Olfactory Reference Syndrome: A Separate Disorder Or Part Of A Spectrum

This article provides a narrative review of the literature on olfactory reference syndrome (ORS) to address issues focusing on its clinical features. Similarities and/or differences with other psychiatric disorders such as obsessive-compulsive spectrum disorders, social anxiety disorder (including a cultural syndrome; taijin kyofusho), somatoform disorders and hypochondriasis, delusional disorder are discussed. ORS is related to a symptom of taijin kyofusho (e.g. jikoshu-kyofu variant of taijin kyofusho) Although recognition of this syndromes more than a century provide consistent descriptions of its clinical features, the limited data on this topic make it difficult to form a specific diagnostic criteria. The core symptom of the patients with ORS is preoccupation with the belief that one emits a foul or offensive body odor, which is not perceived by others. Studies on ORS reveal some limitations. Although there is a lack of data for validators, including symptom profile; familial aggregation; environmental risk factors; cognitive, emotional, temperament and personality correlates; biological markers; patterns of comorbidity; course of illness: and response to treatment, current data suggest that ORS appears different form other disorders, but, this difference is not enough to put this syndrome in a separate diagnosis., in DSM-5, ORS has its place in the “Other Specified Obsessive-Compulsive or Related Disorder” category, in which, this category is for patients who have symptoms characteristic for obsessive-compulsive and related disorder but do not meet the full criteria for any specific obsessive-compulsive or related disorder. Further studies are needed for understanding the nature, prognosis, treatment and morbidity.

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INTRODUCTION:
Olfactory reference syndrome (ORS) is a condition in which individuals erroneously believe that they emit an unpleasant, foul, or offensive body odor. Odors may include almost anything foul smelling and are often believed to originate from an organ or system, including the mouth, genitals, rectum, or skin (APA, 2000). In clinical settings, common specific concerns include halitosis, genital odor, sweat, flatulence or anal odor (Philips KA et al., 2006). Uncommonly, patients have concerns on emitting non-bodily odors such as ammonia (Tilley H, 1895), detergent (Ross CA, 1987), burned rags (Harriman, 1934), or rotten onions (Sutton, 1919). Usually, the belief of emitting an odor is often accompanied by ideas or delusions that the odor is noticeable to other people and they will react negatively, for example, by rubbing their nose in reference to the odor or turn away in disgust. Repetitive behaviors of smelling themselves, showering excessively and attempting to mask the odor are performed by many patients (Pyrs-Philips, 1971).

History

Fig.1: Olfactory reference syndrome has some specific symptoms for obsessive compulsive and related disorders but does not meet full criteria for any disorder in DSM-V

ORS through DSM and ICD Classifications
In both the Diagnostic and Statistical Manual of Mental Disorders, 4th. Edition (DSM-IV) and the International Classification of Disease (ICD), the term ORS is not mentioned or included as a separate disorder. However, ORS symptoms are
considered as delusional disorder, somatic type in DSM IV (Potts CS, 1891); “Somatic delusions can occur in several forms. Most common are the person’s conviction that he or she emits a foul odor from the skin, mouth, rectum, or vagina….” (Potts CS, 1891). Similarly, ICD-10’s text on persistent delusional disorders notes that delusions may “express a conviction that…others think that he or she smells…” (Philips KA, 2007). In DSM-IV, ORS is also referred to as a type or part of social anxiety disorder (social phobia), noting that persons with social phobia may fear to offend with their body odor. Under the Cultural note for ORS in DSM-IV: “taijin kyofusho”.

“In certain cultures (e.g., Japan and Korea), individuals with Social Phobia may develop persistent and excessive fears of giving offense to others in social situations…. These fears may take the form of extreme anxiety that blushing, eye-to-eye contact, or one’s body odor will be offensive to others. Taijin kyofusho, is similar to social phobia in Japan and Korea (Suzuki K, 2004).

Although its clinical features are confusingly mentioned in three different sections of DSM-IV, where they even are not adequately described, for more than a century, ORS has been stated as a discrete syndrome in many cultures. However, given the suffering and impairment associated with it, the term ORS (currently the most widely used term for this syndrome) is still not explicitly mentioned. The questions whether ORS should be mentioned as a part of other disorder such as delusional disorder or social phobia, or should be taken as a separate diagnosis with its own set of diagnostic criteria, remain unclear.

