Is Olfactory Reference Syndrome an Obsessive-Compulsive Spectrum Disorder?: Two Cases and a Discussion

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A number of disorders characterized by intrusive repetitive symptoms and varying degrees of insight may overlap phenomenologically and neurobiologically with obsessive-compulsive disorder (OCD). There is a question as to whether olfactory reference syndrome, a disorder characterized by persistent preoccupations about body odor accompanied by shame and embarrassment, is also an OCD spectrum disorder. Two cases of olfactory reference syndrome, with accompanying phenomenological and neurobiological data, are presented in order to discuss the possible overlap with OCD. A number of phenomenological and neurobiological features in these patients were at least partially reminiscent of OCD. In particular, despite having poor insight, both patients demonstrated significant improvement upon treatment with a serotonin reuptake inhibitor.

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Several authors have described psychiatric patients with persistent preoccupations with personal odor.[1–4] Although these olfactory symptoms can be seen in a number of disorders, including schizophrenia, depression, and medical conditions, there also seems to be a specific clinical entity characterized by such preoccupations. An early case series in the Western literature used the term *olfactory paranoid syndrome*.[5] Japanese patients with similar symptoms have, however, been characterized as having an anxiety condition known as *taijin kyofusho* or *anthropophobia*,[6,7] a term that emphasizes the avoidance of social situations typically seen in such patients.

In perhaps the most extensive investigation of the syndrome to date, Pryse-Phillips[8] introduced the term *olfactory reference syndrome* (ORS) to differentiate the olfactory symptoms seen in this entity from those of schizophrenia, depression, and temporal lobe epilepsy. Patients with ORS held themselves responsible for the odor, and therefore experienced a "contrite reaction," characterized by shame and embarrassment. Such patients "tended to wash themselves excessively, to change their clothes with more than usual frequency, to hide themselves away, and to restrict their social and domestic excursions."[8]

In DSM-IV,[9] delusions about personal odor are described as an example of the somatic subtype of delusional disorder. However, it is conceivable that ORS might also be diagnosed as obsessive-compulsive disorder (OCD) with poor insight. Indeed, a number of so-called obsessive-compulsive spectrum disorders,[10–12] such as body dysmorphic disorder,[13,14] hypochondriasis,[15,16] and pathological jealousy,[17] have been described as having both obsessional and delusional variants. This spectrum of disorders has been defined in terms of a commonality of important phenomenological and neurobiological features, such as preferential response to selective serotonin reuptake inhibitors (SSRIs).[13,15,18]

An immediate question is whether it is also useful to conceptualize ORS as lying on an OCD spectrum. In this article we present two cases of patients with ORS, with accompanying phenomenological and neurobiological data, in order to address the possible overlap of this condition with OCD. Both patients had a Western (rather than Eastern or African) cultural background and were seen in our Obsessive-Compulsive Disorders Clinic, where they were interviewed with the Structured Clinical Interview for the Diagnosis of Axis I Disorders[19] on presentation.

CASE REPORTS

Case 1. Mr. A. was a 17-year-old male who described a persistent preoccupation, which had begun about 6 months

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previously, with the idea that he smelled of urine. Each time he urinated, for example, he worried that he had wet his underwear, and that consequently people would think that he smelled of urine. He stated that these thoughts occupied most of his waking time. As a result of the thoughts he would repeatedly check his underwear for urine stains, would change his clothing excessively often, and would use more deodorant than usual. In addition, shame and embarrassment about the perceived odor gradually led him to avoid more and more social interactions, and he even began to miss days at school. He became increasingly demoralized and at the time of presentation exhibited a number of symptoms of depression, although he did not meet DSM-IV criteria for a major depressive episode. On close questioning, the patient said that he was 95% certain that he did in fact smell of urine, although on occasion he felt that his preoccupations about odor were excessive and unreasonable. There was no history of classic obsessions or compulsions or of definite hallucinations or delusions. There was no history of substance abuse or of an underlying general medical disorder.

The patient consented to SPECT imaging of the brain before beginning a trial of an SSRI. Five minutes prior to injection of 555 MBq (15 mCi) [^{99m}Tc]hexamethylpropyleneamine oxime (HMPAO), the patient was instructed to allow himself to focus on the idea that he might in fact be smelling of urine, in the same way that he usually did when preoccupied with this concern. He continued with these thoughts and actions for 5 minutes after injection.

