

Case report

Acute cutaneous botryomycosis of the hands

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ABSTRACT

Botryomycosis is a rare bacterial infection which is most commonly caused by *Staphylococcus aureus*. It usually affects the skin but can also involve the viscera. The cutaneous form typically presents as ulcerated plaques or nodules that evolve over months to years to form discharging sinuses. As opposed to the norm for this uncommon infection, our case had a relatively acute presentation. A 44-year-old male with a history of relapsed mantle cell lymphoma, stem cell transplant and chronic graft versus host disease on immunosuppressive therapy presented with 2–3 week history of sores on his hands. Punch biopsy and subsequent pathological testing confirmed the diagnosis of botryomycosis. The patient was subsequently treated with clindamycin for 30 days and the lesions completely resolved. In this case report, we highlight the acute presentation of this relatively rare and usually chronic infectious disease. © 2020 The Author(s). Published by Elsevier Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

Introduction

Cutaneous botryomycosis is a rare and chronic suppurative bacterial infection with the most common causative organism being *Staphylococcus aureus* [1,2]. Only a couple hundred cases have been reported worldwide. Skin lesions usually present as papules, nodules, fistulas, abscesses or ulcers with seropurulent discharge [3]. Our case depicts an acute presentation of this rare disease which usually takes months or years to manifest clinically.

Case presentation

A 44-year-old male presented to dermatology clinic with new skin lesions on his hands. Patient had past medical history of mantle cell non-Hodgkin lymphoma diagnosed four years ago which had relapsed three times since then. Patient had been treated with chemotherapy and stem cell transplant during each episode. He was also diagnosed with chronic graft versus host disease. At the time of current presentation, patient's mantle cell lymphoma had been in remission for about 6 months. He was taking daily ibrutinib and prednisolone every alternate day.

The patient had noted lesions on both hands 2–3 weeks prior to the day of presentation. The lesions started in areas of prior trauma and subsequently progressed to plaques that ulcerated and formed crusts. On examination, there were 7 lesions on the right hand and 4 lesions on the left hand (Figs. 1, 2). Two punch biopsies were

obtained, one for staining and the other for culture. Cultures grew *Staphylococcus aureus*. Hematoxylin and eosin stained slides showed pathognomonic sulfur granules comprising of a central solid collection of cocci bacteria surrounded by eosinophilic material also known as Splendore-Hoeppli effect (Fig. 3). A diagnosis of cutaneous botryomycosis was thus made and the patient was started on oral clindamycin 600 mg three times daily for 30 days which resulted in complete resolution of the lesions (Fig. 4).

Discussion

Botryomycosis is a chronic suppurative bacterial infection that can involve the skin and viscera. Around 200 cases of botryomycosis have been reported to date [3]. Risk factors include alcoholism, diabetes mellitus, HIV infection, cystic fibrosis, chronic granulomatous disease, trauma and surgery [4–10]. Like most other reported cases, our patient was immunocompromised. He had a history of recurrent mantle cell lymphoma, bone marrow stem cell transplant and chronic graft versus host disease on immunosuppressive therapy.

The most common etiological agent for cutaneous botryomycosis is *Staphylococcus aureus*. Other pathogens that have been associated with botryomycosis include coagulase negative *Staphylococcus*, *Streptococcus*, *Pseudomonas aeruginosa*, *Escherichia coli*, *Proteus*, *Serratia*, *Actinobacillus* and *Peptostreptococcus* [1]. Cutaneous variant of the disease can present as nodules, sinus tracts, abscesses or ulcers with seropurulent discharge [2,3]. The discharge can sometimes contain bacterial granules. Lesions usually take months to years to develop and ulcerate [12]. But

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Fig. 1. The lesions on right hand before treatment.



Fig. 2. The lesions on left hand before treatment.

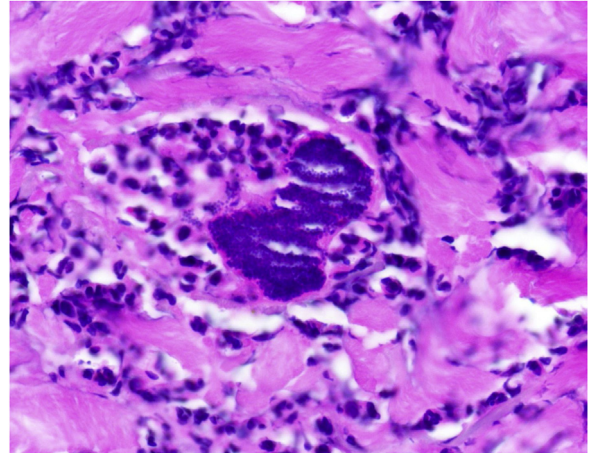


Fig. 3. The Splendore-Hoeppli phenomenon seen on biopsy of the lesions.



Fig. 4. Resolution of lesions on both hands after treatment.

contrary to the norm, our patient presented with ulcerated nodules within 2–3 weeks of initial disease onset. Botryomycosis can also affect various other organs of the body. The visceral variant most commonly affects lungs but it can also involve liver, spleen, kidney and brain [11].

Histological examination of skin lesions in botryomycosis shows formation of a rim of eosinophilia composed of antigen-antibody complexes, tissue debris and fibrin surrounding basophilic bacterial granules and colonies. This is known as the Splendore-Hoeppli phenomenon [11,13]. The characteristic histological findings along with culture are pivotal in making a

diagnosis of botryomycosis and help differentiate it from actinomycosis, nocardiosis and tuberculosis.

Treatment typically involves antimicrobial therapy for prolonged duration with surgical debridement if necessary. Our case was treated with clindamycin which led to resolution of lesions without need for any surgical debridement.

Conclusions

With this report, we aim to illustrate an atypical, relatively acute presentation of this rare chronic suppurative bacterial skin infection. Cutaneous botryomycosis should be considered as a possible differential for acute onset ulcerative skin lesions especially in immunocompromised patients.

CRediT authorship contribution statement

Angad Singh: Writing - original draft, Writing - review & editing.
Courtney Cook: Writing - review & editing. **Kaitlyn Kollmann:**
 Writing - review & editing. **Anand Rajpara:** Supervision.

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