**Statements on ORS through DSM and ICD**

DSM-III-R: “Convictions that the person emits a foul odor…are one of the most common types of delusional disorder, somatic type” (APA, 1987).

DSM-IV: “Somatic delusions can occur in several forms. Most common are the persons’ conviction that he or she emits a foul odor from the skin, mouth, rectum or vagina…” (APA, 2000).

ICD-10: “Express a conviction that…othersthink that he or she smells…” (WHO, 1992).

**Clinical Features of ORS**

**Perceived Odors**

Unpleasant, foul or offensive body odors include almost anything bad smelling and are often believed to originate from an organ or the system (APA, 2000). Consistent with prior reports, ORS patients most commonly focus on bodily smells, such as general body odor, halitosis, genital odor and flatulence/rectal odor. All subjects report at least one odor that may normally be emitted from their bodies (Pryse-Philips W, 1971)(Iwu CO, 1990)(Osman AA, 1991)(Philips KA, 2006)(Begum M, 2010). Rarely, some patients reported urine, sperm, sweat, armpit odor or malodorous from hands and feet (Tee CK, 2014)(Pryse-Philips W, 1971)(Iwu CO, 1990)(Osman AA, 1991). Occasional odors that patients reported are said to resemble non-bodily smells, such as ammonia, detergent, burned rags, candles or rotten onions (Tilley H, 1895) (Sutton RL, 1919)(Harriman PL, 1934)(Ross CA,1987) (Begum M, 2010). Emitting body odor that may smell like rotting fish is reported by the patients with an uncommon metabolic disorder, which is also known as fish malodor syndrome: trimethylaminuria (Mitchell SC, 2001).

**Odor Hypersensitivity or Misinterpretation**

Patients with temporal lobe epilepsy may have complaints of smelling foul odors. Olfactory sensations caused by pituitary tumors may irritate the hippocampus locally, so that foul odors “arise”. ORS symptoms differ from symptoms of other disorders that may cause olfactory hallucinations, including migraine headaches, head injury, intranasal disorders, consisting of a typical bodily odor that emanates from the sufferer; being persistent rather than brief and not being accompanied by other auras, typical of temporal lobe epilepsy (APA-2014) (Pryse-Philips W, 1971) (Acharya V, 1998)(Chen C, 2003). On the other hand, frontal, ethmoidal or sphenoidal sinus inflammations can be causes of subjective sense of offensive odors. Olfactory reference syndrome is characterized by the false belief of patients that he or she has a foul body odor that is not actually perceived by others. The
idea is purely based on their misinterpretation of other people’s behavior (e.g. opening windows or nose rubbing) (APA, 2014) (Alvarez WC, 1958)(Marks IM, 1988)(Iwu CO, 1990). However, a recent study suggests that it is unclear whether such patients are hypersensitive to normal body odors (which they consider noxious or offensive) or whether they experience an olfactory hallucination (Philips KA, 2011).

**Referential Thinking**

In ORS patients, referential thinking involves misinterpreting the meaning of other people’s comments (e.g. about an odor), gestures (e.g. touching their nose), or other behaviors (e.g. clearing their throat, opening a window, or looking or moving away from the patient) (Philips KA, 2006). Nearly all subjects (88%) in a recent study reported lifetime ideas or delusions of reference (Philips KA, 2011). These findings are compatible with Pryse-Phillips’ finding in the early seventies (97%) (Pryse-Phillips W, 1971).

**Repetitive Behaviors**

Preoccupation of patients leads to repetitive behaviors, such as washing the body or changing clothes (APA, 2014). The content of some ORS behaviors is also similar to that of compulsions that may occur in Body Dismorphic Disorder (BDD) and/or Obsessive Compulsive Disorder (OCD) - e.g. repetitive checking, excessive showering and excessive clothes changing. Similar in form to those of OCD and BDD, excessive repetitive behaviors of ORS patients are performed and intended to eliminate, check, obtain reassurance about or mask the perceived odor (i.e. camouflaging) (Prazeres AM, 2010). They spend time with thoughts about their odor and engaging behaviors to check or minimize. These behaviors are usually to check or eliminate odor that is perceived, to obtain reassurance about it, and to prevent others from smelling it. Checking their body for odor; excessive showering or other washing; or repetitive use of deodorant, mouthwash, or perfume are some examples for patients with ORS (Pryse-Phillips W, 1971) (Bishop ER, 1980) (Marks I, 1988) (Malasi TH, 1990). Repetitive behaviors are the result of shame and embarrassment, referential thinking and time consuming preoccupations to eliminate the odor they perceive. However, some ORS repetitive behaviors appear unique to ORS; for example, neither BDD nor OCD involve sniffing ones’ underwear, excessively laundering ones’ clothes or camouflaging their with mints. In a recent study, nearly all subjects (95%) were found to perform at least one excessive repetitive behavior. Moreover, camouflaging behavior was found in all patients with ORS? (Philips KA, 2011).

