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Case Report
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Two Cases of Olfactory Reference Syndrome Treated with Risperidone

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ABSTRACT

Olfactory reference syndrome (ORS) is a rare psychiatric condition severely affecting quality of life and global functioning of affected subjects. Despite its peculiar clinical pattern, its classification as independent entity rather than as delusional disorder or general medical manifestation of neurological diseases, is still debated and far from to be clarified. Indeed, if classifying ORS as a type of delusional disorder according to DSM-5 seems problematic, its inclusion in obsessive-compulsive related disorders seems to be more promising from both a psychopathological and a management point of view. Nevertheless, ORS exhibits an inadequate response to antidepressants and psychotherapy, with increased sensitivity for second generation antipsychotics. As a result, its definition, clinical diagnosis, and adequate treatment management are missing, leading to a poorly evidence-based approach to the topic. Here, we present two ORS cases successfully treated with risperidone after several previous treatment attempts. We aim to spread knowledge about this rare but severe disorder to improve clinicians' awareness adding evidence to the current poor literature available on the topic.

Keywords: Obsessive-compulsive and related disorders, olfaction, olfactory reference disorder, olfactory reference syndrome

INTRODUCTION

Olfactory reference syndrome (ORS) is a delusional fear or false belief of emitting an offensive body odor which is not perceived by others, and preoccupation most often focused on the mouth, armpits and genitals, with bad breath and sweat as the most common odor descriptions.[1] ORS is often accompanied by the characteristic behaviors such as repetitive washing, usage of deodorants/perfumes, frequent changing clothes, avoiding the social situations and gatherings, causing significant social avoidance, depressed mood, anxiety and referential ideation.[2–4] Despite a clear clinical set of symptoms that characterizes this primary psychiatric syndrome, there is no consensus to consider this as a separate diagnostic entity, as also its classification changes based on the diagnostic system used, in addition to the uncertainty regarding the treatment options.[2,3]

Indeed, DSM-IV-TR describes ORS as a somatic-type delusional disorder and classifies it as a type of cultural jiko-syu-kyofu (a subtype of taijin-kyofu).[5] On the other hand, DSM-5 classifies ORS as a disorder related to obsessive compulsive disorder (OCD). Moreover, ORS is classified under the category of "obsessive-compulsive or related disorders" in the ICD-11.

Hereby we present two cases of ORS that had a unique psychiatric history and successfully responded to risperidone. The patients gave their written consent to publish this report anonymously.

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CASE PRESENTATIONS

Case 1

A 32 years old lady who has a positive family chart history of psychosis in two of her 3rd degree relatives of paternal origin, presented to us with an episodic course of illness with 2 previous psychotic episodes over the past 10 years which were diagnosed as brief psychotic disorder with ORS. Main major psychiatric (e.g., schizophrenia spectrum disorder, bipolar disorder, severe mental retardation, drug abuse) and general medicine disorders (e.g., dementia, brain trauma) were ruled out before starting treatment. Each of these episodes were characterized by delusions of persecution, delusions of reference along with the pre-occupation related to her oral hygiene, i.e., "saying that a bad smell is emanating from her oral cavity" even though no such smell is occurring and because of which she used to avoid the social situations. Secondary to this she used to brush her teeth repetitively every hourly often up to 30 times per a day which was presented exclusively during the episodes and she could recall the origin of this belief about bad smell as that once a teacher told her in front of her friends that the bad smell is coming from the patient's mouth and advised regular brushing, since then patient used to be having this smell problem except whenever she was on risperidone which would be stopped completely i.e., this symptom used to improve along with the psychotic symptoms each time with risperidone 6 mg/day. Prior to the risperidone, she was treated with fluoxetine up to 40mg/day along with aripiprazole 30 mg/day for 6 months with good response in psychotic symptoms, but ORS symptoms did not respond to fluoxetine and aripiprazole treatment, with a Yale-Brown Obsessive Compulsive Scale Modified for ORS (ORS-YBOCS) of 41 (range 0-48, positive if ≥20). Because of her concerns related to the pregnancy she had stopped the risperidone and currently presented with the recurrence of the above symptoms. Therefore, she was hospitalized due to her clinical presentation complexity to ensure better treatment management. During the current admission patient olfactory preoccupation did not improve with olanzapine 20mg/day for 3 weeks, which was prescribed by another physician for fear of inducing risperidone-related hyperprolactinemia considering the patient's female sex and young age. Hence, she was restarted on risperidone 6mg/day with which she had shown a dramatic response within a week and then discharged (ORS-YBOCS = 8).

Case 2

Mr. V, 19 years old male, discontinued his graduation and temperamentally slow to warm up during the childhood with a positive family h/o psychosis in two of the 2nd degree relatives from the maternal origin. He applied to us with complaints of a personal history of gradual academic decline, suicide attempts over the past year and a half, along with increasing irritability, para-suicidal gestures, and social withdrawal in the last 6 months and poor response to antidepressant treatment (e.g., selective serotonin reuptake inhibitors (SSRI), and tricyclic antidepressants (TCAs)). Also in this case, other major psychiatric (e.g., schizophrenia spectrum disorder, bipolar disorder, severe mental retardation, drug abuse) and general medicine disorders (e.g., dementia, brain trauma) potentially confounding were ruled out. Upon clarifying his symptoms, the patient reported that a year and a half ago his mother told him that he smelled bad, and he started to believe that his sweat smells bad and that people outside would make fun of him and humiliate him. He gradually started expressing to his father the need to buy him deodorants and perfumes and also, his desire to study in a college located in the coolest place of the country wherein he would not sweat. Patient held this belief with an extraordinary conviction and certainty. He was also reluctant to go out of the house or go shopping to buy something and wanted his father to do these things for him. Otherwise, no other psychotic symptoms/neurotic symptoms were present. A diagnosis of olfactory reference syndrome was made, and in the light of previous failures with antidepressants trials, the patient was started on risperidone 4 mg/day, then gradually increased to 8 mg/day, and showed gradual improvement within 2 weeks, except for a few behavioral symptoms due to his low IQ.

