CASE REPORT

A RARE CASE OF BOTRYOMYCOSIS IN AN IMMUNOCOMPETENT PATIENT

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The term botryomycosis is a misnomer meaning bunch of grapes and fungal infection. It is a chronic, granulomatous infection caused by the bacteria with only a few cases reported worldwide. We present a case of cutaneous botryomycosis in an immunocompetent patient which was identified on histopathology. The culture showed growth of Staphylococcus aureus which was sensitive to oral and intravenous antibiotics. The patient was started on oral linezolid and the lesions improved significantly without the need of surgical debridement and amputation. Botryomycosis serves as a diagnostic dilemma as the disease mimics other disorders like actinomycetoma, eumycetoma, scrofuloderma and nocardiosis. Therefore, it is important to consider botryomycosis as a differential in chronic suppurative infections even in immunocompetent hosts.

Keywords: Botryomycosis; Fungal infection; Immunocompetent patient

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INTRODUCTION

The term botryomycosis is Greek in origin and a misnomer where 'botryos' means bunch of grapes and 'mycosis' means fungal infection.[1] Botryomycosis is a chronic, granulomatous, suppurative infection caused by bacteria.[2] After the discovery of bacterial etiology, many other terminologies have been suggested like actinophytosis, staphylococcic actinophytosis, bacterial pseudomycosis and granular bacteriosis.[3] Botryomycosis is a rare cutaneous infection with only a couple of hundred cases reported worldwide.[4] Cutaneous botryomycosis primarily results from the direct inoculation of the organism at the site of trauma or injury in immunocompromised individuals.[5] The cutaneous lesions may present as papules, nodules, fistulas, abscesses, and ulcers with seropurulent discharge.[6] In most cases, the identified causative organism is *Staphylococcus aureus* and others like *Streptococcus* spp., *Pseudomonas aeruginosa*, *Escherichia coli*, *Serratia* spp., and anaerobes are less common.[4] Very few cases of the infection have been reported in immunocompetent hosts. We report the case of a young immunocompetent male affected by cutaneous botryomycosis caused by *Staphylococcus aureus.*

CASE PRESENTATION

A 16-year-old man, a resident of a rural area of Sindh, Pakistan, a farmer by occupation and with no previous known comorbidities presented via outpatient department of dermatology of a tertiary care hospital in Karachi, Pakistan with a complaint of large swelling over his right foot for last two years. According to the patient the lesions developed after local trauma during farming. There was no history of tuberculosis, or tuberculous contacts, diabetes or immunosuppression. Initially, there was the development of small vesicular lesions which ruptured spontaneously and led to the development of discharging sinuses. The lesions progressed and multiplied over time with secondary draining sinuses with a serosanginous and haemorrhagic discharge. However, the patient denies any expression of yellow, black or white grains. The patient had no previous significant drug, travel or family history.

On examination, he was a young male of average height and build, alert, oriented and vitally stable during the time of examination. His sub-vitals examination was normal except for the presence of pitting oedema of the right foot extending to the ankle joint. The systemic examination for cardiovascular, respiratory, central nervous system and abdominal examinations were all normal. On cutaneous examination multiple, erythematous to hyper-pigmented, hyperkeratotic, papules and nodules were appreciated extending onto the medial border and plantar surface of the right foot (figure 1 and 2). No discharge was appreciated. No grains could be expressed. A few small papules are also appreciated on the dorsum of the right foot. The initial differentials of the patient were considered to be acintomycetoma, eumycetoma, scrofuloderma, botryomycosis, chronic bacterial osteomyelitis and actinomycosis. The absence of grains were against the differentials of eumycetoma and actinomycetoma. Considering the above differentials, a skin biopsy specimen was obtained which was sent for histopathology, fungal smear, fungal and bacterial culture Mycobacterium tuberculosis culture, acid-fast bacilli (AFB) stain and gene Xpert. The fungal smear and cultures were negative. The investigations for Mycobacterium tuberculosis and non-tuberculous

