosis, and that it occurs with other psychiatric conditions such as major depressive disorder (51%), social phobia (24%), and generalised anxiety disorder (24%).

In line with the emerging evidence, the recent revision to the DSM-5 has classified hoarding as a separate disorder under the Obsessive Compulsive Related Disorders section. The diagnostic criteria for HD as delineated by the DSM-5 include the following (American Psychiatric Association, 2013):

- Persistent difficulty in discarding or parting with possessions regardless of their actual value that results in clutter compromising the use of active living spaces;
- Significant distress or impairment in social, occupational, or other areas of functioning;
- Hoarding is not attributable to another medical condition i.e. organic hoarding (Mataix-Cols et al., 2011) (e.g., brain injury, cerebrovascular disease, Prader –Willi Syndrome)
- Hoarding in these cases is not explained by symptoms of other mental disorders (e.g., delusions in schizophrenia, cognitive deficits in dementia etc.)

1.1. Psychological theories of hoarding

According to the cognitive-behavioural model proposed by Frost and colleagues (Frost and Hartl, 1996; Steketee and Frost, 2003), compulsive hoarding has been conceptualized as a multifaceted problem resulting from information processing deficits, emotional attachment problems, behavioural avoidance, and dysfunctional beliefs about the nature of possessions.

Information processing deficits comprise three deficits. Firstly, there is indecisiveness; it is suggested that individuals with hoarding disorder have a tendency to avoid making and/or postponing decisions that may stem from a fear of making mistakes. Saving everything may be a way of avoiding decision-making. Individuals with hoarding disorder may also perceive their possession(s) to be of high value, which includes both an instrumental or material value and a sentimental value. Thus, the decision to save is reinforced. They may also make erroneous decisions about the probability of future use for an item and they may see the item as being highly necessary and the thought of discarding it is unpleasant, as the act could be seen as being harmful to them or even to the possession. All these factors lead to difficulty in discarding and to the saving of all possessions regardless of real value or need. Secondly, individuals with hoarding disorder are unable to categorise and organize their possessions. They tend to have too many categories leading to piling of objects, their inability to categorise items leads to the creation of new piles (a process referred to as churning) rather than the storage/discarding of items. They also tend to mix important with unimportant items due to their inability to categorise and this in turn leads to an inability to discard as they are fearful of losing valuable possessions. This often results in the loss of valuables, including stored currency, during the process of enforced decluttering. Lastly, there might be memory deficits i.e. a lack of confidence in remembering where things are kept and hence everything is kept in easy to spot locations and is accessible when needed.

There are also emotional attachment problems i.e. individuals with hoarding disorder view many of their possessions as extensions of themselves, they may imbue them with human-like properties and thus feel very sentimental towards their possessions and have difficulties discarding them. These possessions may also afford them a sense of comfort or security and hence if other people touch, move or use the possessions they may feel very distressed and violated. Tied in to the indecisiveness and emotional attachment is behavioural avoidance wherein the individuals with hoarding disorder avoid the task of discarding as it is perceived as unpleasant and difficult. They also are fearful of making the wrong decision, and hence not making a decision about where to put the item and what to discard is preferred, leading to clutter and hoarding.

Evidence for socio-demographic correlates of hoarding remains inconclusive, partly due to methodological differences across studies. While the mean clinical onset of hoarding symptoms is often reported during childhood or adolescence, hoarding severity appears to increase with age (Ayers et al., 2010) with hoarding symptoms being almost three times more prevalent in older adults (ages 55–94 years) compared to younger adults (age 34–44 years) (7). With regards to gender differences, Iervolino et al (Iervolino et al., 2009) found a significantly higher prevalence of hoarding behaviour among men, while Mueller et al. (2009) and Timpano et al (Timpano et al., 2011) did not find a gender difference. Samuels et al (Samuels et al., 2008) found hoarding to be more prevalent in the older age groups, in men, and to be inversely related to household income. While some studies have suggested that individuals with hoarding problems are less likely to be married (Pertusa et al., 2010; Frost and Gross, 1993), Bulli et al (Bulli et al., 2014) did not find any differences in terms of gender, age, marital status, level of education, and employment status in an Italian non-clinical sample. Thus overall, studies seem to suggest that hoarding is not associated with any specific gender, ethnicity or socio-economic factor and that the prevalence and severity seems to be higher among older adults.

Although research has suggested genetic, neurological, psychosocial and emotional factors as possible broad causal factors for hoarding disorder, the cause of HD is yet to be determined. Given that research on HD is still in its nascent stages (Brakoulias and Milicevic, 2015) an in-depth understanding of the causes is important to gain insight into the disorder. Furthermore, a clear understanding of the risk-factors of HD could aid in informing evidence-based treatments (which are limited) for this disorder (Brakoulias and Milicevic, 2015; Thompson et al., 2017). This review aimed to determine the possible causal factors of hoarding behaviours.

2. Methodology

The types of studies that were included for this review were cross-sectional, case-control and cohort studies that investigated the possible causal factors of hoarding disorder. Studies were eligible if the i) participants were diagnosed clinically with hoarding disorder based on DSM-5 criteria; ii) included self- reported hoarding behaviours; iii) the hoarding behaviours were assessed using hoarding assessment scales; and iv) they were published in peer-reviewed journals. Studies were excluded if they i) presented insufficient information on causal association; ii) were review articles; iii) were not published in English.

2.1. Search methods for identification of studies

A combination of MESH terms and key words related to hoarding disorder and causes were used to search the electronic databases OVID (Medline), EMBASE, and CINHAL for relevant articles published between January 2000 and November 2018. We hand- searched the bibliography of included reports to identify additional cross-references that could be considered for inclusion in the review.

