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DELUSIONAL INFESTATION. EKBOM'S SYNDROME IN A 53-YEAR OLD WOMAN. CASE REPORT

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Palabras clave: Reporte de caso; Delirio de parasitosis; Delirio de parasitosis de Ekbom.

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ABSTRACT

Introduction: Delusional infestation is a rare psychiatric disorder defined as a condition in which the patient has the unshakable belief and perception of being infested with parasites. Its treatment is difficult, and frequently includes antipsychotic medications (such as olanzapine or aripiprazole). Non-pharmacological treatment, particularly psychotherapy, can be used for less severe cases. Dermatologists and psychiatrists must take a multi-disciplinary approach (preferably in a psycodermatology dedicated clinic) since this type of patients sometimes refuse treatment.

Case description: A 53-year-old female businesswoman describes a clinical history of five years of visual hallucinations, depressive symptoms, and generalized pruritus, along with the use of toxic substances to "clean" her skin and cloths. She reports similar symptoms in some relatives but they were not evaluated. Blood tests and analyses of the "specimen" brought by the patient were performed, yielding negative results. The patient had never been assessed by any specialist, and showed disoriented during the consultation. Follow-up was not possible due to the reluctance of the patient to follow the indications and seek psychiatric treatment. Moreover, the patient did not respond to further communication attempts.

Discussion: Delusional infestation is an uncommon disease that endangers the patients and the people around them. Its treatment is difficult and long, and not conducting proper follow-up is a great risk. Its prevalence and incidence is variable and generally unknown. It can affect the patient, their next of kin, pets or the environment, and the "pathogen" can be a living organism or an inanimate object.

Conclusion: This case is important as it shows the hardships of treatment, adequate follow-up and care, as well as the need to improve how these patients are approached. Additionally, both classical and uncommon signs and symptoms could be observed as the patient stated that her relatives were affected (possible delusional infestation by proxy).

CASE DESCRIPTION

Delusional parasitosis (DP), delusional infestation (DI) or Ekbom's syndrome is an uncommon but not rare (1-3) psychiatric disorder in which patients have a fixed, false belief that they are infected or infested with parasites or other living organisms (4-6). This condition may lead to self-mutilation (5) or affect other members of the family (7). It was first described more than a hundred years ago (8), and is classified as a persistent delusional disorder in ICD-10 (9) or as a delusional disorder of the somatic type in DSM-V (10), although much is left to understand about this disease. The prevalence is estimated between 0.18 and 4.2 per 100,000 (11,12), with an incidence of 1.9 per 100,000 (12). However, its variation is wide: in the United Kingdom, the estimated incidence is about 4.9 per million (11).

Delusional infestation by proxy is estimated in approximately 5-15% of the cases (7, 13). Its onset has been associated with changes in the glucose metabolism of the thalamus and the left putamen, and with alterations in the dopaminergic neurotransmission of the striatum, again in the left putamen (14). There is also evidence of abnormal frontolimbic brain activity (15) and abnormal grey and white matter volume (16,17).

It generally affects women over 50 (30-60) years of age (6, 18), with a mean age of

onset at 56.9 years (19). It can be classified as primary or secondary; the primary form of the disease does not present an organic or psychiatric underlying cause (Table 1) (6,20,21), and patients are otherwise mentally healthy (6,22). A really uncommon form of this disease is DI by proxy, which is a shared psychosis referred as folie à deux or folie à trois, where patients believe that other individuals or pets are infested rather than themselves (18,23,24). This presentation is more frequent in veterinary practice (18).

Table 1. Drugs or conditions associated with the onset of secondary delusional infestation.

Drugs or conditions	References
HIV	(4)
Neuropsychiatric drugs	(25, 26)
Dialysis	(20, 27)
Neuropsychiatric diseases such as schizophrenia and depression	(20, 28)
Stroke	(20)
Psychotropic drugs	(20, 29)
Dementia	(20)
Renal disease	(30)
Iatrogenic	(31, 32)
Occupational hazard for entomologist and healthcare workers	(33)

Source: Own elaboration based on the data obtained in the study.

