CASE REPORT

EBBOM’S SYNDROME: TWO CASE REPORTS TREATED WITH OLANZAPINE

Roberto Poli, Emilia Agrimi

Abstract

Ebbom’s syndrome, a rare psychiatric condition characterized by the delusional belief to be infested with some kind of parasites, may be divided in primary, secondary to another psychiatric condition or caused by a medical illness, medication or substance abuse. An elderly woman, 78, widow, showed abrasions on her body and scratching-induced lesions that dermatologist denied for a parasitosis but the patient did n’t feel reassured neither changed her opinion. Olanzapine, prescribed at the starting dose of 5 mg/day, increased to 10 mg/day well tolerated and after a month reduced anxiety, sleeplessness, lack of appetite while tactile and visual hallucinations disappeared. A woman, 62, was a relapse of frontal and ala temporalis meningioma. The patient was referred to the psychiatric assessment by her dermatologist, as she stated to be affected by diffuse skin parasitosis (report of “parasitophobia” and “leg vascular ulcer”). Delusional thought was well organized. No cognitive deficiencies. Treated successfully with olanzapine.

The cases reported have the typical characteristics of Ebbom’s primary and secondary syndrome. The symptomatic and behavioural manifestations are equally typical: acute onset, tactile and visual hallucinations, scratching-induced lesions and various dermatologic examinations. Olanzapine shows good compliance and remission of psychiatric symptoms after two months of treatment.

Key Words: Ebbom’s syndrome, delusional parasitosis, delusional disorder, dermatozoic delusion

Declaration of interest: None

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Introduction

Ebbom’s syndrome is a rare psychiatric condition, characterized by the delusional belief to be infested with some kind of parasites. This condition has generally an acute or sub-acute onset, usually in senile or pre-senile age, but in absence of mental deterioration. This syndrome is also known with different names such as delusion of infestation or dermatozoic delusion and parasitophobia. Anglo-Saxons define this disorder also delusional parasitosis in the 45 cases described in 1946 (Wilson and Miller 1946). According to the nosographic classification of DSM-IV-TR, the corresponding diagnostic framing is “delusional disorder, somatic type”.

However this clinical picture has its own features which account for the need of a description apart.

Ebbom’s syndrome is named after the Swedish neurologist Karl Axel Ebbom in 1937 described 7 cases of this delusional disorder. In 1674 Sir Thomas Browne described the “Morgellons disease” (Kellet 1935), an entity in which the patient excavates the epidermis and dermis to retrieve the causative “threads”. The major differences between parasitophobia and Morgellons disease is the general absence of malaise, fever or systemic symptoms in parasitophobia and the not unusual finding of these in Morgellons disease, in contrast with the bugs and the “matchbox” sign in parasitophobia (Feller 2009).

Other observations of cases with overlapping characteristics date back to 1894 with Thibierge (Thibierge 1894) who classified them as “acarophobia”.

Ebbom’s syndrome is divided into three subgroups: primary; secondary to another psychiatric condition, usually schizophrenia or depression; and secondary to a medical illness, medication or substance abuse. This disorder is considered rare, even if it is possibly underestimated, since many cases escape the psychiatrists’ observation and are seen by dermatologists (Szepietowski 2007). The referral to a psychiatrist is often ignored by the patient.

Various hypotheses have been formulated about...
the etiopathogenesis of this disorder, which are mostly based on a multi-factorial model where organic, psychological and environmental factors play a role. It has been recently assumed a specific function deficit of dopamine active transporter (DAT) in the corpus striatum, with a consequent increase of extracellular dopamine in the corpus striatum (Huber et al. 2007).

Clinical case A

Woman, 78, suffering for hypertension along 15 years, treated with beta-blocking antihypertensive agents. No other illness or hospitalisation reported. Not previous contact with psychiatry and not familiarity with psychiatric disorders.

The patient comes to the her first psychiatric examination urged by her son, her sister and her primary care physician, but states she does not need any psychiatric help. She is well oriented in space-time parameters and she doesn’t show any cognitive deficiencies; both her short-term and long-term memories are good. Her speech is fluid and consistent and her gestures are proper and lively. She believes that her house and her body are infested with parasites: her main belief is she affected from scabies, but she does not rule out the possibility of other kinds of parasites. She shows various parts of her body attacked, on her opinion, by the infestation: actually some abrasions, ulcerations and hyperpigmented and hypopigmented scars can be observed, all undoubtedly scratching-induced lesions. She admits to feel an intense itch and to scratch herself, and firmly exhibits spread red spots on the skin of the arms, legs and torso, which could be the evidence of a contact with parasites, of which she can even feel the bites by touch. She has already been examined twice by dermatologists, who ruled out a parasitosis, but the patient has not felt reassured and has not changed her opinion.

