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## Case Study

### Persistent Halitosis Can Be A Part Of Olfactory Reference Syndrome – A Case Report.

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#### ABSTRACT

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Some halitosis cases are subjective, that are not confirmed by halitometer or third party individuals despite the patient insistently complaints malodor. They may be clinically related with neurological or psychological disorders. Although, they first appeal to dentist but not psychiatrist. This paper reports a halitosis case is diagnosed olfactory reference syndrome and treated by clomipramine. Oral health care professionals should be aware of such cases.

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**INTRODUCTION:**

Halitosis term defines a chronic condition which is an endogenous malodor that is classified into 5 types; corresponding to physiological (type 0), oral (type 1), airway (type 2), gastroesophageal (type 3), blood-borne (type 4) and subjective (type 5).<sup>1</sup>

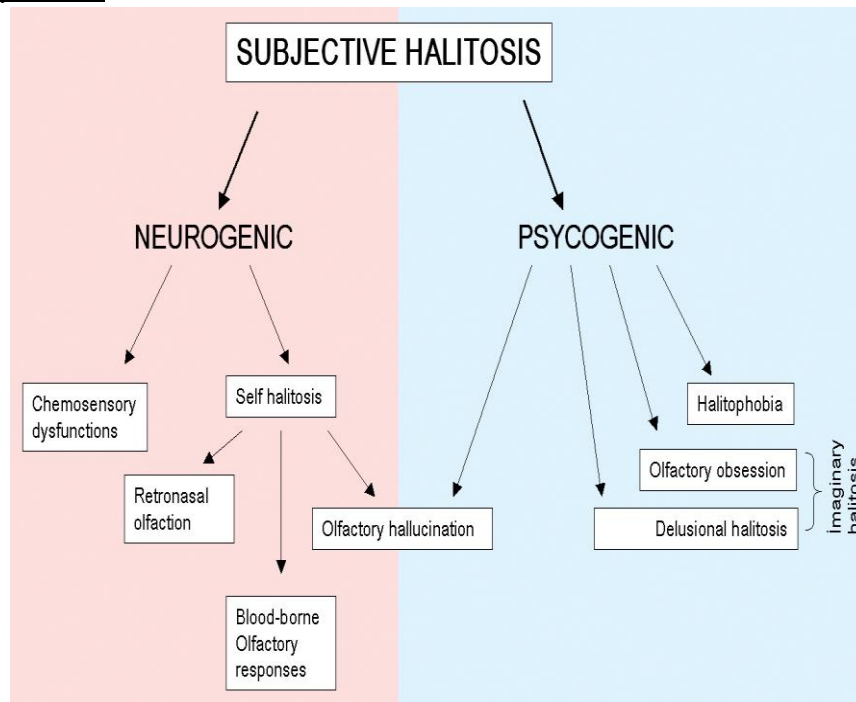
Subjective halitosis is any kind of malodor which cannot be confirmed by others in the environment of the patient, despite the patient’s complaints persist. There is not any defined local or systemic problem. As much as %27 of subjective halitosis cases may be misdiagnosed by the specialists of internal medicine, ear-nose-throat, endocrinology, dentistry.<sup>2</sup>

Two forms of subjective halitosis are defined: psychogenic and neurogenic. Psychogenic forms (chemical stimuli do not exist at sensors) may appear in several conditions, including anxiety disorders, olfactory reference syndrome (ORS), obsessional, phobic or delusional disorders. Secondly, neurogenic forms (chemical stimuli do exist at sensors) may originate from some disorders, including chemosensory dysfunctions or self halitosis (Fig.1).<sup>3</sup>

ORS is defined as the subjective condition in which a patient erroneously believes s/he emits an unpleasant odor although not perceived and confirmed by others. The so-called odor that the patient believes may originate from various body regions naturally associated with an odor like genital/anal regions, armpits, and mouth.<sup>4,5</sup>

This case represents an ORS patient referred by halitosis clinic and evaluated, treated by a psychiatrist with an antidepressant, clomipramine. Case reports and researches on ORS is extremely limited. This is the third ORS case which is reported and treated with clomipramine.