SPECT data were acquired by use of a dual detector gamma camera (Elscint, Helix) equipped with fanbeam collimators, using the step-and-shoot mode and a circular orbit. A 128×128 image matrix was used, acquiring data in 3-degree steps, for 15 seconds per step, through 360 degrees. Data were reconstructed by using a Metz filter (power 5, FWHM 14mm) to provide tomographic images in the transaxial (parallel to the orbitomeatal line), coronal, and sagittal planes. Each image set was normalized to the mean cerebellar counts.

Pretreatment SPECT revealed multiple perfusion defects, including perfusion defects anteriorly in the right and left inferior frontal lobes (higher on the left) and in the left frontoparietal area; irregular perfusion in the parietooccipital regions; and diminished perfusion of both temporal lobes and the left occipital area.

The patient was then treated with citalopram 40 mg daily. He showed a gradual reduction in his preoccupation with odors. Ratings of symptoms related to his odor on the Yale-Brown Obsessive-Compulsive Rating Scale[20] fell from 29 at week 0 to 25 at week 4, 17 at week 8, and 8 at week 12. Ratings of depressive symptoms on the Montgomery-Åsberg Depression Scale fell from 11 at week 0 to 6 at week 4, 5 at week 8, and 1 at week 12. Symptoms remained significantly improved throughout the subsequent course of treatment in our clinic (8 months).

Case 2. A 54-year-old woman presented with a persistent preoccupation that she had a foul body odor. The smell emanated from her armpits, breast folds, feet, and anal region and was reminiscent of rotting fruit. This particular symptom had begun about 30 years earlier. A more recent concern was that drains from her house emitted a similar foul smell. Although the patient was able to admit that these concerns were perhaps unrealistic, she was often concerned about embarrassing herself, and this had led to excessive handwashing, frequent changes of clothing, and repeated cleansing of drains. Furthermore, at times these concerns resulted in marked depressive symptoms that met DSM-IV criteria for a major depressive episode. The patient also suffered at times from checking (of lights, stove, taps) and counting rituals. There was no history of definite hallucinations or delusions. There was no history of substance abuse or of an underlying general medical disorder.

Electroencephalography and computed tomography of the brain revealed no abnormalities. The patient was initially treated with amitriptyline 150 mg daily, which improved mood but not olfactory symptoms. When the patient discontinued medication after 12 weeks, depressive symptoms worsened, and she submitted to a course of electroconvulsive therapy (18 bilateral treatments). This again improved mood but not olfactory symptoms. A 12-week course of fluoxetine, reaching a maximum dose of 80 mg daily, had no further effect on olfactory symptoms. Treatment with pimozide 4 mg daily for 8 weeks was similarly unsuccessful. A combination of clomipramine 250 mg daily and perphenazine 4 mg thrice daily for 12 weeks also had little positive effect. However, after increase of the clomipramine to 300 mg daily and discontinuation of the perphenazine, the patient showed a gradual, partial, but definite improvement in both her preoccupation with foul odors and her depressive symptoms. A retrospective Clinician's Global Impression change score would be 2 (much improved).

DISCUSSION

The two case reports of olfactory reference syndrome (ORS) presented here can be used as a basis for a discussion of the relationship between this entity and OCD. Phenomenologic, neurobiologic, and pharmacologic issues will be considered in turn.

The phenomenology of the symptoms in the two patients is reminiscent of OCD in a number of respects. Although all symptoms were related to body odor, these met the DSM-IV[9] criteria for obsessions (intrusive thoughts about body odor) and compulsions (repetitive cleansing behaviors). Although symptoms were often not recognized to be unreasonable or excessive, they were accompanied by significant shame and distress. Thus, on SCID-I interview, the patients were diagnosed as having OCD with poor insight. Both patients experienced onset of the disorder in adolescence, as well as secondary depressive symptoms, features that are common in OCD.[21] Nevertheless, only Patient 2 described obsessions and compulsions that did not directly relate to complaints about her own body odor (concerns about other odors, checking, counting), whereas OCD patients tend to have a number of different kinds of symptoms over time.[22]