**Insight**

The ORS patient may have good, fair, poor or absent insight into the behavior (APA, 2014). Previous findings suggest that most of the patients have delusional beliefs, while some of the patients have poor insight. However, no prior study assessed insight in ORS using a reliable and valid measurement (Bishop ER, 1980) (Malasi TH, 1990)(Osman AA, 1991)(Suzuki K, 2004) (Begum M, 2010)(Philips KA). In addition, only 21% of patients with delusional beliefs are reported (Prazeres AM, 2010), and findings suggest that although ORS beliefs are often delusional, ORS should not be classified as delusional disorder (Philips KA, 2011).

**Age at Onset, Chronicity and Gender Differences**

Literature reports indicate the mean age of ORS onset is in the early or mid twenties (Philips KA, 2006) (Begum M2010), but the text accepts the mean age at onset of 25 years of age (APA, 2014). Most reports found that ORS symptoms were usually chronic (Pryse-Phillips W, 1971)(Philips KA, 2011). Follow-up of patients over two years demonstrated no changes in the symptoms (Pryse-Phillips W, 1971). The syndrome is predominant in males and in singles (APA, 2014)

**Functioning/Disability**

Data indicate that ORS causes clinically significant limitations in functioning, distress and significant social disability (Pryse-Phillips W, 1971). Strikingly, rate of socially active patients with ORS is very low. Many individuals are socially isolated (Pryse-Phillips W, 1971). Shame, embarrassment, and/or concern about offending others with their odor causes prominent social avoidance, isolation and impairment of work or school functioning (Pryse-Phillips W, 1971)(Bishop ER, 1980)(Davidson M,
Comorbidity

Comorbidity of other mental disorders with ORS is reported. Major depressive disorder (MDD) was the most commonly reported as secondary to ORS, (Phillips KA, 2006)(Prazeres AM, 2010). In a study, nearly three quarters of the sample was considered to have depressive symptoms primarily due to ORS (Phillips KA, 2011). Lifetime substance use disorder is found in nearly half of the patients. Social anxiety disorder, OCD and BDD were also encountered frequently. Although ORS seems to have important differences, high comorbidity with these disorders questions whether ORS is related with them (Stein DJ, 1998)(Lochner C, 2003)(Phillips KA, 2006)(Feusner JD, 2010).

Suicidality

In a study by Pryse-Phillips’ on 36 subjects, 43% experienced “suicidal ideas or action” and 5.6% committed suicide during the follow-up period. The author states that the suicides were attributable to ORS (Pryse-Phillips W, 1971). Additionally, the literature does not provide any evidence or suggestion that ORS is a result of common stressors or losses, or a culturally sanctioned response to a significant/particular event. Lifetime suicidal ideation and suicidal attempts are reported very high among ORS patients (Prazeres AM, 2010).

Treatment Seeking by Patients

Non-psychiatric, medical, surgical or dental treatments are ineffective in all cases of ORS patients (Phillips KA, 2011). As reported in the literature, patients consult dentists, surgeons, and ear-nose-throat specialists for supposed halitosis; proctologists, surgeons, and gastroenterologists for supposed anal odors; and other physicians such as dermatologists and gynecologists without effective results (Phillips KA, 2006; Prazeres AM, 2010). Non-psychiatric consultations and/or treatments appear usually ineffective and are therefore associated with patient dissatisfaction (Forte FS, 1952) (Pryse-Phillips W, 1971) (Iwu CO, 1990) (Prazeres AM, 2010).

