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Ekbom Syndrome; a Dilemma for Gastroenterologists and Dermatologists: Clinical Case of Delirium of Parasitosis Following a Proven Infestation

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Abstract **Case Report**

***Background*:** Described in 1938, Ekbom's syndrome is a disorder in most cases having a psychiatric origin. In addition to being frequent in psychiatry consultations, it I can be presented in dermatology essentially and other medical fields as well (such as gastroenterology). ***Case Report*:** We report the case of a patient who, after a suspected infestation, presented with Ekbom syndrome but the lack of knowledge of the disorder led to a three-year delay in diagnosis and treatment. ***Conclusion*:** Despite the understanding of this disorder is an integral part of the training of any psychiatrist, it is not the case of other medical fields, and the lack of awareness of this disorder can be harmful to the patient, because the delay in diagnosis and the excessive paraclinical explorations can make the management difficult.

Keywords: Ekbom, Oxyuriasis, dead-end diagnosis, ectoparasitosis, folie à deux.

INTRODUCTION

In 1938, the Swedish psychiatrist Karl-Axel Ekbom reported a series of eight cases that presented with a delusional belief of being infested with small organisms.

Ekbom originally called this condition "*Praeseniler Dermatozoewahn*" or *presenile dermatozoic delusion*.

It should not be confused with *Wittmaack-Ekbom* syndrome, which is a restless legs syndrome (Freudenmann & Lepping, 2009).

It is a rare pathology falling within the framework of delusional disorders.

We are reporting the case of a patient seen in psychiatry consultation 3 years after the disorders' onset.

Treating a psychiatric condition years later makes the treatment more challenging, luckily it wasn't the case to our patient, but doctors in other medical fields should be aware of that diagnosis that can look like a physical abnormality, this can help the patient get the best medical attention available.

CASE-PRESENTATION

H is a 68-year-old lady, widow for about 10 years and mother of 3, currently living with her youngest daughter. She has no psychiatric, medical or surgical history.

Her first contact with psychiatry was when she was referred by the gastroenterologist after a long follow-up for a pruritus *sine Materia*.

Indeed, 3 years ago, the patient presented an oxyuriasis revealed by an anal pruritus and the presence of Enterobius Vermicularis worms in her feces, which generated a strong concern. She ended up consulting a general practitioner who put her on Albendazole, then declared her cured.

Four months later, she began to present the same symptoms, generating strong anxiety again, but this time the proctology examination was without any abnomality.

Faced with a therapeutic dead end, she was referred to a gastroenterologist, who did not uncover any oxyuriasis infection and put her on antihistamines as a symptomatic treatment.

As a matter of fact, the patient had several biological tests, particularly liver tests, two colonoscopies, two scotch tests, a parasitological examination of the stools and a search for parasites in a dermatological sample brought back by the patient in a small box, all of which did not reveal any anomaly.

The physical examination reveals some scarification, lichenification which indicates a chronic scratching, especially on the forearms. Thoses injuries were explained by the patient by "*worms that come out of her anus and penetrate her skin, then migrate through her body to her scalp, and she is forced to compress some parts of her skin with her tombs to be able to crush the worms*".

The persistence of the pruritus was the source of a disruption in the family's dynamic in the household; the patient washed her clothes, bedding and those of her grandchildren on a daily basis.

She did not stop there, she also reported self-medication by taking Albendazole and forcing it on the other family members.

She was kept on hydroxyzine. However, given the diagnostic and therapeutic dead-end after 3 years of outpatient consultations, she was referred to a psychiatric facility for an assessment.

Psychiatric examination revealed cenesthetic hallucinations and an unshakable and firm belief that she was infested by parasites. Which caused anxiety and altered interpersonal relationships.

The patient was put on risperidone 2mg/day with distancing from her delusional ideas after 2 months of treatment but without real criticism.

DISCUSSION

This clinical case is indeed consistent with Ekbom's syndrome after a combination of data including being an elderly patient, female gender, belief in being infested by parasites in the absence of any biological evidence (Aït-Ameur *et al.*, 2000; Trabert, 1995), and the matchbox sign (sufferers often compulsively pick up small particles of skin or fibers which they interpret as evidence of infestation) (Bhatia, 2015).

