Case Report

Successful treatment of olfactory reference syndrome with clomipramine

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Abstract

Subjective halitosis is a chronic disorder, and cannot be easily identified or measured by objective methods. The patients who are complaining with subjective halitosis usually refer to dentists, yet they reject to psychiatric consultations. One of the causes is olfactory reference syndrome (ORS) which is accepted as delusional or obsessive thoughts. ORS patients are usually refer to others' behaviours; people who are closing their mouth, coughing and touching their nose, opening window, turning faces to another side to protect themselves. These are the signs as to emitting malodour which patients misinterpret. This paper illustrates a 51-old male who suffers from halitosis since 30 years which is diagnosed as ORS and treated with clomipramine.

Keywords

Olfactory reference syndrome, Clomipramine, Subjective halitosis, Antidepressant

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Introduction

Halitosis is a term that defines chronic, endogenous malodour which is classified from type 0 to 5; which corresponds to physiological, oral, airway, gastroesophageal, blood–borne and subjective, respectively. [1] Subjective halitosis is featured by malodour that cannot be confirmed by others, further, despite the patient's complaints, there is not any local or systemic problem. Subjective halitosis cases may be misdiagnosed by specialists (including internal medicine, ear-nose-throat, endocrinology) as much as 27% indicated.[2] Subjective halitosis is defined in two clinical forms; psychogenic (anxiety, obsessional or delusional disorders including olfactory reference syndrome (ORS) and neurogenic (cacosmia-bad odour sense, phantosmia-imaginary odour sense, chemosensory dysfunctions).[3] Olfactory syndrome is a condition in which a person mistakenly believes that he or she emits an unpleasant body odour, but not perceived by others and is usually described by patients where this belief is usually accompanied by ideas of reference. In case reports and case series, as a result of embarrassment and suffering, individuals with ORS have been shown to engage in time-consuming rituals (repetitive behaviours, including sniffing the body, bathing excessively or attempting to mask the odour) aimed at masking or fixing the odour (usually originating in regions naturally associated with bad odour, like the mouth, armpits and genital/anal regions), avoid social situations, experience impaired work functioning, exhibit significant distress, report suicidal ideation and past suicide attempts and sometimes become housebound and social avoidance. [4] Odours are usually regarding the bodily originated ones. Uncommonly, patients have concerns on emitting non-bodily odours such as burned rags.[5] Or rotten onions.[6]

Despite the marked severity and impairment associated with ORS, empirical research on ORS is extremely limited. Sufferers frequently seek other health professionals (dentists, dermatologists, gastroenterologists, ear-nose-throat specialists), trying to solve the alleged problem.[7,8] At present, ORS is one of the conditions mentioned in the Diagnostic and Statistical Manual of Mental Disorders-5 (DSM-5)9 which is not categorized as a separate disorder.[10]

This case report illustrates a patient representing an ORS patient referred by halitosis clinics and evaluated, treated by a psychiatrist with clomipramine. In the literature, so far, clomipramine is shown to be effective in the treatment of ORS by this paper too. We suggest clomipramine would be useful in treating ORS patients especially with insight. In addition, this clinical situation must be in the attention of medical settings (psychiatrists, ear-nose-throat specialists and dentists) and may be kept in mind during clinical, treatment and differential diagnosisprocesses.

Case presentation

A 51-year-old divorced man who is directed/oriented by his mother to the psychiatry department with an increasing frequency of experiencing abnormal odour from his mouth since his childhood. He complained of – at least last two-year of constant smelling a very unpleasant oral odour that was like 'burning plastic mixed with rotting fish' had gradually enhanced. In the beginning, in first 2 years, the odour would increase in only/inly in leisure time at home. The intensity was quite less than the following years. Then this odour perception and the intensity became long-lasting and at all places he has been (all social and work areas). For the last 4 years, odour perception and restlessness became unbearable. So, he did not share the places where people has been/were. He became a "loner" sometime later. He resigned from his job and began to work on computer at home to earn his money and not to join public places. When he had to join some groups of family members or with people in the market places, he had chosen the latest times when the people were about to leave there. Some gestures of people precipitated the so-called odour he emitted. When he saw any person closing their mouth, itching or caressing their face and nose, giggling of people to whom his eyes contacted to them reminded his feelings and ideas of emitting odour. For the last year, he rejected to join his own family at home. He slept all day long and worked at nights so not to come across with family members. He did not share the places or times with them/his family. Especially, her mother worried about him and insisted to admit a professional help. Before his admittance to the dentist, his mother's persistence lasted for about 6 months. Mother's forceful motivation made him accept to apply, his dentist's evaluation did not reveal any loss of smell or taste. His dentist clearly told that this case was really a psychiatric situation. He accepted to admit our psychiatry department to cure his illness. Although long lasting of his illness (approximately over 30 years) and almost seemingly loss of insight, his willingness to psychiatric treatment was a big puzzling.

