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Olfactory Reference Syndrome

Its Relationship to Comorbidity of Social Anxiety Disorder and Obsessive-Compulsive Disorder

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Abstract: Olfactory reference syndrome (ORS) is known to have the clinical features of both obsessive-compulsive disorder (OCD) and social anxiety disorder (SAD). However, there has been no clear explanation as to why ORS has the characteristics of two different disorders. In the present study, the comorbidity rates of ORS in patients with SAD (without OCD, $n = 83$), ORS in patients with OCD (without SAD, $n = 42$), and patients with SAD and OCD comorbidity ($n = 17$) were compared. Of all 142 patients studied, 11 were diagnosed with ORS. The comorbidity rate of ORS in comorbid SAD/OCD group was significantly higher than those in both SAD and OCD groups. Logistic regression analysis of 100 cases of SAD and selected 69 cases of generalized SAD showed that the risk of ORS was significantly higher in patients with OCD and bulimia nervosa. Of 59 cases with OCD, the risk of ORS was significantly higher in patients with SAD. The results of the present study suggest that the comorbidity of SAD and OCD most likely explains the development of ORS.

Key Words: Olfactory reference syndrome, social anxiety disorder, obsessive-compulsive disorder, comorbidity

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Olfactory reference syndrome (ORS) is a condition in which individuals erroneously believe that they emit unpleasant, foul, or offensive body odor. Odors are often believed to originate from the mouth, genitals, or skin (Begum and McKenna, 2011; Feusner et al., 2010; Phillips and Menard, 2011; Pryse-Phillips, 1971). This belief is often accompanied by ideas or delusions of reference, that is, the belief that other people take special notice of the odor in a negative way, for example, rub their nose in reference to the odor or turn away in disgust. In Japan, ORS has been considered to be a variant of "*Taijin-kyofu-sho*," a diagnosis that has features in common with social anxiety disorder (SAD) (Kasahara, 1995; Suzuki et al., 2004). In fact, ORS and SAD share certain characteristics (Feusner et al., 2010; Phillips and Menard, 2011; Suzuki et al., 2004; Tada and Kojima, 2002). Patients with either disorder become anxious immediately upon exposure to social situations and try to avoid it, worrying about shame or embarrassment, and they have marked anticipatory anxiety. In fact, ORS is referred to social phobia (SP) in the section on "specific culture, age, and gender features" in the *Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition* (*DSM-IV*) (American Psychiatric Association, 1994).

On the other hand, symptomatic similarities between ORS and obsessive-compulsive disorder (OCD) have also been noted (Feusner et al., 2010; Stein et al., 1998; Veale and Matsunaga, 2014). For example, most individuals with ORS perform excessive, repetitive behaviors that could be characterized as compulsive, while thoughts about their odor being intrusive, repetitive, and irresistible could be characterized as obsessive. These symptomatic similarities between ORS and OCD have led to the idea that ORS is a variant of obsessive-compulsive spectrum disorders (Stein et al., 2016).

Currently, ORS is considered a universal syndrome recognized in various cultures, rather than a culture-bound syndrome found in East Asia, and is referred to in the *DSM-5* as a type of "other specified obsessive-compulsive disorders and related disorders (OCRDs)" (American Psychiatric Association, 2013). Although *DSM-5* does not give ORS a diagnosis code, the *International Classification of Diseases, 11th Edition* (*ICD-11*) (World Health Organization, 2019) classifies ORS as OCRDs and gives it a diagnostic code.

Thus, ORS is a unique syndrome that has features of both SAD and OCD. However, previous papers have not adequately explained why ORS resembles these two distinct mental disorders. I hypothesized that ORS is most likely to occur in cases of SAD/OCD comorbidity, in which case ORS would have features of both SAD and OCD. To test this hypothesis, I investigated the comorbidity of ORS in patients with SAD without OCD, OCD without SAD, and SAD/OCD comorbidity.

