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**Research article** 

# Clinical characteristics of hoarding disorder in Japanese patients

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# ABSTRACT

Previous studies have reported clinical characteristics of hoarding disorder (HD), such as early onset, a chronic course, familiality, high unmarried rate, and high rates of comorbidities. However, clinical research targeting Japanese HD patients has been very limited. As a result, there is a low recognition of HD in Japan, leading to insufficient evaluation and treatment of Japanese HD patients.

The aim of the current study was to delineate the clinical characteristics of Japanese HD patients. Thirty HD patients, 20 obsessive-compulsive disorder (OCD) patients, and 21 normal controls (NC) were targeted in this study.

The HD group had a tendency toward higher familiality, earlier onset, and longer disease duration compared to the OCD group. In addition, the HD group showed a significantly higher unmarried rate than the NC group. The top two comorbidities in the HD group were major depressive disorder (56.7%) and attention-deficit/ hyperactivity disorder (26.7%). The HD group had significantly higher scores on hoarding rating scales and lower scores on the Global Assessment of Functioning Scale than the other two groups.

The current study showed a clinical trend in Japanese HD patients similar to previous studies in various countries, suggesting that HD may be a universal disease with consistent clinical symptoms.

# 1. Introduction

Hoarding disorder (HD) is a new disease concept presented in the *Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5;* American Psychiatric Association, 2013). Patients with HD find it difficult to throw away possessions, regardless of their actual value, and to organize those things. According to Frost and Hartl (1996), the symptoms of hoarding have been defined as "(1) the acquisition of and failure to discard a large number of possessions that appear to be useless or of limited value; (2) living spaces sufficiently cluttered so as to preclude activities for which those spaces were designed; and (3) significant distress or impairment in functioning caused by the hoarding." In addition, other previous studies have reported that severe hoarding caused serious problems affecting the patient's neighborhood such as sanitation

problems, fires, and collapse of stacked items (Frost et al., 2000; Lucini et al., 2009; Mataix-Cols, 2014; Mataix-Cols and Fernandez de la Cruz, 2018; Tolin et al., 2008). Thus, hoarding has been suggested to affect not only the patient but also family members and neighbors if symptoms become serious without appropriate intervention or treatment. Although hoarding was conventionally regarded as a subtype of obsessive-compulsive disorder (OCD), recent studies have suggested differences in clinical characteristics, cognitive function, and neuro-imaging findings between the hoarding subtype and other subtypes of OCD (Mataix-Cols et al., 2010; Pertusa et al., 2010). Based on these studies, hoarding became independent as a new diagnostic category called HD in the *DSM-5*.

Several European studies have reported the prevalence of HD to be approximately 2.3%–6.0% (Bulli et al., 2014; Iervolino et al., 2009;

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Mueller et al., 2009; Timpano et al., 2011). As clinical characteristics of HD, previous studies have reported familiality (Iervolino et al., 2009; Pertusa et al., 2008; Samuels et al., 2007; Steketee et al., 2015), high unmarried rate (Kim et al., 2001), early onset (during childhood or adolescence; Grisham et al., 2006; Landau et al., 2011; Tolin et al., 2010a), a chronic course (Ayers et al., 2010; Grisham et al., 2006; Tolin et al., 2010a), and poor insight (Jakubovski et al., 2011; Matsunaga et al., 2010; Tolin et al., 2010b; Torres et al., 2012). Moreover, several recent studies have reported high rates of comorbidities with HD, including depressive disorders, anxiety disorder, impulse control disorders, and attention-deficit/hyperactivity disorder (ADHD; Frost et al., 2001; Frost et al., 2011; Mueller et al., 2009; Samuels et al., 2002; Sheppard et al., 2010; Tolin et al., 2018; Tolin et al., 2012a). Previous studies have suggested that hoarding is associated with ADHD (Grisham et al., 2007; Grisham et al., 2010; Hartl et al., 2005; Park et al., 2016).

Recent studies have also reported the possibility of cognitive impairment in HD (Grisham and Baldwin, 2015; Woody et al., 2014), such as information processing problems in hoarding patients (Blom et al., 2011; Frost and Hartl, 1996; Grisham et al., 2010; Mackin et al., 2011; Mackin et al., 2016; Pushkarskaya et al., 2018; Raines et al., 2014; Steketee and Frost, 2003; Tolin et al., 2011; Wincze et al., 2007) and the impairment of attention, memory, and executive function in HD (Ayers et al., 2016; Ayers et al., 2013; Steketee and Frost, 2003; Tolin et al., 2018; Tolin and Villavicencio, 2011).

