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Hoarding in Children and Adolescents with Obsessive-Compulsive Disorder

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Abstract

Compared to studies in adults, there have been few studies of hoarding in children and adolescents with obsessive-compulsive disorder (OCD). In the current study, we evaluated OCD clinical features, Axis I disorders, and social reciprocity scores in 641 children and adolescents with OCD,

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Authors Samuels, Bienvenu, Fyer, Geller, Grados, Greenberg, Knowles, McCracken, Murphy, Nestadt, Pauls, Piancentini, Pulver, Rasmussen, and Riddle designed the overall study and protocol. Authors Bienvenu, Cullen, Fyer, Goes, Geller, Grados, Greenberg, McCracken, McLaughlin, Murphy, Nestadt, Piacentini, Pauls, Rasmussen, Riddle, and Stewart conducted clinical evaluations and/or final diagnostic case reviews. Authors Samuels, Bienvenu, Maher, Shugart, and Wang contributed to data analysis and interpretation. Author Samuels wrote the first draft of the manuscript. All authors contributed to and have approved the manuscript.

of whom 163 (25%) had hoarding compulsions and 478 did not. We found that, as a group, youth with hoarding had an earlier age at onset and more severe lifetime OCD symptoms, poorer insight, more difficulty making decisions and completing tasks, and more overall impairment. The hoarding group also had a greater lifetime prevalence of panic disorder, specific phobia, Tourette disorder, and tics. As measured with the Social Reciprocity Scale, the hoarding group had more severe deficits in parent-rated domains of social communication, social motivation, and restricted interests and repetitive behavior. In a multivariable model, the overall social reciprocity score, age at onset of OCD symptoms, symmetry obsessions, and indecision were independently related to hoarding in these children and adolescents with OCD. These features should be considered as candidate risk factors for the development of hoarding behavior in pediatric OCD.

1. Introduction

Hoarding behavior is characterized by difficulty discarding possessions, even those that seem useless or of little value to others; such a large number of objects may be accumulated that personal surroundings are cluttered and difficult to use (Frost & Gross, 1993). Hoarding occurs in almost 30% of adults with obsessive-compulsive disorder (OCD) (Steketee & Frost, 2003). Among adults with OCD, those with hoarding symptoms have, on average, an earlier age at onset of OCD, more severe obsessive-compulsive symptoms, greater prevalences of major depression, generalized anxiety, social phobia, and pathological grooming behaviors, more limited insight, and poorer response to treatment. Adults with hoarding also have been found to have greater numbers of obsessive-compulsive, dependent, and schizotypal personality disorder traits (Frost, Krause, & Steketee, 1996; Frost, Steketee, Williams, & Warren, 2000; Mataix-Cols, Baer, Rauch, & Jenicke, 2000; Mataix-Cols, Marks, Greist, Kobak, & Baer, 2002; Mataix-Cols, Rauch, Manzo, & Jenicke, 1999; Samuels et al., 2002; Samuels et al., 2007).

Many adults with hoarding report onset of hoarding behavior in early adolescence or younger (mean age, 11–13 years, in several studies) (Fontenelle, Mendlowitz, Soares, & Versiani, 2004; Grisham, Frost, Steketee, Kim, & Hood, 2006). The factor structure of obsessive-compulsive symptoms in children and adolescents is similar to that in adults, with a distinct hoarding dimension found in almost all studies (Bernstein, Victor, Nelson, & Lee, 2013; McKay et al., 2006; Stewart et al., 2004; Stewart et al., 2007). Moreover, the prevalence of hoarding in a population-based sample of 15-year old twins was found to be 3.7% (excluding the criterion for clutter, which is often prevented by parents) (Ivanov et al., 2013), similar to population-based estimates in adults (Iervolino et al., 2009; Mueller et al., 2009; Samuels et al., 2008). However, there have been few studies of hoarding in children and adolescents with OCD (Storch et al., 2011b).