The duration of the disease ranges from months to years (34). The most affected organ is the skin but other parts of the body can be affected as well (21,35). For its diagnosis, delusion must have a duration of at least 1 month, but patients must remain highly functional (1) and present with two main symptoms: 1) the strong belief of being infested despite medical evidence shows otherwise (100% of the patients), and 2) abnormal sensations as if an infectious agents caused them (88%) (6,36,37). Considering that the delusion of being infected can vary in intensity (38), in less severe cases the belief can be wavered but not reduced (39). Some of the symptoms reported are listed in Table 2 (1,4,5,6,12,19,20,40,41).

Table 2. Possible symptoms of delusional parasitosis described by patients or relatives.

Symptoms described by patients	Symptoms described by relatives
Pruritus	Psychosis
Poor sleep	Confusion
Tactile hallucinations	Strange behavior
Self-mutilation or self-damage	
Visual hallucinations, and other types of hallucinations	
Delusional ideas	
Dysphoria	
Disturbed reasoning and judgement	
Formication and other sensations of movement under the skin	
Intrusive and non- reducible belief of infestation	
Proof of infestation (specimen or matchbox sign)	
Use of toxic products topically or orally to “treat” the condition	

Source: Own elaboration based on the data obtained in the study.

Atypical manifestations appear when the “pathogens” are relatively large, the environment is infested rather than the individual, or the infestation is caused by inanimate objects (3,6,42). The infesting species range from unspecific living beings to microorganisms and small animals. These specimens can be “stored” and handled without disgust (6,43), and are treated as trophies instead; the spec-

imen, however, should be examined (43). This is known as the "Matchbox sign", which is characterized by the patient collecting an inert substance (for example, dead skin) in a container, stating that it contains living specimens of the parasite in different stages of development (12).

The delusion can result in damage to the skin, hair, eyes and other family members, as they try to "clean" the infestation with dangerous substances or elements including fire and electricity (6,44,45). This condition can be affected by a large array of complications as listed in Table 3 (6,12,35).

Table 3. Possible complications of delusional parasitosis

Erosions
Excoriations
Ulcers
Lichenification
Chronic irritant contact dermatitis
Lichen simplex chronicus
Prurigo nodularis
Lichen amyloidosis
Corneal abrasions
Secondary infections
Increased mortality

Source: Own elaboration based on the data obtained in the study.

The recommended treatments are based on anti-psychotic medication such as risperidone, pimozide (36,46), olanzapine, amilsupride, quetiapine and aripiprazole (47-50). Atypical anti-psychotics have a more favorable side-effect profile (50). The therapeutic effect may be observed between one to ten weeks (47). Lepping et al. (51) assessed the efficacy of this drugs, reporting that typical and atypical anti-psychotics achieved a remission proportion of 60-100%. In less severe cases, non-pharmacological treatment, such as psychotherapy, can be used (50,52). Other non-pharmacological therapies include: neurosurgery, transcutaneous electric stimulation and electroconvulsive therapy (50), with a less than optimal success rate (36,50).

Joint management by psychiatrists, psychologists and dermatologists is required (28,53-55), preferably in a clinic dedicated to psychodermatology (56,57). Adequate treatment leads to remission in 75% of the cases, although 25% of those patients may relapse and require longer therapy (58). Treatment should be introduced after obtaining a complete medical history and a systematic evaluation (54), specially to discard secondary causes or differential diagnosis (59-63) (Table 4). Offering antimicrobial or anti-parasitic drug trials reinforces the delusional ideas and is not recommended (6).

Table 4. Differential diagnosis of delusional parasitosis.

Hypocondriasis circumscripta
Dermatitis artefacta
Skin picking disorder
Chronic pruritus
Morgellons disease

Source: Own elaboration based on the data obtained in the study.

Three groups of patients have been suggested for classification purposes: 1) patients with hypochondriac traits, 2) patients with paranoid symptoms and without hypochondriac apprehensions, and 3) patients with hypochondriac and paranoid traits (64). Most patients have other associated psychological disorders including anxiety, depression and appearance-related concerns, among others

(37, 65). More information can be obtained in the review made by Freudenman (6).

