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Exophytic botryomycosis: An unusual clinical presentation☆

Dear Editor,

Botryomycosis is a chronic infectious disease of bacterial origin, granulomatous and suppurative with a worldwide distribution. The incidence and prevalence are unknown, although it is considered an infrequent entity, with approximately 200 cases reported around the world.[1–3]

A 42-year-old male patient, a farmer, arrives at the dermatology service with a slow-growing lesion that had appeared 2-years earlier in the great toe finger of the right foot. The patient reported pain of moderate intensity that was enhanced with daily walking, as well as self-limited bleeding. Physical exam reveals in the dorsal aspect of the great toe finger an exophytic ulcerated tumor, erythematous, with hematic crust on the surface and some areas of bleeding, measuring 5×5 centimeters (Fig. 1).

A biopsy was taken for the clinical hypothesis of squamous cell carcinoma vs. amelanotic melanoma; the result of the histopathological study showed pseudoepitheliomatous hyperplasia with basophilic granular bodies (grains) with numerous neutrophils (Figs. 2 and 3). Microbiological cultures were negative. The diagnosis of the exophytic botryomycosis was made, surgical resection was indicated by the plastic surgery service, and antibiotic management with trimethoprim-sulfamethoxazole was started.

Botryomycosis derives from the Greek ''botrys'' (bunch of grapes) and ''myces'' (fungus) because initially, a fungal etiology was suspected. Two types of the presentation can be described, cutaneous and visceral.[1,4]

The cutaneous presentation represents 75% of the reported cases, the remaining 25% correspond to the visceral type. Botryomycosis can occur at any age, although it rarely occurs in 'children's and adults over 70-years old, it mainly involves areas with greater susceptibility to trauma such as hands, feet, head and neck.[1,3,4]

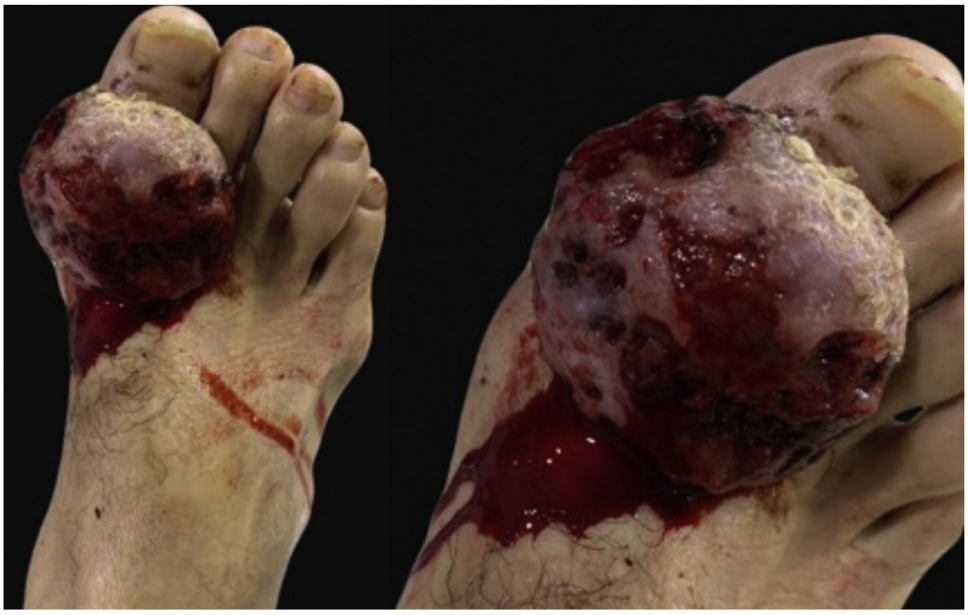

Figure 1 On the right great toe finger exophytic, ulcerated tumor.

☆ Study conducted at the Hospital Universitario La Samaritana, Bogotá, Colombia.

