ABSTRACT: Olfactory Reference syndrome (ORS) is characterized by patients falsely believing that they exude a foul body odor, which is embarrassing and disturbing to the patient. The increased anxiety due to this belief leads to compulsive behaviors, social anxiety, and functional impairment. ORS poses enormous challenges in its diagnosis and treatment. The disorder can be often treatment-resistant or recurrent. A case formulation followed by a classificatory diagnosis is an effective approach to distinguish it from other diagnoses with an overlapping spectrum of symptoms. Here, we present a case of a 42-year-old Australian woman with recurrence of ORS post major stressful triggers. She reported a trial of a series of expensive cosmetic and hydraulic treatments, however, her symptoms persisted, causing significant deterioration in her mood and social functioning, interpersonal relationships, and self-care. Patient was brought to the psychiatric inpatient unit with the complaint of feeling unsafe in her own house. She was started on Lurasidone, along with 12 weeks of intense cognitive behavioral sessions. The use of psychotherapy is underreported even though it significantly reduces ongoing distress. The patient remains asymptomatic along with improved social functioning on subsequent follow-ups.

KEYWORDS: Olfactory reference syndrome, ORS, OCD, Taijin Kyofusho, case report

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Introduction

Olfactory Reference Syndrome (ORS) is a psychiatric condition characterized by an erroneous belief that one is exuding a false body odor, that brings significant distress and sociofunctional impairment to the patient, albeit, is undetectable to others.1 Described as a disorder in a case series of 36 patients for the first time in 1971, the literature since then is permeated with the concerned syndrome, albeit is a topic of ongoing debate when concerned with its classification, the nosology, and treatment.2,3 The diagnosis of ORS can be challenging as the symptomatology often overlaps with various other disorders and the patient may have variable insight.4

Patients with ORS often present with a chronic recurrent course with significant distress, blame themselves for repelling others, and it debilitates the quality of life for themselves and their close ones.6 In recurrent cases, they often develop psychotic-like symptoms or suicidal ideation.6

ORS has not gained popularity in classificatory systems and is currently placed in DSM-5 under “Other Specified Obsessive-Compulsive Disorders” as well as under Taijin Kyofusho, a disease described as “anxiety about and avoidance of interpersonal situations, due to the thought, feeling, or conviction that one’s appearance and actions in social interactions are inadequate or offensive to others.”8,9 The challenges around its classification stem from the relatively frequent response of the syndrome to Selective Serotonin Reuptake Inhibitors (SSRIs) suggesting a possible overlap with Obsessive-Compulsive Spectrum pathology. Few of the earliest studies highlighted mood affective disorders, especially major depression disorder (MDD) as the most frequently associated comorbidity with ORS,2,6,7 also supporting the theory of efficacy of SSRIs in context to ORS. Other comorbid disorders reported include bipolar disorder, personality disorders, depression, schizophrenia, hypochondriasis, alcohol and substance use disorders, and body dysmorphic disorder.2,6,8,9

Despite this preference, evidence also exists for a series of cases successfully treated with antipsychotics. Treatment can be quite complex and treating distressing symptoms often gets the priority. Our case report presents a case of successfully treating ORS with the aid of Lurasidone and Cognitive behavioral therapy (CBT). The use of psychotherapy is underreported even though it significantly reduces ongoing distress.4

Case Description

Here we present a case of a 42 years old female who was referred for assessment Consultation Liaison Psychiatry services. She had harmonious relationships with her family, employed as a translator till 1 week before her presentation to the practice, married, with a long psychiatric history of suffering from ORS. She endorsed working in a stressful work environment which proved to be one of the major triggering factors for relapse during this presentation. For a period of 6 months, she searched for...
the sources of a foul smell coming from her body. She believed that she emitted a smell that disgusted others. She was apprehensive to leave her house for the job because people were looking at her. On a few occasions, when she went out due to persuasion by her husband, she felt that people were talking about her smell and kept their distance from her. She also reported hearing the voices of her deceased family members who talked about the body smell. She lacked any suicidal idea- tion and called her family as a protective factor. She was highly distressed, and she felt like a burden to her family. On the scale of Patient Health Questionnaire (PHQ-9), she was cited as mildly depressed with a score of 10.

She suffered from a similar episode 2 years ago, more intense in severity and duration, for which she opted hydrolysis treatment to reduce the smell. She also spent thousands of dollars on cosmetic procedures for the same. She eventually needed hospital admission as she began to feel suspicious and unsafe amongst her own people. The patient in all her episodes and during remission, lacked enough symptoms to diagnose OCD, while her depression and psychosis stemmed only from her delusion of emitting foul body odor.

The patient did not endorse symptoms of conversion disorder, malingering. While ORS has frequently been classified in literature across the spectrum of OCD and Body Dysmorphic disorder (BDD), the patient in our opinion did not project the symptomatology to diagnose BDD or OCD in her.