MATERIALS AND METHODS

All subjects were outpatients of Jimbocho Mental Health Clinic in Tokyo, and they were followed by the author for a relatively long time (mean follow-up period, 5.4 ± 4.0 years). The study sample was composed of 142 outpatients with a diagnosis of SAD without OCD ($n = 83$; mean age, 40.8 ± 10.8), OCD without SAD ($n = 42$; mean age, 40.0 ± 11.4), and SAD/OCD comorbidity ($n = 17$; mean age, 46.0 ± 12.4). Structured Clinical Interview for *DSM-IV* Axis I Disorders, Clinician Version (First et al., 2007) was used for the diagnosis of SAD and OCD, and other comorbid disorders. All patients were asked if they had a preoccupation of emitting body odor and other specific symptoms of ORS. The diagnosis of ORS was made when the patient was preoccupied by the belief that they emit an unpleasant, foul, or offensive body odor, which is not perceived by others, and the preoccupation caused clinically significant distress or impairment.

The cases which had comorbidity of schizophrenia and other delusional disorders were not included in this study.

The Liebowitz Social Anxiety Scale (LSAS) Japanese version (Asakura et al., 2002) was administered for SAD patients to assess the severity of the symptoms and used for statistical analysis. Yale-Brown Obsessive Compulsive Scale (YBOCS) Japanese version (Nakajima et al., 1995) was also administered for OCD patients to determine the nature of obsessions and compulsion and the severity of the symptoms.

Lifetime comorbidity rates of major depressive disorder (MDD), bipolar I or II disorders (BP), panic disorder with agoraphobia or without agoraphobia (PD), and bulimia nervosa (BN) in SAD without OCD, OCD without SAD, and comorbid SAD/OCD groups were compared. As other disorders were relatively rare, they were not included for statistical analysis. Age of onset of SAD, OCD, ORS, and other comorbid disorders were assessed retrospectively.

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Differences in demographic, clinical variables, and comorbidity rates between the 3 groups were compared by means of chi-square analysis for categorized variables and *t*-tests for continuous variables.

Logistic regression analysis was performed separately on 100 cases with SAD (generalized and nongeneralized), 69 cases with generalized SAD and 59 cases with OCD to examine any association between relevant independent demographic and clinical variables and ORS. The following factors were introduced as independent variables: sex, present age, age of onset of SAD and OCD, diagnoses of MDD, BP, PD, BN, and hereditary factors (diagnosis of mood disorder or schizophrenia in first-degree relatives of the patients).

Statistical significance was set at $p < 0.05$, and SPSS (SPSS 11.0, 2001) was used for all statistical analyses.

RESULTS

Clinical Features of ORS

Of the 142 patients who met the diagnostic criteria for SAD, OCD, or both, 11 (male = 3, female = 8) had typical ORS symptoms. All 11 cases complained of unpleasant odors coming from the mouth, armpits, or skin, which made people around them uncomfortable. They had repetitive behaviors such as repeatedly checking their own odor, repeatedly checking with family members to see if they could smell it, changing their clothes frequently, and showering excessively. Insight regarding the perceived body odor was variable. The degree of conviction was higher in the three patients who had seen a dermatologist or dentist before the psychiatric visit and in the one who had avoided interpersonal relationships for a long time due to ORS symptoms. In the remaining seven patients, the degree of conviction was not always high.

The diagnostic criteria for schizotypal personality disorder include symptoms that predispose to developing ORS, such as the idea of reference, and it is said that schizotypal personality disorder coexists in 10% of OCD cases (Attademo and Bernardini, 2021). However, none of the 11 patients with ORS symptoms met the diagnostic criteria for schizotypal personality disorder.

Onset of ORS

Ten of the 11 patients developed ORS (mean age of ORS onset, 19.7 ± 8.2) after an average of 7.6 ± 8.8 years after the onset of SAD. All SADs with ORS were generalized type. The temporal relationship between OCD and ORS varied: OCD and ORS occurred simultaneously in two cases, OCD preceded ORS in two cases, and ORS preceded OCD in two cases.

Reason for Visit

Of the 142 patients, 56 visited the clinic for depressive symptoms, 55 for SAD symptoms, and 15 for OCD symptoms. Of the 11 ORS patients, only two sought treatments for ORS symptoms in psychiatric clinics.