Figure 1. Subjective halitosis classification. In neurogenic forms of subjective halitosis, there is a real olfactory stimulation that can be at sensor level (self-halitosis) or perception level (chemosensor dysfunctions). This is lacking in the psychogenic forms that sequentially appear halitophobia, obsessional, or delusional disorders, including olfactory reference syndrome



*(The Figure has been copied from 3rd reference Özen ME Aydin M., Compend Contin Educ Dent. 2017;38 10):*

**Case Description and History:**

A 28-year-old single girl has applied to halitosis clinic with repulsive odor from her mouth. She complained non-stop smelling a very unpleasant oral malodor that was like 'rotting fish', sometimes dead rat, urine-like or garbage had continued since her childhood. Recent ten year, her halitosis became unbearable by her social environment despite nobody had verbally complained or warned her about halitosis. She did not feel malodor herself, but strongly believed to other people felt her malodor. According to her, other people were having touch or close their nose with fingers, or people turned their faces far from her. On the other hand, other people were not having halitosis despite they have never brush their teeth, she thought.

So, she avoided to be in crowds, whereby socially isolated from environment including her boy friend. She had visited more than a dozen medical professional for halitosis. Gastroscopy, rectoscopy, rhinoscopy had been made; probiotics, vitamins, colloidal silver, proton pump inhibitors, antibiotics, Candida diet, carbohydrate diet, numerous rinses have been prescribed; but all failed.

She brushed her teeth and tongue with a middle hard dental brush and correct brushing technique regularly twice daily. She did not take any medications, smoke or drink alcohol at all. She did not have any systemic health problems, including postnasal drip, enteric parasite, constipation, gastroesophageal reflux, allergy, bad taste on her tongue, sinusitis, xerostomia, pneumonia, neither degenerative diseases nor surgery anywhere on her body.

*Examination and diagnostic protocol*

Intraoral examination: any tongue coating, bad dental restoration, dental caries, gingival bleeding, pathologic periodontal pocket were not detected. Regular contours for 4 amalgam fillings are followed. No radiological abnormality has been found among all teeth. Her lips normally close her mouth within rest position, she did have nasal respiration habit. Unstimulated saliva pH, 7, volume 2 ml/min was measured.

She distinguished numerous kind of odor without interruption even one minute in the mouth. Her complains continued malodor even when her lips closed or when she took an ice piece in her mouth. She felt lesser intense halitosis when she was alone at home.

Measurement for halitosis level was made with a previously described procedure.<sup>6</sup> Oral gas concentrations were found 1.1, 0,0,0.4,8 ppm, intranasally 1.2, 0,0,0.4,10 ppm, alveolar breath 2.2, 1, 0, 0.4, 12 ppm for organic gases, NH<sub>3</sub>, SO<sub>2</sub>, H<sub>2</sub>S, H<sub>2</sub> respectively. H<sub>2</sub>S peak concentration was found 2.2 ppm by a cysteine (20 mMol) rinse (cysteine challenge test). This reflects the individual's oral halitosis production capacity. Cysteine challenge test is most accurate evidence of Type 1 halitosis than momentary halitosis reading.<sup>6</sup> The H<sub>2</sub>S peak shows 8 or 10 fold increments of initial H<sub>2</sub>S level in patients with Type 1 halitosis. In this case, initial H<sub>2</sub>S level increased only 5.5 fold (2.2 / 0.4). This eliminates Type 1 halitosis. Already, If this case had been Type 1 halitosis, her malodor perception would be decreased when put an ice piece in the mouth or when closed the mouth.<sup>7</sup> On the other hand, lack of bad taste on the tongue mostly eliminates chemosensor dysfunctions.<sup>7</sup> Organoleptic or chemical examinations for halitosis have not been attempted because of they may easily cause misdiagnosis.<sup>8</sup>

Uninterruptedly continuing more than one kind of malodor throughout long period (since childhood), malodor that is perceived by all people except herself, lesser intense halitosis when loneliness at home, lack of tongue coating and measurable malodor in oral, nasal and alveolar air, increment of initial halitosis less than 8-10 fold by cysteine challenge test, the core ideas "other people do perceive her halitosis except herself", "to have halitosis since childhood", "all people pay attention to keep far from me due to my severe halitosis", "people close their nose", avoiding participating in social activities due to halitosis, presence body malodor aside from halitosis, are having perfectly fit to Type 5 halitosis. The patient was diagnosed Type 5 (psychogenic form) halitosis and referred to a psychiatrist.