During her previous episode, she had a treatment trial with Risperidone followed by Amisulpride. Risperidone was discontinued after a trial of 1 week because of worsening condition. She responded to Amisulpride, however, had developed galactorrhea. She discontinued the treatment on her own after 6 months because of the side effects.

During the second episode, we started her on Amisulpride but changed to Lurasidone due to side effects. She responded to 80 mg of Lurasidone without any significant side effects. She also received cognitive behavioral therapy from a qualified clinician focusing on behavioral modifications, adaptive coping skills, interpersonal training with modeling, and thought-stopping exercises. She was taught various grounding techniques to reduce the associated distress. She reported no symptoms after 3 months of intensive treatment, felt well about herself and resumed work with reduced working hours.

| NAME OF STUDY; COUNTRY; YEAR | NATURE OF STUDY | ASSOCIATED COMORBIDITY | INTERVENTION | OUTCOME |
|-------------------------------|-----------------|------------------------|--------------|---------|
| Takenoshita et al.10 Japan, 2021 | Case report | • Anxiety | 1 mg Aripiprazole | 1-year follow-up, she reported she was doing well and was continuing her medication of 1 mg of ARP. At 2-year follow-up, she was in complete remission with medication. |
| Özsoy et al11; Turkey, 2021 | Case Report | • MDD with psychotic features | clomipramine (150mg/day), clonazepam (4mg/day), escitalopram (20mg/day), quetiapine (200mg/day), Pimozide (2mg/day) | At follow up 5 months later, was beginning to begin taking care of self, and perform activities of daily living independently. |
| Vani and Arumugam12 India, 2020 | Case Report | • Anxiety and Polysubstance use | Psychoeducation, CBT | After 10 sessions, patient’s daily functioning improved. OVIS Score reduced from 5.7 to 4.4 |
| Jegede et al16; USA, 2018 | Case Report | • Depression | Pimozide 1 mg/BD, Fluvoxamine 25mg/day | 70% symptomatic improvement in patient’s condition was noted after a few days of starting treatment. Her affect was brighter, she went outside her room for meals and group therapy and started socializing too. |
| Albers et al13 USA, 2018 | Case Report | • Depression • Chronic Hepatitis C | Olanzapine (2.5mg) | Patient showed marked improvement in odor identification deficit and odor-evoked parosmia. |
| Zantvoord et al14; The Netherlands, 2016 | Case Report | • generalized anxiety disorder (GAD) | CBT | In the following months, ORS and GAD symptoms remained in remission. |
| Lim and Wan15 Singapore, 2015 | Case series | Case B: Anxiety | Case A: CBT only Case B: Low dose Antipsychotic | Case A: After CBT patient showed a remarkable recovery. Her social anxiety also reduced significantly and she no longer avoided social interaction. Case B: After few doses patient stopped taking medications herself and defaulted further treatment. |

(Continued)
to effectively target OCD symptoms. In other severe cases, delusional disorder, in a few reported cases, SSRIs have shown orders including obsessive-compulsive disorder (OCD) and psychosis, or cognitive behavior therapy (CBT) to be effective for ORS.

The exact causal factor of ORS is not known. It is hypothesized to be multifactorial, a culmination of genetic, neurobiological, and environmental risk factors. The prevalence of ORS is also not well established and there is a dearth of epidemiological research on ORS. However, the literature has reported a community prevalence in a range of 0.5% to 2.1%. The current existing literature majorly consists of case reports or case series and mentions treatment either with selective serotonin reuptake inhibitors (SSRIs), tricyclic antidepressants (TCA), antipsychotics, or cognitive behavior therapy (CBT) to be effective for ORS.

Since most ORS symptoms overlap with several other disorders including obsessive-compulsive disorder (OCD) and delusional disorder, in a few reported cases, SSRIs have shown to effectively target OCD symptoms. In other severe cases, where insight is absent, antipsychotics alone or in combination have shown to be an effective modality. Even though, ICD-10 classifies this syndrome of major significance as a delusional disorder, not many previously reported cases mentioned auditory hallucinations or paranoia. Where cases present with somatic preoccupations, body dysmorphic disorder (BDD) can be considered a differential diagnosis. However, patients with BDD show more concern with physical appearance than with unpleasant smells. Henceforth, due to the overlapping nature of ORS with social phobia, ORS cases get misdiagnosed as social anxiety, and the patient may get inappropriate medical care. It is also important to note here that in most of these reported cases, CBT has been shown to be underreported and an untapped, potentially effective treatment modality. A meticulous search of the literature, however, proves CBT to be effective both alone and in combination with pharmacotherapy.