Pryse-Phillips' pioneering report[8] described 36 patients with ORS and documented extensive support for

the existence of this entity in previously published case material. The report emphasized that ORS is characterized by a "contrite" reaction to the perception of odor. That is, the patients are "deeply ashamed, embarrassed, self-abasing." In this series there was common use of deodorant or excessive washing (82%) and excessive changing of clothes (68%). Patients were able to construct a hierarchy of feared situations, and avoidance behavior was typical (97%). Fifty-two percent had developed continuous depression, and 50% had obsessional features. Although many patients continued to work and did not meet criteria for schizophrenia, there was often marked functional impairment. They had frequently presented to medical specialists but had rarely received psychiatric treatment. These findings of psychosocial dysfunction are consistent with our cases and with an overlap between ORS and OCD.[23]

On the other hand, Pryse-Phillips[8] found that average age at onset of ORS was 25.4 years, 78% of patients were males, 60% had lost insight, and 75% had olfactory "hallucinations." This age at onset and male:female ratio were higher than is typical for OCD, and psychotic symptoms are uncommon in OCD. However, demographic discrepancies may reflect the fact that Pryse-Phillips drew many subjects from inpatient settings. Furthermore, degree of insight varies in OCD and related disorders, and patients with poor insight have been classified as psychotic by early authors. Indeed, given the focused nature of these patients' symptoms, with no evidence of obsessions and compulsions unrelated to odor, the entity is particularly reminiscent of disorders such as body dysmorphic disorder, hypochondriasis, and obsessional jealousy, where there is frequently limited insight about a single obsessional focus.

SPECT findings in Case 1 described here differed partially from the classic findings of increased prefrontal/orbitofrontal cortex activity often seen in OCD.[24] However, a number of SPECT studies have also noted reduced blood flow in patients with OCD.[25] Furthermore, the behavioral challenge used during our SPECT procedure differed from the resting scan methodology used in most OCD studies. Unfortunately, it was not possible to obtain a repeat SPECT scan after treatment to determine whether the medication had led to improvement in perfusion deficits.

There is little in the literature on the neurobiology of ORS. Alliez and Dongiers[26] described one patient with right temporal spike activity, but the phenomenology of olfactory hallucinations in temporal lobe epilepsy appears rather different from that seen in ORS.[8] Certainly it has been suggested that central mechanisms in psychiatric disorders such as depression and schizophrenia result in alterations to olfactory threshold. However, once again the phenomenology of olfactory symptoms in these disorders differs from that in ORS,[8] and it is not clear that the latter actually involves alterations in perceptual rather than cognitive-affective processes.

Both patients reported here showed a gradual but significant response to treatment with an SSRI. The SSRIs, including clomipramine[27] and citalopram,[28] are well known to be effective in OCD.[29,30] There are a number of previous reports of pharmacotherapy for olfactory reference syndrome, with Videbech[5] describing patients who failed to respond to neuroleptics and Pryse-Phillips[8] noting that one patient did not respond to treatment with tranquilizers and a monoamine oxidase inhibitor. Pryse-Phillips[8] does indicate, without providing more details, that there may be fair reduction in symptoms after treatment with psychotherapy and tricyclic antidepressants. Monosymptomatic hypochondriasis, which includes some patients with ORS,[31,32] has been linked with response to pimozide,[33] and a number of authors have also noted that this condition may respond to tricyclics.[32,34] Finally, there are reports of ORS responding to clomipramine.[35]

Although connections between olfactory and obsessive-compulsive symptoms have long been drawn,[36] there is little in the contemporary literature about this topic. However, the cases of ORS described here provide tentative evidence of at least a partial overlap with other patients with obsessive-compulsive spectrum disorders such as body dysmorphic disorder, hypochondriasis, and obsessional jealousy. Given the separate classifications of ORS and possibly related conditions in our current nosology, perhaps further changes to the DSM-IV are necessary to emphasize that a number of conditions previously conceptualized as psychotic in nature may have similarities with OCD. In addition, given the similarities between ORS and entities that have been conceptualized as "culture-bound,"[6,7] such nosological changes should perhaps reflect the possible universality of these conditions.

Systematic study of a larger sample of patients may be helpful in determining whether ORS is best viewed as an OCD spectrum disorder, as part of a social phobia spectrum, or as a delusional disorder. A putative overlap between ORS and OCD would suggest that ORS patients might also respond to SSRIs and behavioral therapies; further basic and clinical work to determine the selectivity of the serotonergic mediation of ORS and similar symptoms would thus be of particular interest.[37,38]

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