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Apropos of an Olfactory Reference Syndrome case

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Olfactory Reference Syndrome (ORS) is one of the varieties of the somatic type of the Delusional Disorder, and it is characterized by the mistaken statement of a patient who declares the issuance of a foul odor coming from his own body and that others may notice. In the upcoming edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-V) it has been proposed to break off ORS as an independent pathology. From an illustrative case report, we review the relevant literature and discuss this proposal.

Key words: Delusional disorder somatic subtype, Olfactory hallucination, Hypochondriasis, olfactory reference syndrome

Actas Esp Psiquiatr 2012;40(4):234-8

Acerca de un caso de síndrome referencial olfatorio

El Síndrome Referencial Olfatorio (SRO) es una de las variedades del tipo somático del trastorno delirante y es definido por el juicio falso que el individuo afectado defiende respecto a la emisión de un olor fétido proveniente de su propio organismo y que los demás pueden advertir. En la venidera edición del Manual Diagnóstico y Estadístico de las Enfermedades Mentales (DSM-V), se ha propuesto desgajar al SRO como una patología independiente. A partir del reporte de un caso ilustrativo, revisamos la respectiva bibliografía y comentamos tal propuesta.

Palabras clave: Trastorno delirante tipo somático, Alucinación olfativa, Hipocondriasis, Síndrome referencial olfatorio

INTRODUCTION

It is assumed that smell, on the contrary to sight and hearing which are more developed sensorial capacities, has less importance. Thus, the psychopathology related to this sensory sphere is usually overlooked.¹ A conspicuous example is the Olfactory Reference Syndrome (ORS) that was first reported in 1891 and whose presence has been verified in all of the geographic latitudes. However, it currently does not have an independent status in the diagnostic classifications.² Although Kraepelin had doubts about the existence of a somatic type among the paranoidias, since the middle of the last century, the literature on this type has been the most abundant regarding that of other delusional disorders.³ In the ICD-10, ORS is mentioned among the disorders of delusional ideas. However, in the North American classification edited in 1980 (DSM-III), it was considered as part of an atypical somatomorphic disorder. In the DSM-III-R edition, ORS was transferred to the somatic type of delusional disorders, where it has remained up to now.⁴

The name of Olfactory Reference Syndrome is owed to Pryse-Phillips who, in 1971, published a large series of cases where he individualized it in regards to other pictures in which there were similar symptoms but within schizophrenic, affective or organic psychoses.⁵ Although there is consensus on the core symptoms of ORS (the vehement concern about bad body order), there is still controversy regarding whether this proposal has a delusional character in all of the cases and if there is a hallucinatory correlate to such idea. In fact, in the most recent review on the subject, a meager minority of cases was found (22%) in which the undeniable presence of olfactory hallucination is reported (versus 75% in the original series of Pryse-Phillips). Furthermore, delusional conviction was only observed in little over half of the cases (52%), while in the remaining patients, such idea oscillated between from overvaluation and obsessiveness.⁴

The smells observed by the patients are commonly body ones or those of natural emission: axillary, genital smells, those from perspiration, urine, stools or burps, and halitosis, but they are described as extremely intense.⁶ Another

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prominent symptom of ORS is feeling embarrassed: more than 75% of the patients interpret gestures and words of others as referential - up to the most outlandish type. This finally leads them to withdrawal and occupational and social incapacity in a considerable proportion. In the Pryse-Phillips series, hardly 3% of those affected had an active social life.⁴ The long periods of time they use to check the supposed odors and other compulsive behaviors, such as dissimulating their presence, also influences the severe incapacity of the patients.⁷

Regarding the demographic data, the prevalence of ORS is usually greater among males with predominance of single persons, and the age of onset covers from the end of adolescence to the beginning of adulthood. In almost half of the cases, a precipitating event is described before the onset of the symptoms, generally some disapproving reprimand, although this could be an early event of already-established morbid course.⁸