Storch et al. (2007) studied 80 children and adolescents treated at an OCD clinic (age range, 7–17 years) and found that 21 % had significant hoarding symptoms, with "worse insight, more magical thinking obsessions, ordering/arranging compulsions, higher levels of anxiety, aggression, somatic complaints, and overall externalizing and internalizing symptoms" than the non-hoarding patients. Mataix-Cols, Nakatani, Micali, & Heyman (2008) factoranalyzed OCD symptoms in 238 children and adolescents (age range, 8–18 years) referred

to a pediatric OCD clinic and found that scores on the hoarding/checking dimension were associated with longer duration of illness, increased levels of pervasive slowness, responsibility, indecisiveness, pathological doubt, depression and greater emotional problems, both self-rated and parent-rated. Masi et al. (2010) studied 257 children and adolescents (age range, 6–18 years) referred to a pediatric mood and anxiety service; compared to the other patients, the 15 in whom hoarding was the most severe, stable, and impairing obsessive-compulsive symptom had the most severe and impairing illness, and the greatest prevalence of panic disorder and bipolar disorder. Frank et al. (2014) studied 68 children treated at a pediatric anxiety clinic (age range 4–10 years); compared to the non-hoarding patients, the hoarding group at an earlier age at onset of their primary diagnosis, a greater prevalence of attention deficit/hyperactivity disorder (ADHD), and a greater prevalence of anxiety disorders (generalized anxiety, social anxiety, or separation anxiety disorders).

The etiology of hoarding is complex, with evidence of involvement of genetic factors (Ivanov et al., 2013; Samuels et al., 2007), adverse life experiences (Cromer, Schmidt, & Murphy, 2007; Landau, Iervolino, Pertusa, & Santo, 2011; Przeworski, Cain, & Dunbeck, 2014), and deficits in executive functions (Grisham et al., 2010; Mathews, Perez, Delucchi, & Mathalon, 2012; McGuire et al., 2014; Morein-Zamir et al., 2014). Moreover, there is evidence for an association between hoarding behavior and autism. Hoarding behavior occurs relatively frequently in children and adults with autism spectrum disorders (Bejerot, 2007; McDougle et al., 1995; Ruta, Mugno, D'Arrigo, Vitiello, & Mazzone, 2010), and a distinct hoarding dimension has emerged from factor analyses of obsessive-compulsive symptoms in children with autism spectrum disorders (Anagnostou et al., 2011; Scahill et al., 2014). In addition, compared to healthy controls (although not psychiatric controls), adult patients with hoarding disorder are more likely to have autistic traits (Pertusa et al., 2012). However, the relationship between hoarding and autistic traits has been little studied in children and adolescents with OCD.

In the current study, we addressed several of the gaps in the previous literature by investigating hoarding in a large sample of children and adolescents with OCD, and who were not selected for treatment. We employed a social reciprocity questionnaire to evaluate parent-reported autistic traits in the participants. We also used multivariable modeling to evaluate independent risk correlates for hoarding in this sample. We proposed that, as in adults, OCD-affected children and adolescents with hoarding are clinically distinct from those without hoarding. We tested this hypothesis by comparing OCD clinical features, Axis I disorders, and scores on social reciprocity scales between those with and without hoarding.

2. Methods

2.1 Participants

The individuals included in the current analyses had participated in one of two multisite, collaborative family/genetic studies of OCD, which have been described in detail elsewhere. In brief, the OCD Collaborative Genetics Study (OCGS) (2001–2006), targeted recruitment on families with OCD-affected sibling pairs, and extended these when possible through affected first- and second-degree relatives (Samuels et al., 2006). The OCD Collaborative

Genetic Association Study (OCGAS) (2007–2012) targeted recruitment on trios (i.e., an affected proband and both parents), but also included pedigrees with a proband and unaffected sibling, as well as families with multiple-affected members (Mattheisen et al., in press). Participants were recruited into the studies from outpatient and inpatient clinics, referrals from clinicians in the community, web sites, media advertisements, self-help groups, and annual conventions of the International Obsessive Compulsive Foundation.

To be considered affected, a participant had to meet DSM-IV OCD diagnostic criteria at any time in his/her life (American Psychiatric Association, 1994). Probands were included if, in addition to meeting DSM-IV criteria, their first onset of obsessions and/or compulsions occurred before 18 years of age. Probands with schizophrenia, severe mental retardation, Tourette disorder, or secondary OCD (i.e., OCD occurring exclusively in the context of depression) were excluded. Individuals had to be at least 6 years old to participate in the study. Written, informed consent (for adults and adolescents) or assent (for children) was obtained prior to the clinical interview. The protocol was approved by the institutional review board at each study site.