Treatment Results

Psychotropic medications are used by many patients but it is not clear whether these drugs are used for ORS or for the comorbid disorders (Phillips KA, 2011). Case series and anecdotal reports suggest that serotonin-reuptake inhibitor (SRI) monotherapy, or combination with antipsychotics, or an antipsychotic monotherapy may all be effective treatments (Beary MD, 1981) (Marks I, 1988) (Malasi TH, 1990) (Osman AA, 1991) (Gomez-Perez JD, 1994) (Dominquez RA, 1997) (Stein DJ, 1998) (Kobayashi T, 2005) (Phillips KA, 2006) (Feusner JD, 2010) (Prazeres AM, 2010). However, although ORS beliefs are often delusional, treatments with SRIs were found more efficacious than with antipsychotics (Phillips KA, 2006) (Begum M, 2010) (Phillips KA, 2011). Limited data show that behavioral approaches, consisting of exposure to avoided social situations and ritual prevention, may be efficacious as well (Phillips KA, 2006) (Begum M, 2010). However, treatment research is extremely limited for ORS (Phillips KA, 2011).

Relation with Obsessive-Compulsive Spectrum Disorders

Although ORS may be related to one or more of the disorders mentioned, in DSM, it is proposed to be an obsessive-compulsive spectrum or an anxiety disorder. However, lack of quality research and the inexistence of direct comparison studies between ORS and obsessive-compulsive spectrum disorders, makes this disorder to be unique in DSM (Phillips KA, 2011).

Conflicts

In many cases, the response to psychotropic agents or to behavioral therapy suggest that ORS has many features of an internalizing disorder, rather
than reflecting a social deviance or conflict. But in the literature, this exact evidence or suggestion is not pointed out as a result of social deviance, other conflicts with society.

**Awareness**

Many patients with ORS do not seek psychiatric treatment at all, which may be due to the lack of public awareness that these symptoms represent a treatable entity. Usually, ORS patients visit non-mental health professionals such as dentists, gastroenterologists, dermatologists, or gynecologists, who may not be aware that ORS is a known form of mental illness. But also in psychiatric settings, many patients with ORS receive no diagnosis or an inaccurate diagnosis or even misidentification may occur.

**Differential Diagnosis: Similarities and Boundaries with Other Psychiatric Disorders**

**Social Anxiety Disorder**

Some clinical features of ORS appear to be common with social anxiety disorders. In Japan and Korea, ORS is considered to be a form of *taijin kyofousho*, which is a culturally bounded syndrome. Individuals with *taijin kyofousho* fear that their body or bodily functions embarrass, displease or be offensive to others; in terms of facial expressions, odor, appearance, or movements (APA, 2000). One of the several fears of individuals with *taijin kyofousho* is emitting body odor (17%) (Matsunaga H, 2001). Most individuals with ORS are concerned about the social implications of emitting a foul odor, with patients commonly experiencing shame, embarrassment, and anxiety in social situations, as well as avoidance of social situations (Bourgeois M, 1972) (Lochner C, 2003). Comparison of individuals with ORS to those with social anxiety disorder found similarities in demographics and also a comorbidity with depression. However, the key characteristic of social anxiety disorder is rather different: patients with social anxiety disorder have fear that they will act in a way that will be embarrassing or humiliating. Thus, social anxiety patients are typically primarily concerned about their actions or how they speak, eat or write etc., rather than how they smell. Most individuals with ORS perform excessive, repetitive behaviors that are compulsive, which is apparently different from patients with social anxiety disorder. These behaviors are usually to check or eliminate odor that perceived, to obtain reassurance about it, and to prevent others from smelling it.

**Obsessive Compulsive Disorder**

The repetitive behaviors observed in cases of ORS raised question whether ORS is related to Obsessive Compulsive Disorder (OCD). Individuals with ORS usually report repetitive, troubling and intrusive thoughts about their “odor,” which some describe as obsessive (Alvarez, 1958) (Hawkins C, 1987) (Osman AA, 1991). Similar with OCD patients, ORS patients may spend many hours per day being preoccupied with these thoughts (Hawkins C, 1987). Other than that, the Serotonergic drug response of ORS patients is an additional similarity with OCD (Domínguez RA, 1997) (Stein DJ, 1998) (Lochner C, 2001). However, in contrast to ORS, presence of delusional belief is less common in OCD (Insel TR, 1986) (Kozak MJ, 1994) (Eisen JL, 1999).