In approximately 40% of the ORS cases, comorbidity has been declared with anxiety or depression (mostly of subsequent appearance to the complaint of unhealthy odor). Furthermore, up to 30% of the patients make some suicide attempt and more than 50% require psychiatric hospitalization.⁴ In regards to personality traits, the most frequent correspond to group C of the DSM-IV (avoidant, dependent, and anancastic).⁴ One section that has been little explored is the presence of psychiatric family backgrounds in patients having ORS, although it could be indicated that there is meager evidence linking ORS with schizophrenia as a precipitating factor.³

It is known that the prevalence of delusional disorder of all types ranges from 0.05 to 0.1%. However, a vast subregistry is presumed, even more so in cases of somatic delusion disorder and in developing countries.⁹ It is obvious that the majority of patients having ORS do not consult a psychiatrist but rather odontologists, dermatologists, gastroenterologists and even surgeons. This distorts the frequency figures from our psychiatric medical samples.¹⁰ There are no numbers on the populational prevalence of ORS. An interesting datum comes from Japan where, as part of a picture called *taijin-kyofu* ('*taijin*': interpersonal; '*kyofu*': fear), a variety is described (*jiko-shu-kyofu*) in which fear of having offensive personal odors predominates. Close similarities have been described between *taijin-kyofu* and generalized social phobia.¹¹ There are no epidemiological data on these conditions, although the diagnosis of *taijin-kyofu*¹¹ prevails in 7.8% of the Japanese neurotic patients.

Hypotheses have been made on serotonin and dopamine dysfunctions secondary to glutamate and oxytocin control as a cause of ORS, by analogue with some repetitive behaviors of cleaning and checking similar to some of those of the obsessive-compulsive disorder in addition to disorders of regulator genes such as Hoxb8 and SAPAP3, translated to

predominance of basal lymph nodes and limbic lobe and that govern grooming behavior in animal models.¹² There are isolated and unequal reports of images obtained by SPECT.^{7, 13} Without counting the reports of cases secondary to cerebral lesions and epilepsy of the temporal lobe,^{14, 15} currently there is no certainty about the neurobiological bases of the primary ORS. The psychodynamic sign that is consistently linked to the olfactory hallucinations with guilty feeling have also not been studied.¹⁶

Because of its chronicity and progressive worsening, the prognosis of ORS was usually conceived as unfavorable.¹⁷ However, the recent review by Begum and McKenna -which only included 84 cases of the 180 collected, reported that up to two thirds of their patients partially improved or recovered. The positive outcome of ORS was recorded in 33% of the patients who received antipsychotics and 55% of those who received antidepressants. These data, at first, would contradict the identity of ORS as a delusional-rooted disorder. Even in the same review, most of the successes occurred with psychotherapy alone (78% of those who received it).⁴ Along this line, another interesting piece of data is the successful treatment of five cases of ORS by Desensitization and Reprocessing of Ocular Movements since this therapy has not been useful previously in psychotic pictures.¹⁸

It has recently been suggested that ORS should be separated as an independent disorder and *ad hoc* criteria have been proposed for its diagnosis in the fifth edition of the North American Classification of mental disorders (DSM-V).^{2, 6, 19} (Table 1).

CLINICAL CASE

A 35-year-old male, computer technician, married and father of two children. His wife usually reprimanded him about his hygiene and about two years ago he began to observe an especially foul-smelling odor from his feet. At first, he only thought that this was perceived by the persons in the home, but he gradually observed that this odor was more intense. Since nine month ago, he has observed that in the street and in his work center, several persons have felt uncomfortable about the odor from his feet. They demonstrated this by shifting their eyes away, touching their nose, clearing their throat or moving away from him. He even believed that they whispered behind his back. On the bus, he sat in the back seats and refused to attend social affairs. In his work site, he generally went to the bathroom to wash and change his socks, since the odor from his feet was more repulsive when he wore shoes. On the other hand, with sandals and at home, it was milder, and also when he slept. He also noticed that when he was with women, because he felt more anxious, the odor was more intense.