CASE DESCRIPTION

Female, 53 year-old, Hispanic businesswoman who attends, by her own volition and alone, an outpatient medical appointment at the Parasitology Department of Instituto Nacional de Salud (National Health Institute), referring a clinical history of 5 years characterized by the perception of macroscopic parasites that crawl over her body, biting her face, head and anterior thorax, and leaving white eggs which evolve to brown adults in about eight days. She also referred generalized pruritus and shows excoriations due to scratching (Figures 1 and 2), which she "treated" with Vicks Vaporub, domeboro (calcium acetate and aluminum sulphate), crotamiton and Canesten (clotrimazole) cream. At some point, she also used thinner and varsol to wash her clothes and skin.

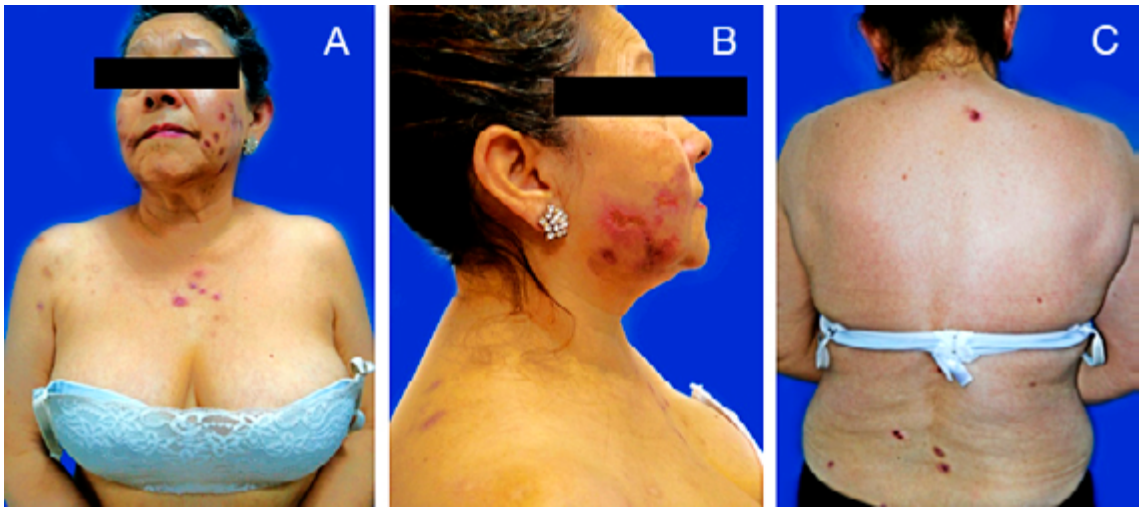

Figure 1. A. Front of the patient. The lesions can be observed in the cheeks, arms, and chest. B. Profile of the patient portraying a close up of the cheek lesions. C. Back of the patient showing the extent of the lesions.
Source: Own elaboration based on the data obtained in the study.

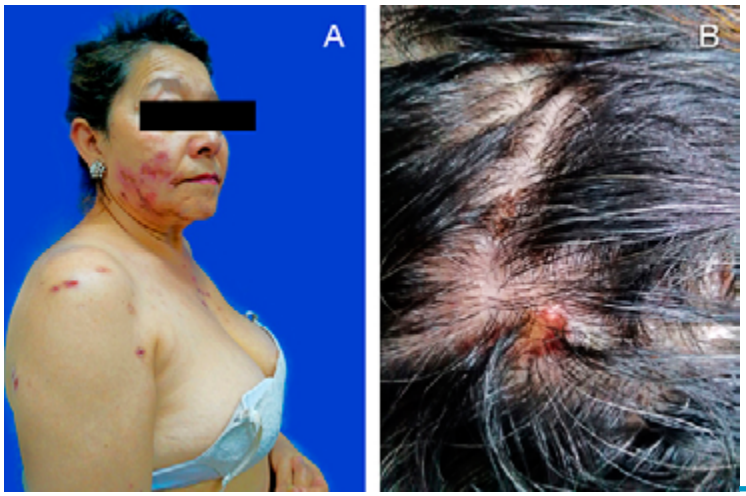

Figure 2. A. Profile of the patient showing the lesions on both cheeks B. Picture showing some of the multiple lesions on the scalp.
Source: Own elaboration based on the data obtained in the study.