2.2 Measures

As described previously (Samuels et al., 2006), diagnostic assessments were conducted by psychiatrists or PhD-level psychologists, who interviewed participants directly using a semi-structured format for the evaluation of psychopathology. The Schedule for Affective Disorders and Schizophrenia for School-Age Children-Present and Lifetime Version (K-SADS-PL) was used to ascertain and record lifetime psychopathology in children and adolescents (age range, 6–17 years) according to DSM-IV criteria (Kaufman et al., 1997). The OCD section was adapted from the Schedule for Affective Disorders and Schizophrenia-Lifetime Anxiety version (SADS-LA-R) (Mannuzza, Fyer, Klein, & Endicott, 1986) and included detailed screening questions. A similar section was developed for assessing tics, Tourette disorder, and other tic disorders. The Child Yale-Brown Obsessive Compulsive Scale and Symptom Checklist (CY-BOCS) were used for the assessment of OCD symptoms and severity in children (Scahill et al., 1997).

For each obsession and compulsion endorsed by an individual, the examiner asked the individual to rate, for the worst period, the time occupied by the symptom, from 0 (none), 1 (mild, less than 1 hour per day or occasional intrusion), 2 (moderate, 1–3 hours per day or frequent intrusion, 3 (severe, 3–8 hours per day), and 4 (extreme, more than 8 hours per day or near constant intrusion). The examiner also rated the level of distress of the individual when experiencing symptoms, from 0 (no distress), 1 (mild, infrequent, not too disturbing, and still manageable distress), 2 (moderate, frequent, and disturbing, but still manageable), 3 (severe, frequent, and very disturbing distress), and 4 (extreme, near constant, and disabling distress).

To evaluate poor insight, indecision, task difficulty, doubt, and global impairment, examiners rated participants on 5-point scales, each ranging from "none" to "extreme", with clear descriptors of each rating provided.

The Family Informant Schedule and Criteria (Mannuzza et al., 1985) was used to obtain additional information about each participant from a knowledgeable parent. Examiners completed a narrative formulation for each case. The Johns Hopkins Diagnostic Assignment Checklist was used to collate all the clinical information (the semi-structured direct interview, case formulation, informant interview, and medical records). All psychiatric diagnoses were made according to strict DSM-IV criteria (American Psychiatric Association, 1994). Pathological nail biting and pathological skin picking were operationalized as described previously (Bienvenu et al., 2000; Cullen et al., 2000). At each site, each case was reviewed independently by two expert diagnosticians who reviewed all case materials and assigned final best-estimate diagnoses. Final diagnoses were reviewed by diagnosticians at Johns Hopkins University.

As with other obsessive-compulsive symptoms, hoarding obsessions and compulsions were assessed with the CY-BOCS Symptom Checklist. To be assigned, these symptoms had to be clinically significant; i.e., the clinician determined that the individual recognized that his/her symptoms were excessive or unreasonable, and the symptoms caused marked distress, were time consuming, or significantly interfered with normal routine, occupational functioning, or social activities and relationships (American Psychiatric Association, 1994). For the current study of hoarding behavior, classification into hoarding and nonhoarding groups was based on the presence or absence of hoarding compulsions; only 18 of the participants without hoarding compulsions were diagnosed with hoarding obsessions.

Beginning in July 2008, the Social Responsiveness Scale (SRS) Parent Report was used, to evaluate social responsiveness, restricted interests, and repetitive behaviors. This instrument is a 65-item questionnaire, completed by a parent, to assess these behaviors in children and adolescents between the ages of 4 and 18 years. In addition to the total score, scores on five subscales are derived, viz.: "social awareness" (ability to recognize social cues); "social cognition" (ability to interpret social cues once they are recognized); "social communication" (expressive social communication); "social motivation" (motivation to engage in social-interpersonal behavior); and "restricted interests and repetitive behavior" (stereotypical behaviors and restricted interests). T-scores were determined from the raw scores using published norms which incorporate different reference means and standard deviations for men and women. Each T-score distribution has a mean of 50 and standard deviation of 10. The total score serves as the overall measure of the severity of deficits in social responsiveness. For the total score, T-scores of 59 or less are considered in the "normal" range. Scores ranging from 60–75 are considered in the "mild to moderate" range, indicating clinically significant deficiencies in reciprocal social behavior. Scores greater than 75 are considered in the "severe" range and are strongly associated with a clinical diagnosis of autistic disorder, Asperger's disorder, or more severe cases of pervasive developmental disorder (Constantino & Gruber, 2005).