In Japan, there have been very limited clinical studies targeting HD patients as newly defined by the *DSM-5*. Toward appropriate intervention and treatment for Japanese HD patients, we have proposed that it is necessary to delineate the clinical characteristics of Japanese HD patients under the new definition of HD and clarify whether the clinical characteristics of HD differ among Japanese patients vs Western patients. Matsunaga et al. (2010) investigated hoarding symptoms in Japanese OCD patients, and the results were similar to those of previous studies mainly in Western countries, suggesting that hoarding in OCD is a consistent symptom regardless of cultural characteristics. Therefore, we hypothesized that Japanese HD patients may have similar clinical characteristics to Western patients.

The aim of the current study was to accurately identify the clinical characteristics of Japanese HD patients. Based on the aforementioned background of HD independence from OCD in the DSM-5 and the clinical characteristics of HD reported in Western countries, we hypothesized that Japanese HD patients may also show early onset, a chronic course, high unmarried rate, and high rates of comorbidities, etc., compared to OCD patients and/or normal subjects. We established a clinical control group of OCD patients and a normal control (NC) group to understand the clinical characteristics of the current Japanese HD patients. We conducted detailed background examinations, structured psychiatric interviews, and evaluated the psychiatric symptoms of each group. We also conducted neuropsychological tests as supplementary background data for each group to reinforce the clinical characteristics of the participants. Finally, we discussed whether the clinical characteristics of Japanese HD patients are different from those of Western patients by comparing the results of the current study with previous studies mainly in Western countries.

#### 2. Materials and methods

#### 2.1. Participants and procedures

Participants in the HD group were recruited from the Department of Psychiatry of Kyushu University Hospital, related local psychiatric clinics and hospitals, and through newspaper advertisements targeting the general public issued by a local mental health institution in Japan. The inclusion criterion for the HD group was to meet the HD diagnostic criteria of the *DSM-5*; trained psychiatrists conducted the structured interviews and made the diagnosis. Participants in the OCD group were recruited from among outpatients at the Department of Psychiatry of Kyushu University Hospital. Patients were included in the OCD group if they met the OCD diagnostic criteria in the *DSM-IV*. The exclusion criterion for the OCD group was having a clinical level of hoarding symptoms. Participants in the NC group were recruited from healthy people who had no clinical psychiatric symptoms and adequately maintained their social and occupational functions. The exclusion criteria for the NC group were those who satisfied the diagnosis of Axis I disorders in the *DSM-IV* and had a history of psychiatric disorder and treatment. All of the participants were recruited from August 2013 to October 2017.

In the current study, after acquiring consent, psychiatrists who specialize in OCD treatment conducted psychiatric interviews with the participants to obtain clinical information including their background, medical history, and past/current symptoms. The presence or absence of hoarding in first-degree relatives was obtained from the participants in the interview about their clinical information. The evaluators also conducted neuropsychological tests. For the participants in the HD group, the evaluators also listened to details about their saved items and their reasons for difficulty in discarding. All of the participants were confirmed to be without neurological disorders, brain injury, serious medical condition, history of drug or alcohol addiction, or mental retardation. The institutional research and ethics committee of Kyushu University approved the study (No. 28-101). Written informed consent was obtained from each participant before the assessments began.

# 2.2. Clinician-administered measures

#### 2.2.1. Structured interview for hoarding disorder (SIHD)

For the assessment of HD, we conducted the SIHD (Nordsletten et al., 2013a), a semi-structured interview based on the *DSM-5* criteria for HD, which consists of questions about each of the diagnostic criteria and specifiers and assists the diagnosis of HD. Based on the *DSM-5*, the degree of insight in the HD and OCD groups was evaluated as "good/fair," "poor," or "absent/delusional" in our study. The SIHD used in the current study was the Japanese version that we translated from the original English version.

# 2.2.2. Structured clinical interview for DSM-IV Axis I disorders (SCID-I)

The SCID-I (First et al., 1996) Japanese language edition, a structured interview to diagnose Axis I disorders in the *DSM-IV*, was used to assess OCD and other diagnoses. The *DSM-5* Japanese language edition was not yet published when we recruited the OCD patients in this study; therefore, we assessed OCD and other diagnoses using the *DSM-IV*.

# 2.2.3. Conners' adult ADHD diagnostic interview for DSM-IV (CAADID)

Comorbid ADHD was diagnosed using the CAADID (Epstein and Kollins, 2006).

#### 2.2.4. Clutter Image Rating (CIR)

We employed the CIR (Frost et al., 2008) to assess the severity of the participants' hoarding symptoms. The CIR is a visual assessment consisting of nine photographs showing stepwise degrees of clutter in each of three rooms (kitchen, living room, and bedroom), and the participants choose photographs closest to the states of their homes. The chosen photographs were converted into points, and the CIR score was calculated by averaging the total points of the three rooms. The cut-off score for clinical level hoarding was 4 points (Frost et al., 2008).

# 2.2.5. Yale-Brown Obsessive-Compulsive Scale (Y-BOCS)

The Y-BOCS (Goodman et al., 1989a, 1989b) measured the severity of obsessions and compulsions.

