DOI: I0.5350/DAJPN20I0230209t

Olfactory Reference Syndrome: a Case Report

Emine Ebru Aksu¹, Haluk Aksu², Abdulkadir Tabo², Ömer Saatçioğlu³

¹Ass't Dr., ²Sp. Dr., ³Assoc. Prof. Dr., Bakırköy Prof. Dr. Mazhar Osman Research and Training Hospital for Psychiatry, Neurology and Neurosurgery

ÖZET

Olfaktör referans sendromu: Olgu sunumu

Olfaktör Referans Sendromu (ORS), kişinin bedeninin çok kötü bir koku yaydığını düşündüğü, bu yüzden insanların kendisi hakkında olumsuz düşünceler taşıdığına inandığı, sosyal ve iş yaşantısında sorunlara yol açan psikiyatrik bir bozukluktur. ORS, DSM-IV de sanrılı bozukluk somatik alt tipi altında ele alınmaktadır. ORS'si olan hastalar, sanrılı bozukluktan farklı olarak kokudan kendilerini sorumlu hisseder ve büyük utanç yaşarlar. Bu makalede, vücudunun kötü koku yaydığı hezeyanı ve bunun sebep olduğu belirgin depresyon belirtileri olan 32 yaşında bir erkek olgu sunulmuştur. Paroksetin ve olanzapin kombinasyonu ile gözle görülür düzelme göstermiştir. ORS, literatür doğrultusunda tanı ve tedaviye yönelik tartışılmıştır.

Anahtar kelimeler: Olfaktör referans sendromu, monosemptomatik hipokondriyak psikoz, sosyal fobi, sanrılı bozukluk, psikotik bozukluk, major depresyon

ABSTRACT

Olfactory reference syndrome: a case report

Olfactory Reference Syndrome (ORS) has been defined as a psychiatric condition characterized by persistent preoccupation about body odor accompanied by shame, embarrassment, significant distress and avoidance behavior. Patients often limit their public appearances and restrict their social and occupational encounters. In DSM-IV, delusions about personal odor are described as an example of the somatic subtype of delusional disorder. However, patients with ORS are different from delusional disorder. These patients feel themselves responsible from the smell and live a shame. In this article, we present a case of a 32 year old single male whose delusion of emitting a foul body odor has caused significant depressive symptoms. He showed remarkable improvement with a combination of paroxetine and olanzapine therapy. ORS is discussed in the light of literature with regard to diagnosis and treatment.

Key words: Olfactory reference syndrome, monosymptomatic hypochondriac psychosis, social phobia, delusional disorder, psychotic disorder, major depression

Address reprint requests to: Dr. Ömer Saatcioğlu, Ataköy 9. Kısım, A5-A Blok, D:6l İstanbul - Türkiye

Phone: +90-212-543-6565/1552

E-mail address: osaatcioglu@yahoo.com

Date of acceptance: April 30, 2010

INTRODUCTION

Ifactory reference syndrome (ORS) is a condition in which a person believes that his or her own body emits a foul odor and that others have a negative opinion about him or her because of this foul odor (1). Patients with ORS have a constant preoccupation that they emit a foul body odor, and are inclined to feel embarrassed and blame themselves. The main difficulty in the clinical evaluation of ORS is distinguishing delusions from preoccupations and odor hallucinations (2). ORS frequently begins at an early age and is more often encountered in single males (1). Pryse-Philips defined the disease as ORS and differentiated it from olfactory symptoms encountered in schizophrenia, depression, and temporal lobe epilepsy (1).

The state of thinking that the body emits a foul odor can also be encountered in other diseases such as schizophrenia, depression, and temporal lobe epilepsy.

Patients with these symptoms were said to have olfactory paranoid syndrome in early series of Westernorigin literature (3). Japanese patients with similar symptoms were said to have a condition known as tai-jin kyofusho or anthropophobia (4,5).