2.2. Data collection and analysis

Two researchers (AH & VS) independently screened the titles and abstracts retrieved using the search strategy. The full texts for the shortlisted references were retrieved and assessed for eligibility based on the inclusion and exclusion criteria. The articles that qualified after full text screening were considered for data-extraction and narrative synthesis. Any discrepancies when selecting the studies were resolved by discussing with the third reviewer (MS). We used a standardized form to extract the data from the selected studies. Data from each

Table 1
Characteristics of included studies for causes of hoarding.

Study Id	Sample characteristics	Study design	Hoarding Assessment	Variables evaluated
Przeworski et al. (2014) (31)	212 non-clinical sample who responded to announcements on websites, blogs, listservs and social media sites for OCD or Hoarding	Cross-sectional study	SI-R (cut-off score of 41)	LEC
Cromer et al. (2007) (28)	180 Primary OCD diagnosis based on SCID-IV	Cross-sectional study	SCID-IV, SI-R	TLEs
Tolin et al. (2010) (33)	751 adults with self- reported hoarding who provided at least 2 severity ratings over time	Cross-sectional study	HRS-SR, HRS-I.	Study authors developed a checklist consisting of 25 life events and categorized each event into 1 of 4 categories.
Hartl et al. (2005) (29)	26 potential hoarding participants were solicited from a self-help organization for clutters and area agencies. 36 controls	Cross-sectional study	SI-R PCS	TES-L
Landau et al. (2011) (30)	HD: N = 24 HD without comorbid OCD OCD (HD + OCD): N = 20 HD with comorbid OCD OCD: N = 17 OCD without hoarding symptoms Non clinical control: N = 20	Cross-sectional study (retrospective)	SI-R Semi-structured clinician-administered interview to identify the temporal association between life events and symptom onset	Clinician-administered semi-structured interview: Temporal relation between life events and symptom onset
Shaw et al. (2016) (73)	N = 80 Undergraduate students	Cross-sectional study	SI-R Hypothetical hoarding paradigms: Hypothetical discarding task Hypothetical acquiring task	THQ
An et al. (2009) (27)	29 OCD patients (with hoarding symptoms 13; without hoarding symptoms 16) and 21 healthy controls	Cross-sectional study with control group	SI-R Y-BOCS OCI-R	fMRI measured regional blood oxygen level dependent responses in the brain during the experiments that triggered hoarding-related and symptom-unrelated symptoms
Tolin et al. (2012) (47)	N = 107 adults. HD = 43 (Diagnosed on DSM-5 and clinical criteria by Frost and Hartl) OCD = 31 HCs = 33	Cross-sectional study	HRS-I OCI-R	fMRI- Functional magnetic resonance imaging measured regional hemodynamic response in the brain during decisions made to keep or discard possession.
Saxena et al. (2004) (74)	N = 45 who met DSM-IV criteria for OCD (12 of whom had compulsive hoarding as a prominent OCD symptom factor) N = 17 Normal subjects	Cross-sectional study	Hoarding severity/ saving symptoms were measured retrospectively on 0-4 rating scale with Raunch et al. method.	Flurodeoxyglucose PET- regional blood glucose metabolism
Mataix-Cols et al. (2004) (34)	N = 16 OCD patients (OCD assessed with YBOCS and the symptom checklist) N = 11 Healthy volunteers	Cross-sectional study	SI-R, PI-R	fMRI measured regional blood oxygen level dependent responses in the brain during the symptom provocation experiment.
Medley et al. (2013) (35)	279 college students from the southern USA 210 individuals from the general community who presented to an anxiety clinic to participate in various research and or treatment studies.	Cross-sectional study Cross-sectional study	Study 1: SI-R Study 1: SI-R	ASI-3 ASI-3
Timpano et al. (2014) (52)	213 unselected, nonclinical sample recruited through introductory psychology courses at a large South-Eastern university	Cross-sectional study	SI-R	DTS, PTEQ
Oglesby et al. (2013) (36)	279 college students from a large southern university recruited from the undergraduate psychology research pool and signed up for a study titled “Behaviour and Personality study” through an online experiment database	Cross-sectional study	SI-R	IUS
Timpano and Schmidt (2013) (40)	484 undergraduate students Clinical sample of 135 individuals presenting at a specialized anxiety disorder research program and university clinic over the course of approximately 18 months 102 undergraduate students: 64% of participants were preselected for falling above the mean on a short saving behaviour screening questionnaire	Cross-sectional study Cross-sectional study Cross-sectional study	Study 1: SI-R Study 2: SI-R Study 3: SI-R	SCS SCS SCS, Discarding Task

(*continued on next page*)

Table 1 (*continued*)