# 2.2.6. Global Assessment of functioning (GAF) scale

The overall psychological, social, and occupational functions were assessed using the GAF Scale (Endicott et al., 1976).

#### 2.2.7. Assessment of educational history

We assessed the participants' educational history based on a previous study (Okada et al., 2014). We classified the educational history into seven stages and quantified them by converting the scores to evaluate the educational history instead of educational years in our study. This was because, for example, it is not appropriate to adopt the absolute value of the number of years of education of subjects who have enrolled in multiple universities as their educational history. In our study, elementary school graduation was quantified as "1," junior high school graduation was "2," high school dropout was "3," high school graduation was "4," four-year university dropout or junior college, vocational school dropout/graduation was "5," four-year university graduation was "6," and a master's degree or higher was "7."

#### 2.3. Self-reported measures

# 2.3.1. Saving Inventory-Revised (SI-R)

We employed the SI-R (Frost et al., 2004) to assess the severity of the participants' hoarding symptoms. The SI-R is a self-reported questionnaire consisting of three subscales: excessive acquisition, difficulty in discarding, and clutter. Each of the questions was scored from 0 point to 4 points, and the total SI-R score was in the range of 0–92 points. The cut-off score for clinical level hoarding was 41 points (Frost et al., 2004). The SI-R used in the current study was the Japanese version (SI-R-J), whose reliability and validity were verified for Japanese students at undergraduate and vocational colleges in Japan (Tsuchiyagaito et al., 2015).

#### 2.3.2. Beck Depression Inventory-Second Edition (BDI-II)

The BDI-II (Beck et al., 1996; Kojima et al., 2002) assessed the severity of depression.

#### 2.3.3. Autism-Spectrum Quotient (AQ)

Autistic traits were assessed with the AQ (Baron-Cohen et al., 2001; Wakabayashi et al., 2004).

# 2.3.4. Adult ADHD Self-Report Scale (ASRS)

The ASRS (Kessler et al., 2005; Takeda et al., 2017) assessed the frequency of ADHD symptoms.

## 2.3.5. Conners' adult ADHD rating scales (CAARS)

The CAARS (Conners et al., 1999; Conners et al., 2012) measured the severity of ADHD symptoms. To evaluate the CAARS score, the following eight subscales were used: "Inattention/Memory Problems," "Hyperactivity/Restlessness," "Impulsivity/Emotional Lability," "Problems with Self-Concept," "DSM-IV: Inattentive Symptoms," "DSM-IV: Hyperactive-Impulsive Symptoms," "DSM-IV: ADHD Symptoms Total," and "ADHD Index."

#### 2.4. Neuropsychological assessments

#### 2.4.1. Trail making test (TMT)

We used the Japanese version of the TMT (Kashima et al., 1986) to evaluate attention function. The TMT has two types of tests, Part A and Part B. Part A uses a test sheet with numbers 1 to 25 randomly placed, and the subject connects the numbers with a line in order from 1. Part B is a test that connects randomly arranged numbers 1 to 13 and Kana characters "A" to "Shi" alternately in order from 1. We evaluated the sustainability and selectivity of attention, cooperativeness of visual and exercise, and speed of information processing from the times required for Part A and Part B. Furthermore, we evaluated the ability to switch attention and executive function from the time required for Part B. We also calculated the time required for Part B minus the time required for Part A, which is an indicator reflecting the ability to suppress reactions, switch actions, and employ working memory.

# 2.4.2. Modified Stroop test

We used the Japanese version of the Modified Stroop test (Part I and Part III; Kato, 1988), which is a task to detect frontal lobe dysfunctions, especially stereotype inhibition dysfunction. Part I uses an illustration with a total of 24 dots in four colors: red, blue, green, and yellow. Part III uses an illustration with Chinese characters written in a color different from the meaning of the word; for example, the Chinese character with the meaning "blue" is written in red ink. The subject answers the "color" of the writing ink as soon as possible in Part I and Part III of the test. We evaluated reading fluency by the time required for Part I and the ability to suppress daily habitual or stereotypic activity from the time required for Part III.

#### 2.4.3. Iowa gambling task (IGT)

We evaluated decision-making abilities using the IGT (Bechara et al., 1994). The IGT is believed to simulate decision-making in real-life situations and evaluate the process of action selection based on decision-making that is not influenced by general knowledge or past experiences. In the IGT, the subject selects a total of 100 cards with rewards or fines from four piles of cards (decks A, B, C, and D) to increase the total amount as much as possible. Decks A and B are set as "risky decks," which are both high in rewards and fines and ultimately lead to a reduction in the amount earned. Conversely, decks C and D are set as "safe decks," which are both low in rewards and fines and eventually lead to an increase in the amount earned. We evaluated the decision-making abilities from "the number of times a safe deck (C or D) was chosen minus the number of times a risky deck (A or B) was chosen" and the total amount of money earned.