Moreover, she reports to have seen small unidentified fibres spread all over the house and even on mattresses, steadily interpreting this fact as an evidence of the presence of insects moving the textile.

The patient is highly worried about this “invasion” of parasites into her house and has already arranged disinfections by part of specialized companies for three times, with unsuccessful results for her. She spends a great part of the day to look for parasites and uses insecticides rashly. She isn’t absolutely critical about what is occurring and accuses her relatives and physicians not to understand the situation and to consider her “crazy”. Her son states the situation has become intolerable since when her mother started to call for him continuously, saying the house is more and more invaded by insects and that she can no longer stay there. In the moments of greatest discomfort, she has even thought that the only solution is to go away from that house. The idetic polarization on this experience has also induced serious problems of sleeplessness and hyporexia.

In spite of the initial refusal for the psychiatric referral, the patient shows some confidence, due to the attention she receives and for the absence of critics or contestations to the facts she is reporting. The patient is then advised – in order both to rule out a secondary disorder and to find a better therapeutic alliance – to carry out hematoclinical tests, a brain CT scan and a neuropsychological assessment. The patient accepts to come back for a second interview with the results of the tests. The brain CT scan is negative and the neuropsychological tests give age-related normal results. The hematoclinical tests do not show any significant abnormalities.

The patient, though retaining a steady position about her delusional belief, accepts a drug therapy following the assumption that some “emotional factors” could support the accompanying symptoms she is complaining of.

Olanzapine is prescribed at the starting dose of 5 mg/day, with following increases up to 10 mg/day after 2 weeks. After one month of treatment, anxiety, agitation, sleeplessness, and lack of appetite have completely disappeared, as well as the total polarization of the patient’s thought on parasitosis. The patient has gone back to her activities of daily living, and conflicts with her relatives have ceased. She has resumed regular sleep and nutritional patterns. She does not use disinfectant agents any more and does not think about leaving her house. In addition, tactile and visual hallucinations have disappeared; the patient has actually admitted she does not see any signs of parasites either on herself or in the environment and she does not feel any insect bites any more. However, she has not been able to review the incident critically, keeping the belief she had been infested with parasites.

The follow-up visits after 2, 4 and 6 months confirmed the patient’s perfect well-being.

Clinical case B

Woman, 61, unmarried, any previous contact with psychiatry; a brother suffering for schizophrenia. She lives alone in a small village in the Province of Cremona, Italy. Educational level: Degree in Foreign Languages; she has been a speech therapist till the age of 57. Her pathologic history shows that the patient was submitted to surgical procedure for pituitary gland adenoma when she was 20 and subsequently to radiotherapy; then she started hormone replacement therapy.

In 2003 the patient was submitted to another neurosurgical procedure for multiple brain myelomas and subsequently to radiotherapy with an outcome of almost complete blindness. In 2005 there was a relapse of frontal and ala temporalis menigioma, presently under monitoring. The patient is referred to the psychiatric assessment by her dermatologist, as she states to be affected by diffuse skin parasitosis (report of “parasitophobia” and “leg vascular ulcer”).

At the interview the patient seems quite perplexed in describing her symptoms to the psychiatrist, but she is anyhow cooperating; she is oriented in space-time parameters, and she does not show any cognitive deficiencies. She looks shabby, with scratching-induced skin lesions on her face and limbs; moreover, some pieces of paper soaked wit water can be observed on her skin – the patient explains that she rubs her whole body with them, as an effective method “to clean out parasites”. She reports she has been suffering from such
a parasitosis for two years, since when, after an accidental fall, she got an abrasion on her leg, which became “infested” – according to her words - with soil contaminated with parasites which “spread all over her body”.