Study Id	Sample characteristics	Study design	Hoarding Assessment	Variables evaluated
Timpano and Shaw (2013) (38)	72 young adults who received research familiarization credit for participation	Cross-sectional study	SCI-R SCI CAS	IDAQ, OAQ
Timpano et al. (2009) (37)	720 undergraduate students	Cross-sectional study	Study 1: OCIR [Sum of 3 hoarding items of the OCIR]	ASI-3
	265 undergraduate students who received research credit for participation	Cross-sectional study	Study 2: Obsessive-OCIR	DTS
	409 undergraduate students who received research credit for participation	Cross-sectional study	Study 3: SI-R	DTS, ASI-3
Timpano and Rasmussen (2013) (39)	372 undergraduate students at a large university in the United States who participated in exchange for research-familiarization credit	Cross-sectional study	Study 1: SI-R	BIS
	160 young adults at a German university who participated in exchange for course credit	Cross-sectional study	Study 2: GCHI	UPPS
Mathews et al. (2007) (48)	11 multigenerational families with a minimum of 2 individuals affected with OCD (Families were originally ascertained through probands with DSM-IV OCD whose symptoms began before age 18)	Cross-sectional study	YBOCS LOICV	Family history, DNA
Seedat and Stein (2002) (53)	15 subjects assessed as having clinically significant hoarding [10 were recruited from an OCD clinic-Tygerberg Hospital, Cape Town and five were recruited via newspaper advertisement)	Cross-sectional study	Semi-structured 'hoarding questionnaire'	Family History
Perroud et al. (2011) (45)	Large cohort of Caucasian twins (Of the 3410 participants, 2350 were singletons)	Cross-sectional study	HRS-SR	Genotyped using either Illumina 317 K (n = 1348) or 610 K (n = 2062) Bead Chips
Ayers et al. (2010) (22)	18 older adults recruited throughout San Diego County, University of California, San Diego Psychiatric Clinics, and the VA San Diego Healthcare System	Cross-sectional study	SI-R, UHSS	Psychosocial and medical histories
Winsberg et al. (1999) (54)	20 patients with OCD and hoarding identified at an OCD clinic (Stanford, California)	Cross-sectional study	Semi-structured Interview	Family history of hoarding
Lochner et al. (2005) (51)	315 adult OCD patients (162 male; 153 female)[referred from the OCD Association of South Africa, community-based primary care practitioners, and specialist psychiatrists]	Cross-sectional study	YBOCS	DNA(Genotyping)
Samuels et al. (2007a,b) (49)	624 participants recruited from outpatient and inpatient clinics, referrals from clinicians in the community, websites, media advertisements, self-help groups, and annual meetings of the Obsessive Compulsive Foundation	Cross-sectional study	YBOCS	FHI
Nordsletten et al. (2013a,b) (44)	Community sample of 5022 adult, female twins (2529 pairs) from the TwinsUK adult twin registry, based at St Thomas' Hospital in London, England	Cross-sectional study	HRS-SR	Peas in a Pod Questionnaire (Zygosity) DNA
Mathews et al. (2014) (42)	Adult twin pairs (n = 7906) and their family members from the Netherlands Twin Register (N = 15,914)	Cross-sectional study	HRS-SR	Phenotypes (hoarding & OCS) were derived from self-symptom-based data
Iervolino et al. (2009) (4)	5022 twins from the Twins UK adult twin registry	Cross-sectional study	HRS-SR	Phenotypes (hoarding & OCS) were derived from self-symptom-based data
Ivanov et al. (2013) (41)	Population-based sample of 15 year old twins (N = 3974) enrolled in the Swedish Twin Registry and taking part in the ongoing Child and Adolescent Twin Study in Sweden (CATSS)	Cross-sectional study	HRS-SR	Twin zygosity was determined either by using single nucleotide polymorphisms (SNPs) or (if DNA was unavailable) an algorithm based on twin similarity with predictive value greater than 95% compared to DNA testing
Samuels et al. (2007b) (50)	219 multiplex OCD families collected as part of the OCD Collaborative Genetics Study [recruited from outpatient and inpatient clinics, referrals from clinicians in the community, websites, media advertisements, self-help groups, and annual meetings of the Obsessive Compulsive Foundation]	Cross-sectional study	YBOCS	DNA

(*continued on next page*)

Table 1 (*continued*)

Study Id	Sample characteristics	Study design	Hoarding Assessment	Variables evaluated
Monzani et al. (2014) (43)	5409 female members of the TwinsUK adult population-based twin register	Cross-sectional study	HRS-SR	Zygosity was determined using the Peas in a Pod Questionnaire and further established via DNA confirmation methods in uncertain cases
Samuels et al. (2002a,b) (17)	126 subjects with OCD	Cross-sectional study	YBOCS	NA

ASI: Anxiety Sensitivity Index; ASI-3: Anxiety Sensitivity Index-3; BIS: Barratt Impulsivity Scale; CAS: Compulsive Acquisition Scale; DNA: Deoxyribonucleic acid; DSM-IV: Diagnostic and Statistics Manual of Mental Disorders version IV; DTS: Distress Tolerance Scale; FHI: Family History Interviews; HRS-SR: Hoarding Rating Scale- Self-Report; IDAQ: Individual Differences in Anthropomorphism; IUS: Intolerance of Uncertainty Scale; LEC: Life Events Checklist; LOICV: The Leyton Obsessional Inventory, childhood version; OAQ: Object Attachment Questionnaire; OCD: Obsessive Compulsive Disorder; PTEQ: The Perception of Threat from Emotion Questionnaire; SCS: Self-Control Scale; SCI: Savings Cognition Inventory; SI-R: Savings Inventory- Revised; TES-L: Traumatic Events Scale- Lifetime; THQ: Traumatic History Questionnaire; TLEs: Traumatic Life Events; UPPS: Urgency Premeditation Perseverance Sensation Seeking Impulsive Behaviour Scale; YBOCS: Yale Brown Obsessive Compulsive Scale.

included study was independently extracted by two reviewers (AH & VS). The results were organized based on four main research areas that were identified after screening the full-text articles. These four areas were: (i) traumatic life events; (ii) genetic association; (iii) emotional variables; and (iv) neural mechanisms.