# 2.4.4. Keio version of the Wisconsin card sorting test (KWCST)

We evaluated executive function using the KWCST (Kashima et al., 1985), which consists of 48 trials. The KWCST is a test of conceptual or "set" conversion failure; that is, the test detects the difficulty of moving from one particular concept or mind set once held or manipulated to another concept or mind set. The following six subscales were used to evaluate the KWCST score: categories achieved (CA), perseverative errors of Nelson (PEN), difficulties of maintaining set, numbers of response cards until the first category achieved, total errors (TE), and non-perseverative errors of Nelson.

# 2.5. Statistical analyses

All of the statistical analyses were conducted using JMP Pro 13 (SAS Institute Inc., Cary, NC, USA). For continuous quantitative variables, normality was first evaluated for each group using the Shapiro-Wilk test. For normally distributed continuous variables, the mean (standard deviation) in each group was estimated, and multiple comparisons were conducted using Dunnett's test. For non-normally distributed continuous variables, the median (interquartile range) was calculated for each group, and comparisons of two groups were conducted using the Wilcoxon rank sum test. Steel's test was performed for multiple comparisons. For binary variables, the frequency in each group was expressed using % (*n*) and compared between groups using the  $\chi^2$  test and Fisher's exact test. The significance levels were set for a *p* value < 0.05.

#### 3. Results

Thirty participants with HD, 20 participants with OCD, and 21 healthy adults who met the inclusion and exclusion criteria of each group participated in the current study. The range of their ages was from 18 to 65 years old.

# 3.1. Hoarded items and reasons for hoarding

As shown in Table 1, very frequently hoarded items were books/ magazines/comics/textbooks (76.7%), clothes (76.7%), plastic bags/

Та	ıbl	e 1	. 1	Hoarde	ed i	tems	and	reasons	for	hoard	ing
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Hoarding by a First-Degree Relative, % (n)

	Hoarding Disorder ( $n = 30$ )		
oarded Items	n	%	
Books/Magazines/Comics/Textbooks	23	76.7	
Clothes	23	76.7	
Plastic Bags/Paper Bags	16	53.3	
Documents	14	46.7	
Boxes/Cardboard Boxes	9	30.0	
Receipts	8	26.7	
Flyers	7	23.3	
Items Related to Their Children	6	20.0	
Videos/DVDs/CDs	5	16.7	
Newspapers	4	13.3	
Postal Items	4	13.3	
Notes	4	13.3	
Containers	4	13.3	
Dolls	4	13.3	
Hobby Items	4	13.3	
Garbage	4	13.3	
Gifts	4	13.3	
Album/Photos	3	10.0	
Free Paper	3	10.0	
Food Packages	3	10.0	
Bottles	3	10.0	
PET Bottles	3	10.0	
Bags	2	6.7	
Dishes	2	6.7	
easons for Hoarding		l.	
Future Use	19	63.3	
Sentimental Attachment	18	60.0	
Not Good at Organizing	8	26.7	
Avoid Waste	6	20.0	
Too Much Time to Prepare before Discarding	6	20.0	
Procrastination	4	13.3	
Compulsive Shopper	4	13.3	
Never Available	4	13.3	
Lose Motivation	3	10.0	
Part of Personal Identity	2	6.7	
No Reason for Discarding	2	6.7	
Personality	2	6.7	
Feel Sorry	2	6.7	
ep as Records	2	6.7	

paper bags (53.3%), and documents (46.7%). Various other items were also hoarded, and most were common items that are found in daily life. Regarding the reasons for hoarding, "future use" (63.3%) and "sentimental attachment" (60.0%) were the most frequent, in almost equal proportions.

# 3.2. Demographic characteristics and psychiatric profiles

As shown in Table 2, no significant differences were found in some demographic information (for example, gender ratio, age, educational history, and the proportion living alone). However, the HD group showed a significantly higher proportion of patients with first-degree relatives who hoarded than the OCD and NC groups (p = 0.002, p < 0.001, respectively). Moreover, the unmarried rate was significantly higher in the HD group than the NC group (p = 0.03).

As shown in Table 3, the participants in the HD group had a significantly lower age at onset and longer disease duration compared to the OCD group (p = 0.002, p = 0.03, respectively). Moreover, the HD group showed a tendency to have a higher proportion of patients with a history of psychiatric comorbidities than the OCD group (p = 0.06). The main comorbidities in the HD group were major depressive disorder (MDD; 56.7%) and ADHD (26.7%). In addition, the HD group had a significantly higher comorbid rate of ADHD than the OCD group (p = 0.02). The high rate of 73.3% of patients in the HD group were taking psychiatric medication. Conversely, no significant difference in the proportions with poor insight was found between the HD and OCD groups (p = 0.37), and no patient in either the HD or OCD groups showed absent or delusional insight.

