Delusional infestation complicated by gangrene and osteomyelitis requiring finger amputation

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INTRODUCTION

Delusional infestation (DI) is an uncommon entity defined by an individual's belief of microbiological infestation despite absence of evidence.[1] The presumed infestation could be that of a living or inanimate pathogen, and the cardinal symptom is dysesthesia. The disease frequently drives patients to seek various interventions and leads to impaired quality of life.[2]

The delusions may be of primary or secondary origin.[1] The secondary causes include neurogical diseases, psychiatric disorders, nutritional deficiencies, medications, infections, intoxication, neoplasms, and metabolic imbalances.[3] The mechanism underlying the development of DI is not well understood. Many authors propose it to be of psychiatric origin. However, others hypothesize it to be a type of immune-related pruritic dermatosis or an infection.

Herein, we describe the case of a man with cutaneous dysesthesias due to what he presumed to be a parasitic infestation. The disease led him to self-mutilate his left second finger until the development of osteomyelitis, which required amputation.

Abbreviations used:

DI: delusional infestation
MD: Morgellons disease

CASE REPORT

The patient is a 55-year-old man with a 6-year-history of dysesthesia that he described as sensation of movement underneath his skin accompanied by pruritus, erythema, burning, and pain. The patient noted "flies and worms" coming out of his skin and believed that his symptoms were due to a parasitic infection.

His travel history was notable for camping, which he believed might have put him at risk for an infection. He lived in South Carolina when the symptoms began, and he had traveled to South Florida, Bahamas, Kansas, Louisiana, Texas, New York, Colorado, and Nevada. His medical history was significant for primary immunodeficiency of unknown etiology diagnosed 5 years prior and treated with immunoglobulin. The patient believed his immunodeficiency rendered him susceptible to infection. No history of diabetes or vitamin B12 deficiency was documented.

Over the course of his illness, the patient was afebrile and was treated with antiparasitic (ivermectin, albendazole, permethrin, and praziquantel), antibacterial (doxycycline, ciprofloxacin), and antifungal medications without resolution of symptoms. He developed nodules, erosions, and ulcers on his upper and lower extremities, including a nonhealing ulcer on his left second finger, secondary to scratching and attempting to extract the presumed pathogen (Fig 1). Multiple skin biopsies demonstrated chronic spongiotic dermatitis, acute and chronic folliculitis, and panniculitis. Bacterial, mycobacterial, and

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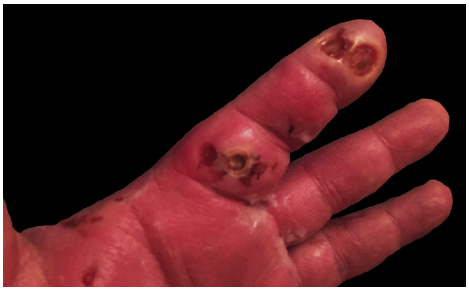

Fig 1. The patient developed erosions and a nonhealing ulcer on his left second finger that was worsening and concerning for osteomyelitis.

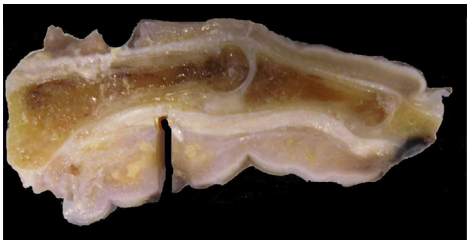

Fig 2. The patient consulted with his amputated finger in a specimen container. The amputation site was through the metacarpal phalangeal joint with a smooth resection margin of subchondral bone. The finger was covered by white-tan skin. 2 ulcers with necrotic borders were present at the level of the tip and ventral aspect of the proximal phalanx. The first ulcer extended to the remaining proximal third of the distal phalanx.

fungal cultures were negative. Serologic studies failed to provide evidence of infection. Duplex ultrasound of the upper extremities ruled out vascular disease. While many of his wounds healed, the ulcer of the left second digit developed cellulitis and osteomyelitis, requiring surgical amputation. The patient presented to the dermatology clinic with his amputated finger in a glass container, requesting evaluation to identify the organism causing his disease (Fig 2). On physical exam, the patient presented with erythematous, eczematous patches involving the face, chest, abdomen, back, and arms, as well as excoriated papules and generalized xerosis. His left second digit was amputated with a healing stump.

Histopathology of the amputated digit revealed lichen simplex chronicus prurigo nodularis, ulceration, cellulitis, and acute and chronic osteomyelitis (Figs 3 and 4). Periodic acid-Schiff, Grocott methenamine-silver, and Warthin-Starry stains were all negative.

During the last follow-up, the patient manifested persistence of his symptoms in other body sites with similar characteristics, and suicidal ideation. Pimozide was started with partial improvement of his symptoms.

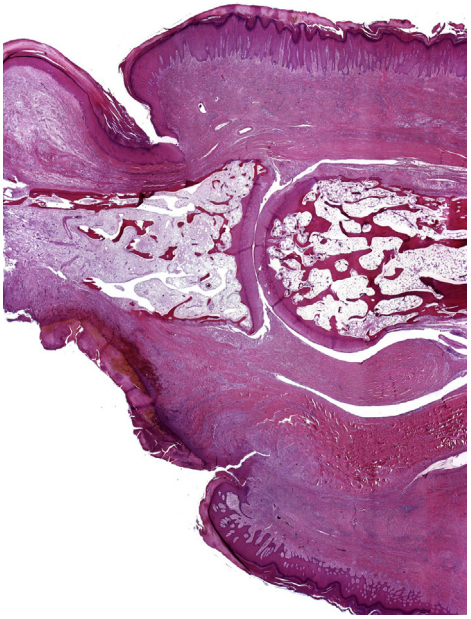

Fig 3. Histology demonstrated a deep skin ulcer with acute and chronic osteomyelitis of the distal phalange (Hematoxylin-eosin stain, original magnification: ×1).