In our case, the patient experienced foul body odor, anxiety, and paranoia after a stressful triggering event. We believe that our patient might have lost insight early on in her illness. She also went on to take extreme measures where she spent thousands of dollars on several cosmetic procedures. A mix of safety and avoidance behavior was noted too, such as not going out of her house or checking her own body for the source of foul odor. Such behaviors can be attributed to compulsion or paranoia. The embarrassment associated with a foul smell might have been the reason, our patient acquired an avoidance coping style. Therefore, we conceptualized it to be a schizo-obsessive disorder as she had symptoms of both, during the evolution of her illness. Her presentation was similar in both episodes. In such concurrent illness, serotonin, and dopamine become a critical focus therefore we chose Lurasidone due to its high affinity for both, dopamine and serotonin receptors.

### Discussion

| NAME OF STUDY; COUNTRY; YEAR | NATURE OF STUDY | ASSOCIATED COMORBIDITY | INTERVENTION | OUTCOME |
|-----------------------------|-----------------|------------------------|--------------|--------|
| Michael et al16 Australia, 2014 | Case series | Case 1: Anxiety | Case 1: CBT | Case 1: 2 months later, patient had no ORS symptoms. Case 2: Complete resolution of symptoms, significant improvement in functioning. |
|                          |                | Case 2: Anxiety | Case 2: Citalopram 40 mg CBT Aripiprazole titrated to 10 mg/day |                  |
| Muffati et al16; Italy, 2008 | Case Report | MDD with psychotic features | imipramine 100mg/daily Aripiprazole 2.5 mg | The patient showed a significant improvement with aripiprazole. |
| Miranda-Sivelo et al16; Spain, 2013 | Case Report | • Axillary, palmar, and plantar bromhidrosis • paranoid schizophrenia | low-dose risperidone (3mg/d) psychotherapy | • extended follow-up of this patient for 7 year • Negative symptoms persisted • ORS resolved. |
| Chernyak et al19 USA, 2020 | Case Report | • Hyperhidrosis | Psychoeducation psychotherapy with behavior exposure based technique Fluoxetine 60 mg Neurology consult | • 2 years after treatment, her functioning markedly improved • she was able to complete her education and return to work • relationship with family members also improved |
| Senkal et al20 Turkey, 2015 | Case Series | Case 3: MDD Case 4: Depression | Case 1: Fluoxetine 60 mg/day Aripiprazole 15 mg/day Case 2: Sertraline 100mg/day Aripiprazole 10 mg/day Case 3: Sertraline 50 mg/day Case 4: Venlafaxine 75 mg/day Aripiprazole 15 mg/day | Case 1: returned to normal life with significant improvement in symptoms Case 2: responded well with significant improvement in her symptoms Case 3: Symptoms diminished after starting treatment and increase in the level of functioning Case 4: Marked improvement in functioning, decreased depressive symptoms. |
In addition, lurasidone has been shown to be highly effective in improving mood, memory, and cognition, has a low incidence of extrapyramidal symptoms, and has a very little side effect profile. It has previously been reported in the literature to effectively treat anxiety, depression, and schizophrenia. In our case, it effectively targeted both anxiety and psychotic symptoms. Additionally, CBT was offered to our patients as well. CBT has proven to be helpful not only for anxiety, depression, OCD and BDD, but it has shown to substantially benefit ORS patients with full remission after only 2 months. Similarly, in our case patient showed exceptional improvement and full remission after 3 months of offering treatment.

Albeit, a good case formulation can direct physicians toward an appropriate and patient-specific treatment approach. There still exists a need for quality randomized control trials to guide treatment trajectory. We believe that this case not only adds significantly to the existing literature available for ORS but will also be essential in order to improvise the diagnosis and treatment strategies for such a debilitating disorder. This disorder might find an independent place in future DSM with more research and data.

Conclusion

Olfactory Reference Syndrome is a psychiatric condition characterized by an erroneous belief that they exude a false body odor, bringing significant distress and socio-functional impairment to the patient, however, is undetectable to others. Despite being reported in the literature for over a century, olfactory reference syndrome has received little attention in terms of research, diagnosis, treatment guidelines, classification, and recommendations. Evidence on treatment and management is relatively less for ORS with antipsychotics and CBT as compared to SSRIs. Our case report attempts to add to the effectiveness of antipsychotic lurasidone and CBT in the management of ORS with psychotic elements.

Author Contributions

SD and LS were responsible for conception and supervision, data proofing and writing the final manuscript. SP and AAR wrote the initial two drafts and was responsible for obtaining data images and incorporating laboratory data into the manuscript. NW and AB wrote the third draft, peer review changes and approved the final draft after editing. All authors approved the final manuscript and agreed to be equally accountable for this work.

Ethical Approval

Ethical approval was waived by the patient for this case report.

Informed Consent

Written informed consent was obtained from the patient(s) for their anonymized information to be published in this article. The subject had the decision capacity to provide written informed consent.

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