#### 3.3. Hoarding scales and other clinical variables

Scores on the hoarding scales and other clinical variables are summarized in Table 4. The HD group had significantly higher scores on the SI-R-J and CIR than the OCD and NC groups (all p < 0.001), and the scores on both hoarding scales in the HD group were above the original cut-off score for each scale. Interestingly, most of the scores on the ADHD evaluation scales (the ASRS and subscales of the CAARS, such as "Inattention/Memory Problems") were significantly higher in the HD group than the OCD and NC groups (all p < 0.05). In addition, the GAF score in the HD group was low and significantly lower than in the other two groups (all p < 0.001).

# 3.4. Neuropsychological tests

0.002

We conducted neuropsychological tests as supplementary background data for each group (Table S1). Contrary to predictions, no significant difference between the HD and NC groups was seen in the results of all of the tests performed. Conversely, the results of a test of attention function (TMT) were poorer in the OCD group than the HD group with

0.0(0)

p

0.39

0.80

0.96

0.03

0.46

< 0.001

HD vs. NC

#### Table 2. Demographic characteristics of HD, OCD, and NC groups. HD OCD NC D (n = 20)(n = 30)HD vs. OCD (n = 21)Female, % (*n*) 73.3 (22) 65.0 (13) 0.53 61.9 (13) Age, mean (SD) 42.7 (12.6) 44.5 (10.6) 0.81 44.5 (8.1) Educational History, median (IQR) 6.0 (4.0-6.0) 5.0 (4.0-5.0) 0.34 5.0 (5.0-6.0) Unmarried, % (n) 63.3 (19) 65.0 (13) 0.90 33.3 (7) Living Alone, % (n) 33.3 (10) 20.0 (4) 0.30 23.8 (5)

46.7 (14)

Abbreviations: HD, Hoarding Disorder; OCD, Obsessive-Compulsive Disorder; NC, Normal Control; SD, standard deviation; IQR, interquartile range. We classified the educational history into the following seven stages and quantified them by converting the scores to evaluate the educational history. Elementary school graduation was quantified as "1," junior high school graduation was "2," high school dropout was "3," high school graduation was "4," the four-year university dropout or junior college, vocational school dropout/graduation was "5," the four-year university graduation was "6," and the master's degree or higher was "7."

5.0 (1)

# Table 3. Psychiatric profiles.

	HD ( <i>n</i> = 30)	OCD ( $n = 20$ )	р
Age at Onset, median (IQR)	14.0 (9.3–21.3)	28.5 (20.0-33.8)	0.002
Duration of Disease (IQR)	18.5 (14.3–32.3)	15.0 (8.5–21.8)	0.03
Any Psychiatric Medication, % (n)	73.3 (22)	100.0 (20)	0.02
Antidepressants, % (n)	50.0 (15)	95.0 (19)	< 0.001
Poor Insight, % (n)	23.3 (7)	35.0 (7)	0.37
Comorbid Disorders (lifetime), % (n)	80.0 (24)	55.0 (11)	0.06
MDD, % ( <i>n</i> )	56.7 (17)	45.0 (9)	0.42
ADHD, % (n)	26.7 (8)	0.0 (0)	0.02
OCD, % ( <i>n</i> )	23.3 (7)	-	-
Pervasive Developmental Disorders, % (n)	20.0 (6)	0.0 (0)	0.07
Bipolar II Disorder, % (n)	10.0 (3)	0.0 (0)	0.27
Panic Disorders, % (n)	10.0 (3)	5.0 (1)	0.64
Specific Phobia, % ( <i>n</i> )	10.0 (3)	5.0 (1)	0.64
Psychotic Disorders, % (n)	6.7 (2)	0.0 (0)	0.51
Social Phobia, % (n)	3.3 (1)	5.0 (1)	> 0.99
Post-traumatic Stress Disorder, % (n)	3.3 (1)	0.0 (0)	> 0.99
Eating Disorders, % (n)	3.3 (1)	5.0 (1)	> 0.99
Generalized Anxiety Disorder, % (n)	0.0 (0)	10.0 (2)	0.16
HD, % ( <i>n</i> )	-	0.0 (0)	-

**Abbreviations:** HD, Hoarding Disorder; OCD, Obsessive-Compulsive Disorder; IQR, interquartile range; MDD, Major Depressive Disorder; ADHD, Attention-Deficit/Hyperactivity Disorder.

The duration of disease was calculated from "age" and "age at onset."

respect to the TMT Part A (p = 0.02). Moreover, on a test of executive function (KWCST), the participants in the OCD group also had significantly poorer scores on its subscales such as CA, PEN, and TE than those in the HD group (all p < 0.05).