2.3 Data analysis

Demographic features and clinical characteristics were compared between hoarding and non-hoarding participants, using the chi-square test for categorical variables, or Student's t-test for continuous variables. Correlation among SRS scales was evaluated using the Pearson

r statistic. Logistic regression by the method of General Estimating Equations (GEE), which provides correct estimates when cases may occur in the same families, was used to evaluate the relationship between Axis I disorders and hoarding (Liang & Zeger, 1986). GEE also was used to evaluate the relationships between the SRS total score and hoarding, adjusting for covariates that were found to be associated with hoarding. Given the exploratory nature of this study, each test was two-sided, with p<0.05.

3. Results

3.1 Demographic characteristics

A total of 641 children and adolescents (age 6–17 years) with definite OCD were evaluated during these studies and included in the current analyses. Three hundred and thirty (52%) were boys, and three hundred and eleven (49%) were girls. Thirty-eight (6%) were 6–8 years old; 161 (25%) were 9–11 years old; 214 (33%) were 12–14 years old; and 228 (36%) were 15–17 years old.

A total of 163 (25%) of the participants were diagnosed with definite hoarding compulsions, while 478 (75%) were not. Hoarding and non-hoarding participants had similar age distributions (mean age, 13.1 and 13.0 years, respectively). However, a greater proportion of hoarding than non-hoarding participants were female (56% vs. 46%; $\chi^2_1 = 4.68$; p=0.03).

3.2 Hoarding features

Among the hoarding individuals, the mean age of onset of hoarding compulsions was 7.7 years of age (range, 5–16 years). The time occupied by hoarding compulsions during the worst period was reported to be 1–3 hours daily in 23% of these individuals, and more than 3 hours daily in 15%. The level of distress associated with hoarding compulsions during the worst period was reported to be moderate (frequent and disturbing) in 21% of these individuals, and severe or extreme (near constant and disabling) in 28%.

3.3 OCD features

Hoarding participants had, on average, an earlier age at onset of obsessive-compulsive symptoms (mean, 6.6 vs. 7.5 years; $t_{636} = 3.71$, p<0.001), and more severe symptoms, as assessed by the mean YBOCS score for the worst symptomatic period (28 vs. 26; $t_{599} = 2.60$, p=0.01). The YBOCS score was not significantly related to the age at onset of hoarding (Pearson r= -0.06, p=0.17).

Several categories of obsessions were more prevalent in the hoarding participants, including contamination obsessions (75% vs. 66%, χ^2_1 = 5.0, p=0.03); symmetry obsessions (57% vs. 42%, χ^2_1 = 10.9, p<0.001); and somatic obsessions (38% vs. 29%, χ^2_1 = 4.9, p=0.03). In addition, several types of compulsions were more prevalent in the hoarding participants, including repeating compulsions (71% vs. 56%, χ^2_1 = 11.0, p<0.01); counting compulsions (46% vs. 33%, χ^2_1 = 8.5, p<0.01); and ordering compulsions (57% vs. 41%, χ^2_1 = 12.1, p<0.01).

A greater proportion of hoarding than non-hoarding individuals were rated by the examiners as having poor or lacking insight into their symptoms (33% vs. 19%, $\chi^2_1 = 13.4$, p<0.001).

A greater proportion of hoarding participants were rated as having moderate to extreme indecision (52% vs. 38%, χ^2_1 = 8.2, p<0.01) and moderate to extreme difficulty starting or completing tasks (60% vs. 47%, χ^2_1 = 7.6, p<0.01). In addition, a greater proportion of hoarding participants were judged by the examiners to have marked to extreme impairment (44% vs. 31%, χ^2_1 = 8.0, p<0.01). Also, a greater proportion of hoarding participants had received treatment for OCD (93% vs. 87%, χ^2_1 = 4.2, p=0.04)(Table 1).