Demographic Features

Demographic features of the SAD without OCD, OCD without SAD, and comorbid SAD/OCD groups are shown in Table 1. The three groups did not differ significantly with respect to sex, present age, and hereditary factor. There was no significant difference in the age of onset of SAD between the SAD without OCD and the comorbid SAD/OCD groups. Likewise, there was no significant difference in the age of onset of OCD between the OCD without SAD and the comorbid SAD/OCD groups. In the comorbid SAD/OCD group, the percentage of generalized type (94.1%) was significantly higher than that in the SAD without OCD group (63.9%) ($p = 0.019$).

LSAS scores of the comorbid SAD/OCD group (76.4 ± 37.3) were significantly higher than that of SAD without OCD group (50.3 ± 22.3) ($p = 0.001$). YBOCS scores did not differ between OCD without SAD and comorbid SAD/OCD groups (Table 1).

Comorbidity

Table 2 shows the lifetime comorbidity of ORS and other disorders in SAD without OCD, SAD/OCD, and OCD without SAD. The comorbidity rate of ORS in comorbid SAD/OCD group (29.4%) was significantly higher than those in both SAD without OCD (6.0%) and OCD without SAD groups (2.4%) ($p = 0.003$, $p = 0.002$, respectively), but the comorbidity rate of ORS did not differ between SAD without OCD and OCD without SAD groups. The comorbidity rates of BP in both SAD/OCD and OCD without SAD groups (23.5%, 21.4% respectively) were significantly higher than that in SAD without OCD group (3.6%) ($p = 0.003$, $p = 0.001$, respectively). The comorbidity rates of MDD, PD, and BN were not different between the three groups.

Factors Associated With ORS Comorbidity

The results of the logistic regression analysis of variables were shown in Table 3. Of 100 cases with SAD, ORS was significantly associated with OCD ($p = 0.015$; adjusted odds ratio [aOR], 6.31; 95% confidence interval [CI], 1.43–27.87) and BN ($p = 0.012$; aOR, 11.92; 95% CI, 1.73–82.30). Because all SADs comorbid with ORS as generalized SAD, logistic regression analysis was performed for the 69 cases with generalized SAD, excluding nongeneralized SAD, as well. Results showed that, in 69 cases with generalized SAD, as in all SADs, ORS was significantly associated with OCD ($p = 0.026$; aOR, 6.09; 95% CI, 1.24–29.84) and BN comorbidity ($p = 0.008$; aOR, 18.21; 95% CI, 2.13–155.59).

Of 59 cases with OCD, SAD comorbidity was significantly associated with ORS ($p = 0.013$; OR, 17.08; 95% CI, 1.82–160.54).

Comorbidity of other disorders, MDD, BP, and PD were not associated with ORS in logistic regression analyses. Likewise, sex, present age, the onset of SAD or OCD, hereditary factor, LSAS, and YBOCS scores were not related with ORS.

DISCUSSION

The present study, which followed patients with SAD or OCD for a relatively long time, revealed that ORS mainly appears during the course of SAD and many years after the onset of SAD. Since only a few cases sought treatment due to ORS symptoms, long-term observations of patients are necessary to find ORS patients.

Previous studies have reported a 2%–19% comorbidity of OCD with SAD (Koyuncu et al., 2014; Schneier et al., 1992) and an 8%–42% comorbidity of SAD with OCD (Torres et al., 2006). In the present study, the percentage of OCD in SAD was 17%, and the percentage of SAD in OCD was 28.8%, which is consistent with previous studies.

Lochner and Stein (2014) reported that ORS was comorbid in 2 of 65 patients with SAD and 1 of 106 patients with OCD. Their study included patients with a primary diagnosis of SAD and OCD and used a cross-sectional evaluation, so the rate of ORS comorbidity is likely lower than in this study.