# 4. Discussion

We discussed whether the clinical characteristics of Japanese HD patients differ from those of Western patients by comparing the results of the current study with previous studies mainly in Western countries.

# 4.1. Hoarded items and reasons for hoarding

Hoarded items in the current HD group were commonplace items, such as "books/magazines/comics/textbooks," "clothes," "plastic bags/ paper bags," and "documents." As in our research, a previous study (Nordsletten, et al., 2013b) showed that frequently hoarded items of HD participants were commonplace items such as "old clothes," "letters," "videos/CDs, etc.," and "magazines." Moreover, in many cases in the current study, the objects of HD patients' interest and the bags or boxes containing such items were mixed and hoarded together. In addition, the reasons for hoarding depended on each item, and "future use" and "sentimental attachment" were the top two reasons for hoarding. Interestingly, Nordsletten et al. (2018) also reported a result similar to our study in that the majority of HD participants in London and Rio de Janeiro showed a belief that the items would be useful in the future or had sentimental attachment. In conclusion, our study suggests that Japanese HD patients hoard similar items for reasons similar to those of previous studies in Western countries.

# 4.2. Demographic characteristics and psychiatric profiles

The trend of early onset and long disease duration in the current HD group seems to suggest that HD is a disease with a chronic course. Regarding the chronicity of HD, recent studies also showed that the

prevalence of HD and hoarding symptom severity increased with age (Ayers et al., 2010; Cath et al., 2017). These findings including the current study suggested that HD patients rarely show spontaneous remission or improvement due to treatment (Nakao and Kanba, 2019).

We found a significantly higher rate (approximately 50%) of the symptoms of hoarding in first-degree relatives in the HD group compared to the OCD group. Conversely, previous studies on OCD reported that the proportions of definite and subclinical OCD in first-degree relatives were approximately 16.0%–18.2% (Grados et al., 2001; Nestadt et al., 2000; Pauls et al., 1995). Our study and previous evidence suggested that HD is more likely to be familial than OCD. Although the current study did not directly evaluate the participants' first-degree relatives, we believe that our results are valuable because the clinical background data of Japanese HD patients has been very limited until now.

In addition to the familiality of HD, the current HD group had a significantly higher unmarried rate (approximately 60%) than the NC group. Similar to our research, a previous study reported the low marriage rate of hoarders, and interestingly suggested that the unmarried status of HD may be associated with more severe hoarding and worse outcomes of intervention (Kim et al., 2001).

Previous studies reported poor insight in patients with hoarding (Frost and Hartl, 1996; Matsunaga et al., 2002; Samuels et al., 2007; Tolin et al., 2012b). However, a recent transcultural study by Nordsletten et al. (2018) reported that the proportion of poor insight was not significantly higher in three cities (London, Fukuoka, and Rio de Janeiro) other than Barcelona and suggested that the reason for this was that patients with poor insight were less likely to participate in such a study in reality. Also, most of the participants in the current HD group were recruited from medical institutions rather than the general population. Therefore, the reason why the proportion with poor insight in this HD group was not significantly higher than in the OCD group seems to be the same reason as the study by Nordsletten et al. In other words, the HD group in our study may include a considerable number of mildly to moderately severe patients with HD whose insights were retained to a certain extent.

The participants in the current HD group had high rates of comorbidities with other psychiatric disorders, which may affect the high proportion taking psychiatric medication. The top two comorbidities and their comorbid rate (MDD 56.7%, ADHD 26.7%) in the current HD group were similar to those of previous studies (Frost et al., 2011; Grisham et al., 2010; Tolin et al., 2018). It is noteworthy that the HD group in our study had a high rate of comorbidity with ADHD, similar to previous studies (Frost et al., 2011; Sheppard et al., 2010), despite not being recruited in medical institutions where many ADHD patients were treated. The current results on comorbidities are considered to support the possibility of a close association between ADHD and hoarding.

Most of the HD patients in our study were evaluated for and diagnosed with HD for the first time. Despite the trend of early onset, long disease duration, visiting psychiatric medical institutions with comorbid mental disorders, and taking psychiatric medication for such disorders, the patients in the current HD group had never been diagnosed with HD prior to this study. This fact suggests a low recognition of HD in the medical milieu in Japan. However, the Japanese mass media often discuss the topic of "trash houses (gomi-yashiki)," which have become a growing social problem due to bad smells, risks of fire and infection, and unpleasant appearances (Nakao and Kanba, 2019). We hypothesize that the cultural characteristics unique to the Japanese might be one of the important factors of hoarding. Especially, the spirit expressed in the word "mottainai" has been frequently used by Japanese to mean that it is good to cherish things and not to throw them away. In addition, aging of the population, called a "super-aging" society, and a decreasing marriage rate in Japan could be other important hoarding factors (Ayers et al., 2010; Cath et al., 2017; Kim et al., 2001; Nomura et al., 2019). In future Japanese HD studies, it would be meaningful to explore the relationship between such Japanese cultural and social characteristics and HD.