3.4 Axis I Disorders

Several psychiatric disorders were more prevalent in hoarding than non-hoarding participants. These include panic disorder (8% vs. 3%, χ^2_1 = 4.7, p=0.03), specific phobia (22% vs. 15%, χ^2_1 = 4.4, p=0.04), and Tourette's disorder (7% vs. 3%, χ^2_1 = 7.7, p<0.01). Hoarding participants also had a greater prevalence of social phobia (17% vs. 12%, p=0.07) and tics (36% vs. 28%, p=0.07), at the margin of statistical significance (Table 3). The relationship between hoarding and these disorders were similar in girls and boys, except that the prevalence of panic disorder was substantially greater in hoarding than nonhoarding girls (11% vs. 4%; odds ratio=3.23, 95% CI=1.2–8.5, p=0.01) but not boys (2.9% vs. 3.1%; odds ratio=0.91, 95% CI=0.2–4.4, p=0.90).(Table 2)

3.5 Social Reciprocity Scores

SRS questionnaires were returned for 78 (48%) of hoarding and 200 (42%) of non-hoarding participants. Participants with and without returned questionnaires were not significantly different (p<0.05) on prevalence of hoarding compulsions, gender distribution, age distribution, YBOCS score, prevalence of poor insight, indecision, task completion, and most Axis I disorders. However, completers had an older age at onset of OC symptoms (7.6 vs. 7.0 years, p<0.01), and lower prevalences of specific phobia (12% vs. 21%, p=0.003), Tourette's disorder (0.7% vs. 6.1%, p<0.001), and tics (24% vs. 35%, p<0.01).

The SRS total score was strongly correlated with each of the SRS subscales: social awareness (Pearson r= 0.71), social cognition (r = 0.87), social communication (r = 0.95), social motivation (r = 0.81), and restricted interests and repetitive behavior (r = 0.85).

Higher mean scores (i.e., more social reciprocity deficits) were found for the hoarding than non-hoarding groups on social communication (59 vs. 55, $t_{265} = 2.14$, p=0.03), social motivation (65 vs. 59, $t_{262} = 2.90$, p<0.01), and repetitive interests and behavior (70 vs. 64, $t_{266} = 2.80$, p<0.01), as well as the SRS total score (63 vs. 58, $t_{245} = 2.70$, p<0.01) (Table 3).

Using logistic regression, the relative odds of hoarding, per unit increase in the SRS total score, was 1.03, (95% CI=1.01–1.05, p<0.01). The magnitude and significance of this relationship did not appreciably change in separate models that included the SRS total score and, one-by-one, those variables that were found in previous analyses to be associated with hoarding at p<0.10. Seven of these variables (i.e., age at onset, contamination obsessions, symmetry obsessions, repeating compulsions, counting compulsions, ordering compulsions, and indecision) remained related (at p<0.10) to hoarding, in addition to the SRS total score. Therefore, we fit a logistic regression model that included the SRS total score and, simultaneously, these seven variables. In this multivariable model, the SRS total score (OR= 1.03; 95% CI = 1.01 - 1.05, p=0.02), age at onset of obsessive-compulsive symptoms (OR =

0.87; 95% CI=0.76-0.98, p=0.03), symmetry obsessions (OR = 2.38, 95% CI = 1.11-5.07, p=0.03, and indecision (OR = 1.91, 95% CI=1.02-3.60, p=0.04) were significantly, independently related to hoarding in the children and adolescents with OCD (Table 4).

4. Discussion

4.1 Major findings

We found that a substantial proportion (25%) of children and adolescents with OCD in the study sample had hoarding compulsions. As a group, the hoarding participants had earlier age at onset of, and more severe, obsessive-compulsive symptoms, as well as a greater prevalence of panic disorder, specific phobia, and Tourette disorder. These findings are generally consistent with several previous studies of pediatric OCD, which found that hoarding individuals had, on average, longer illness duration (Mataix-Cols et al., 2008), poorer insight (Storch et al., 2007), higher levels of anxiety, depression, other internalizing and externalizing symptoms, and emotional difficulties (Storch et al., 2007; Mataix-Cols et al., 2008), and greater prevalence of specific anxiety disorders (Masi et al., 2010; Frank et al., 2014).