The most important finding of the present study was that the comorbidity rate of ORS was highest in comorbid SAD/OCD group compared with both SAD without OCD and OCD without SAD groups. This high rate of ORS comorbidity in cases of the SAD/OCD group is not likely to be explained by the additive effect of ORS comorbidities in SAD and OCD, and the lack of a significant difference in ORS comorbidity between the SAD without OCD and OCD without SAD groups in the present study is not consistent with the *DSM-5* and *ICD-11* view of ORS as most closely related to OCD.

Furthermore, logistic regression analysis showed that OCD comorbidity was significantly associated with ORS in all cases of SAD. Similarly, in cases of generalized SAD, OCD comorbidity was significantly associated with ORS. In all cases of OCD, comorbidity of SAD

TABLE 1. Demographic Features and Psychological Test Scores in SAD Without OCD, SAD/OCD, and OCD Without SAD

	A. SAD Without OCD (n = 83)	B. SAD/OCD (n = 17)	C. OCD Without SAD (n = 42)	A vs. B	B vs. C	A vs. C
Male/female	47/36	6/11	21/21	$p = 0.108$	$p = 0.304$	$p = 0.482$
Present age	40.8 ± 10.8	46.0 ± 12.4	40.0 ± 11.4	$p = 0.08$	$p = 0.08$	$p = 0.73$
Age of onset of SAD	19.4 ± 10.3	15.1 ± 9.2 (SAD)		$p = 0.113$		
Age of onset of OCD		22.4 ± 11.8 (OCD)	25.3 ± 11.8		$p = 0.392$	
Generalized: nongenrealized; % of generalized type	53:30; 63.9 (%)	16:1; 94.1 (%)		$^{**}p = 0.019$		
Heredity factor	12/83 (14.5%)	5/17 (29.4%)	9/42 (21.4%)	$p = 0.135$	$p = 0.513$	$p = 0.325$
LSAS	50.3 ± 22.3	76.4 ± 37.3		$^{**}p = 0.001$		
YBOCS		15.0 ± 8.0	19.1 ± 6.7		$p = 0.052$	

was also significantly associated with ORS. These results suggest that ORS is most likely to appear when SAD and OCD are comorbid.

On the other hand, ORS was not associated with MDD or BP, suggesting that ORS is not simply a symptom associated with depression or bipolar disorder. Note that BP comorbidity was higher in OCD without SAD and SAD/OCD comorbid groups than in SAD without OCD, but this will not be discussed here.

In addition, LSAS values that are related to SAD severity were not associated with ORS comorbidity. Thus, it is unlikely that the elevated severity of SAD leads to the manifestation of ORS.

Since the comorbidity rates of PD and BN were not significantly different among three groups, the high rate of ORS comorbidity in SAD/OCD group seems to be a specific phenomenon. Taken together the results of the present study suggest that the comorbidity of SAD and OCD most likely explains the development of ORS. If ORS appears primarily when SAD and OCD are comorbid, it is quite natural that ORS has phenomenal features common to both disorders and is not completely consistent with either one.

Phillips and Menard (2011) studied 20 patients with ORS and reported that 65% of them had SP, SP is considered almost equivalent to SAD in *DSM-IV,* and 30% had OCD. Although they did not investigate cases of SP and OCD comorbidity, it is noteworthy that ORS is more prevalent in SP than in OCD patients. Likewise, Lochner and Stein (2014) found more ORS in SAD patients than in OCD patients in their study.

Prazeres et al. (2010) examined 14 ORS comorbidities, and their table shows that there were 4 cases of OCD and SP comorbidity, 2 cases of SP without OCD, and 3 OCD without SP. This suggests that the comorbidity of SAD and OCD is an important factor in the development of ORS.

As with ORS, body dysmorphic disorder (BDD) has been assigned to the category of obsessive-compulsive and related disorders in *DSM-5*, and again, like ORS, BDD is known to have the clinical features of both SAD and OCD (Fang and Hofmann, 2010; Kelly et al., 2013; Phillips et al., 2007). Phillips et al. (2007) compared the clinical characteristics of BDD without OCD, OCD without BDD, and comorbid cases of OCD and BDD. As they showed the comorbidity rates of SP in each group, the comorbidity rates of BDD in OCD without SP and comorbid SP/OCD groups could be known. The values calculated from their table are as follows: the comorbidity rate of BDD in OCD without SP group was 11.6% and in comorbid SP/OCD group was 26.0%, respectively.