In addition to the cenesthetic hallucinations, patients typically experience visual hallucinations or illusions in which the individual perceives common materials such as skin debris as being the pathogens or parasites.

The sex ratio is 2 females to 1 male, but the dermatological damage from attempts to remove parasites is more marked in males (Aw *et al.*, 2004; Trabert, 1995).

In this case study, the psychiatric presentation was not clear at the initial stage of the follow-up, which was justified by the existence of an infection confirmed by a professional and the consistency of the patient's statements. Her daughter's endorsement of her complaints only reinforced the delay in psychiatric care (in fact, *Folie a deux* is present in one third of cases) (Daniel & Srinivasan, 2004; Kim *et al.*, 2003).

A psychiatric cause could be retained only after having carried out on several occasions all the necessary complementary assessments.

Ekbom syndrome or parasitosis delirium is a differential diagnosis to ectoparasitosis and to chronic pruritus.

In the literature, its detection and management are extremely difficult because the diagnosis is often delayed due to the fact that patients are frequently evaluated by several physicians before seeing a psychiatrist. The patient often refuses a psychiatric consultation and subsequent a psychotropic treatment because he or she continues to believe that his or her symptoms have a physical origin. Therefore, the physician (primarily a dermatologist or infectious disease specialist) must first establish a healthy physician-patient relationship to allow for a psychiatric consultation.

A thorough evaluation should include the following:

A. Exclusion of any dermatologic pathology (actual infestation),
B. A search for a recent history of travel and/or exposure to known infected/infested individuals (scabies...),
C. A complete physical examination with complementary tests (blood count for eosinophilia, thyroid function, metabolic profile, liver function, C-reactive protein, urinary toxicology, syphilis screening, human immunodeficiency virus test, viral hepatitis panel, vitamin deficiencies, allergy tests),
D. A search for an underlying neurological disorder (including a complete neurological examination, full mental status evaluation, EEG, head CT and/or MRI, if necessary),
E. A search for another psychiatric disorder (comprehensive mental status examination, supplemented by clinical rating scales and personality assessment). In addition, clinicians should carefully evaluate potentially involved prescribed medications (e.g., antibiotics, corticosteroids, topiramate, etc.), substance abuse and/or alcohol abuse. Some authors also recommend taking a hair sample from patients suspected of being intoxicated.

In the DSM-5TR and ICD-11, primary Ekbom syndrome is included in delusional disorders

(*Diagnostic and Statistical Manual of Mental Disorders*, s. d.; *ICD-11*, s. d.).

In general, the affected patients should always be treated with empathy, providing a comfortable place for them to express their distress without being stigmatized or contradicted, and ensuring that the history of their condition is carefully explored.

It is particularly important to ask whether there are other family members with the same symptomatology (i.e., to investigate "*folie a deux*", shared delusions).

A systematic review of the efficacy of first and second-generation antipsychotics in the treatment of Ekbom syndrome in the setting of delusional disorder found no clinically relevant differences in efficacy, although the authors noted that the evidence was weak because it was derived only from case reports and case series (Lepping *et al.*, 2007).

Some recommend second-generation antipsychotics, including Aripiprazole (2-30 mg per day), Olanzapine (2.5-20 mg per day), Quetiapine (25-600 mg per day) and Risperidone (0.5-6 mg per day) (Bennàssar *et al*., 2009).

A systematic review of multiple case series and observational studies of primary delusional infarction/parasitosis documented a 60-100% efficacy rate with antipsychotics, including Pimozide, Olanzapine and Risperidone (Trabert, 1995).

CONCLUSION

Ekbom syndrome causes considerable suffering for the patients themselves and for their relatives. Due to the lack of understanding of this disease, family members are often unable to seek appropriate treatment for the subject.

Delayed treatment is often the rule, making remission difficult. The medical approach is multi-disciplinary.

There is little research into understanding or treating this illness. Clinically, the stigmatizing term delirium is avoided by calling the disease Ekbom syndrome.

DECLARATION

- **Ethics Approval and Consent to Participate:** There is no ethical issue.
- **Consent for Publication:** The patient has given consent for publication
- **Competing Interest:** The authors declare that they do not have any competing interests.
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- **Availability of Data and Materials:** Data sharing in not applicable to this article as no data sets were generated or analyzed during current study.

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