mycobacteria such as Mycobacterium avium complex (AFB, gene Xpert and cultures) were negative. The histopathology report revealed an intact epidermis with overlying hyperkeratosis. Focal ulceration is also noted. The dermis shows foci of abscess formation and giant cell reactions. No fungus or sulphur granules were seen. No evidence of tuberculosis or malignancy was documented. The bacterial culture showed growth of a golden-yellow pigmented, opaque colony that was diagnosed as *S. aureus* by Gram staining, catalase and coagulase tests. Upon yielding the above-mentioned results a diagnosis of primary cutaneous botryomycosis caused by *S aureus* was made. An initial X-ray of the right foot (anteroposterior and lateral views) was done (figure 3). A magnetic resonance imaging (MRI) with contrast was also obtained to rule out osteomyelitis. The report summarised ongoing marked severe infective cellulitis with secondary involvement of talar, cuboid and lateral cuneiform bones with reduced joint space identified between the intertarsal bones multiple developing abscesses were seen within the soft tissues, the largest one was seen along the medial malleolus and the planter aspect of the midtarsal bone measuring 4.5×1.4 cm and 2.2×1.0 cm respectively. The orthopaedic department was taken on board and an initial consensus to commence treatment with oral antibiotics was put in motion keeping the risks of amputation and disability in a young immunocompetent male. The patient was started on oral linezolid 600 mg twice a day, based on the culture and sensitivity report for 4 weeks. An initial response was noted with significant improvement in the clinical examination of the patient (figure 4). The therapy was continued for another four weeks after the clinical cure. The patient has been under surveillance for the last 6 months with no reoccurrence of his disease and symptoms.