In contrast to our finding of a significantly greater CY-BOCS severity score in hoarding participants, Storch et al. (2007) did not, although it should be noted that their sample size was considerably smaller than that of the current study. We also did not replicate previous reports of an association between bipolar disorder and hoarding symptoms in pediatric OCD (Masi et al., 2010; Joshi et al., 2007); in our study, bipolar disorder was diagnosed in only 4 cases in the study sample (1 hoarding and 3 non-hoarding individuals).

To our knowledge, the current study is one of the first to investigate the relationship between a dimensional measure of social reciprocity and hoarding in pediatric OCD. We found that difficulty with social reciprocity was associated with hoarding in these individuals, independent of other risk correlates of hoarding. In contrast to our findings, Anholt et al. (2010) found that, in 109 adults OCD outpatients, the YBOCS hoarding dimension was significantly correlated with poor social skills, lack of imagination, and poor attention switching, as measured by dimensions of the Autism Spectrum Quotient (AQ) (Baron-Cohen et al., 2001); however, in multivariable models that adjusted for age, hoarding was related only to the ADHD inattention subscale, whereas other OCD dimensions were related to AQ dimensions as well as ADHD dimensions. In support of our findings, Mito et al. (2014) found that, in 81 adult OCD outpatients, hoarding obsessions and compulsions were the only OC symptoms that were significantly more prevalent in those with high vs. low AQ total scores, and that scores on hoarding obsessions and compulsions were significantly correlated with several AQ subscales, including social skills and communication. Morevover, we recently reported a significant relationship between social and communication difficulties, using the Pragmatic Rating Scale (Landa et al., 1992), and hoarding behaviors in adults with OCD (Samuels et al., in press).

A relationship between autistic traits and hoarding is not specific to OCD, since hoarding has been observed in a variety of developmental disorders, including Prader-Willi syndrome and ADHD (Storch et al., 2011b). In these disorders, as well as in OCD, hoarding may be a

consequence of restricted interests, preoccupation with parts of objects, and resistance to environmental change that characterize many individuals with autistic traits. Moreover, although collecting rituals are developmentally normative, with peak age of occurrence about 2 years of age (Evans et al., 1997), children with high levels of fear, shyness, and need for order and predictability may maintain and extend these rituals beyond the age when they are adaptive (Zohar & Felz, 2001). Alternatively, hoarding and autism may be neurodevelopmental consequences of deletions of the same, specific chromosomal regions, as suggested by recent findings (Lionel et al., 2014; McGrath et al., 2014).

We also found that early age at onset of OCD, symmetry obsessions, and indecision were independently related to hoarding in youth with OCD. Along with difficulties in social reciprocity, these features should be considered as candidate risk factor is for the development of hoarding behavior in pediatric OCD. Previous studies have found a strong relationship between difficulty making decisions, deficits in executive functioning, and hoarding in adults (Morein-Zamir et al., 2014; Tolin et al., 2012).

4.2 Strengths and Limitations

To our knowledge, this is the largest study to date of hoarding behavior in children and adolescents with OCD. Participants were recruited from a variety of sources, were not selected for treatment, and were thoroughly examined and rigorously diagnosed. However, several potential limitations of the study should be considered. First, given the genetic focus of the OCGS and OCGAS studies, the study sample reflects more familial cases of OCD and hoarding and may be less generalizable to non-familial cases. Furthermore, as all hoarding participants in the study sample had OCD, the results may not be applicable to pediatric hoarding participants without OCD (Mataix-Cols et al., 2010); moreover, assessing hoarding behavior with the CY-BOCS instead of with hoarding-specific measures may not capture hoarding that is independent of OCD (Storch et al., 2011). The DSM-5 now construes hoarding disorder as a condition that is related to, but distinct from, OCD (American Psychiatric Association, 2013).

Second, hoarding may be challenging to identify, especially in children. On the one hand, parent informants may misidentify developmentally normative collecting behavior as pathological; conversely, clutter and other manifestations of hoarding may be more difficult to recognize in children, since parents often have at least some control over the acquisition and discarding of their child's possessions (Storch et al., 2011a). Furthermore, although a substantial proportion of the hoarding participants had hoarding compulsions that were severe and time consuming, the proportion of cases that would meet strict criteria for DSM-5 hoarding disorder, as specified for adults, is unclear (American Psychiatric Association, 2013). Further refinement of the definition and evaluation of hoarding in pediatric cases is needed (Frank et al., 2014).