The same group of investigators showed the comorbidity rates of OCD in SP without BDD, BDD without SP, and comorbid cases of SP and BDD in a different study (Kelly et al., 2013). From their table, the comorbidity rate of BDD in SP without OCD group was 12.8%, and in the comorbid SP/OCD group, the rate was 33.3%. Conceição Costa et al. (2012) compared the comorbidity of other mental disorders in OCD with and without BDD in a large OCD sample. Calculations from their table showed that the comorbidity rate of BDD in OCD without SAD was 6.7% and 21.9% in comorbid SAD/OCD groups, respectively. All these three studies showed that the comorbidity rate of BDD in SAD/OCD comorbid patients seems to be higher than the additive comorbidity of BDD in each disorder. Thus BDD, like ORS, may be a syndrome that is more likely to appear when SAD and OCD coexist. However, it requires future research to know whether BDD is the disorder that is most likely to emerge when SAD and OCD coexist.

Previous studies have been attempting to classify ORS into one category, and the results were still confusing (Feusner et al., 2010; Phillips and Menard, 2011). The results of this study suggest that ORS does not necessarily need to be classified into one category. Rather, ORS is better described as a group of symptoms that often appear when two different mental disorders are combined.

In the field of medical diagnosis, a special condition called "silent ischemia or asymptomatic myocardial infarction" is well known (Cohn et al., 2003). When a diabetic patient with peripheral neuropathy has a myocardial infarction, the symptoms of heart failure occur without the strong chest pain that is characteristic of acute myocardial infarction. In other words, the coexistence of the two conditions results in a different symptomology.

We should pay more attention to the specific symptoms that appear against the background of the coexistence of different mental disorders. To the best of our knowledge, there were no studies that compared the comorbidity rates of ORS among patients with SAD without OCD, OCD without SAD, and comorbid SAD/OCD patients. This method is essential to know the clinical condition in which ORS develops.

The theory of the inference-based approach (IBA) seems to successfully explain the mechanism by which ORS symptoms emerge

TABLE 2. Life Time Comorbidity of ORS and Other Disorders in SAD Without OCD, SAD/OCD, and OCD Without SAD

	A. SAD Without OCD (n = 83)	B. SAD/OCD (n = 17)	C. OCD Without SAD (n = 42)	A vs. B	B vs. C	A vs. C
ORS	5/83 (6.0%)	5/17 (29.4%)	1/42 (2.4%)	$^{**}p = 0.003$	$^{**}p = 0.002$	$p = 0.368$
MDD	38/83 (45.8%)	10/17 (58.8%)	20/42 (47.6%)	$p = 0.327$	$p = 0.436$	$p = 0.846$
BP	3/83 (3.6%)	4/17 (23.5%)	9/42 (21.4%)	$^{**}p = 0.003$	$p = 0.860$	$^{**}p = 0.001$
PD	7/83 (8.4%)	2/17 (11.8%)	8/42 (19.0)	$p = 0.662$	$p = 0.499$	$p = 0.085$
BN	4/83 (4.8%)	2/17 (11.8%)	6/42 (14.3%)	$p = 0.272$	$p = 0.499$	$p = 0.065$

