



## Case Study

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### Cutaneous Botryomycosis: A Rare Case Report

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Botryomycosis is a rare chronic suppurative granulomatous bacterial infection, which can present as cutaneous or visceral disease. We present a case of cutaneous botryomycosis in a 68-year-old female patient, who presented to the Skin outpatient clinic with indurated nodular masses along with discharging sinuses, on the lower abdomen, persisting for the past 10 years. The patient underwent various treatments elsewhere with no improvement. Mycetoma, botryomycosis, and actinomycosis were considered as differential diagnoses. On culture, *Staphylococcus aureus* was isolated, which is the most common cause of botryomycosis. As per the sensitivity report, treatment was started with co-trimoxazole and the patient responded well to the antibiotic treatment given, hence the aetiological diagnosis is important for the management of such cases.

#### Introduction

Botryomycosis is a rare chronic suppurative granulomatous bacterial infection, involving the skin and viscera, in which the organisms form granules composed of bacterial masses that are bound to each other [1]. The cutaneous form may present as nodules, abscesses, and sinuses with purulent discharge and grains, that may heal after several months to leave atrophic scars [2]. In 1870, botryomycosis was originally discovered by Otto Bollinger, as a post-castration complication in a horse and in 1884, the name was coined by Sebastiano Rivolta, which refers to its grape like granules

(Gr. botryo = bunch of grapes, mykes = fungus) and the mistakenly implied fungal aetiology. The first paper on botryomycosis in humans was published in 1919, which stated *Staphylococcus aureus* as the aetiological agent, contrary to the initial thoughts and the granules were not composed of mycotic masses, but were the bacteria that adhered to each other [3]. The nomenclature is a misnomer as it is caused by true bacteria and not by fungus [4]. *Staphylococcus aureus* has been incriminated in 40% cases, followed by *Pseudomonas aeruginosa* in 20% cases, while other organisms have also been reported which include coagulase negative

staphylococci, *Streptococcus spp.*, *Escherichia coli* and *Proteus spp* [5].

We present a case of cutaneous botryomycosis in a 68-year-old female patient, who presented to the Skin outpatient clinic with indurated nodular masses along with discharging sinuses, on the lower abdomen, persisting for the past 10 years.

### Case Report

In May, 2019, a 68-year-old female, resident of Nepal, presented to the Skin outpatient clinic of Sarojini Naidu Medical College, Agra, having indurated nodular masses with discharging sinuses, on the lower abdomen above the right groin that was of 10 years duration (Fig. 1). She also gave past history of trauma 10 years back at the same site. The patient underwent various treatments elsewhere with no improvement. There was no history of diabetes mellitus, tuberculosis and immunosuppression. She was non-reactive for HIV (Human immunodeficiency virus) and there was no evidence of any other disease or abnormality. Her routine investigations were within normal limit. Mycetoma, botryomycosis, and actinomycosis were considered as differential diagnoses, and the patient was then referred to the Microbiology Department for further confirmation. In the laboratory, pus was collected from the lesion and few fine sandy yellowish coloured grains were visible in the pus. Pus was then transferred to a sterile petridish and few yellowish grains were picked up with a sterile blunt forceps and transferred to a drop of sterile saline in another petri dish. On examination, the grains were found to be 0.5 to 1 mm in size and round to oval in shape with fairly regular margins. Two grains were crushed between two sterile glass slides. One of the slides was used for Gram staining and the other for 10% KOH wet mount preparation. The Gram-stained smear of the

crushed granule revealed a compact, granular, gram-positive mass, made of Gram-positive spherical cocci, which were about 1  $\mu\text{m}$  in size. KOH wet mount did not show any fungal element. Culture was done on sheep blood agar at 37°C and Sabouraud Dextrose Agar (SDA) with antibiotics at room temperature and at 37°C. After overnight incubation, culture on blood agar yielded pure and heavy growth of golden yellow, beta hemolytic colonies (Fig. 2). Gram stain, positive catalase test, mannitol fermentation and positive slide and tube coagulase test confirmed it to be *Staphylococcus aureus*. On antimicrobial susceptibility testing by Kirby-bauer disc diffusion method, the isolate was found sensitive to co-trimoxazole (1.25/23.75 $\mu\text{g}$ ), clindamycin (2 $\mu\text{g}$ ), cefoxitin (30 $\mu\text{g}$ ), gentamicin (10 $\mu\text{g}$ ), ciprofloxacin (5 $\mu\text{g}$ ) and linezolid (30 $\mu\text{g}$ ) while resistant to penicillin (10 units), erythromycin (15 $\mu\text{g}$ ) and tetracycline (30 $\mu\text{g}$ ), thus the isolate obtained was methicillin-sensitive *Staphylococcus aureus* (MSSA). No growth was obtained on SDA.

As per the sensitivity report, treatment was started with co-trimoxazole for 2 weeks. After 2 weeks, the clinical examination showed drying of few sinuses. He was asked to continue co-trimoxazole for 1 more month and was advised to come for follow up.

### Results and Discussion

Botryomycosis is a rare disease with few cases being reported worldwide. Review of the literature has shown around 140 published cases on botryomycosis, and cutaneous form of the disease was addressed in 28 articles [6]. The pathogenesis of botryomycosis is not completely understood and is believed to be associated with defective cellular immunity, particularly with low lymphocyte counts. The major predisposing factors include skin trauma, postoperative complications,

treatment with steroids, diabetes mellitus, liver disorders, alcoholism and cystic fibrosis. Less common factors are malnutrition, AIDS (Acquired Immunodeficiency Syndrome), glomerulonephritis and bronchial asthma.<sup>7</sup> The primary cutaneous form is most common,

presenting with nodules, abscesses and multiple sinuses or fistulae with serous or purulent discharge with or without granules [7, 8]. The prognosis of cutaneous botryomycosis is generally good and much better than the visceral type [9].

**Fig.1** Lower abdomen showing indurated nodular masses with discharging sinuses



**Fig.2** Blood agar showing beta hemolytic, golden yellow colonies of *Staphylococcus aureus*



Botryomycosis should be differentiated from conditions like mycetoma, actinomycosis and tuberculosis that have similar clinical features. Microscopy of the discharge and culture and sensitivity tests help in confirming the diagnosis. In the present case, *Staphylococcus aureus* has been isolated,

which is the most common cause of botryomycosis. This case has been reported due to its rarity and the likelihood to be mistaken for diseases such as mycetoma and actinomycosis, which differ in their etiology and treatment. The patient responded well to the antibiotic treatment given, hence the

aetiological diagnosis is important for the management of such cases.

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