# Table 4. Hoarding scales and other clinical variables.

	HD	OCD	р	NC	р
	(n = 30)	(n = 20)	HD vs. OCD	(n = 21)	HD vs. NC
SI-R-J, median (IQR)	65.0 (57.8–73.3)	16.5 (11.3–26.5)	< 0.001	12.0 (7.5–25.5)	< 0.001
CIR, median (IQR)	4.5 (3.2–5.8)	1.5 (1.1–1.7)	< 0.001	1.0 (1.0–1.3)	< 0.001
Y-BOCS, median (IQR)	13.0 (0.0–18.0)	19.0 (16.3–23.5)	0.003	0.0 (0.0–0.5)	< 0.001
BDI-II, median (IQR)	26.0 (15.0-38.0)	18.0 (9.5–29.5)	0.20	4.0 (2.0–10.5)	< 0.001
AQ, mean (SD)	27.9 (9.5)	25.3 (8.0)	0.43	16.9 (6.1)	< 0.001
ASRS, median (IQR)	4.0 (3.0–5.0)	1.0 (0.0–2.8)	< 0.001	0.0 (0.0–1.5)	< 0.001
CAARS					
Inattention/Memory Problems, median (IQR)	74.0 (60.0–80.0)	53.0 (45.0-65.0)	< 0.001	47.0 (43.0–53.5)	< 0.001
Hyperactivity/Restlessness, median (IQR)	58.0 (53.0-72.0)	48.0 (42.0–56.5)	0.005	47.0 (41.0–54.0)	< 0.001
Impulsivity/Emotional Lability, median (IQR)	59.0 (46.0–78.0)	50.0 (38.0-63.5)	0.07	50.0 (43.5–54.5)	0.04
Problems with Self-Concept, median (IQR)	62.0 (52.0–70.0)	57.5 (50.0-71.3)	0.61	50.0 (46.5-56.0)	0.004
DSM-IV: Inattentive Symptoms, median (IQR)	63.0 (55.0-83.0)	50.0 (42.5-65.3)	0.009	50.0 (44.0-57.0)	< 0.001
DSM-IV: Hyperactive-Impulsive Symptoms, median (IQR)	61.0 (50.0-80.0)	47.0 (42.5–58.5)	0.005	47.0 (43.0–56.0)	0.001
DSM-IV: ADHD Symptoms Total, median (IQR)	72.0 (53.0–79.0)	50.5 (44.3-61.5)	0.003	49.0 (43.5–56.0)	< 0.001
ADHD Index, median (IQR)	70.0 (59.0–79.0)	57.0 (47.3-68.0)	0.05	49.0 (44.0–54.0)	< 0.001
GAF, median (IQR)	60.0 (50.0-60.0)	65.0 (60.0–70.0)	< 0.001	90.0 (90.0–90.0)	< 0.001

Abbreviations: HD, Hoarding Disorder; OCD, Obsessive-Compulsive Disorder; NC, Normal Control; SD, standard deviation; IQR, interquartile range; SI-R-J, Japanese version of the Saving Inventory-Revised; CIR, Clutter Image Rating; Y-BOCS, Yale-Brown Obsessive-Compulsive Scale; BDI-II, Beck Depression Inventory-Second Edition; AQ, Autism-Spectrum Quotient; ADHD, Attention-Deficit/Hyperactivity Disorder; ASRS, Adult ADHD Self-Report Scale; CAARS, Conners' Adult ADHD Rating Scales; DSM, Diagnostic and Statistical Manual of Mental Disorders; GAF, Global Assessment of Functioning.

# 4.3. Hoarding scales and other clinical variables

The current HD group showed higher scores on two hoarding scales (that is, the SI-R-J and CIR) than the original cut-off score for each scale, and these scores were equivalent to those in previously reported studies (Frost et al., 2004; Frost et al., 2008; Mataix-Cols et al., 2013; Tolin et al., 2010c). However, to the best of our knowledge, there has been no study that validates the cut-off score for these hoarding scales in Japanese HD patients. Thus, further studies with larger samples are needed to verify the assessment ability of the hoarding scales for a Japanese population including HD patients.

Our study showed that the scores on most of the ADHD symptom evaluation scales were significantly higher in the HD group than the OCD group. Similar to previous studies (Frost et al., 2011; Sheppard et al., 2010), this result suggested that HD shows a higher comorbid rate with ADHD compared to OCD.

The low GAF score in the current HD group suggests that the hoarding symptom causes serious damage to daily life. As previously discussed, the HD group in our study may be composed mainly of mildly to moderately severe HD patients. However, the OCD group in this study was possibly composed mainly of relatively severe OCD patients (see *Limitations*). Despite the possibility that the overall severity of the current HD group was mild to moderate, the fact that the GAF score in the HD group was significantly lower than that of the OCD group suggests that the hoarding symptom directly affects the patient's life. Therefore, we believe that HD is a disease requiring appropriate intervention and treatment as soon as possible without severe symptoms.