The patient takes to the appointment, in a small plastic jar, pieces of skin and coagulated blood which she says contain the eggs and two adults (Figure 3). This specimen is handled without disgust or contempt. She also describes how one of her sons have seen the parasites fly after one of the eggs hatched, while other members of the family do not have parasites but suffer from pruritus. Unfortunately, the relatives were not present during the interview and refused to be interviewed or examined afterwards, so this may be another delusory idea of the patient.

She denies travelling to other places in the past five years. Medical background includes diabetes mellitus diagnosed twelve years ago, and pharmacological background includes metformin 850mg/day, glibenclamide 5mg/day, ivermectine 51 drops (which she has used in repeated doses since the onset of the disease), difenhidramine 50mg every 8 hours and, occasionally, amoxicillin 500mg every 12 hours; she denies using other medications. Her gynecological background is G4P3C0V3.

Physical examination revealed an arterial pressure of 100/60, heart rate of 84, respiratory rate of 18, temperature at 35.5°, weight 51.3kg, height 145cm, multiple excoriations on the scalp and neck, and cicatrized lesions in the inferior third of the face, chest and back. The patient had normo-reactive isocoria and eye bags; the rest of the physical examination was normal.

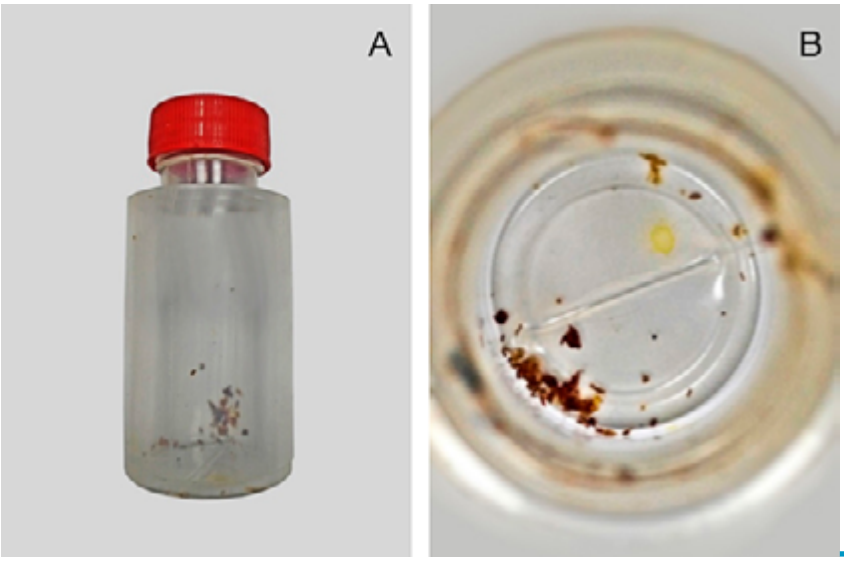

Figure 3. A. Jar brought to the appointment by the patient. B. Close up of the jar contents.
Source: Own elaboration based on the data obtained in the study.

Regarding mental aspects, the patient had a negative attitude, was passive, cried easily, but had good visual contact with the interviewer. She had a slight psychomotor retardation and a sad attitude, with anxiety traits. Delirious thoughts were present, as well as sad mood, with semi-structured suicidal ideations, non-existent introspection, uncertain prospection, and good interpersonal relationships. Her speech was not slurred, and her behavior was organized and non-catatonic. Even though she was depressed, she did not show any signs of alogia or affective flattening, and the depressive symptoms started long after the delusions. She did not hear voices.

Samples were taken, and cultures from feces and lesions were obtained, yielding negative results for microorganisms. Hemo-

gram was normal, tests for liver and renal function were normal, and glycemic control was optimal. Toxicological exam was normal. Complete studies of the skin and fecal matter were made, yielding negative results as well. The specimens brought by the patient were analyzed providing a negative result. The patient was examined for needle marks which were absent. She did not present alterations in the rhinal mucosa nor respiratory or cardiac problems in the physical exam or medical tests.

