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Research Paper

A case series of atypical presentations of Olfactory Reference Syndrome

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Abstract:
*Olfactory Reference syndrome (ORS) is a rare syndrome and is quite challenging to diagnose. It is characterised bythe obsessional and inaccurate belief of having a foul odour coming from the body leading to significant dysfunction in day-to-day living. ORS patients first seek treatment froma physician, dermatologist, dentist etc. to alleviate the perceived odour, which frequently leads to misdiagnosis and later seek psychiatric opinion often leading to unnecessary treatment delay. Also,patients with ORS can have limited insight and ideas of reference which may mimic psychotic or delusional disorders, though show worsening with antipsychotic agents.ICD-11 (*International classification of diseases*) has incorporated Olfactory Reference Syndrome with poor insight (6B22.1) as a separate entity. We present a case series of ORS with varied atypical presentationsand different psychiatric diagnoses coming to our OPD, where proper evaluation and serial assessments helped to diagnose them as a case of ORS. Hence, a significant improvement was seen in these cases on propertreatment with anti-obsessional agents.*

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I. Background:

Historically, Potts (1891) first described the phenomena of an individual becoming convinced that he or she gives off an unpleasant smell. Later on, thesephenomena was given many names such as *bromidrosiphobia* (Sutton, 1919), chronic olfactory paranoid syndrome (Videbech, 1966) and olfactory phobic syndrome (Walter, 1965) in various reported cases over the subsequent years.Despite gradually knowing the descriptions of ORS symptomatology for more than a century, itsnosological status remained undecided. In the classification of DSM IV TR as well as ICD 10, ORS patients with poor insightwere considered to be delusionaland it was categorised under delusional disorder (somatic subtype).But, with newer classificatory systems like in DSM-5 ORS has been categorized under another specified obsessive-compulsive and related disorder[1]while, ICD-11 incorporated Olfactory Reference Syndrome with poor insight (6B22.1) as a separate entity.

ORS, is a syndrome that presents with the belief that an offensive odour emitting from one's body leads to significant distress or impairment in daily functioning, yet no such odour is detected by others[2]. Its prevalence ranges from 0.5-2.1% [3, 4] and is attributed to a combination of behavioural, neurobiological, cognitive, genetic, and environmental factors[5]. ORS is often associated with repetitive and excessive behaviour to camouflage this perceived odour, feeling of embarrassment and avoidance of social situations[6].

A majority of patients with ORS have a long latency for the first consultation with psychiatry doctorsdue toinitially extensive evaluation and treatment from dermatologists, otolaryngologists, and dentists to alleviate the perceived odour[7, 8, 2]. The perceived negative emotions, repeated failed attempts to correct the odour and the self-experienced negative cognitions have a serious psychological impact including hopelessness and suicidal ideation[2].

Case presentations:

Case-1:A 28 years old graduate male employed in a private company belonging to Middle Socioeconomic Status from urban background living in government shelter home (*Rain basera*) presented in OPD with complaints of feeling that people around talk about him and make faces (look at him with disgust) for past 3 years. This gradually increased over thelast 2 years and patient would avoid getting close to others including his family members and friends. He stopped going to work and would prefer to stay alone and isolated. Around a year back he even left his home and started staying in a *shelter home.*
Even after changing his place of living his belief continued and he founda similar experience with strangers. Moreover, he would also report smelling sewage wherever he went. At initial contact, considering the presence of referential delusions and olfactory hallucinations leading to significant socio-occupational dysfunction and normal neuroimaging (MRI Brain to rule out brain pathology), a provisional diagnosis of Schizophrenia, Paranoid subtype was made. The patient was started on tablet Olanzapine 5mg which was further built up to 15mg over 4 weeks.

On follow-up, the patient reported further worsening of his stated symptoms. The subsequent cross-sectional assessment revealed fidgety, restlessness anddysphoric affect. He was found to be sitting with his arms tightly pulled towards his body for which he expressed that a foul odour would come out if he would sit relaxed. On further evaluation, he reported that for the past 3 years he would feel that foul smell of sewage and faeces would come from his genitalia and armpit which over a period of 1 year started to come from the whole of his body. He would bathe multiple times with soaps with good aroma or use body deodorants but the malodour would still continue. Though, others would not report the malodour, he would still experience this because of which he'd feel people don't come near him and make faces. This led to avoiding interaction with others and covering his armpits by tightly squeezing his arms close to his body.

In view of features of repetitive and compulsive behaviourto camouflage the perceived malodour, lack of any other psychotic phenomenon and poor response with worsening of symptoms to antipsychotic medication; the patient's diagnosis was revised to Olfactory Reference Syndrome with poor insight (6B22.1) as per ICD-11. He was started with Capsule Fluoxetine 20mg which was gradually increased to 60mg over 4 weeks. The patient reported a good response to the treatment. Along with this, tablet Olanzapine was tapered and stopped. The improvement was noted onthe CGI scale, where scores in the severity of illness improved from 6 (baseline score) to 2 and scores in global improvement decreased from 7 (baseline score) to 1 on every follow-up.