**Body Dysmorphic Disorder**

ORS’s clinical features have many similarities to body dysmorphic disorder (BDD); the primary symptoms of both disorders involve a belief of a bodily defect which leads to anxious avoidance of relevant (often social) situations (Lochner C, 2003). Preoccupation and repetitive behaviors to check or remediate the perceived problem are other similarities (Bishop ER, 1980) (Beary MD, 1981) (Davidson M, 1982) (Brotman AW, 1984) (Marks I, 1988) (Phillips KA, 2006). Both ORS and BDD are characterized by frequent seeking of medical treatment in an attempt to alleviate the symptoms (e.g. treatment from dentists or gastroenterologists in ORS, surgery or dermatologic treatment in BDD) (Bishop ER, 1980) (Davidson M, 1982) (Iwu CO, 1990) (Malasi TH, 1990) (Osman AA, 1991).

**Somatoform Disorders and Hypochondriasis**

There are also some apparent similarities to other somatoform disorders, primarily to hypochondriasis. Although both disorders involve preoccupation with the body, they are often
marked by obsessional thinking and include repetitive behaviors such as checking and seeking medical diagnoses and treatments (APA, 2000). In hypochondriasis, the core symptom is the fear about having a serious disease, whereas in ORS, ideas or delusions of reference and social avoidance characteristically are prominent.

**Delusional Disorder**

Reports suggest that beliefs in ORS may not always be delusional and in such cases does not meet the criteria for delusional disorders. In addition, reports of pharmacotherapy treatment responses make ORS different from a delusional disorder. Some reports describe improvement/response to antipsychotics (Riding J, 1975) (Osman AA, 1991), while some show response to serotonin reuptake inhibitor (SRI) (Stein DJ, 1998) (Lochner C, 2003) (Kobayashi T, 2005). Others seem to respond to tricyclic antidepressants (TCA) (Brotman AW, 1984) (Fernando N, 1988), or to combinations (Malasi TH, 1990) (Osman AA, 1991) (Luckhaus C, 2003). Depressive episodes, with or after ORS, are more common than in delusional disorders. Patients with ORS often have prolonged depressive episodes (Pryse-Phillips W, 1971) (Malasi TH, 1990). Depressive episodes are often considered secondary to ORS. In the majority of cases, these episodes appear after the development of odor concerns (Pryse-Phillips W, 1971).

In a study to assess delusionality or insight of ORS beliefs, most of ORS (84.6%) patients had delusional ORS beliefs, less (15.4%) had non-delusional beliefs (Philips KA, 2011). The belief of a subjective sense of smell that does not exist externally may rise to the level of a somatic delusion, in which case a diagnosis of delusional disorder should be considered. The syndrome has been well documented in the psychiatric literature, usually classified as a delusion of perception. Whether or not it deserves a special diagnostic category is open to question (APA, 2014).

**CONCLUSION**

Finally, in DSM-5 so far, ORS has been included in the Other Obsessive-Compulsive or Related Disorder category. This category is for patients with obsessive-compulsive and related disorder-specific symptoms, but who do not fully meet the criteria for any specific obsessive-compulsive or related disorder.

This diagnosis is appropriate under three situations: (1) an atypical presentation, (2) another specific syndrome not listed in DSM-5, or (3) the information presented is insufficient to make a full diagnosis of an obsessive-compulsive or related disorder. In assessing a patient with olfactory reference syndrome, it is important to exclude somatic causes. (APA, 2014)

In the psychiatric literature, ORS has been described in multiple regions of the world and it has long been recognized as a discrete syndrome that occurs in individuals. Data indicate that ORS causes significant limitations in functioning or distress and significant social disability (Pryse-Phillips W, 1971). Although the exact underlying mechanism of ORS is unclear, preliminary (uncontrolled) reports of improvement in ORS with pharmacotherapy or psychosocial treatment (behavioral therapy - Marks I, 1988; cognitive-behavioral therapy - Bizanzer AN, 2008; paradoxical intention - Milan MA, 1982) indicate indirect results of support to the existence of disturbances and psychobiological processes in ORS.

As seen in the literature, studies on ORS reveal some limitations, including relatively small sample groups, lack of control-comparison groups, non-psychiatric medical reports, medical conditions, seizure histories, efficacy of psychotropic medications on ORS and each of the ORS symptoms, use of standardized measures. Further studies are needed for understanding the nature, prognosis, treatment and morbidity.

For validators, there is a lack of data containing the symptom profile; family union; environmental risk factors; cognitive, emotional, temperament and personality relations; biological markers; patterns of comorbidity; The course of the disease: and in response to treatment, available data indicate that ORS appears to be different from other disorders such as social anxiety disorder, BDD, OCD, hypochondriasis or delusional
disorder. Differences in terms of symptom profile, comorbidity, and response to treatment can be identified.

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