Third, the SRS was completed for only a subset of OCD-affected youth; it was not included in the OCGS study, and was introduced over a year after the initiation of the OCGAS study. A spurious estimate of the magnitude of the relationship between SRS and hoarding could have occurred if parents were more likely to complete the questionnaire for those children with both hoarding behavior and high SRS scores (e.g., children with more severe OCD).

However, we found that the children and adolescents with SRS scores were not significantly different from SRS non-completers on prevalence of hoarding compulsions, demographic characteristics, or most clinical features, apart from an older age at onset of obsessive-compulsive symptoms and lower prevalences of specific phobia, Tourette's disorder, and tics.

4.3 Clinical implications

Although much has been learned in the past decade about the treatment of adult hoarding patients (Mataix-Cols, 2014), there have been few reports of the treatment of children and adolescents with hoarding behavior (Storch et al., 2011b). Clinicians should be aware that, among children and adolescents with OCD, those with hoarding may be more severely ill, with poorer insight, and greater co-occurrence with other psychiatric symptoms. These patients also may have more difficulty making decisions, completing tasks, and possibly with other executive functions, which may interfere with their adherence to treatment. Given possible impairments in social reciprocity, they also may have more difficulty engaging and interacting with clinicians. Clinical studies are needed to determine the prognosis of these patients, and to develop more effective strategies for their treatment.

4.4 Future research directions

Several future research directions are suggested by the results of this study. First, there is need for longitudinal follow-up of these cases, to determine if the course of OCD is different in those with and without hoarding symptoms, and if the relationships between social reciprocity, the other independent risk correlates (early age at onset, symmetry obsessions, and indecision) and hoarding persist into adulthood. Second, given the poorer treatment response of many adult OCD patients with hoarding behavior, there is need to determine if treatment of hoarding and social reciprocity deficits, symmetry obsessions, and indecision earlier in the course of illness improves the prognosis. Third, there is need to elucidate potential neurobiological mechanisms underlying pediatric hoarding, through neuropsychological, neurophysiological, and functional neuroimaging studies. Finally, there is need to identify genetic and environmental factors that increase the risk of developing hoarding behavior during the pediatric period.

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Highlights

• We compared clinical features in pediatric OCD cases with (N=163) and without (N=478) hoarding.

- The hoarding group had earlier age at onset and more severe OCD, poorer insight, and more impairment.
- The hoarding group also had more severe deficits in social reciprocity.
- After adjusting for other factors, social reciprocity was independently related to hoarding.

Table 1
Clinical features in children and adolescents with OCD, by hoarding status

	Hoarding (N=163) n (%)	Non- Hoarding (N=478) n (%)	Test Statistic	p-value
Obsessions				
Aggressive	98 (61)	268 (57)	$\chi^2_{1} = 0.5$	0.49
Contamination	121 (75)	301 (66)	$\chi^2_1 = 5.0$	0.03
Sexual	34 (21)	110 (24)	$\chi^2_{1} = 0.5$	0.50
Religious	62 (39)	150 (32)	$\chi^2_{1} = 2.5$	0.11
Symmetry	93 (57)	197 (42)	$\chi^2_1 = 10.9$	< 0.01
Somatic	61 (38)	131 (29)	$\chi^2_1 = 4.9$	0.03
Hoarding	142 (92)	18 (4)	$\chi^2_1 = 450.9$	< 0.001
Compulsions				
Cleaning	116 (71)	310 (65)	$\chi^2_1 = 1.9$	0.17
Checking	101 (62)	264 (56)	$\chi^2_{1} = 2.2$	0.14
Repeating	115 (71)	267 (56)	$\chi^2_1 = 11.0$	< 0.01
Counting	74 (46)	158 (33)	$\chi^2_1 = 8.5$	< 0.01
Ordering	93 (57)	196 (41)	$\chi^2_1 = 12.1$	< 0.01
Other clinical features				
Insight into symptoms, poor or lacked	51 (33)	84 (19)	$\chi^2_1 = 13.4$	< 0.001
Indecision, moderate to extreme	79 (52)	171 (38)	$\chi^2_1 = 8.2$	< 0.01
Difficulty starting or finishing tasks, moderate to extreme	92 (60)	212 (47)	$\chi^2_1 = 7.6$	< 0.01
Doubting, moderate to extreme	57 (37)	152 (34)	$\chi^2_1 = 0.4$	0.52
Global impairment, marked to extreme	65 (44)	137 (31)	$\chi^2_1 = 8.0$	< 0.01

