Botryomycosis presented as chronic non-healing ulcer

Botryomycosis is a chronic suppurative infection characterized by a granulomatous inflammatory response to bacterial pathogens which may present with cutaneous or visceral involvement (Winslow et al, 1959). The name "botryomycosis" was coined in 1884, but its bacterial nature was not discovered until 1919 (Bersoff-Matcha et al, 1998). The term "botryomycosis" is derived from the Greek word "botrys" (meaning "bunch of grapes") and "mycosis" (a misnomer, due to the presumed fungal aetiology in early descriptions).

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The most common causative bacteria is *Staphylococcus aureus* and occasionally *Pseudomonas spp*, *Escherichia coli*, *Proteus spp*, and *Streptococcus spp* (Devi et al, 2013).

Botryomycosis can mimic mycetoma — a chronic infection of the skin and subcutaneous tissue — in an immunocompetent individual. It is multifocal in its development, characterized by granulomatous bacterial infection which also can occur in an immunosuppressed patient (Rit et al, 2015).

Case report

We report a case of cutaneous botryomycosis in an immunocompetent 11-year-old patient, a primary school student, living at Hutan Melintang, Perak.

He presented to our orthopaedics clinic with a non-healing ulcer at his left foot, which he had for a duration of 1 year *[Figure 1]*. He described that the skin lesion had an insidious onset and was associated with itchiness. The patient reported no preceding history of trauma, animal bite or insect bite. Otherwise the patient did not complain of any pain in the left foot or associated fever. He was able to ambulate well with the lesion. One previous antibiotic course was prescribed by a general practitioner, however, this only partially resolved the condition.

Upon review at the clinic, the patient's general condition was well and his systemic examination unremarkable.

On examination of his left foot, however, there was an ulcer measuring 6 x 5 cm with an hypertrophic edge and central depression. There was also minimal pus at the base of the ulcer with minimal erythematous and warmness of the skin surrounding the ulcer.

Examination of the right foot was normal. The range of movement of left ankle and foot are normal. No palpable lymph node present over his inguinal or any other sites.

In view of chronic wound, initially squamous cell carcinoma of the left ankle was considered.

The patient and his mother were asked for their consent in taking an urgent biopsy of the skin lesion, which they agreed to. The patient was scheduled for urgent four-quadrant skin biopsy over the left ankle.

Intra-operative finding: regular skin ulcer with well-defined border and asymmetric anterior ankle swelling.

A four-quadrant biopsy was done at 3, 6, 9, 12 o'clock (marked as A, B, C, D respectively). Some superficial pus discharge was noted, and culture and sensitivity tests were taken. The tissue underneath the ulcer was healthy with no marked tissue attachment.

The patient was given oral cloxacillin empirically for 1 week to as a precaution.

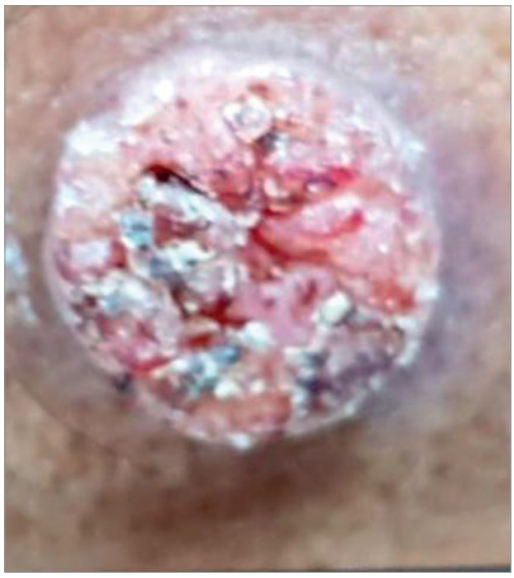

Figure 1. Initial lesion during first presentation

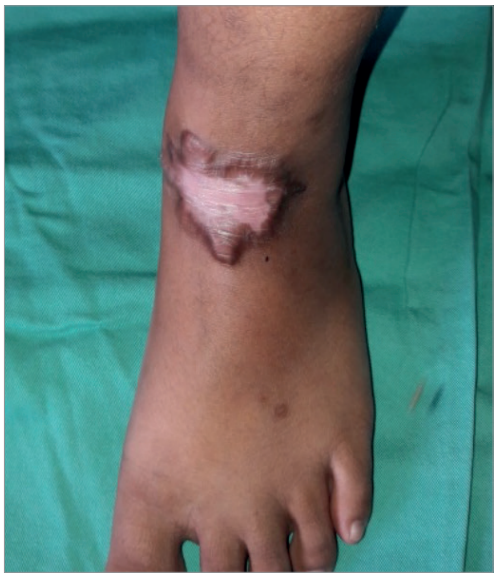

Figure 2. Well-healed scar post wound debridement

Four quadrant histopathology result
Samples A, B, C, D showed marked pseudoepitheliod hyperplasia with multiple intraepidermal and dermal micro-abscesses. A few vague granulomatous and foreign body type giant cell responses, which are associated with ruptured follicles, could be seen but no necrosis. The Periodic acid-Schiff (PAS) and Gomori methenamine silver (GMS) stains were negative with regards to fungal elements. The Ziehl Neelsen stain was negative for acid fast bacilli. The Gram stain showed numerous gram positive cocci within the dilated follicles filled with lamellated keratin.

Interpretation: Botryomycosis
Swab culture and sensitivity: mixed growth.

Upon review of the HPE result, wound debridement of the left foot was planned.

Intraoperative finding: Left ankle (dorsum) wound overgrowth, 6 x 5 cm, thorough wound debridement was done, no pus discharge noted.

The patient was discharged and given oral cefuroxime for 1 week. He recovered well post operatively. The wound was dressed with a hydrogel (DermaSyn) dressing daily.

On the subsequent follow up, the patient was found to be well. The wound fully healed after 1 month post surgical debridement *[Figure 2]* and therefore discharged from the authors' clinic.

Discussion

Cutaneous botryomycosis is a rare disease, scarcely described in the international literature. The incidence rates of botryomycosis in Malaysia are not known, probably due to under-reporting or misdiagnosis of the condition.

The pathogenesis of botryomycosis is not well-known. It is associated with defects of cellular immunity, particularly with low lymphocyte counts. Botryomycosis requires a delicate balance between number of microorganisms inoculated, their low virulence, and host tissue response for it to occur (Rit et al, 2015).

Botryomycosis of the skin should be differentiated from mycetoma, actinomycosis, chronic abscess, tuberculosis and skin malignancies.

Conclusion

Botryomycosis is a rare condition, but nowadays the incidence is increasing. Cases may be missed or mistakenly identified and mainly confused with fungal infections. The authors strongly suggest that an urgent skin biopsy should be to be done in suspected cases of cutaneous botryomycosis. In this case, the patient responded well to thorough wound debridement and antibiotic therapy. WAS

References

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