

Ekbom syndrome treated with olanzapine: a case report

Sindrome di Ekbom trattata con olanzapina: un caso clinico

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Summary

Objectives

Ekbom syndrome is a rare psychiatric disorder also known as delusional parasitosis or delusional infestation or dermatozoic delusion. There is no consensus on the most appropriate treatment. Traditionally, typical antipsychotic pimozide has been considered as first choice. Due to the side effects of typical antipsychotics, atypical antipsychotics (AA) can be taken into consideration as a valid alternative.

Method

We report a case of Ekbom syndrome treated with olanzapine. We examined the case of a 69-year-old woman. About three years ago, the patient began feeling as if infested by worms and eggs after a butterfly landed on her hair. She referred that the worms were spread throughout her body and amassed under her skin, and in her eyes, mouth, nose and genitals. The patient perceived materials such as lint or skin debris as worms and eggs, and collected them in pieces of paper as "proof" of infestation. The patient believed to have infected her husband and was worried about infecting other family members. The patient

had a history of recurrent depressive episodes with many admissions to psychiatric wards.

Results

The patient agreed to be hospitalized and to be treated with antipsychotics. Olanzapine was administered. The initial dose was 5 mg/day, then increased stepwise to 20 mg/day. After about 4 weeks, the patient showed improvement regarding her delusion: she improved her critical ability about the infestation; appetite and sleep also improved.

Conclusions

This case report shows the efficacy of olanzapine in this type of disturbance, and that can be used as a first-line agent in delusional parasitosis. In addition to drug therapy, establishing a therapeutic alliance was crucial. In fact, while at the beginning the patient was reluctant to accept psychiatric help, at the end a good therapeutic alliance was established and she was fully adherent to pharmacotherapy.

Key words

Delusional parasitosis • Classification • Pimozide • Atypical antipsychotics • Olanzapine

Introduction

Ekbom syndrome is a rare psychiatric disorder also known as delusional parasitosis (DP)¹ or delusional infestation or dermatozoic delusion. Initially described by Thiberge in 1894, the syndrome was defined in 1938 by Ekbom¹. The syndrome is characterized by the conviction of being infested by invisible mites or insects, despite clear evidence of the contrary. These pathogens, often but not always, cause itching, and the patient obtains skin lesions by scratching. Additional symptoms such as illusions or hallucinations may be present².

According to the aetiological classification of Lepping and Freudenmann³, primary and secondary DP can be distinguished (Table I). In primary DP, the delusion occurs independently of any medical condition; in secondary DP, the delusions arise in the setting of another major medi-

cal (infection, somatic illnesses associated with itching or para-aesthesia, e.g. diabetes mellitus, uraemia, jaundice, cancer), neurological disorder (e.g. dementia, brain tumour, stroke) or psychiatric disorder such as schizophrenia, major depressive disorder or mania; finally, DP can be induced by psychotropic and non-psychotropic substances. Establishing the prevalence of this psychiatric disorder is difficult because patients usually consult only dermatologists or general practitioners instead of psychiatrists; those who do end up consulting a psychiatrist usually have low insight regarding their condition, thus leading to poor adherence. Women are more often affected than men. Trabert et al. show that the mean age onset is 57.02 ± 14.6 years².

There is no consensus on the most appropriate treatment. Traditionally, typical antipsychotic pimozide has been considered as first choice⁴. Due to the side effects of typ-

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TABLE I.
Ekbohm syndrome: aetiopathogenetic classification. *Sindrome di Ekbohm: classificazione eziopatogenetica.*

Aetiopathogenetic classification*	
I. Primary	
A. No organic or psychiatric cause	
II. Secondary	
A. Organic cause	
1.	B12 deficiency, folic acid deficiency
2.	Heart failure, arrhythmias
3.	Meningitis, syphilis, encephalitis, leprosy, tuberculosis, HIV
4.	LES, rheumatoid arthritis
5.	Vascular dementia, multiple sclerosis, cerebrovascular accidents, tumours, trauma
B. Substance	
1.	Cocaine, amphetamine
2.	Corticosteroid, ciprofloxacin, IMAO, captopril, bleomycin, clonidine
C. Psychiatric	
1.	Schizophrenia
2.	Mood disorders
3.	Anxiety
4.	Folie à deux

* Adapted from Lepping et al.³.

ical antipsychotics, atypical antipsychotics (AA) can be taken into consideration as a valid alternative. We report a case of primary DP treated with olanzapine.