Due to the absence of criteria to diagnose other psychiatric disorders or substance abuse or physical disease, the patient met the criteria of a delusional disorder of the somatic type (at least 1 month of delusions, visual hallucinations consistent with the disease, social function not markedly impaired, and brief mood episodes compared to the delusional period), thus Ekbom syndrome was diagnosed. The patient did not accept psychiatric treatment, and neither dermatology nor psychiatry services were able to assess the patient. Follow-up was not possible due to the refusal of the patient to start psychiatric treatment, even when it was suggested and advised for her depression.

Even though the patient did not accept treatment or referral, medical indications and recommendations were given. Prognosis is not optimal without pharmacological and dermatological follow-up or treatment, therefore, it is highly unlikely that the condition of the patient will be resolved.

Written informed consent was obtained from the patient for the publication of this case and the photographs.

DISCUSSION

Ekbom's syndrome, better referred as delusional infestation, is an uncommon disease that generally affects woman above 50 years of age. Physicians must be cautious when approaching this kind of patients, considering the words used and even the proposed therapies, in order to generate a good patient-physician relationship and help patients to understand the importance of treatment with a psychiatrist. Not conducting proper follow-up is dangerous; for instance, this case is a prime example of that, since the patient presented a typical primary delusional infestation that not only affects her life and self-esteem, but her family's life as well. Here, the information provided by the patient allowed to infer (although it was not confirmed) that some members of her family may suffer from delusional infestation by proxy. This presentation is similar to other cases reported in the literature (63).

Treatment is difficult and must be carried out based on an interdisciplinary approach. One way of improving treatment is through the use of psycodermathology, which is a new subspecialty emerging from dermatology, and includes interventions such as psychoeducation and cognitive behavioral therapy or joint interviews with dermatologists and psychiatrists (66) that have proven to be cost-effective (57).

Patients generally blame parasites, but can also consider other small organisms or inanimate objects. This condition can affect the individual, someone or something close to the individual or the environment, and there are even cases of shared psychosis.

Usually, sick patients are not violent, but they can be a threat to themselves and others since they may use dangerous substances during the "cleansing" process, and may even mutilate themselves. This case, as other cases (37), shows the relationship between underly-

ing psychiatric diseases and DI, and the long process involved to diagnose this disease. Physicians must first discard and find evidence that the patient is not infested, looking for symptoms or signs of underlying psychiatric disorders or the use of psychotropic drugs or conditions associated with this disease as well. Considerations about pets or individual victims of DI by proxy and their protection are outside the scope of this report.

Some of the most important aspects of this study include, first, initial assessment and referral, even though the patient did not follow the instructions, and second, the thorough clinical and physical examination alongside a complete laboratory analysis done to the patient. Some of the limitations are that the patient was never assessed by a specialist, that the outcome of the case is unknown, and that the relatives could not be evaluated, therefore, delusional infestation by proxy could not be confirmed.

CONCLUSION

This case clearly exposes the hardships of treating this type of psychiatric patients, as well as the difficulties for adequate follow-up and care. A better way of following and delivering care to these patients could possibly include domiciliary visits or clinics dedicated to psycodermatology. However, this case is important since it shows both classical and uncommon signs and symptoms, that the disease can affect patients and possibly their relatives as they can also suffer from similar symptoms (delusional infestation by proxy), and that they should also be included in the treatment. A comprehensive approach to the family and its involvement should be considered when approaching this type of patients.

PATIENT'S PERSPECTIVE

Follow-up was difficult, but during the few interviews that could be conducted, she expressed that her situation was dire, that she was desperate. Her mood had not improved and the negative thoughts were worse over time. At first, her work nor her relationships were affected, but they were compromised as the disease progressed, to such an extent that she was seeking advice from multiple physicians, but she thought that they did not believe her and never returned.

TRANSPARENCY

The authors declare that all the information contained in these pages is true, honest and transparent, that no important aspect of the case was omitted, and that every relevant characteristics or differences have been exposed.

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CONFLICT OF INTERESTS

None declared by the authors.

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