From a temporal point of view, there is a clear correlation between the meningioma relapse and the onset of the psychiatric condition. According to the clinic documentation and objectiveness, the patient results to have a leg vascular ulcer with serious inflammatory reaction in the surrounding tissues, which is worsened by the cleaning practices with antiparasitic products and camphor carried out by the patient herself. She sought the advice of various dermatologists who gave her a number of indications of treatment but who, as the patient comments, “were not able to find the parasites”. She tells the parasites are continuously moving under her skin, and that only by rubbing she can take them off; sometimes they go out from her ears in great numbers and she can’t have rest by night. Moreover, she states she is really surprised that “the others” cannot see such a large amount of parasites that she picked up in a small box to show to her dermatologist in the past. She is tired and wants to be hospitalized in order to solve such a troublesome problem. She accepts the hospitalization in the psychiatric ward, without making questions on the ward itself. When her attention is drawn about the strangeness of the symptoms she describes and about the absence of objective and clinical findings of a parasitosis, she does not seem to perceive the meaning of the statement and ritualizes her own delusional beliefs.

The psychopathologic picture is characterized by a well-organized delusion, which doesn’t apparently affect the other domains of psychic functioning. The emotional involvement with delusion is inadequate.

In the ward the patient is submitted to the routine hematochemical tests, which are within the normal range and to neurosurgical assessment for the relapse of brain meningiomas. The brain MR recently carried out at the out-patient’s service is seen by a specialist and a follow-up is advised after 6 months.

A drug therapy is started with olanzapine at the dose of a 10 mg/day that the patient accepts, interpreting it as a “support to stand that annoyance”.

The follow-up examination after 2 months confirms the good compliance with the drug therapy and the disappearance of the delusional thought concerning the parasite infestation.

Discussion

The cases here reported have the typical characteristics of Ekbom’s primary and secondary syndrome (Caimi et al. 2003). Both patients are women, live alone, have no previous psychiatric history and show a late onset of psychotic symptoms without signs of cognitive impairment. Advanced age of onset, feminine gender, absence of psychiatric history and social withdrawal are all elements indicated in the literature as typical of Ekbom’s syndrome. The symptomatologic and behavioral manifestations of the disorder are equally typical: acute onset, tactile and visual hallucinations, scratching-induced lesions, various dermatologic examinations and frequent requests of disinfestations by part of specialized companies. In the case B was also present the typical “matchbox sign” while in both cases was absent the “contagiousness” in which the patient is so convincing that he shares delusion of infestation by his familiar (Bourée et al. 2007). The lacking elements – which was not possible to detect in case A – is the presence of a triggering factor.

In the face of such dermatologically defined symptomatologic picture, we think however it is always advisable to accurately investigate and explore the patient’s history and accomplish the in-patient workup in order to rule out other psychiatric conditions or disorders caused by a medical illness, medication or substance abuse. The recent literature (Steinert and Studemund 2006), in fact reports a case of Ekbom’s syndrome acutely induced by a ciprofloxacin treatment, with regression of the disorder at the drug discontinuation.

The adherence to the delusion was complete in both cases and both patients were very reluctant to accept the psychiatric examination and a psychopharmacological therapy. However, after a first stage of diffidence, a good therapeutic alliance was established, which determined a good compliance to the psychopharmacological therapy.

About the choice of the antipsychotic therapy, though retaining pimozide as a reference benchmark, case-reports and case-series of patients successfully treated with atypical antipsychotic agents: olanzapine (Freudemann et al. 2007, Mehan 2006), but also risperidone (Pacan et al. 2004), aripiprazole (Rocha and Hara 2007) and amisulpride (Lepping et al. 2005) have been published in the last years. Olanzapine effectiveness is notoriously associated to a better side-effect profile versus neuroleptics: mainly absence of extrapyramidal effects and QTc non-prolongation. On the other side it is necessary not to underestimate and to assess metabolic risks carefully, in particular bodyweight gain and glucose intolerance.

In both cases our choice was olanzapine due to its good sedative action and its good effectiveness in the control of sleeplessness, since neither patient showed signs of metabolic risk.

Olanzapine effectiveness resulted to be excellent at the dose of 10 mg/day, while in literature cases have been reported with symptom resolution at lower dosages, in consideration of the usually advanced age of the patients suffering from this kind of pathology.

In conclusion, we can state that, from the effectiveness point of view, the cases we have described seem to indicate, like others described in literature, that olanzapine can be a first-choice drug in the treatment of Ekbom’s syndrome. Further data and randomized clinical trials are obviously required to confirm this observation.

References


Ekbohm’s syndrome