For each selected article, the review team summarized the following:

- Study characteristics
- Sample / participant characteristics
- Outcome measures used to establish and/or study hoarding behaviours under the four key research areas
- Causes of hoarding
- Interventions for hoarding

3. Results

In all, 396 published articles were identified, and after excluding the duplicates, 379 were screened for eligibility. Firstly, titles and abstracts were screened to further select those papers to be extracted for detailed assessment. After reading the full-texts, 31 reports were included and 89 reports were excluded. The characteristics of all the included studies are summarized in Table 1. Details of the process of screening and selecting studies are described in Fig. 1 as a PRISMA flowchart

3.1. Characteristics of participants included in the studies

The age of the participants ranged from 6 to 95 years. The total number of participants included in each study ranged between 15 and 15,914. Of the included studies, 15 studies were conducted in non-clinical populations, 12 studies consisted of participants with the primary diagnosis of OCD who were further categorized as having predominant hoarding and non-hoarding symptoms, two studies included participants with self-reported hoarding behaviour and two studies included participants with clinically diagnosed hoarding symptoms.

3.2. Type of outcome measures used in studies on hoarding

3.2.1. Hoarding symptoms

Most of the studies used the Savings Inventory-Revised (SI-R) (Ayers et al., 2010; An et al., 2009; Cromer et al., 2007; Hartl et al., 2005; Landau et al., 2011; Przeworski et al., 2014; Shaw et al., 2016; Tolin et al., 2010; Mataix-Cols et al., 2004; Medley et al., 2013; Oglesby et al., 2013; Timpano et al., 2009; Timpano and Shaw, 2013; Timpano and Rasmussen, 2013; Timpano and Schmidt, 2013) to differentiate between individuals with hoarding symptoms and individuals without hoarding symptoms. Other scales used were Hoarding Rating Scale-Self-Report (HRS-SR) (Iervolino et al., 2009; Tolin et al., 2010; Ivanov et al., 2013; Mathews et al., 2014; Monzani et al., 2014, Nordsletten2013b; Perroud et al., 2011), University of California Los Angeles Hoarding Severity Scale (UHSS) (Ayers et al., 2010), German Compulsive Hoarding Inventory (GCHI) (Timpano and Rasmussen, 2013), Saving Cognitions Inventory (SCI) (Timpano and Shaw, 2013), and the Compulsive Acquisition Scale (CAS) (Timpano and Shaw, 2013).

Few studies used Functional Magnetic Resonance Imaging (fMRI), and Flurodeoxyglucose Positron Emission Topography (PET) scans to measure the regional hemodynamic response and cerebral glucose metabolism in various regions of the brain during hoarding related experiments (An et al., 2009; Mataix-Cols et al., 2004; Saxena et al., 2004; Tolin et al., 2012).

3.2.2. Traumatic life events

Traumatic life events (TLE) are defined as incidents that cause physical, emotional, or psychological harm in the course of a person's life. These TLE were measured using the Traumatic Life Events List (TLE) (Cromer et al., 2007), Trauma History Questionnaire (THQ)

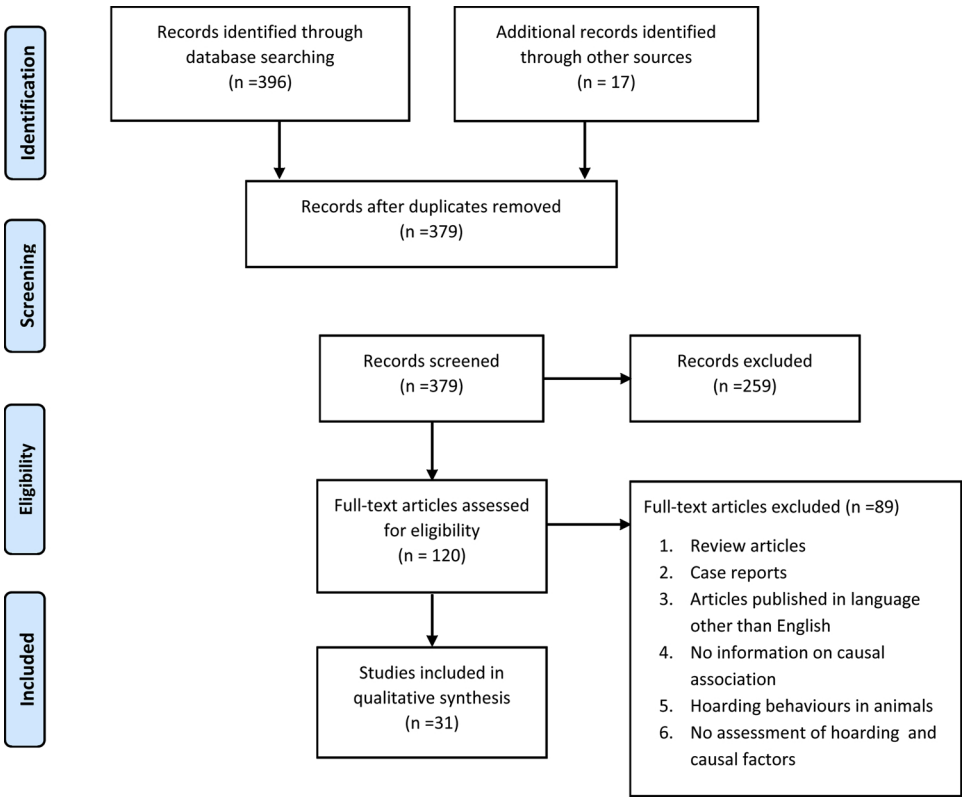

Fig. 1. PRISMA Flow Diagram for Causes of Hoarding.