ORS phenomenology has been often discussed and examined under various terms in the literature: ORS bromhidrosisphobia (6), chronic olfactory paranoid syndrome (3), monosymptomatic hypocondiasis (7), and monosymptomatic hypochondriac psychosis (MHP) (8). MHP was first identified by Munro and is characterized by dysmorphophobia, delusions of parasitic infestation, or delusions of foul odor emanating from the body (9). There is no absolute distinction between ORS and MHP, and MHP falls under the somatic sub-type of delusional disorder in DSM-IV (10).

Recently, ORS has been said to be a variant of obsessive compulsive disorder with little insight or to resemble social anxiety disorder (11). Patients in

the OCD spectrum (12,13) include those with body dysmorphophobia (14,15), hypochondriasis (16) and pathological jealousy (17), with both obsessions and delusions of various types. Similar phenomenological and neurobiological properties are considered to exist in the response of disorders in this spectrum to Selective Serotonin Re-uptake Inhibitors (SSRI) (18).

In this article, a case of ORS in a young male patient, starting with the delusion of emitting odor and later accompanied by depressive symptoms, is presented and discussed in light of the literature.

CASE

The patient is a single 32-year-old male, university graduate, and the eldest of three siblings. It was learned that he had a minimum-wage job in a relative's workshop, was living with his mother and his siblings, and that his father had died of lung cancer eight years ago. He appeal to the Bakırköy Prof. Dr. Mazhar Osman Research and Training Hospital for Psychiatry, Neurology and Neurosurgery psychiatry outpatient clinic with the complaint that he was emitting a foul odor from his genital area and that for this reason he must have a sexually transmitted disease.

The patient's complaints began after he contracted molluscum contagiosum following a dubious sexual encounter a year and a half ago. Even though his sexually transmitted disease has been cured with treatment, he said that his disease persisted, he refused to accept physicians' reassurances and was constantly visiting several physicians. When no pathology was detected as a result of examinations and tests in urology, internal medicine, and dermatology, he was referred for psychiatric evaluation.

He stated that since he was emitting an odor from his genital area, he could not meet up with people, he could not leave his home, and he even had to quit his former job and was only able to work in a relative's workshop. He said that he could not use public transportation to get to work and could only go to work by walking 20 to 25 kilometers every day and that he had lost about 12 kilograms. He said he had a bath and changed his underwear five to six times a day and constantly put on perfume. He said that the odor was coming from his geni-

tal area, that he could not smell this odor but he guessed it to be like that of rotten eggs, and he thought that people could smell this odor and hinted to him about it somehow with their body language. He said that he constantly asked members of the household if could smell an odor and that his family was now fed up with these questions. He further said that people implied with their actions and words that he was emitting an odor, and when they did he immediately left the location. He complained that he could not have an emotional relationship, although he would very much like to. The patient said that he was basically experiencing a psychiatric condition connected to a physical disease and he was not surprised that he was emitting a foul odor and was constantly doing research about this on the Internet.

A psychiatrist he had previously gone to with similar complaints had started him on sertraline 50 mg/day and risperidone 2 mg/day, but he could not see any benefit therefrom. He had appeal to our polyclinic for depression, sleep disturbance, increased troubles, and continuation of his strong concerns about the odor.

No pathology was detected in his physical and neurological examinations. In his psychiatric examination, he was average for his age, he took care of himself well, and his speech was fluent and intelligible. His affect was depressive and troubled. His associations were normal and relevant. The content of his thoughts included references, ideas of worthlessness, and somatic delusions. No perceptual disorders were detected. There was no insight.

Concerning the patient's family history, it was learned that his sister had been treated in our hospital with a diagnosis of panic disorder. He stated that he did not use any alcohol or psychoactive substances, other than one or two cigarettes a day. The patient's hemogram, biochemical, thyroid function, hepatitis, VDRL and HIV tests were normal. Urine examination was normal and metabolite substances were not detected. No pathology was detected in the EEG and brain MR imaging. Inadequate and insecure personality features were in the foreground in the MMPI carried out, while obsessive personality features and psychotic findings with distinct inadequacies in relationships between persons were detected in the Rorschach test protocol. The Hamilton Depression

Scale (HDS) and Hamilton Anxiety Scale (HAS) points were 28 and 29, respectively.