TABLE 3. Logistic Regression Analysis of Variables Associated With ORS

Variable	*p*		95% CI
Univariate Logistic Regression Analysis of Variables Associated With ORS for SAD (Generalized and Nongeneralized SAD, *n* = 100)			
		Odds Ratio	
Sex	0.138	0.34	0.08–1.41
Present age	0.299	1.03	0.97–1.10
SAD onset	0.114	1.09	0.98–1.20
OCD	0.008**	6.50	1.63–25.83
MDD	0.156	2.79	0.68–11.47
BP	0.114	4.25	0.71–25.53
PD	0.219	2.96	0.53–16.74
BN	0.005**	12.43	2.10–73.39
Hereditary factor	0.260	2.33	0.54–10.10
LSAS	0.010	0.97	0.95–0.99
From the variables selected in the univariate regression analysis, multivariate regression analysis was performed and odds ratios were adjusted			
		aOR	
OCD	0.015**	6.31	1.43–27.87
BN	0.012**	11.92	1.73–82.30
Univariate logistic regression analysis of variables associated with ORS for SAD (generalized SAD, *n* = 69)			
		Odds Ratio	
Sex	0.270	0.44	0.10–1.88
Present age	0.564	1.02	0.96–1.09
SAD onset	0.351	1.05	0.96–1.15
OCD	0.039*	4.37	1.074–17.73
MDD	0.409	1.84	0.43–7.81
BP	0.119	4.67	0.67–32.36
PD	0.191	3.44	0.54–21.91
BN	0.012*	12.21	1.73–86.18
Hereditary factor	0.266	2.38	0.52–10.97
LSAS	0.145	0.98	0.96–1.01
From the variables selected in the univariate regression analysis, multivariate regression analysis was performed and odds ratios were adjusted			
		aOR	
OCD	0.026*	6.09	1.24–29.84
BN	0.008**	18.21	2.13–155.59
Univariate logistic regression analysis of variables associated with ORS for OCD (*n* = 59)			
		aOR	
Sex	0.523	0.56	0.09–3.32
Present age	0.460	0.98	0.91–1.05
OCD onset	0.452	1.03	0.95–1.12
SAD	0.013*	17.08	1.82–160.54
MDD	0.965	0.96	0.18–5.21
BP	0.101	4.30	0.75–24.54
PD	0.984	0.98	0.10–9.40
BN	0.815	1.31	0.13–12.98
Hereditary factor	0.130	3.82	0.68–21.58
YBOCS	0.439	1.06	0.91–1.24

(Julien et al., 2016). According to IBA, OCD is a disorder of the imagination that is characterized by pathological doubt. In this model, obsessions are not normal intrusions but rather occur in inappropriate contexts. IBA considers that obsessional doubt is created through a reasoning process termed inferential confusion, where an internal narrative leads an individual to distrust the senses and invest in a remote possibility rather than reality. The narrative is hypothesized to be coherent with vulnerable self-themes, which could explain the selective nature of OCD. SAD has vulnerabilities in the context of interpersonal relationships, including low self-esteem and sensitivity to criticism and rejection. In patients with comorbid SAD and OCD, the doubt "What if I odor?" that is based on SAD's vulnerability to self may arise. This is followed

by characteristic avoidance and compulsive behaviors associated with ORS.

The results of logistic regression in cases of SAD showed that BN was significantly associated with ORS, which means BN comorbidity may be related to the development of ORS in SAD patients as well as OCD comorbidity. From family studies of eating disorders, including BN, it is proposed that they are a phenomenal variant of OCD (Bellodi et al., 2001). However, it is still unclear why BN comorbidity is associated with ORS in SAD.

In this study, only a few ORS patients visited a medical institution for the treatment of their odor, the rest were those who complained of ORS symptoms during treatment for SAD. Thus, the cases in this study may be less severe than the cases reported in the past. In severe cases, other mechanisms may better explain the development of ORS.

This was a relatively small study, conducted in one region and at a single institution. Future studies should be conducted across different cultures, at multiple institutions, and with a larger number of patients to obtain reliable statistical data.

CONCLUSIONS

The reasons why ORS has features of both SAD and OCD are not fully understood. This study is characterized by its relatively long-term observation of patients with SAD and OCD. Among the 142 patients studied, 11 ORS patients were found, and most of them had ORS symptoms during the course of SAD. Statistical analysis showed that the most relevant factor for the diagnosis of ORS was the coexistence of generalized SAD and OCD. The idea that psychiatric comorbidity produces specific psychiatric symptoms has not been previously thought of. Therefore, it is significant that the present study found that the coexistence of SAD and OCD is associated with ORS.

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DISCLOSURE

The author declares no conflict of interest.

This study was approved by the Ethics Committee of Nihon University Hospital in November 2013 (approval number 131103).

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