(Shaw et al., 2016), Traumatic Events Scale-Lifetime (TES-L) (Hartl et al., 2005), and Life Events Checklist (LEC) (Przeworski et al., 2014).

3.2.3. Genetics

Information on genetics and family history were collected using the Diagnostic Interview for Genetic Studies (DIGS) (Mathews et al., 2007), Family History Interviews (Samuels et al., 2007a), Family Informant Schedule and Criteria (Samuels et al., 2007b), Johns Hopkins Diagnostic Assignment Checklist (Samuels et al., 2007a, b), and the Peas in a Pod Questionnaire (Iervolino et al., 2009; Nordsletten et al., 2013b). Genotyping was done using either Illumina 317 K or 610 K BeadChips (Perroud et al., 2011).

3.2.4. Emotional and neurocognitive factors

Emotional factors that are hypothesized to be associated with hoarding include anxiety, depression, distress, self-control etc. The following measures were applied to assess emotional variables: Beck Anxiety Inventory (BAI) (Ayers et al., 2010; Timpano et al., 2009; Timpano and Rasmussen, 2013; Timpano and Schmidt, 2013), Beck Depression Inventory (BDI) (Ayers et al., 2010; Medley et al., 2013; Oglesby et al., 2013; Timpano et al., 2009; Timpano and Rasmussen, 2013; Timpano and Schmidt, 2013), Temperament and Character Inventory (Lochner et al., 2005), State-Trait Anxiety Inventory (STAI) (Timpano and Rasmussen, 2013), Anxiety Sensitivity Index -3 (ASI-3) (Timpano et al., 2009), Depression, Anxiety, and Stress Scale -21 (DASS-21) (Timpano et al., 2009; Timpano and Shaw, 2013; Timpano et al., 2014), Distress Tolerance Scale (DTS) (Timpano et al., 2009, 2014), Spielberger Trait Anxiety Inventory (STAI) (Timpano et al., 2009), Individual Differences in Anthropomorphism Questionnaire (IDAQ) (Timpano and Shaw, 2013), Object Attachment Questionnaire (OAQ) (Timpano and Shaw, 2013), Self-Control Scale (SCS) (Timpano and Schmidt, 2013), Social Interaction Anxiety Scale (SIAS) (Timpano and Schmidt, 2013), Penn State Worry Questionnaire (PSQW) (Oglesby et al., 2013; Timpano and Schmidt, 2013), Intolerance of Uncertainty Scale (IUS) (Oglesby et al., 2013), Perception of Threat from Emotion Questionnaire (PTEQ) (Timpano and Schmidt, 2013).

3.3. Causal factors of hoarding behaviour

Of the included studies, six studies (Cromer et al., 2007; Hartl et al., 2005; Landau et al., 2011; Przeworski et al., 2014; Shaw et al., 2016; Tolin et al., 2010) studied an association between TLE and hoarding behaviours, four studies (An et al., 2009; Mataix-Cols et al., 2004; Saxena et al., 2004; Tolin et al., 2012) examined the neural correlates of hoarding disorder, seven studies reported an association between emotional variables and hoarding symptoms (Medley et al., 2013; Oglesby et al., 2013; Timpano et al., 2009; Timpano and Shaw, 2013; Timpano and Rasmussen, 2013; Timpano and Schmidt, 2013; Timpano et al., 2014); fourteen studies found genetic associations with hoarding symptoms (Iervolino et al., 2009; Samuels et al., 2002a; Ayers et al., 2010; Ivanov et al., 2013; Mathews et al., 2014; Monzani et al., 2014, Nordsletten2013b; Perroud et al., 2011; Mathews et al., 2007; Samuels et al., 2007a, b; Lochner et al., 2005; Seedat and Stein, 2002; Winsberg et al., 1999).

3.4. Traumatic life events and hoarding disorder

In general, individuals with hoarding symptoms experienced greater number of lifetime traumatic events as compared to those without hoarding symptoms (Cromer et al., 2007; Hartl et al., 2005; Landau et al., 2011; Przeworski et al., 2014; Tolin et al., 2010) with the number of TLE found to positively correlate with the severity of hoarding (Cromer et al., 2007; Hartl et al., 2005; Landau et al., 2011; Przeworski et al., 2014; Tolin et al., 2010). In contrast, Shaw et al (Shaw et al., 2016) and Tolin et al (Tolin et al., 2010) found no association between the number of TLE and severity of hoarding symptoms. There is some evidence that linked the onset of hoarding symptoms to specific TLE such as interpersonal trauma including assault, accidental or tragic loss

of a loved one, or childhood neglect (Przeworski et al., 2014; Tolin et al., 2010); other stressful life events (natural or man-made disasters and actual or feared injury/illness/death of self or others); and environmental factors (general negative parenting style, relationship difficulties (Landau et al., 2011); and loss or change in relationships).

Grisham et al (Grisham et al., 2006), Landau et al (Landau et al., 2011) and Tolin et al (Tolin et al., 2010) found a higher rate of stressful or traumatic events at time of onset for respondents with a later age (age 16 or higher) of onset of symptoms. Later onset hoarding was hypothesized to be etiologically distinct from the more early childhood-onset disorder, and that stressful or traumatic events play a greater role in the onset of late-onset hoarding.

3.5. Neural correlates of hoarding disorder

Studies using sophisticated imaging techniques have found that individuals with predominant compulsive hoarding symptoms have a unique pattern of lower metabolism in the posterior cingulate cortex and occipital cortex (cuneus) areas of the brain. They also had significantly reduced glucose metabolism in the dorsal anterior cingulate gyrus; the severity of hoarding was negatively correlated with metabolism in this region of the brain across all OCD patients (Saxena et al., 2004).