The patient was diagnosed with major depression and olfactory reference syndrome and started on paroxetine 20 mg/day, pimozide 4 mg/day and biperidene 2 mg/day. Because an extrapyramidal system side effect developed with the use of pimozide and the patient could no longer tolerate the drug and the drug harmony was ruined, pimozide was discontinued and olanzapine 2.5 mg/day was started. As a full response was not seen within two weeks, the dose was increased to 5 mg/day. In an examination of the patient six weeks later, a distinct improvement was seen in his symptoms and his HDS and HAS points were detected as 11 and 13, respectively. The patient, whose odor delusion had lessened considerably, stated that he could now take long trips and felt more comfortable communicating with people.

DISCUSSION

In this case, the presence of odor delusions and the development of clinical symptoms with this delusion suggested olfactory reference syndrome. ORS has been a controversial disease since it was first identified, and has quite different etiologies in biopsychosocial terms (9). The diagnostic checklist prepared for ORS states that ORS is neither monosymptomatic nor monodelusional, and may be accompanied by hallucinations, depression, and delusions with paranoid content (6,10,19). The literature stresses, however, that these symptoms are encountered secondarily, rather than primarily (20).

Although Pryse-Phillips accepts that the odor symptom in ORS is a hallucination, it can be evaluated as a hallucination or a delusional disorder in certain cases. Recently, opinions that ORS is a delusion have become predominant. Just as seen in this case, reference ideas that can develop secondarily may accompany ORS (1,10). Our patient believed that he was emitting a foul odor and insisted that people hinted about this to him. Although isolated symptoms may be encountered in some patients (9), the literature states that ORS may be included within different well-known diagnosis categories over time and may also be encountered

in other psychiatric diseases (20). Given that different psychiatric cases may also develop in our case, the patient must be followed up in the long term.

ORS is said to be difficult to distinguish from depressive disease with basic symptom analysis. Depression can develop reactively to delusions in ORS (1). Depression developed secondarily to ORS in our case as well. Patients with ORS consider themselves responsible for the odor and thus experience a feeling of shame and embarrassment. These patients take a bath or shower and change their clothes more often than necessary and avoid social interactions (1). This situation is generally chronic and the patient's quality of life worsens dramatically. Some patients with untreated ORS may commit suicide (1,6,21). Consequently, cases with co-morbid depression must be evaluated properly in terms of suicide and treatment aimed at depression must not be overlooked. Furthermore, since obsessions may reach a level of delusion in obsessive compulsive disorder with little insight, it is important to exclude OCD in differential diagnosis (22). ORS differs from social anxiety disorder to the extent that the patient believes he or she is disturbing people by emitting a foul odor and does not consider these fears excessive or meaningless (23). The degree of the negative impact of ORS on daily functioning can be understood from the fact that our patient traveled long distances by foot, could not communicate with people even though he very much wanted to, and worked in a job that did not match his higher education degree.

Although ORS is a rarely seen somatic-type delusional disorder, it can also be encountered as one of the rare symptoms of a variety of diseases. The delusional structure of this syndrome, most frequently accompanied by depression, responds particularly well to pimozide treatment (24,25). Some patients respond to combination treatment (tricyclic antidepressant and phenothiazines) (9), while others respond only to antidepressant treatment (20). One must not forget that depression has an important place in ORS. If the drug prescribed causes side effects that the patient cannot tolerate, one must not forget that atypical anti-psychotic neuroleptics are an option. The clinician's full knowledge and evaluation of this syndrome will ensure effective treatment.