DISCUSSION

In 1946, Wilson and Miller introduced the term 'delusional parasitosis' to describe patients convinced of infection by parasites.[4] The first known clinical description of this condition dates back to 1938, when Ekbom used the term "Dermatozoewahn," from the Greek "derma" (skin) and "zoon" (living-being), and the German "wahn" (delusion). Early names for this disease include Ekbom syndrome, acarophobia, parasitophobia, and dermatophobia.[1,4] More recently, the term 'delusional infestation (DI)' was proposed to encompass delusions of animate and inanimate pathogens, which has been accepted to describe this condition.[5] In 2002, Leitao coined the term 'Morgellons disease (MD)' to describe her son's itching illness. Leitao based this name on "The Morgellons", coined by Browne in *A Letter to a Friend*.[2]

The prevalence of DI is unknown. However, recently, after mass media coverage of MD, the number of consultations has increased.[6] One study reported that the prevalence of DI is 3.65 per 100,000 people with a Caucasian predominance, and other studies estimate approximately 5.5 cases per million.[4,5] DI typically affects women aged 62 years or older.[4,7] DI has been associated with psychiatric comorbidities (68.5%), smoking (32%), hypothyroidism (22.1%), and a positive family history of DI (18%).[8]

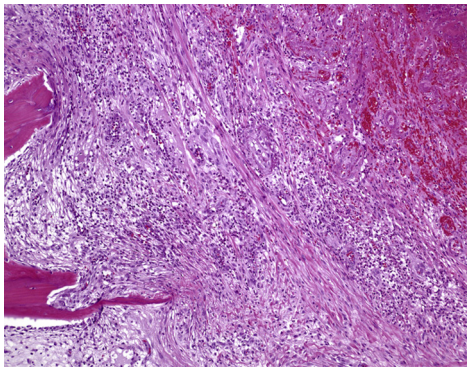

Fig 4. The cutaneous ulceration was deep and showed marked acute and chronic inflammation and osteomyelitis (Hematoxylin-eosin stain, original magnification: ×20).

There is an ongoing debate regarding the pathogenesis of DI. Some authors hypothesize that the disease is not a delusional disorder and refer to the condition as MD.[8] Other authors consider DI and MD as separate illnesses and discourage using the two terms interchangeably. Indeed, many online, non-empirically supported treatment suggestions for MD are readily available and may lead to patient self-harm. Furthermore, MD has been suggested to be associated with *Borrelia burgdorferi* infection.[9] Some suggest that the filaments that project or are embedded in the skin of MD patients are composed of keratin and collagen, resulting from spirochete-induced keratinocyte and fibroblast proliferation.[9] However, other studies have failed to support an infectious origin.[10]

Patients frequently attempt to extract the 'pathogen' or fibers and use a plethora of tools or agents to do so. Consequently, excoriations, prurigo nodularis, ulcerations, scars, and secondary infections develop. The manifestations may be asymmetrical due to the dominant hand effect. A large proportion of patients (25%-75%) try to demonstrate proof of their illness collecting specimens and bringing them to physician visits.[5,8] This scenario is known as the 'specimen sign', 'matchbox sign', or 'baggies sign', as patients may use different containers.[5]

Non-cutaneous manifestations include psychiatric (depression and hallucinations), neurological (cognition problems), and general symptoms (fatigue and myalgia).[7,8] Patients develop complex stories to explain their illness.[9] They are often reluctant to be stigmatized by a diagnosis of mental illness, resulting in social isolation, loss of employment, and loss of custody of children. Therefore, DI may be underdiagnosed.[9] The duration of the symptoms varies from less than 1 year to decades, and many patients become anxious due to the chronicity of DI, and some even develop suicidal behavior.[10]

Histopathologically, the skin reveals nonspecific inflammatory changes and no identifiable pathogens.[1] The fibers or filaments are biofilaments derived from the patient; keratin and collagen with variable amounts of pigment.[3] Chronic lesions develop features of lichen simplex chronicus and prurigo nodularis.

Primary DI is diagnosed when no other condition explains the patient's delusions. Secondary causes include medical conditions, psychiatric illness such as schizophrenia or bipolar mania, intoxication, medications, and neurologic impairment.[7] The differential diagnoses include true infestation, medical conditions with pruritus, trichotillomania, hypochondriasis, dermatitis artefacta, and psychosis.[5,11]

Treating DI patients is challenging. There is often patient-physician distrust due to disagreement on the origin of the disease. Patients often feel hopeless and abandoned, and they are prone to rejecting therapeutic proposals.[11] Acknowledgment of the patient's suffering and detailed work-up and collaborative management are imperative to exclude other conditions and to build trust.[8,9] Having a delusional disorder does not necessarily qualify the patient as incompetent. Thus, their decision-making capacity must be formally assessed. There are documented cases of successful treatment with antipsychotics, and psychotherapy. Medications currently used include anti-histamines and antipsychotics, particularly second-generation antipsychotics such as risperidone and olanzapine.[3,4] Antidepressants have also been used to manage associated depression. A peculiar finding is the recovering after using antibiotics.[4,6] Possible mechanisms of action are placebo effect and anti-inflammatory activity. In a few patients, electroconvulsive therapy demonstrated beneficial results.[12]

The patient presented here suffered from severe, unrelenting symptoms of DI resistant to traditional therapies and ultimately led to auto-amputation. Earlier diagnosis of DI and appropriate antipsychotic treatment may have prevented his irreversible morbidities.

Conflicts of interest

None declared.

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