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Case Report

A Novel Perspective on Olfactory Reference Syndrome and Associated Specified Obsessive-Compulsive Disorders

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Abstract

In the DSM-V, Olfactory Reference Syndrome (ORS) is categorized as a specified obsessive-compulsive and related disorder characterized by fear of having an offensive body odor (also termed Jikoshu-kyofu: a variant of taijin kyofusho). Anxiety and body dysmorphia are often present in this disorder and patients frequently believe that others are negatively responding to them and their odor. Originally described by Pryse-Phillps in 1971, the underlying etiology of ORS remains highly controversial. Like many disorders, patients with ORS and associated disorders have not been tested for olfactory dysfunction, even though such dysfunction could be associated with their symptoms. In this paper, we describe four patients with ORS and associated disorders who had their olfactory function measured using a well-validated psychophysical test. All were found to have genuine smell impairment analogous to that commonly seen in non-ORS cases of dysosmia or phantosmia secondary to damage to the olfactory epithelium. These novel observations beg the question as to whether many patients with ORS actually have objective alterations in their smell perception, which may play a role in or underlie how their obsessive-compulsive disorders manifest. Our findings shed entirely new light on this poorly understood syndrome and provide novel alternatives for patient management and therapy.

Keywords: Depression; Olfactory Reference Syndrome; Olfaction; Dysosmia, Phantosmia; UPSIT

Introduction

It is well established that olfactory hallucinations, usually of an unpleasant character, can be a sign of epilepsy, migraine, schizophrenia, and other serious medical conditions [1,2]. In 1971, Pryse-Phillips identified a cohort of patients who experienced olfactory hallucinations of unpleasant body odor and who simultaneously held the belief that people were reacting negatively towards them because of it. He named this set of symptoms the "Olfactory Reference Syndrome" (ORS) because he felt that these symptoms were distinct from those related to their diagnoses of schizophrenia, depression, and paranoid states. A key element of this syndrome was the patients' belief that an unpleasant offensive odor emanated from their immediate environment or person, such as from the skin, mouths, genitals, or recta [3]. One patient came to believe that people around him were talking about the malodour, closed their noses while near him, or opened windows in trains

and buses to rid themselves of the stench. The 'contrite' reaction is a remarkable feature of many such patients, with a number repeatedly washing their hands, overusing perfumes, changing their clothes multiple times a day, and restricting their social life and work. DSM-III classified ORS as a typical somatoform disorder that concerns body functioning or appearance largely devoid of the symptoms of other mental disorders (e.g., so-called "monosymptomatic hypochondriacal psychosis") [4]. DSM IV reclassified ORS as a delusional disorder, somatic subtype, whereas DSM V lists ORS as a specified obsessive-compulsive and related disorder characterized by fear of having an offensive body odor. DSM V also links ORS to "taijin kyofusho," a disturbance of personal interaction listed under "other specified obsessive compulsive and related disorders."

It should be noted many ORS patients who believe they are the source of the odor regularly seek medical help and frequently undergo unfruitful diagnostic tests. Such patients often complain of ineffective medical care due to the recurrent negative tests and their inability to gain effective medical help through traditional approaches. In one study of ORS patients, more than 44% looked for non-psychiatric medical, surgical, or dental treatments for their perceived odor, and as many as a third actually received some form of ineffective medical or surgical intervention. Interestingly, eight ORS patients in this study looked for treatment from 20 different clinicians and six ORS patients received treatments from a total of 10 non-psychiatric health professionals [5]. One case report details the "unnecessary" surgical lumbar sympathectomy on a patient with ORS who, the authors suggest, should have begun psychiatric treatment much earlier [6]. Another case report details a number of surgeries, including turbinoplasties and a tonsillectomy, prior to the establishment of an accurate diagnosis [7]. It is clear that the "costs" of this condition can be great to the health care system and to the patients themselves. In fact, as many as 40% of the ORS patients have been found to be housebound for at least one week due to their ORS, 68% had a history of suicidal ideation, and 32% attempted suicide [5].