During the process of making decisions related to discarding their personal possession, those individuals with hoarding disorder demonstrated greater activity of functional Magnetic Resonance Imaging (fMRI) signals in the Anterior Cingulate Cortex (ACC) and right insular cortex. On the contrary, lower activity was observed during discarding of experimenter's possessions (apparent biphasic pattern). The HD group also demonstrated lower fMRI signal in the left fusiform gyrus, left thalamus and right thalamus than in the healthy controls. The scores on the saving inventory that measure hoarding were positively correlated with activity in the right insula and right superior parietal lobule and negatively correlated with activity in precentral gyrus and cuneus (Tolin et al., 2012). Furthermore, the ratio of saved to discarded items was positively correlated with activity in the right fusiform gyrus and pons (Tolin et al., 2012).

OCD patients with hoarding demonstrated significantly greater activation than non-hoarding patients and controls did in a large bilateral cluster situated in the anterior part of the ventromedial prefrontal cortex (VMPFC) during provocation of hoarding-specific anxiety. The cerebellum was significantly more activated bilaterally in the hoarding group than in the control group (An et al., 2009). Hoarding-related anxiety also correlated inversely with activation in bilateral dorsal prefrontal regions, basal ganglia, temporal cortex and parieto-occipital regions.

3.6. Emotional and neurocognitive factors

Emotional factors associated with hoarding symptoms include impulsivity, Anxiety Sensitivity (AS) (i.e., fear of anxiety-related sensations) (Reiss et al., 1986), distress tolerance, anthropomorphism (i.e., see human-like qualities in non-human entities) (Timpano and Shaw, 2013), self-control, and intolerance of uncertainty (Oglesby et al., 2013).

It has been suggested that hoarding may have some overlap with impulse control disorders (Frost et al., 2011; Steketee and Frost, 2003). Individuals with hoarding similar to those with impulse control disorders report experiencing pleasure or relief upon engaging in the behaviour (acquiring and saving behaviours among individuals with characteristic features of hoarding) (Steketee and Tolin, 2011). (Frost et al., 2011) found that up to 78.3% of participants in a primary hoarding sample met clinical criteria for one impulse control disorder. Timpano et al (Timpano and Rasmussen, 2013) found that more severe hoarding symptoms were associated with greater levels of impulsivity in two separate samples.

Hoarding severity has been found to be significantly correlated with higher AS scores (Medley et al., 2013; Timpano et al., 2009). The study by Medley et al (Medley et al., 2013) also found that the physical concerns aspect of AS (referring to the fear of behaviours or sensations associated with the experience of anxiety) was most significantly associated with hoarding severity; while overall AS was associated with the difficulty discarding subscale of the SI-R and not the acquisitioning and clutter subscales. This suggests that individuals who hoard may avoid decisions to discard in an effort to avoid the unpleasant emotions associated with the task of discarding (Steketee and Frost, 2003).

Hoarding was also shown to be associated with difficulties experiencing and tolerating negative emotions (known as distress tolerance) (53). Distress tolerance was associated with all subscales of SI-R except clutter; SI-R total, difficulty discarding, and acquiring scores were also associated with threatening appraisals of sadness, guilt, anger, anxiety, and disgust. Acquiring was linked with greater emotional intensity for sadness, disgust, frustration, anxiety, and irritability. In contrast, difficulty in discarding possessions was linked with greater intensity of fear and anxiety (Steketee and Frost, 2003).

The tendency to anthropomorphize objects has been observed clinically in adults with predominant hoarding symptoms (Timpano and Shaw, 2013). In a case study of severe hoarding, Savoie et al (Savoie, 2008) noted that the individual used anthropomorphic descriptions of garbage, such that she "felt bonded with it and felt sorry for it" (p. 256). Tolin et al (Tolin, 2011) similarly described how some hoarding patients experience anthropomorphic thoughts related to making sure their discarded possession remains unharmed or is given to a good home. A study by Timpano and Shaw (Timpano and Shaw, 2013) found that Individual Differences in Anthropomorphism Questionnaire (IDAQ) scores (that measures the extent to which respondents believe a nonhuman entity has human characteristics) were significantly correlated with greater hoarding symptoms. Higher IDAQ scores were significantly associated with greater difficulty in discarding and marginally correlated with higher rates of acquisition.

Low self-control has also been suggested to be a contributing factor of hoarding. Adults with hoarding disorder are unable to resist the urges to acquire and save possessions and report feeling powerless to control the urges even when motivated for treatment. Christensen & Greist et al (Christensen and Griest, 2001) found that individuals with hoarding while possessing the intent to manage their saving and acquiring behaviours, lacked the necessary behavioural follow-through. Timpano & Schmidt (Timpano and Schmidt, 2013) found that hoarding was strongly associated with low levels of self-control in both non-clinical and clinical samples. In the non-clinical sample, the authors also found that lowered levels of self-control remained a significant predictor for all three facets of hoarding - clutter, difficulty discarding and acquiring. Using an experimental paradigm, the authors also demonstrated that those in the depletion condition (i.e., those participants with less self-control available) experienced more difficulties in the compliance task i.e. they saved more items (could take the item home with them), waited to decide for more items (i.e. decide to revisit the item at the end of the experiment), and discarded fewer items (wherein item was immediately shredded by the experimenter).