Case report

We present the case of a 69-year-old woman hospitalized at the Psychiatry Department of Policlinico "Tor Vergata" Hospital in Rome. About three years ago, the patient began feeling as if infested by worms and eggs after a butterfly landed on her hair. She reported that these worms were spread throughout her body and amassed under her skin, and in her eyes, mouth, nose and genitals. The patient consulted general practitioners, dermatologists, gynaecologists and ophthalmologists; she accused physicians of not understanding, while at the same time she refused to consult psychiatrists due to lack of insight. She then spent most of her time trying to remove the parasites from her skin, showering multiple times a day and using disinfectants and lotions. Insomnia and hyporexia were soon added to the symptomatology. Illusions were present: the patient perceived materials such as lint or skin

debris like worms and eggs, and collected them in pieces of paper as "proof" of infestation. The patient believed to have infected her husband and she was worried about infecting other family members. The patient had a history of recurrent depressive episodes with many admissions to psychiatric wards. Eleven years ago the patient underwent surgery for carotid stenosis, and she suffers from advanced osteoporosis that developed in the years after a hysterectomy when she was 39 years old. She was on treatment with aspirin, citalopram and benzodiazepines. The patient agreed to be hospitalized and to be treated with antipsychotics. Laboratory tests were within normal range. The mental state examination showed no cognitive deficit (MMSE = 30); the clock drawing test was normal. Cerebral CT showed initial brain atrophy compatible with age. At the moment, the patient's mood was euthymic. A diagnosis of primary delusional parasitosis was made, according to diagnostic criteria for delusional disorder, somatic type in DSM-IV-TR and ICD-10 criteria for persistent delusional disorder. According to the upcoming DSM-5 criteria, the patient would also be diagnosed a delusional disorder. Following the current NICE guidelines⁵, we treated the patient with olanzapine. The initial dose was 5 mg/day, then increased stepwise to 20 mg/day. After about 4 weeks the patient showed improvement regarding her delusion: she improved her critical ability about the infestation; appetite and sleep also improved as.

Discussion

This case is a typical example of primary DP. The patient is a woman, age of onset of 66 years, with a duration of disease of about 3 years. Our data are consistent with literature data which shows a major prevalence in the female population, mean age of onset at 55 years and mean duration of illness to be 3.31 years². Collecting bits of skin and other small particles as evidence of infestation is characteristic, and is referred to as "matchbox sign"⁶. Our patient was married and had a know and old history of psychiatric disease. This is in contrast with literature findings for Ekbohm syndrome's patient.

DP is usually treated with the antipsychotic pimozide^{7,8}. Pimozide, like other typical antipsychotics, is associated with extrapyramidal side effects. Moreover, pimozide therapy can cause prolongation of the QT interval, requiring baseline and periodic electrocardiographic monitoring. Due to these serious adverse effects, the use of atypical antipsychotics (AA) has been suggested. Comprehensive reviews of the clinical efficacy of AA in the treatment of both primary and secondary DP have been reported^{3,8-13}. Other studies have shown full or partial remission with olanzapine¹⁴⁻¹⁶. We performed a review of the recent literature to select cases of DP (primary or secondary) treated with olanzapine. Studies were selected

TABLE II.
DP cases treated with olanzapine. *Casi DP trattati con olanzapina.*

References	Dose	Outcome
Kumbier et al., 2002	10 mg	++
Nicolato et al., 2006	5 mg + citalopram 20 mg	++
Meehan et al., 2006	2.5- 20 mg	++
Freudenmann et al., 2007	2.5 mg - 7.5 mg	++
Bosman et al., 2007	10 mg	++
Mercan et al., 2007	10 mg	++

++ Remission

through PubMed. We used the following keywords: delusional parasitosis and olanzapine; delusion of parasitosis and olanzapine; dermatozoic delusion and olanzapine. We selected studies published since 2002 (Table II). This case report shows the efficacy of olanzapine at a 20 mg/day dosing, although there has been evidence of effectiveness at lower doses.

In conclusion, this case report shows the efficacy of olanzapine in this type of disturbance and that it can be used as a first-line agent in delusional parasitosis. In addition to drug therapy, establishing a therapeutic alliance was crucial. In fact, while at the beginning the patient was reluctant to accept psychiatric help, at the end a good therapeutic alliance was established and she was fully adherent to pharmacotherapy.

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