REFERENCES

- Pryse-Philips W. An olfactory reference syndrome. Acta Psychiatr Scand 1971: 47:484-509.
- 2. Thomas CS. Dysmorphobia and monosymptomatic hypochondrisis (letter). Am J Psychiatry 1985; 142: 1121.
- 3. Videbech T. Chronic olfactory paranoid syndromes. A contribution to the psychopathology of the sense of smell. Acta Psychiatr Scand 1966; 42:182-213.
- 4. Tanaka-Matsumi J. Taijin kyofusho: Diagnostic and cultural issues in Japanese psychiatry. Cult Med Psychiatry 1979; 3: 231-245.
- Takahashi T. A social club spontaneously formed by ex-patients who had suffered from anthrohobia. Int J Soc Psychiatry 1975; 21: 137-140.
- Sutton RL. Bromidrosiphobia. J Am Med Assoc 1919; 72:1267-1268
- 7. Bishop ER. Monosymptomatic hypocondriasis. Psychosomatics 1980; 21:731-747.
- Munro A. Delusional hypocondriasis: A description of monosymptomatic hypocondriacal psychosis (Monograph Series 5). Toronto: Clarke Institue of Psychiatry, 1982.
- 9. Munro A. Monosymptomatic hypocondriacal psychosis. Br J Psychiatry 1988; 153 (Suppl. 2): 37-40.
- American Psychiatric Association. Diagnostic and Statistical Manual of Mental Disorders. Fourth ed. (DSM-IV). Washington DC: APA. 1994.
- Lochner C, Stein DJ. Olfactory reference syndrome: Diagnostic criteria and differential diagnosis. J Postgrad Med 2003; 49: 328-331.
- 12. Jenike MA. Obsessive-compulsive related disorders: a hidden epidemic. N Engl J Med 1989; 321:539-541.
- Stein DJ, Hollander E. The spectrum of obsessive-compulsive related disorders: In Hollander E (editor). Obsessive-compulsive related disorders. Washington, DC: American Psychiatric Press, 1993, 241-271.

- 14. <u>Bizamcer AN, Dubin WR, Hayburn B. Olfactory Reference Syndrome</u>. Psychosomatics 2008; 49:77-81.
- McElroy SL, Phillips KA, Keck PE Jr, Hudson JI, Pope HG Jr. Body dysmorphic disorder: Does it have a psychotic subtype? J Clin Psychiatry 1993; 54:389-395.
- 16. Fallon BA, Javitch J, Liebowitz MR. Hypochondriasis and obsessive compulsive disorders: overlaps in diagnosis and treatment. J Clin Psychiatry 1991; 52:457-460.
- Stein DJ, Hollander E, Josephson SC. Serotonin reuptake blockers for the treatment of obsessional jealousy. J Clin Psychiatry 1994; 55:30-33.
- 18. Zohar J, Insel TR. Obsessive-compulsive disorders: psychobiological approaches to diagnosis, treatment and pathophysiology. Biol Psychiatry 1987; 22:667-687.
- Munro A. Monosymptomatic hypocondriacal psychosis. Br J Psychiatry 1982; 141: 344-349.
- Cashman FE, Pollock B. Treatment of monosymptomatic hypocondriacal psychosis with imipramin. Can J Psychiatry 1983: 28: 85.
- 21. Bebbington PE. Monosymptomatic hypocondriasis abnormal behaviour and suicide. Br J Psychiatry 1976; 128:475-478.
- Stein DJ, Le Roux L, Bouwer C, Van Heerden B. Is olfactory reference syndrome an obsessive-compulsive disorder?: Two cases and a discussion. J Neuropsychiatry Clin Neurosci 1998; 10:96-99.
- 23. Tada K, Tojima T The relationship of olfactory delusional disorder to social phobia. J Nerv Ment Dis 2002; 190:45-47.
- 24. Sayar K, Aksu G. Olfaktör referans sendromu: Bir olgu sunumu. Klinik Psikofarmakoloji Bülteni 2003; 13:183-187.
- Ulzen TP. Pimozide-responsive monosymptomatic hypocondriacal psychosis in an adolescent. Can J Psychiatry 1993; 38:153-154.