#### 4.4. Neuropsychological tests

Previous studies showed that HD may be characterized particularly by deficits in the areas of attention, memory, and executive function, although mixed results were reported in neuropsychological studies on hoarding (Grisham and Baldwin, 2015; Mataix-Cols et al., 2011; Woody et al., 2014). Therefore, we conducted neuropsychological tests as supplementary background data for the participants in the current study, but no significant difference was observed between the HD and NC groups in all of the neuropsychological tests. The neuropsychological abilities of the current HD group may be preserved because of the possibility that the severity of this HD group was mild to moderate. However, the results of the TMT part A and CA/PEN/TE of the KWCST in the current OCD group were significantly poorer than those of the HD group. The OCD group's attention and executive function detected by the TMT part A and the KWCST is suggested to decline due to the possibility of the population being skewed to relatively severe OCD patients. We believe that our results could be helpful for future studies on cognitive functions in HD and OCD.

#### 4.5. Limitations

There are three limitations to the current study. First, this study had a small sample size. However, to the best of our knowledge, research focused on Japanese HD patients is very limited. In addition, despite the small sample size, we successfully detected several statistically significant data. Further studies with larger sample sizes should be conducted to clarify the clinical characteristics of HD patients in Japan.

Second, there is a possibility that a selection bias occurred in the target groups. Most of the participants in the current HD group were recruited from the psychiatric department of Kyushu University Hospital and related organizations rather than the general population. Therefore, most of the participants had other psychiatric diseases and were already under treatment for those diseases, which seems to affect the high rates of comorbidities and psychiatric medications in the current HD group. In addition, several of the patients themselves saw the advertisements for our study and hoped to participate. Thus, the severity of the HD patients in this study who could take such action may be relatively mild, although they were on a chronic course. The current result that there were no significant differences between the HD and NC groups on the neuropsychological tests may be due to the selection bias of the HD group. However, the participants in the current OCD group were recruited from patients visiting a specialized OCD outpatient clinic at the Department of Psychiatry at Kyushu University Hospital, and it may be possible that recruitment focused on relatively severe OCD patients compared to general outpatient OCD patients. The recruitment problem in the current OCD group seems to affect the poor performance of the OCD group on the neuropsychological tests. Furthermore, the previously described selection bias of the HD and OCD groups in our study may affect the result that there was no significant difference in the degree of insight between the HD and OCD groups. However, this study is a valuable report of the very

limited clinical studies targeting Japanese HD patients, and thus we believe that our findings will contribute to future HD studies.

Third, there is a problem of the high rate of participants taking psychiatric medications in the current HD and OCD groups. Both the HD and OCD groups in our study had high comorbid rates of psychiatric diseases, and approximately 70% of the patients in the HD group and all of the participants in the OCD group were taking psychiatric medications. Especially for the OCD group, in addition to the possibility of the population being skewed to relatively severe OCD patients, the possibility that the high rate of participants taking psychiatric medications affected the poor performances on the neuropsychological tests may be considered. In contrast, despite the high rate of participants taking psychiatric medications in the current HD group, there was no significant difference in the neuropsychological tests compared with the NC group. From this result, it is suggested that the cognitive function detected by the neuropsychological tests in the current HD group was preserved because of the possibility that the severity of this HD group was biased toward relatively mild. In future cognitive function studies, it is desirable that participants should be taking as few psychiatric medications as possible.

#### 5. Conclusions

Our study revealed the clinical characteristics of Japanese HD patients. As a result, a clinical trend of Japanese HD patients similar to previous studies mainly in Western countries was suggested in many areas including early onset, a chronic course, familiality, high unmarried rate, and high rates of comorbidities such as MDD and ADHD. HD may be a universal disease with consistent clinical symptoms. Future HD studies should be conducted for a deeper understanding of the clinical characteristics including the relationship between Japanese cultural and social characteristics and HD.

# Declarations

#### Author contribution statement

Masumi Kuwano: Conceived and designed the experiments; Performed the experiments; Analyzed and interpreted the data; Wrote the paper.

Tomohiro Nakao: Conceived and designed the experiments; Analyzed and interpreted the data.

Koji Yonemoto: Analyzed and interpreted the data.

Satoshi Yamada, Keitaro Murayama, Kayo Okada, Shinichi Honda, Keisuke Ikari, Hirofumi Tomiyama, Suguru Hasuzawa: Performed the experiments.

Shigenobu Kanba: Conceived and designed the experiments.

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#### Competing interest statement

The authors declare no conflict of interest.

#### Additional information

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#### M. Kuwano et al.

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