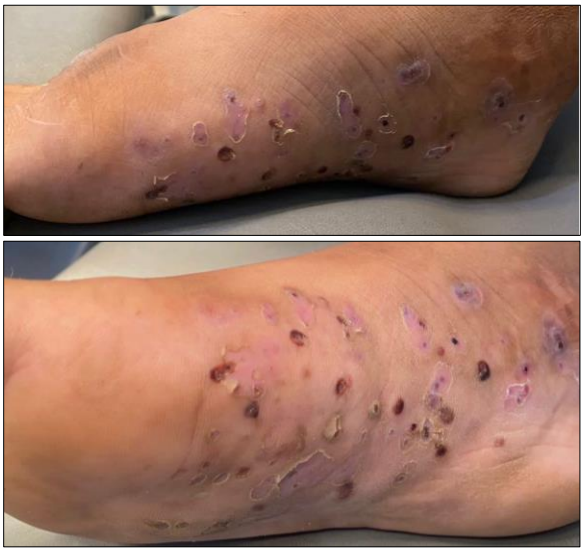

Figure 1

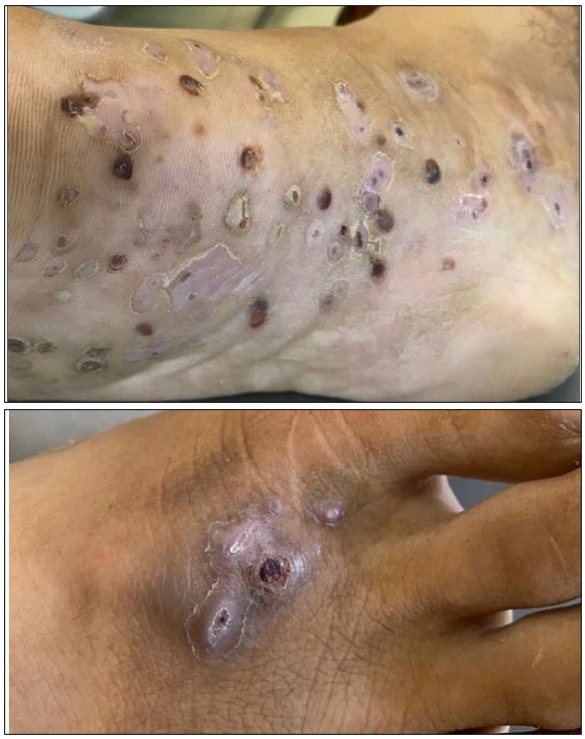

Figure 2

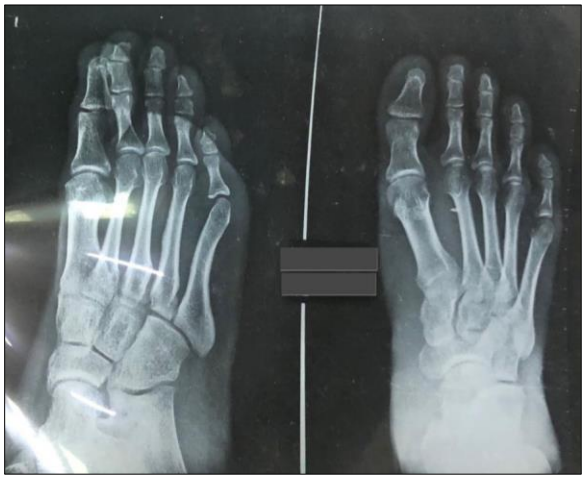

Figure 3

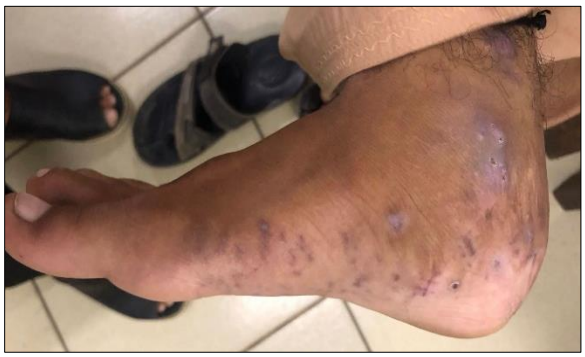

Figure 4

DISCUSSION

Botyomycosis exists in two clinical forms, cutaneous and visceral. The cutaneous form presents in the form of the development of papules, nodules, ulcers and draining sinuses, quite similar to the presentation of our patient. However, the visceral form usually manifests as pulmonary involvement in patients with underlying lung diseases like cystic fibrosis.[7] It can also involve other organs like the liver, spleen, kidney and brain.[4] Various comorbidities and immunocompromised conditions have been linked to botryomycosis such as diabetes mellitus, liver disease, alcoholism, lupus, cystic fibrosis, asthma, malnutrition, and immunoglobulin deficiency, long-term treatment with corticosteroids and human immunodeficiency virus /acquired immunodeficiency syndrome.[6] Most cases are seen to be triggered by trauma and direct inoculation of the organisms.[8] The trauma can be of varied aetiology ranging from burns, surgery, road traffic accidents, manual labour or disruption of the protective skin barrier.[9] The history of trauma may date back many years before the actual clinical presentation of the disease.

The pathogenesis is based on the direct inoculation of the organism. In cases of small inoculum, the organism is eradicated by the immunocompetent host, however, if the inoculum is large it can result in a robust immunological response resulting in the formation of papulonodular lesions rupturing into abscesses and draining sinuses.[9] In most cases, *S aureus* is the identified organism. Speedy recovery with clinical improvement is noted with appropriate antibiotic therapy in most cases. However, the diagnosis of botryomycosis can be clinically challenging due to its ability to mimic other granulomatous chronic cutaneous infections like actinomycosis, tuberculosis, nocardiosis, and mycetoma.[4] Therefore, it is important to obtain histopathology along with bacterial cultures to determine antibiotic culture and sensitivity for the treatment of the cutaneous disease. In the review of the literature, it is seen that an appropriate antibiotic regimen can result in swift recovery without the need for surgical debridement or amputation. It is important to consider the differential of botryomycosis in other chronic granulomatous cutaneous infections in both immunocompetent and immunocompromised hosts. Cases may be mistaken or inappropriately diagnosed resulting in aggressive management with debridement or amputation. Therefore, it is important to perform the necessary investigations to reach a correct diagnosis and avoid hostile management of botryomycosis which improves significantly with antibiotic therapy.

REFERENCES

1. Padilla-Desgarennes C, Vázquez-González D, Bonifaz A. Botryomycosis. Clin Dermatol 2012;30(4):397–402.
2. Rit K, Saha R, Chakrabarty P. A Case Report of Staphylococcus aureus Induced Cutaneous Botryomycosis in a Patient with Acquired Immune Deficiency Syndrome. MAMC J Med Sci 2015;1(2):108–10.
3. Chintaginjala A, Harshavardhan K, Kumar AS. Cutaneous botryomycosis: a rare case report. Indian J Dermatol 2016;61(1):126.
4. Singh A, Cook C, Kollmann K, Rajpara A. Acute cutaneous botryomycosis of the hands. IDCases 2020;19:e00709.
5. Mechow N, Göppner D, Franke I, Kolesnik M, Bonnekoh B, Gollnick HP, *et al*. Cutaneous botryomycosis diagnosed long after an arm injury. J Am Acad Dermatol 2014;71(4):e155–6.
6. Sirka CS, Dash G, Pradhan S, Naik S, Rout AN, Sahu K. Cutaneous botryomycosis in immunocompetent patients: a case series. Indian Dermatol Online J 2019;10(3):311–5.
7. Devi B, Behera B, Dash ML, Puhan MR, Pattnaik SS, Patro S. Botryomycosis. Indian J Dermatol 2013;58(5):406.
8. Follows GA, Mathew J, Lucas S, Black MJ, Goodship TH. Cutaneous botryomycosis in a patient with lupus nephritis. Nephrol Dial Transplant 1998;13(12):3200–1.
9. 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.

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