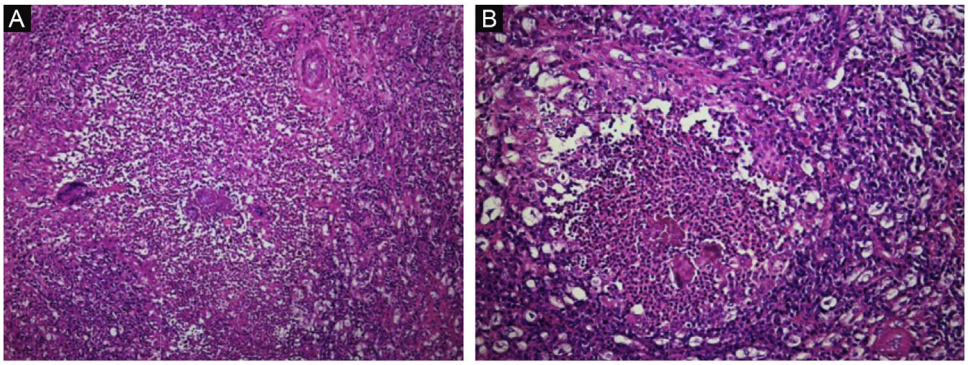

Figure 2 Inflammatory infiltrate with neutrophils and granular basophilic bodies (grains). (A), Hematoxylin & eosin, x10. (B), Hematoxylin & eosin, x40.

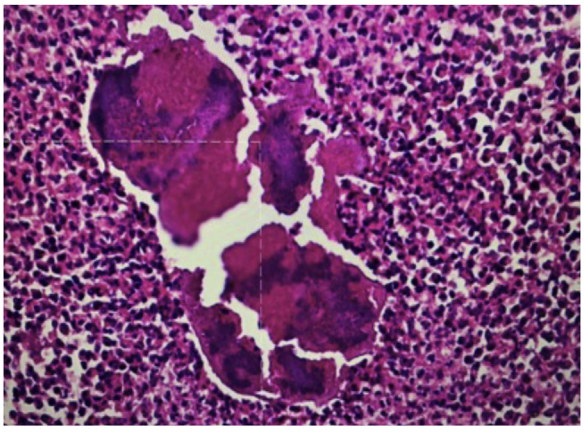

Figure 3 Granular basophilic bodies, grains (Hematoxylin & eosin, x100).

The history of trauma is the most important risk factor, in the case presented in this article, this was the probable method of inoculation since the patient works in agriculture. Other risk factors linked are immunosuppression, diabetes *mellitus*, liver disease, alcoholism, systemic lupus, cystic fibrosis, malnutrition, immunoglobulin deficiency, glomerulonephritis, HIV/AIDS, or surgery history.[1,4]

The pathogenesis of this entity is not well understood, and many authors agree that this reaction corresponds to a Splendore-Hoeppli phenomenon, in which antigen-antibody complex, immunoglobulin G and C3 are precipitated, a process in which phagocytosis and intracellular bacterial destruction is prevented.[1]

The 'patient's present nodules, fistulas, abscesses, and ulcers with seropurulent exudate in which 3–5 mm white-yellowish granules can be seen and the systemic infection has not been observed.[5] The diagnosis is made by isolating the causative agent; however, it is not easy to isolate.[1,4]

The differential diagnosis is other infectious granulomatous diseases such as mycetoma, actinomycosis, sporotrichosis, cutaneous tuberculosis, and malignant tumor diseases such as squamous cell carcinoma and amelanotic melanoma.[1]

Antibiotic treatment should be directed to the causative agent, in the case of extensive lesions, failure of systemic treatment, or severely immunocompromised patients, excision and drainage of the lesions are recommended.[1,2,4] In our case, the treatment was tumor excision by the plastic surgery service with reconstruction with partial-thickness graft, additionally, empirical treatment with trimethoprim-sulfamethoxazole was given due to the epidemiological profile and possible causative agent.

We present the clinical case of exophytic botryomycosis, an unusual clinical presentation previously not reported in the literature.

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Authors' contributions

Lina Paola González-Cardona: Critical literature review; critical manuscript review; preparation and writing of the manuscript; study conception and planning.

Adriana Mercedes Alejo Villamil: Data collection, analysis and interpretation.

Carolina Cortés Correa: Approval of the final version of the manuscript; effective participation in research orientation.

Elkin Omar Peñaranda Contreras: Approval of the final version of the manuscript; intellectual participation in propaedeutic and/or therapeutic; management of studied cases.

Conflicts of interest

None declared.