He was regularly brushing her teeth and tongue; he was also doing a 3-min mouth washing after every brushing. He was not a smoker, a drinker or taking any medications regularly. Any systemic problem, including postnasal drip, enteric parasite, constipation, gastroesophage al reflux, allergy were not detected. Halitosis examination was made with a previously described procedure.[11] Saliva volume was 2.5 ml/min, pH was found 6.5 and H_2S level was less than 100 ppb was found. Any dental caries, bad dental restoration, pathologic periodontal pocket, tongue coating were not detected. Cranial computerized tomography scan showed no pathology. Neither nasal nor alveolar air found odorous. He is diagnosed as Type 5 (subjective halitosis).

All workups with blood screen, cranial magnetic resonance imaging and electroencephalogram; neurological examinations did not reveal any pathological signs

indicating neurological disorders such as epilepsy. No discordance was disclosed by her family members.

The case was diagnosed as ORS regarding his history, symptoms and signs. So far, he has not applied to a psychiatrist before and had not taken any psychotropic medications.
His insight, easily convinced to have a psychiatric disorder and to accept the treatment made us to choose an antidepressant. Our experience with clomipramine[12] and case reports[13,14] with this agent made us chose this drug choice again. Clomipramine's twice Daily 75 mg was initiated and tolerated well. Although he showed approximately 40% relief after 1-month interval with this treatment. Addition of 75 mg clomipramine during the 2nd month interval resulted with 60% improvement. After 2-months, 225 mg/day dosage reached a good result, as a consequence he began to socialize. By the end of 3rd month, treatment reached 70% healing. An addition of 75 mg/day to 225 mg/day dosage (totally 300 mg/day), following the end of the 4th month, he felt very well indeed. Monitoring of him showed us; in two months, the odour so-called he had emitted diminished. During 3rd month he began to go outside with other friends, and gathered with his family in their meetings. By the end of 4th month, he did not think of others' gestures to remind him as he emits an odour and annoys people around him. Loneliness became a boring situation, he confessed.

After a follow-up of 10-months with 300 mg/day of clomipramine, revealed that his thoughts about others' behaviours (closing their mouth, itching or caressing their face nose, giggling of people when his eyes contacted) chanced thoroughly and did not remind him any of his illness.

Discussion

First description of ORS is made with 36 patients.[4] Core symptom was contrite reaction as emphasized. ORS begins nearly age of 20s. Those patients with ORS are usually deeply ashamed, embarrassed, self-abasing and sensitive reaction of people who believe that their bodies emit odour and think they are source of displeasure or disgust to people around them. They are restricted to their own area, avoid the social places, some show excessive behaviour to dispel the odour they emanate by changing clothes, using deodorants.15

Some other reports of ORS have indicated that the majority of cases have poor insight, while some cases feel that their beliefs and reactions are excessive.[16] In our case, in 30-year duration, patient had poor insight regarding his olfactory symptoms. Previous studies of pharmacotherapy for ORS have reported the effectiveness of antidepressants including tricyclics and serotonergic agents, including selective serotonin reuptake inhibitors (SSRIs) [16,1]7 and clomipramine. [12–14] However, although ORS beliefs are often delusional, treatments withSSRIs were found more efficacious than with antipsychotics.[7,18] In a study with

ORS patients, SSRIs are shown to be effective. On the other hand, some patients with such a condition have been shown to be treatment-resistant.[15]

In our case, symptomatic features are consistent with ORS definition and clinical presentation in other reports. The patient was restricted to his social and domestic excursion because of hisavoidance behaviour. Brushing teeth was not a ritual as seen OCD, since this behaviour was only to dispel odour. Although observation of depression or depressive symptoms in ORS patients, our case did not represent any of the depressive features. His avoidance and choosing to be a "loner" was accepted due to his complaints, not to depression. As indicated in the literature and in the nature of ORS, our patient has not admitted any specialist before. They had frequently consulted medical specialists,[4] a reported case underwent unnecessary surgery and was referred twice by surgeons,[8] but rarely received psychiatric treatment.

Most of the patients with ORS are males,[4] as our patient was. Many patients reported in literature were significantly younger and symptoms begin around age of 20 years. Our patient's symptoms also began 30 years before his admittance when he was 51 years old, in accordance with the onset of ORS clinics in reports and literature. Progress to schizophreniawas also described but our patient did not represent any psychotic social regression or withdrawal. His avoidance was only due to his odour problem, not a result of any psychotic feature.

Indeed, the diagnosis of ORS remains a challenge due to its overlapping features with other psychiatric disorders and is only mentioned briefly in DSM-V. More epidemiological and clinical studies are needed to determine ORS prevalence and diagnostic status, which is the first step to achieving evidence-based treatment approaches. In summary, further studies regarding symptoms, biomarkers and outcomes are needed to fully disentangle ORS from existingdepressive, anxiety and obsessive compulsive spectrum disorders. Thus further studies are needed to establish its diagnostic criteria and also to understand the illness better in order to treat as well as reduce the morbidity due to disease.

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