DISCUSSION

There is some evidence of ORS cases treated with a good response using antidepressants including clomipramine,[6] citalopram,[7] paroxetine,[8] and overall SSRIs have better evidence compared to antipsychotics, considering the limited available literature at present[9]. However, there are successful cases had been treated with antipsychotic monotherapy such as amisulpiride,[10] aripiprazole or risperidone have also been reported.[11] In some instances aripiprazole along with cognitive-behavioral therapy was also tried with some success.[9] In treatment-resistant cases, other antipsychotics such as aripiprazole[12] or blonanserin[13] have been tried as an augmentation strategy to antidepressants.

The significant clinical response to risperidone in both reported cases in only partially surprising, if considering the similarities between ORS and OCD. Indeed, among second-generation antipsychotics, risperidone appears to have the strongest clinical effectiveness in OCD, and therefore a similar clinical efficacy in ORS is plausible.[14]

Our cases are noteworthy in some respects, such as the improvement with risperidone monotherapy; moreover, in case-1 the onset of ORS preceded the psychosis, which is also another unique finding in accordance with the existing literature, according to which ORS can culminate into schizophrenia spectrum disorders.[10] In our case-1, fluoxetine and aripiprazole did not benefit in the ORS initially, hence she was started on risperidone. Subsequently, on re-admission, olanzapine was also a failed trial. In her case, only risperidone had shown clinical efficacy in reducing ORS symptoms along with an improvement in the psychotic features. Hence the importance of reporting such peculiar and rare cases, with the goal to increase clinicians' awareness about ORS and spreading knowledge sharing own experiences.

Informed Consent: Written informed consent was obtained from patients who participated in this study.

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REFERENCES

1. Phillips KA, Menard W. Olfactory reference syndrome: demographic and clinical features of imagined body odor. *Gen Hosp Psychiatry*. 2011;33(4):398-406. [Crossref]

2. Begum M, McKenna PJ. Olfactory reference syndrome: a systematic review of the world literature. *Psychol Med*. 2011;41(3):453-461. [Crossref]
3. Arenas B, Garcia G, Gómez J, et al. Olfactory reference syndrome: a systematic review. *Rev Neurol*. 2013;56(2):65-71.
4. Tee CK, Suzaily W. Unremitting body odour: A case of Olfactory Reference Syndrome. *Clin Ter*. 2015;166(2):72-73.
5. Suzuki K, Takei N, Iwata Y, et al. Do olfactory reference syndrome and Jiko-shu-kyofu (a subtype of Taijin-kyofu) share a common entity? *Acta Psychiatr Scand*. 2004;109(2):150-155. [Crossref]
6. Dominguez RA, Puig A. Olfactory Reference Syndrome Responds to Clomipramine But Not Fluoxetine. *J Clin Psychiatry*. 1997;58(11):497-498. [Crossref]
7. Stein DJ, Le Roux L, Bouwer C, Van Heerden B. Is Olfactory Reference Syndrome an Obsessive-Compulsive Spectrum Disorder? *J Neuropsychiatry Clin Neurosci*. 1998;10(1):96-99. [Crossref]
8. Teraishi T, Takahashi T, Suda T, et al. Successful Treatment of Olfactory Reference Syndrome With Paroxetine. *J Neuropsychiatry Clin Neurosci*. 2012;24(1):E24-E24. [Crossref]
9. Bizamcer AN, Dubin WR, Hayburn B. Olfactory Reference Syndrome. *Psychosomatics*. 2008;49(1):77-81. [Crossref]
10. Yeh YW, Chen CK, Huang SY, Kuo SC, Chen CY, Chen CL. Successful treatment with amisulpride for the progression of olfactory reference syndrome to schizophrenia. *Prog Neuro-Psychopharmacology Biol Psychiatry*. 2009;33(3):579-580. [Crossref]
11. Michael S, Boulton M, Andrews G. Two cases of olfactory reference syndrome responding to an atypical antipsychotic and SSRI. *Aust New Zeal J Psychiatry*. 2014;48(9):878-879. [Crossref]
12. Muffatti R, Scarone S, Gambini O. An Olfactory Reference Syndrome Successfully Treated by Aripiprazole Augmentation of Antidepressant Therapy. *Cogn Behav Neurol*. 2008;21(4):258-260. [Crossref]
13. Takekita Y, Kato M, Sakai S, et al. Olfactory reference syndrome treated by blonanserin augmentation. *Psychiatry Clin Neurosci*. 2011;65(2):203-204. [Crossref]
14. Erzegovesi S, Guglielmo E, Siliprandi F, Bellodi L. Low-dose risperidone augmentation of fluvoxamine treatment in obsessive-compulsive disorder: a double-blind, placebo-controlled study. *Eur Neuropsychopharmacol*. 2005;15(1):69-74. [Crossref]