He often washed his feet with kerosene and bleach, as recommended in internet and applied boric acid, alcamphor

Table 1

Diagnostic criteria proposed for the Olfactory Reference Syndrome⁶

- A. Preoccupation with the belief that one emits a foul or offensive body odor. Said odor, in reality, is not perceived by others.
- B. The preoccupation causes clinically significant distress (e.g.: anxiety, dysphoric mood, shame) or occupational, social impairment or of other important areas of functioning.
- C. Preoccupation is neither a symptom of schizophrenia nor of another psychotic disorder and it is not due to a direct physiological effect of a substance (e.g. a drug or of a medication) or to a general medical condition.

Specify if:

1. Good or regular disease awareness (insight): The individual admits that the belief of ORS is definitively or probably false, or that it would be equally false or true.
2. Poor disease awareness (insight): The individual states that the belief of the ORS is probably true.
3. Delusional belief regarding the bad body odor. The patient is completely convinced that the belief of the ORS is true.

or zinc oxide on his feet. He even put his shoes in the refrigerator at night to "kill the fungi." Our patient stated with total conviction that he could perceive his odor - "of dirty socks, as old cheese," which this could be extended up to six meters away and he assumed that it was due to some skin condition of his feet.

He was so overwhelmed by this problem that it decreased his work performance. He was distracted as he was besieged by the idea that his foot smelliness mortified others. His mood changes were generally fluctuating but one month prior to his admission, his significant other cancelled the sentimental relationship and left with one of their children. The patient became tearful and jumped from a second floor on some junk, apparently during a twilight state. He was hospitalized.

On examination, we found a well-dressed, alert person who appeared younger than his real age and whose speech was fluid and coherent. Affectively, he was concerned and even helpless but with an easily reactive mood during the interview. The suicidal ideation had disappeared. His speech was monotone about the false opinion on the odor of his feet, an opinion that displayed delusion belief, and on his olfactory hallucination - both phenomena that went uncriticized by him. He did not detail other hallucinations or show formal thought disorders. He also had no disorders of the self-awareness. Cognitively, he was unharmed and had normal level of awareness. There were no abnormalities in the physical examination or routine analysis, no in the electroencephalogram and brain tomography. In the psychological study, a dependent and insecure personality was observed with affective immaturity. His I.Q was 89.

The patient was the youngest of six siblings on his mother's part (of these, one sister became schizophrenic and another alcoholic with recurrent depressions and several

suicide attempts). He came from a fatherless home, where the mother was a rude and dominant figure. His personality was signed by submission and lack of self-confidence. He never left his mother's home and lived there for 15 years with his significant other, described as "obsessive for cleanliness." She made all the decisions for him.

Given that he did not fulfill the criteria for schizophrenia or affective disorder and also organicity, he was diagnosed of somatic-type delusional disorder (according to the DSM-IV), and treatment was initiated with 600 mg. of sulpiride and 20 mg. of fluoxetine daily. At five weeks, he was discharged with the desire to reincorporate into his activities and he manifested that he almost did not perceive the supposed odor of his feet. In the follow-up at six months, he had remission of the olfactory delusion, although he preferred to refer to it as "I was much better." He had returned to the relation with his significant other and to work.

DISCUSSION

Although reports have been made over the last century that provide a consistent description of ORS, its nature - described as being halfway between delusional disorders, social phobia, obsessive-compulsive disorder and body dysmorphic disorder, requires a clearer definition.²⁰ A fundamental problem is to outline the nature of delusion. In Anglo-American psychiatry, its conception is restricted to a mistaken judgment while Continental European Psychiatry also includes ideo-affective and perceptive phenomena (intuitions, interpretations, illusions, passion and imaginative exaltation) within the delusional phenomena.¹⁶ In this sense, North American psychiatry stresses the essence of the delusion in the convictional certainty and absence of insight,

thus eluding other characteristics not related with the above such as extension, extravagance, pressure, affective response and influence on the behavior.²¹ In such a concise way, delusion easily overlaps with the overevaluated idea. In this respect, only as an example, a recent study could be mentioned. This study did not report any difference of conviction or insight between overevaluated ideas and delusion.²² Up to what degree could the description of the delusional forms and non-delusional ones of body dysmorphic disorder or those of ORS be due to this confusion - and to the lack of attention to the heterogeneous nature of the delusions, that are assumed to be homogeneous - is a pending question to be clarified. For such purpose, correct ideas of overevaluated ideas and delusion should be considered.²³