Workups with whole blood screen and electroencephalogram; neurological examinations did not reveal any pathological signs indicating neurological disorders such as a tumour and epilepsy. Menstrual cycle, sleeping time, feeding, cooperation, insight, emotions, cooperation and mental activities were found normal. When evaluated with her medical history by a psychiatrist she is diagnosed as ORS regarding her symptoms, and clinical signs. Easily convincing to have a psychiatric disorder and accepting the psychiatric treatment made us choose an antidepressant as a psychotropic agent. Although the long duration of illness

and social avoidance, it was recorded that her insight into the illness was well enough.

Case reports<sup>9-12</sup> with clomipramine experiences encouraged to chose this antidepressant in this patient. Initiation of clomipramine 75 mg twice daily was well tolerated. At the end of 4-week, she showed approximately 50% relief. Addition of 75 mg clomipramine (totally 225mg/day) in the second session resulted in 80% improvement. After 8-weeks, 225mg/day dosage resulted in a good response as if she had never emitted an odour and as a consequence, she began to socialize. Following the end of the 12th and 16th weeks, she recovered very well. Observation by her family and evaluation during check-ups showed; in 8-months, so-called odor she had emitted ended totally, she began to socialize and established social contact with other friends. She looked forward to family meetings. By the end of 10-months, she did not think of others' gestures to remind her as she annoys with the odor once she had emitted. She stopped to take clomipramine after 12 months. The drug dosage was decreased gradually in 2 months. She has no doubts of her bodily odors and does not think she emits any malodor.

## DISCUSSION

At present, ORS is classified in the "Other Specified Obsessive-Compulsive or Related Disorder" in DSM-5, This category patients who do not meet the full criteria of any specific obsessive-compulsive or related disorder, but with symptoms characteristic for these disorders.<sup>5,13-15</sup> ORS related halitosis cases can be classified Type 5 (subjective) halitosis,<sup>3</sup> falls between delusional halitosis (of imaginary group) and any other psychogenic forms (Fig.1).

Patients with ORS are usually deeply embarrassed, ashamed, self-abasing and sensitive reaction of people around them. The core symptom of this disorder is the contrite reaction. They believe that they emit bodily odors and think they are the source of disgust and displeasure to public and people in their around. They avoid social places and are seen to be restricted to their own area, exhibit significant distress, report suicidal ideation and past suicide attempts.<sup>16,17</sup> These signs just fit the case presented here.

The pattern of the symptoms was having very good accordance with previously defined psychogenic forms of subjective halitosis. All of above characteristics are the prominent symptoms seen in the psychogenic

forms of subjective halitosis cases led us to obtain exact diagnose for this case.

ORS may be confused with halitophobia. To separate both, these criteria should be aware, patients with halitophobia are not sure about their halitosis, they have a doubt whether their halitosis exists or not, and their halitosis has characteristics some interruptions along hours or days, another characteristic that is they usually close their own mouth but not refer other people close their nose. In this case the patient pretentiously asserted her halitosis uninterruptedly continued, and other people closed their nose. Halitophobia patients' halitosis does not decrease when they alone at home.

Another close syndrome to ORS is an olfactory obsession (OO). They have repetitive behaviors (changing clothes, using deodorants, rinsing or brushing teeth ten times every day) are prominent signs while ORS patients referring others, is the prominent sign. According to our clinic experiences, type 5 halitosis patients are found usually single or divorced people who wish to get married. They describe their halitosis by using this tems: "Other people (except patient) feel more than one malodor has never been interrupted even 1 minute, since my childhood" or " date i know myself"

Recently, a descriptive and comparative table has been published to lead dental professional to separate all these subjective halitosis forms from each other with a questionnaire.<sup>7</sup> Halitosis patients always firstly visit dental professions. They should be well-trained in and aware of subjective halitosis.

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