Another vulnerability factor for hoarding that has been identified is Intolerance of Uncertainty (IU) which has been defined as "a cognitive bias that affects how a person perceives, interprets, and responds to uncertain situations on a cognitive, emotional, and behavioural level" (Drury et al., 2014). Individuals with high IU find uncertainty stressful and upsetting, believe that uncertainty is negative, and have difficulty functioning in uncertain situations, which leads to avoidance of situations where they feel uncertain (Buhr and Dugas, 2002). Oglesby et al (Oglesby et al., 2013) found that IU was independently and significantly associated with hoarding severity in a large, non-clinical sample. IU was significantly and robustly associated with the discarding, acquiring and clutter subscales of the SIR. The authors suggested that individuals with hoarding symptoms with high IU may want

to avoid the uncertainty associated with making a mistake while discarding and thus IU may contribute to the difficulty in decision making that is observed in hoarding disorder. It is also possible that individuals with hoarding symptoms may acquire unneeded objects in an effort to avoid the negative emotional states and uncertainty associated with not acquiring an item of potential value. The authors hypothesized that clutter would be an invariable result of this significant association of IU with the inability to discard and the desire to acquire thus explaining the association of high IU with clutter in their sample.

3.7. Genetic factors

The role of genetic factors in the causation of hoarding behaviours/symptoms is suggested by studies that have established a family history of hoarding (Samuels et al., 2002a; Ayers et al., 2010; Mathews et al., 2007; Samuels et al., 2007a, b; Seedat and Stein, 2002; Winsberg et al., 1999). Genetic studies have identified several chromosomes to be associated with hoarding traits.

Perroud et al (Perroud et al., 2011) found two genomic loci on chromosomes 5 and 6 that showed suggestive association with hoarding traits. Lochner et al (Lochner et al., 2005) found the L/L genotype of the COMT Vall58Met polymorphism to be significantly more common in the OCD hoarding group, compared with non-hoarding patients and controls in subjects of Afrikaner descent (but not among Caucasian subjects with hoarding). Suggestive linkages to chromosomes 14 and 3 have also been found; Samuels et al (Samuels et al., 2007b) found a significant linkage of OCD to chromosome 14 in families with 2 or more hoarding relatives and a suggestive linkage to chromosome 3 in families with fewer than 2 hoarding relatives.

Twin studies on heritability have found both genetic and environmental factors to play a role in hoarding behaviours; with correlations for monozygotic (identical) twins being higher than dizygotic (non-identical) twins (Mathews et al., 2014; Monzani et al., 2014; Nordsletten et al., 2013b). Phenotypic variance in these studies is generally divided into three factors: additive genetic (variance in a trait that is attributable to genetic factors), common/shared environment (variance attributed to environmental factors shared by family members or twins), and unique/non-shared environment (environmental effects unique to each family member or twin) (Nordsletten et al., 2013b). Two studies which examined the relative contributions of genetic and non-shared environmental factors on hoarding found the variance accounted for by genetic factors to range between 36%–50% and that by non-shared environmental factors to range between 50%–64% for hoarding in general (Iervolino et al., 2009; Mathews et al., 2014). Two studies looked at the genetic and environmental contributions of the individual facets of hoarding (Mathews et al., 2014; Nordsletten et al., 2013b). Additive genetic factors accounted between 22%–49% and non-shared environmental factors accounted for 51%–78% for excessive acquisition; additive genetic factors accounted between 37%–45% and non-shared environmental factors accounted for 63%–64% for difficulty discarding. Mathews et al (Mathews et al., 2014) found 0.20 of the total variance for clutter to be due to additive genetic factors, 0.11 due to common environmental factors, and 0.69 to be due to non-shared environmental factors.

Ivanov et al (Ivanov et al., 2013) found sex differences in relative contributions of hoarding symptoms. Among adolescent boys, 32% of the variance was accounted for by genetic factors, 4% by shared environment, and 64% by non-shared environment whereas, among adolescent girls, genetic contributions accounted for 2% of the variance, shared environment accounted for 32% of the variance and non-shared environment factors accounted for 65% of the variance; Mathews et al (Mathews et al., 2014) in contrast, did not find any evidence for sex-specific contribution for hoarding.

4. Discussion

The current systematic review collated evidence from studies examining the association between various putative causal factors (i.e. genetic, emotional, neural correlates, TLE) and hoarding symptoms. The findings of this review are in line with another comprehensive review that also postulated that hoarding behaviour could be due to a combination of factors (Dozier and Ayers, 2017).

In general, lifetime traumatic events were found to be positively correlated with hoarding behaviours (by 4 studies) with the exception of two studies which did not find an association. Explanations for this association include the hypothesis that TLEs lead to certain individuals feeling less safe, and thus they rely on their possessions for a sense of security and protection (Frost and Hartl, 1996; Sartory et al., 1989). Frost et al (Frost et al., 1995) have suggested that individuals with hoarding symptoms may perceive that by protecting their possessions, they are protecting themselves. Since possessions are associated with security, discarding them leads to anxiety about potential harm. As a result of TLEs these beliefs could be strengthened leading to hoarding behaviour. This association also has implications for treatment, as it emphasises the need for a comprehensive assessment of TLEs in patients with hoarding and the need for trauma work subsequently if needed. However, more research is needed to understand the association between hoarding and TLEs. All the included studies have evaluated the influence of lifetime traumatic events retrospectively. The role of recurrent stressful or traumatic events on hoarding is unknown.