Although sometimes framed as a uniform entity, ORS exhibits considerable heterogeneity, which calls into question whether it should be viewed as a single syndrome. Moreover, although not mentioned in the literature, the possibility exists that some persons classified as having ORS may have a genuine dysomic or phantosmic problem secondary to damage to the olfactory neuroepithelium, as from viruses and other xenobiotics. This dysfunction could play a role in the manifestation of these obsessive-compulsive disorders as preoccupations with body odor, suggesting that underlying biological deficits could influence the ways in which psychiatric disorders such as obsessive-compulsive disorders symptomatically manifest. Dysosmias and phantosmias are well documented in the general population and often reflect perceptual aberrations secondary to incomplete regeneration or prolonged delays in degeneration of the olfactory epithelium [8]. Non-ORS patients who experience such problems complain of an acute or chronic smell sensation, frequently of a non-descript unpleasant chemical-like or smoke-like character, that can occur in the absence of an external stimulus. The cases reviewed in this report meet the diagnostic criteria of ORS proposed by Phillips et al and other specific obsessive compulsive and related disorders associated with body odor [9]. In all four cases, objective olfactory testing found evidence of altered olfactory function.

Case-1

This is a 20-year old man who worked full time at a mail distribution centre. He reported experiencing halitosis for the last seven months and that he had lost confidence in himself due to his worries about others' perception of his halitosis. This case had a noteworthy psychiatric history of anxiety and depression with no suicidal ideation. He complained of feeling moderately lightheaded and mildly nervous, with severe difficulty breathing and severe sweating not due to heat. He described feeling irritable all the time and that he had lost most of his interest in other people and things. He reported smoking marijuana occasionally, but refrained from alcohol and tobacco use. He had a history of sinus infections, which had been treated with antibiotics. His score on the University of Pennsylvania Smell Identification test (UPSIT) [7] was 29/40, indicating moderate loss of smell function. He had previously undergone dental, gastroenterological, otolaryngological, and other evaluations to no avail. His problems began one year prior to his appointment at the University of Pennsylvania Smell and Taste Center. Although psychophysical testing clearly demonstrated a deficit in his function [10], he still felt that he was experiencing halitosis despite the fact that others, including one doctor at the Smell and Taste Center, found his breath not to smell abnormal.

Case-2

Case 2 is a 71-year old fashion designer who reported experiencing a bad smell attributed to a chemical that entered his apartment via the ventilation system. At the time when this was first being experienced, he noted that his lips were full of blisters and that he was very weak and had shortness of breath. He indicated that he repeatedly washed his hair and person to minimize the odor, largely for social reasons. However, when he moved from the 'infected apartment', he continued to experience the bad odor and even went so far as to attribute its source to his neighbour. He reported in his intake interview questionnaire that the exposure has led to "endless suffering." As a result of the perceived lingering odor, he no longer was able to hold a job. Because of his belief that the odor permeated objects around him, he threw out multiple pieces of art and memorabilia from his career, including pictures taken with celebrities and leading political figures. His score on the University of Pennsylvania Smell Identification Test (UPSIT) was 15/40, indicating severe loss of smell function. However, he had

difficulty believing, even after visiting the Center, that his sense of smell was distorted and that the bad odor he was perceiving was not due to contamination of his apartment and person. Interestingly, his initial symptoms of weakness, blisters, and shortness of breath that he attributed to the bad odor were consistent with a viral infection. Such infections are the most common cause of olfactory disturbances, including phantosmias, in the general population [11,12].

Case-3

This is a college-educated woman in her 60's with no medical history of psychiatric disorder or other medical problems. She was referred by her neurologist to the Center because of considerable distress related to her belief that her spouse emits a chronic unpleasant body odor. She felt that he had emitted this odor for more than six months. When she experienced the stench, she became nauseous. She indicated that no one else perceived the odor, which became stronger to her when he was showering, using an electric toothbrush, exhaling excessively, or when his mouth was open. Her spouse was a middle-aged businessperson who had no noteworthy medical or psychiatric history. In light of her complaints, he initially questioned his friends to find out if they could smell the putative odor, although no one responded affirmatively. In attempts to diminish this putative odor, he showered many times a day, frequently checked his breath and armpits for odor, changed his eating habits, and chewed various brands of gum. As a result of his wife's delusional symptoms, he became socially isolated, feeling embarrassed or fearful of offending others with his smell. Upon her insistence, he sought medical help from multiple specialists, including a dermatologist, dentist, otolaryngologist, gastrologist, and a proctologist. None, however, was able to identify a basis for the purported body odor or halitosis. Eventually, the couple fought over this perceived problem and separated. It should be pointed out that steam or warm air can induce dysosmic episodes in susceptible individuals [13]. In this case, her spouse's warm breath, as well the shower vapour could have played a role in inducing the dysosmia she experienced. Although psychophysical testing clearly demonstrated a reduction in her olfactory function (UPSIT score = 26/40), she still felt that her spouse was emitting the bad odor. She had little insight, even after visiting the Center and receiving counselling, that the problem most likely stemmed from her olfactory deficit.