Another point of debate is the postulated belonging of ORS to the "obsessive-compulsive spectrum."⁷ Reviewing the case series, concern about bad body order has been categorized as obsessive in only a minority of patients, so that the malaise could be generalized based on the exception. Bürgy, in an astute analysis,²⁴ criticized the trivialization of the concepts of obsession and compulsion and defended the necessary distinction of obsessions per se. It is well to remember at this point that in order to establish a sign at the expense of another as representative of a nosographic category (delusion or hallucination for the category of psychosis, obsession or compulsion for the obsessive spectrum) that the psychopathology cannot be easily understood analogue to a medical semiology. The reason is that in this, the "signs" refer to as natural objects that retain their meaning independently of the context of the patient while in psychopathology the concepts refer to fragments of human experience that lose their meaning on being taken out of their context.²⁵ It is sufficient to review detailed series such as those of Pryse-Phillips and Videbech^{5, 26} in which most of the ORS cases corresponded to sensitive delusions of Kretschmer: developments per se in which the mind of the patients is shifted from the reality to the influence of the intentionality prevalent in the spirit of the subject. Unfortunately, great advances in the study in this line have not occurred.

It has been stated that the proliferation of the entities in the diagnostic classifications, where each disorder becomes a disease by its own right and differentiation is even presumed in pathophysiology, epidemiology and response to treatment, is inadequate. Such frenzy ends up divesting the diagnoses of validity.²⁷ In this sense, it seems recommendable that the criteria proposed for ORS should be limited to the appendix of the DSM diagnostic classification as a tool for future investigations.^{4, 28}

Other sections having heuristic value correspond to the overlooked pseudoperceptive component of the ORS. The impossibility of representing olfactory sensations (olfactory

imagery) shared in common by the individuals has been described. This is coupled to the fact that olfactory hallucinations are the most prevalent in the normal population.²⁹ In addition, there is a latent role of the olfactory neurons in the social interaction since they are unique, among the sensory, in having direct access to the prefrontal cortex and in deeply participating in such circuits.¹

We consider that the case of our patient, whose delusion and supposed olfactory hallucination globally dispelled in less than one month in spite of their initial severity, is illustrative of the peculiar psychopathology of the ORS³⁰ and similar pictures (such as dysmorphophobia delusions and delusional parasitosis). It is necessary at this point to reflect on hypochondriasis as a core nucleus of the somatic-type delusional disorders. Keep in mind that in European psychiatry, it was called monosymptomatic hypochondriacal psychosis before returning conceptually to the diagnostic classifications with their current names.³¹

Attention is drawn, to mention only some of the differential characteristics, to the fact that somatic delusions globally affect the life of those who suffer them while in the other types of delusional disorders, the affection is limited. Equally, these patients generally seek medical care repeatedly - however not psychiatric, of course.³ Additionally, in the somatic delusional pictures, the suspicious or irritable affectivity characteristic of the rest are not verified, but rather feelings of humiliation or shame predominate, for which greater phenomenological scrutiny is needed.^{32, 33}

Hypochondriasis, more than mere somatomorphic disorder, is a special form of experiencing life, of being in the world, a transformation of vitality and qualities of corporality. This can be conceived as a category of pathological attitude towards "oneself."³⁴ Jaspers correctly points out that the essential criterion that distinguishes the different forms of belief lie not in their conviction and certainty not in the impossibility to correct them or impossibility of their content but in their origins within the experience of the patient. Thus, our body, which is the only part of the world that we can feel from within and on its surface and can also be perceived by us, has complex and irreplaceable phenomenological qualities.³⁵ We conclude that ORS, as the rest of somatic delusional disorders, merits a close and critical review prior to giving it diagnostic emancipation.

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