With regards to neural correlates of hoarding symptoms, the present body of evidence remains inconclusive with regards to the specific regions in the brain that are implicated in hoarding behaviour. While some studies have found unique patterns of metabolic activity in various areas of the brain including the dorsal ACC and thalamus, others have noted differential activity of fMRI signals in areas such as the right insular cortex, left fusiform gyrus, right insula, right superior parietal lobule, precentral gyrus, and cuneus in the frontal pole extending ventrally to the anterior part of the orbito-frontal cortex and dorsally to the medial frontal gyrus and cerebellum. The brain regions associated with hoarding symptoms in the study by An et al (An et al., 2009) are anatomically very close to those associated with hoarding behaviours in human lesion studies (specific patterns of brain damage) (Anderson et al., 1999). Studies suggest that these regions i.e. VMPFC, amygdala and nucleus accumbens are involved in decision making (Bechara et al., 2000; Clark and Manes, 2004). The study by Saxena et al (Saxena et al., 2004) found that severity of hoarding symptoms was significantly correlated with lower activity in the dorsal part of the anterior cingulate gyrus, as well as lower activity in the right posterior cingulate cortex. It is similarly suggested that this lower activity in both the anterior and posterior parts of the cingulate gyrus may mediate the difficulty in making decisions, attentional problems, and other cognitive deficits seen in individuals with compulsive hoarding behaviours. Abnormal regions identified by Tolin et al (Tolin et al., 2012) such as the ACC are also associated with problems in decision-making processes that play a role in the difficulty related to discarding items. Thus while studies identified different neural correlates of hoarding, there is convergence in terms of the role played by these correlates in decision making and it being affected in hoarding. However, further research should confirm the findings in individuals with pure hoarding disorder (without OCD).

Emotional factors, especially AS, were found to be consistently linked with hoarding severity. Other cognitive factors associated with hoarding symptoms include impulsivity, distress tolerance, anthropomorphism, self-control and IU. High levels of AS may lead to avoidance of discarding as it may trigger unpleasant emotions. Research has shown that AS is amenable to treatment (Feldner et al., 2008; Keough and Schmidt, 2012). Single session brief interventions

have been shown to be effective at reducing AS (Schmidt et al., 2007). Thus incorporating interventions in the treatment of those with hoarding disorder could lead to favourable outcomes. Neurocognitive deficits such as IU associated with avoidance, as well as inattention could be addressed by using pharmacotherapy or behavioural therapy, although further research is needed.

Evidence for the genetic basis of hoarding is also varied. Genomic studies have identified several chromosomes in relation to hoarding traits including two genomic loci on chromosomes 5 and 6, chromosome 14, and the L/L genotype of the COMTVall58Met polymorphism. Nevertheless, there is substantial evidence from twin studies supporting the heritability of hoarding symptoms. Genetic factors, non-shared environmental factors, as well as common environmental factors were found to account for the variance in hoarding traits. It is important to note that replication of findings from the genetic studies in large sample and in individuals with hoarding disorder is needed to ascertain the findings.

While this review attempted to collate evidence for the possible causal factors of hoarding, some limitations of the studies needs to be kept in mind. These include a small sample size, being conducted in a predominantly non-clinical population or among individuals who had OCD as a primary diagnosis; absence of standardized diagnostic methods, use of different measures for assessing hoarding outcomes; reliance on self-report assessments leading to reporting bias and they may also be influenced by lack of insight; and inclusion of patients with comorbid depression and anxiety. All the included studies in this review were cross-sectional in nature, therefore only an association not causation of risk factors could be established for hoarding disorder. Future studies can address some of the limitations identified by conducting prospective studies with long-term follow-up, establishing a control group for comparison between patients with hoarding disorder and healthy participants, and using the DSM-5 criteria as a guideline to identify participants with HD. Given the recent classification of hoarding as separate disorder in the DSM-5, and the negative outcomes on the well-being of individuals with clinical hoarding symptoms, a clear understanding of the etiology of hoarding disorder is not only timely but also imperative to better inform interventions targeted to address this disorder. The evidence suggests the involvement of certain genetic factors (which are non-modifiable factors), the presence of emotional traits, environmental factors like TLE, and abnormal brain activities in hoarding behaviour. It is likely that hoarding is the manifestation of a complex interplay of these various factors but the exact nature of this interaction remains to be elicited.

Contributors

Author AH and VS conducted this review. Author AH and VS wrote the first draft of the manuscript. The authors MS, JV, CSA, WMT and YHW conceptualised the topic and developed the protocol. All the authors contributed to and have approved the final manuscript.

Conflict of interest

All authors declare that they have no conflict of interest.

Funding

This work was supported by the Agency For integrated Care, Community Mental Health Division, Singapore; Housing & Development Board, Singapore; and Ministry of National Development, Policy & Planning Division, Singapore. Funding bodies were not directly involved in conducting this systematic review and in the decision to submit the manuscript.

Acknowledgement

Authors would like to thank Louisa Picco for proof reading the previous version of this manuscript.

Appendix 1

SEARCH STRATEGY
EMBASE
#1 'hoarding disorder' OR (hoarding AND behaviour)
#2 etiology OR 'predisposing factor'
#3 'risk factor
#4 #2 OR #3
#5 #1 AND #4
OVID (Medline)
1. exp Hoarding Disorder/
2. hoarding behavi*.mp.
3. 1 or 2
4. causes.mp.
5. exp Causality/
6. etiology.mp.
7. exp Association/ph [Physiology]
8. 4 or 5 or 6 or 7
9. 3 and 8
OVID (PsychINFO)
1. exp Hoarding Disorder/
2. exp Hoarding Behavior/
3. 1 or 2
4. exp Causality/ or exp Risk Factor/ or exp Etiology/
5. 3 and 4
CINAHL
S1 hoarding disorder
S2 Hoarding behaviour
S3 S1 OR S2
S4 causes OR etiology OR predisposing factors OR risk factors
S5 S3 AND S4

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