Case-4

Case 4 is a woman in her 60's is who complained of smelling a persistent mold-like smell. She originally felt that the odor, which others do not notice, emanated from her home, so she left her house periodically in an effort to get rid of the odor, but to no avail. Whatever its perceived source, she felt that the bad smell clung to her body and clothes. At some point she came to believe, as did Case 2, that her next-door neighbour was responsible for the odor, although she could not clearly articulate how or why the neighbour caused the odor. She reported that her body and hands started to shake when she smelled the odor. She also noted that often she tried to blow her nose or to spit to rid herself of the odor, but that this was rarely effective. She reported never being a cigarette smoker and that she refrained from alcohol use. She had no noteworthy psychiatric or medical history or suicidal ideation, and did not experience any other hallucinations or show any evidence of other psychiatric disorders. Like the other cases mentioned above, she lacked self-awareness regarding her problem even after her chemosensory testing revealed a significant alteration of smell function (UPSIT = 22/40).

Discussion

In this case series, we present evidence that the olfactory delusions of some patients with ORS and associated specified obsessive-compulsive type disorders involving delusions of body odor appear to have a biological basis, as measured by olfactory testing. Similar to other documented ORS patients, those described in this report are convinced of the presence of a bad smell that others fail to notice. All four were upset with physicians and friends who do not perceive the putative odor and lacked insight into the likely origin of their problem. In spite of the fact that quantitative olfactory testing documented their aberrant olfactory function, this fundamental lack of insight persisted along with the sensory complaints even after counselling. As shown in these cases, underlying olfactory deficits could play a role in these psychiatric disorders, perhaps influencing how these specified obsessive-compulsive disorders manifest symptomatically in patients. For example, Case 3 was highly suspicious of other people and projected her problem onto her spouse, ultimately resulting in the end of their relationship. Case 2 projected his suspicions onto his neighbours, a similar phenomenon seen with Case 4. While these are fundamentally aggressive ways of dealing with the underlying anxiety generated by these disorders, there are many cases, such as that of Case 1, where patients actually retreat from social interactions, reflecting introversion and compounded insecurities about how they are perceived by others. ORS has been highly associated with other mood disorders, anxiety disorders, and substance use disorders in addition to the potential for suicide [14]. Indeed, Major Depressive Disorder has been reported as the most common comorbid ORS condition [5,15]. It is important to note that depression is prevalent among non-ORS patients with quantifiable olfactory disturbances; however, the severity of the depression appears to depend on the type of olfactory disturbance. For example, non-ORS patients with distorted smell function (dysosmias, phantosmias) score higher on the Beck Depression Inventory than patients with only smell loss (anosmia) [11]. Based on the patients described in this report, true smell dysfunction may play a role in a number of symptoms associated with ORS and related disorders, including reduced body weight, loss of appetite,

and diminished psychological well-being. A similar pattern of problems is observed in post-traumatic/post-infectious dysosmic patients [16]. The partial impairment of smell function in both of these groups of patients may help to explain the mechanisms behind this association. Central mechanisms may also be playing a role. For example, one report found that an ORS patient exhibited deficits in frontotemporal 99mTc HMPAO perfusion on a SPECT scan [17].

The few studies and case reports that suggest effective treatments for ORS patients are quite limited in scope and often fail to control for comorbid influences of multiple medications. Serotonin-reuptake inhibitor antidepressant monotherapy, antipsychotic monotherapy and SSRI/ antipsychotic combination therapies have all been reported to have some efficacy [5,18,19]. Psychotherapy, as well as Cognitive-Behavioural approaches, has received limited success [5]. If smell dysfunction is a significant element of ORS and associated disorders, treating such dysfunction could potentially play an important role in a multidisciplinary approach to this delusional syndrome. Although such treatments are limited, there are a number of therapies reported in the literature. Among the less invasive therapies are the use of alpha-lipoic acid [20], Vitamin B-12 [21], and exposure training to multiple odorants [22]. In cases where suicide appears eminent, lessoning of the tissue producing the aberrant sensations within the neuroepithelium [23] or removal of the olfactory bulbs could be viable options [24,25]. The current report suggests that olfactory dysfunction may play a role in ORS and related obsessive-compulsive type disorders in some patients. Olfactory testing of patients in this case series suggests a measurable decline in olfactory function may underlie and/or perpetuate the psychiatric manifestations of at least some patients with ORS and related disorders. This indicates that olfactory testing in the course of management of patients with ORS and related disorders could provide insight into the pathophysiology of this enigmatic syndrome, as well as facilitate the development of novel approaches to patient care.

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