 Table 2

 Lifetime psychiatric disorders in children and adolescents with OCD, by hoarding status

	Hoarding (N=163) n (%)	Non- Hoarding (N=478) n (%)	Test Statistic	p-value
Panic disorder	12 (8)	16 (3)	$\chi^2_1 = 4.7$	0.03
Agoraphobia	7 (4)	13 (3)	$\chi^2_1 = 1.0$	0.31
Social phobia	27 (17)	54 (12)	$\chi^2_{1} = 3.2$	0.07
Specific phobia	36 (22)	70 (15)	$\chi^2_1 = 4.4$	0.04
Generalized anxiety disorder	37 (24)	94 (21)	$\chi^2_1 = 0.8$	0.38
Separation anxiety disorder	44 (28)	123 (27)	$\chi^2_{1} = 0.1$	0.83
Major depression	50 (31)	119 (26)	$\chi^2_1 = 1.9$	0.17
Body dysmorphic disorder	6 (4)	17 (4)	$\chi^2_1 = 0.01$	0.91
Trichotillomania	4 (3)	16 (3)	$\chi^2_1 = 0.3$	0.58
Nail biting	12 (8)	44 (10)	$\chi^2_1 = 0.4$	0.52
Skin picking	18 (12)	48 (10)	$\chi^2_1 = 0.2$	0.63
Tourette's Disorder	12 (7)	12 (3)	$\chi^2_1 = 7.7$	< 0.01
Tics	58 (36)	131 (28)	$\chi^2_1 = 3.4$	0.07

Table 3
Social Reciprocity Scale (SRS) t-scores in children and adolescents with OCD, by hoarding status

	Hoarding (N=78) Mean (SD)	Non- Hoarding (N=200) Mean (SD)	Test Statistic	p-value
Social Awareness	52.6 (12.0)	51.0 (11.5)	$t_{268} = 1.0$	0.32
Social Cognition	58.3 (13.6)	56.1 (13.8)	$t_{259} = 1.1$	0.27
Social Communication	58.9 (14.5)	54.9 (13.4)	$t_{265} = 2.1$	0.03
Social Motivation	64.8 (15.0)	59.1 (14.3)	$t_{262} = 2.9$	< 0.01
Restricted Interests and Repetitive Behavior	70.0 (18.3)	63.8 (15.7)	$t_{266} = 2.8$	< 0.01
SRS Total Score	63.1 (15.0)	57.7 (13.8)	$t_{245} = 2.7$	< 0.01

 $^{^{}I}\mathrm{Higher}\,\mathrm{SRS}$ subscale and total scores indicate greater dysfunction in social reciprocity.

Table 4

Relationship between clinical variables and hoarding compulsions in children and adolescents with OCD Multivariable General Estimating Equations model

Variable	Odds ratio (95% Confidence Interval)	p-value
Social Reciprocity Scale (SRS) Total score	1.03 (1.01 – 1.05)	0.02
Age at onset of obsessive-compulsive symptoms, years	0.87 (0.76 – 0.98)	0.03
Contamination obsessions	1.89 (0.91 – 3.95)	0.09
Symmetry obsessions	2.63 (1.21 – 5.69)	0.01
Repeating compulsions	1.47 (0.72 – 3.02)	0.29
Counting compulsions	1.73 (0.90 – 3.33)	0.10
Ordering compulsions	0.92 (0.44 – 1.93)	0.82
Indecision	1.86 (1.01 – 3.45)	0.04

 $^{^{}I}\mathrm{Higher}\;\mathrm{SRS}\;\mathrm{scores}$ indicate greater dysfunction in social reciprocity.