References

1. Padilla-Desgarennes C, Vázquez-González D, Bonifaz A. Botryomycosis. Clin Dermatol. 2012;30:397–402.
2. John K, Gunasekaran K, Kodiatte TA, Iyyadurai R. Cutaneous Botryomycosis of the Foot: A case report and review of literature. Indian J Med Microbiol. 2018;36:447–9.
3. Sirka CS, Dash G, Pradhan S, Naik S, Rout AN, Sahu N. Cutaneous Botryomycosis in Inmunocompetent Patients: A case series. Indian Dermatol Online J. 2019;10:311–5.
4. Bailey P, Raybould JE, Sastry S, Bearman G. Cutaneous Botryomycosis and *Staphylococcus aureus*: Diagnosis, Management, and a Systemic Literature Review. Curr Treat Options Infect Dis. 2018;10:347–61.

5. Singh A, Cook C, Kollmann K, Rajpara A. Acute cutaneous botrymycosis of the hands. IDCases. 2020;19:e00709.

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Infectious dermatitis associated with HTLV-I: uncommon case in southern Brazil simulating refractory atopic dermatitis☆

Dear Editor,

HTLV-I (human T lymphotropic virus type-I), a human retrovirus discovered in the 1980s,[1] infects preferentially CD4 T lymphocytes. The worldwide prevalence is uncertain, with an estimated 5 to 10 million infected individuals,[2] mainly in Japan, Iran, Latin America, and Africa.[3,4]

Infectious dermatitis associated with HTLV-I (IDH) was described in Jamaica in 1966, and associated with HTLV-I in 1990, being a rare and treatment-resistant form of exudative dermatitis.[1,3–5]

We describe a case of a seven-year-old girl, from the south of Brazil, born through vaginal delivery, with severe recurrent eczema since she was 18 months of age, when she stopped being breastfed.

On examination, she had macerated, exudative, and foul-smelling eczematous lesions on the scalp and retroauricular, cervical, antecubital, and intergluteal regions; temporal alopecia; crusts in the umbilical, perioral and nasal regions (Figs. 1 and 2). Laboratory tests were normal, except serology for HTLV-I/II which was reactive, confirming the diagnosis of IDH according to the criteria described in Table 1.[5] The other viral serologies were negative. The neurological examination was normal. Her mother also had positive serology for HTLV-I/II. Treatment with oral sulfamethoxazole and trimethoprim was started, with significant clinical improvement.

IDH usually starts in childhood and is considered an early clinical marker of HTLV-I infection.[3,4] The main route of transmission is through breastfeeding.[3,5] Its pathogenesis involves individual susceptibility, immune dysregulation, bacterial superinfection, environmental antigenic stimulation and persistent inflammation.[4] The pro-inflammatory state may be related to the proliferation of T lymphocytes and high levels of IL-1, IL-6, TNFα and IFNα; elevated IgE levels increase susceptibility to *S. aureus* and *S. beta-haemolyticus*.[4]

Patients should be screened for HTLV-I in cases of severe, resistant, recurrent eczema with secondary infection.[4] Atopic dermatitis (AD) and seborrheic dermatitis are the main differential diagnoses.[4] Histopathology is non-specific and CD8 T lymphocytes predominate in immunohistochemistry.[4] Approximately 10% of those infected develop adult T-cell leukemia/lymphoma and HTLV-I-associated myelopathy/adult tropical spastic paraparesis.[2–4] Symptoms tend to show remission at puberty but persist if they start at the adult age.[1,4]

IDH does not have a specific treatment or vaccine; however, it usually responds to antibiotics such as sulfamethoxazole and trimethoprim, and cephalexin, for long periods, with recurrence being common.[3,4] Infected individuals must be monitored due to the possibility of severe neurological and lymphoproliferative complications.

The interruption of the transmission involves screening blood donors, using condoms, family counseling, avoiding breastfeeding, and avoid sharing needles.[4]

IDH is relevant in the practices of dermatologists, infectologists, hematologists and neurologists and, despite its absence from the lists of neglected diseases, the perception is that it is very close to that situation.[4] It is not compulsorily notified, and there are not even policies for the prevention or care for the virus carriers.

We emphasize the importance of this case, as it occurred outside the endemic areas in Brazil – which are the northern and northeastern regions – and because it was managed as a recalcitrant AD for a long period.

☆ Study conducted at the Ambulatório de Dermatologia Sanitária do Rio Grande do Sul; Porto Alegre, RS, Brazil.