Case-2:Twenty-three year old single male presented with complaints of persistent and pervasive depressed mood, disturbed sleep, irritability, agitation, decreased appetite, feeling of worthlessness and hopelessness with crying spells between 2 years. He was taking tablet Escitalopram20mg and tablet Olanzapine 10mg for the past 2 months from a private practitioner with minimal improvement. On further evaluation of history, the patient reported about his treatment for halitosis from a dentist for thepast 2 years. The patient would chew scented flavoured gums, use mouth freshener sprays and rinse his mouth frequently with prescription strength mouthwash as prescribed by his dentist. On inquiry, he did not report perceiving any bad oral odour himself but thought that the other people around him notice it. He made this inference from the gestures the people would make on seeing him like closing their mouths, coughing, touching and rubbing their nose or ridiculing him by turning their faces away. The patient would hesitate in contacting others even when his mouth was closed and would avoid the company of others.He would remain distressed about it leading to significant social dysfunction and that there had been not much change despite dental treatment.

On oral examination, no foul odour, dental caries, plaques or gum abnormality were noted.His oral hygiene was good and there was an absence of any plaques, nicotine pigmentation or ulcer. A diagnosis of Olfactory reference syndrome was kept after assessment. Tablet Olanzapine tapered and stopped and the patient was started on capsuleFluoxetine which was increased gradually up to 80mg. After a plateau of response to 50-60% improvement, the treatment was augmentedwith tablet Risperidone 1mg at night, he reported a significant improvement in symptoms along with improved social and biological activities.

Case-3: A 20-year-old single femalestudent preparing for thepre-medical entrance examinationpresented to our OPD with chief complaintsofdecreased confidence, anxiety and awkwardness in social situations due to which shewas not attending coaching classesand avoiding the company of her friends. As per the patient, her complaints started 2 years back when she gave her first attemptatthe 12^{th} class examination.During that period, she was very distressedand reported noticing a foul smell coming from herself mainly from her underarm area which later she started feeling to be coming from the whole of the upper body. This smell would be more apparent

in the company of others.Many a time, she would ask her family membersif they would perceive the smell but was always denied it. This would give a transient sense of relief to her.She used a number of deodorants but of no help even after using them many times a day.As per the patient, this would be evident by a change in the behaviour of others rather than being actually perceived by her.For the past 2 years, she continued to have a variety of behaviour like taking frequent baths, using deodorants and fragrance soaps, and changing clothes multiple timesin an attempt to camouflage the smell but according to her, all of these are of no use. For the last 6 months, she has enrolled in a coaching centre but she is not currently not attending it because of fear that other students ridicule her for her smell. The patient doesnot report any direct confrontation or abuse by othersbut rather would interpret this from others' gestures. Gradually, over time she also became more withdrawn.The patient was treated with capsule of Fluoxetine 20mg/daywhich was gradually increased to 60mg patient showed a significant improvement in her symptoms.

II. Discussion:

Olfactory Reference Disorder is characterized by persistent preoccupation with the belief that one is emitting a perceived foul or offensive body odour or breath that is either unnoticeable or only slightly noticeable to others. Individuals experience excessive self-consciousness about the perceived odour, often with the ideas of reference (i.e., the conviction that people are taking notice, judging, or talking about the odour). In response to their preoccupation, individuals engage in repetitive and excessive behaviours such as repeatedly checking for body odour or checking the perceived source of the smell, repeatedly seeking reassurance, excessive attempts to camouflage, alter, or prevent the perceived odour, or marked avoidance of social situations or triggers that increase distress about the perceived foul oroffensive odour. The symptoms are severe enough to result in significant distress and impairment in personal, family, social, educational and occupational functioning[9].

A bad odour can be a presentation seen in psychotic disorders like delusional disorder (somatic type) and schizophrenia suggestive of olfactory hallucination which would be ego-syntonic unlike the cases of ORS. These become difficulty to differentiate if the patient have poor insight with ORS symptoms. Moreover, such behaviour can also be a part of other psychiatric disorders like: social anxiety disorder, anxious avoidant personality, body dysmorphic disorder, hypochondriasis and culture bound syndromes like *taijin-kyofushu*[6].

Among the medical conditions know, most common differential for ORS are- axillary, plantar, and/or genital bromhidrosis and hyperhidrosis. Skin conditions like dermatitis may also be present similarly due to irritation from excessive and improper use of hygiene products. Hyperhidrosis can co-occur with ORS and should be treated if present[8]. In a largest study by Greenberg et al. (2016) (N=253), the most commonly reported symptom was preoccupation of bad smells coming from two or more body areas. The perceived smells could be somatic (i.e., sweat, stool) as well as non-somatic (e.g., garbage, ammonia, metal, or stool from an unlikely source). The non-somatic smells may be associated with neurological conditions such as migraine headaches and seizure disorders though are seen to be associated briefly and not persistent for a prolonged time.

III. Conclusion:

Our cases also had a variable presentation with differentials of psychosis, depressive disorder and social anxiety disorder having normal physical examination and neuroimaging. There wasan absence of any dermatological or neurological abnormality which may present as a foul smell in some associated conditions. Moreover, in our first case, apart from presenting like a homeless person with severe mental illness, a social drift that was seen[10] which is majorly seen in chronic psychotic patients. Although, ORS is difficult to diagnose, they show a good response to anti-obsessional serotonergic agents and hence, should be considereda mainstay for the treatment.

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