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**Case Report**

# A Case of mistreated Botryomycosis as Nocardiosis, in a patient with MDR-TB

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**ABSTRACT**:
**Introduction**: Botryomycosis is a chronic purulent, granule-producing granulomatous infection affecting the skin, subcutaneous tissue, and visceral organs, primarily caused by *Staphylococcus aureus* and other bacterial species. It is commonly seen in individuals with impaired cell-mediated immunity and mimics mycetoma and actinomycosis, necessitating accurate diagnosis for effective treatment. **Case History**: A 26-year-old male presented with a gradually enlarging left cheek swelling following facial trauma six months prior. The swelling was associated with pus discharge and nodular formation, who was misdiagnosed and being treated as nocardiosis. The patient had a history of multidrug-resistant tuberculosis (MDR-TB) and alcohol dependence. Material & Methods: Pus samples were sent for microbiological examination, including Gram's stain, ZN stain, and culture on Blood agar, MacConkey agar, and SDA slope, and also for molecular studies. Growth was observed, and preliminary biochemical reactions were performed. **Results**: Gram-positive cocci in clusters were identified, with positive catalase and slide coagulase tests. Biochemical and antimicrobial susceptibility testing confirmed *Methicillin-resistant Staphylococcus aureus* (MRSA). Histopathological staining supported the diagnosis. Conclusion: Accurate diagnosis of botryomycosis is essential for targeted antibiotic therapy. Long-term treatment is required based on bacterial susceptibility patterns, ensuring effective management.

***Keywords: Botryomycosis, Methicillin resistant staphylococcus aureus, Splendore-Hoeppli reaction, granulation***

## INTRODUCTION:

Botryomycosis is a chronic purulent, granule-producing, granulomatous lesion of the skin, subcutaneous tissues and visceral organs, caused by several bacterial species. It occurs in patients with deficient cell-mediated immunity. Clinically and pathologically, it resembles mycetoma and actinomycosis. The bacterial species involve are *Staphylococcus aureus*, *Escherichia coli*, *Actinobacillus lignieresii*, *Pseudomonas aeruginosa*, Proteus, Rhodococcus and other species.

## CASE REPORT:

A 26-year-old male, factory worker, who was apparently normal 5 months back came with complaints of swelling over the left cheek which was insidious on onset and gradually progressive following a trauma to his face 6 months ago, and gradually increased in size to present size. Patient also complained of pus discharge for the same duration, with subsequent development of nodules. He was a known case of multi drug resistant tuberculosis (MDR-TB) and alcoholic dependant syndrome. The patient was under treatment for both MDR-TB and alcohol dependant syndrome at the time of admission. Current status for the same is unknown.

Patient was being diagnosed and treated as nocardiosis for his symptoms at a local hospital at that time. However, there was no improvement of the condition and hence the treatment was stopped after coming to our hospital. His father was also a known case of MDR-TB, who was under treatment as well. On local examination, a diffuse swelling over the left cheek, below the left eye, was palpated, with erythema, multiple pus discharges and one solitary ulcer over the swelling with unhealthy granulation over the base with irregular margins. Swelling was soft on touch, with pus discharge from the sinuses on pressing.

Diagnostic nasal endoscopy with biopsy under local anaesthesia was done. Histopathological reports showed a abscess cavities with bacterial colonies of mixed organisms. Colonies demonstrated eosinophilic

"clubbing" Splendore-Hoeppli reaction. Foreign-body giant cell reaction to granules was seen.

Radiological findings (MRI) showed heterogeneously enhancing soft tissue thickening in the left pre-septal, infraorbital, premaxillary region and buccal mucosal space with adjacent fat stranding and overlying cutaneous thickening with adjacent bony changes. Features were suggestive of infective etiology.

The patient showed dramatic improvement after treatment with parenteral clindamycin and amikacin for a cycle of 2 months with an additional course of oral doxycycline.

## MATERIALS AND METHODS:

The pus discharge, along with the dressing material were sent to Department of Microbiology, JNMC which were subjected to Gram's stain and Ziehl-Neelsen stain. Samples were inoculated onto different culture medias following standard protocols and guidelines. Dressing material was sent in view of finding any granules, to rule out other diagnosis. Once growth was seen in culture media, they were further identified using conventional biochemical reactions and tests, using standard positive and negative controls, to validate the tests. Antibiotic susceptibility tests for various drugs were done using disk diffusion method, according to CLSI guidelines. The pus sample was also sent for Gene-Xpert, which was Negative for MDR-TB.

## RESULTS:

**Table 1: Table showing observation of different tests/ growths**

| Sl. No | Test performed / Growth seen | Observation |
|---|---|---|
| 1. | Primary smear: *Gram's stain* | Pus cells (++), epithelial cells (+), gram positive cocci in singles were seen |
| 2. | Primary smear: Ziehl-Neelsen *stain* | *Negative for acid fast bacilli* |
| 3. | Blood agar: | Small circular golden β-haemolytic colonies were seen. |
| 4. | MacConkey agar | Small circular convex lactose fermenting colonies seen |
| 5. | Grams stain from colonies | Gram positive cocci, approximately 1 μm in diameter, arranged characteristically in grape like clusters were seen. |
| 6. | Catalase test | Positive |
| 7. | Slide Coagulase | Positive |
| 8. | Tube Coagulase | Positive |
| 9. | Urease | Hydrolysed |
| 10. | Mannitol Motility Test | Fermented, Non-Motile |
| 11. | Methyl red | Positive |
| 12. | Voges-Proskauer | Positive |

The microbiological examination of the pus sample revealed *Staphylococcus aureus* as the causative organism. Primary Gram's staining showed Gram-positive cocci in clusters. Ziehl-Neelsen stain was done to rule out other differential diagnosis. The culture on Blood agar displayed small circular β-haemolytic golden colonies, while MacConkey agar revealed convex lactose-fermenting colonies. The isolate tested positive for catalase, slide coagulase, and tube coagulase, confirming its identification as *Staphylococcus aureus*. Additional biochemical tests, including urease hydrolysis, mannitol motility, methyl red, and Voges-Proskauer, further supported this identification.

**Table 2: Table showing the antibiogram (acc. to CLSI guidelines)**

| Drugs | Sensitivity |
|---|---|
| Cefoxitin | Resistant |
| Erythromycin | Resistant |
| Clindamycin | **Sensitive** |
| Cotrimoxazole | **Sensitive** |
| Azithromycin | Resistant |
| Ampicillin | Resistant |
| Ciprofloxacin | Resistant |
| Levofloxacin | Resistant |
| Linezolid | **Sensitive** |
| Gentamicin | Resistant |

Antimicrobial susceptibility testing using the Kirby-Bauer disk diffusion method showed resistance to cefoxitin, erythromycin, azithromycin, ampicillin, ciprofloxacin, levofloxacin, and gentamicin, indicating Methicillin-resistant *Staphylococcus aureus* (MRSA). However, the isolate remained sensitive to clindamycin, cotrimoxazole, and linezolid, suggesting these as potential therapeutic options. This highlights the importance of antimicrobial stewardship to ensure effective treatment strategies for MRSA-associated botryomycosis.


**Figure 1: Multiple nodules with few discharging sinuses seen over the face pre-treatment (A); Post – treatment after 2 months shows resolving of most lesions (B)**

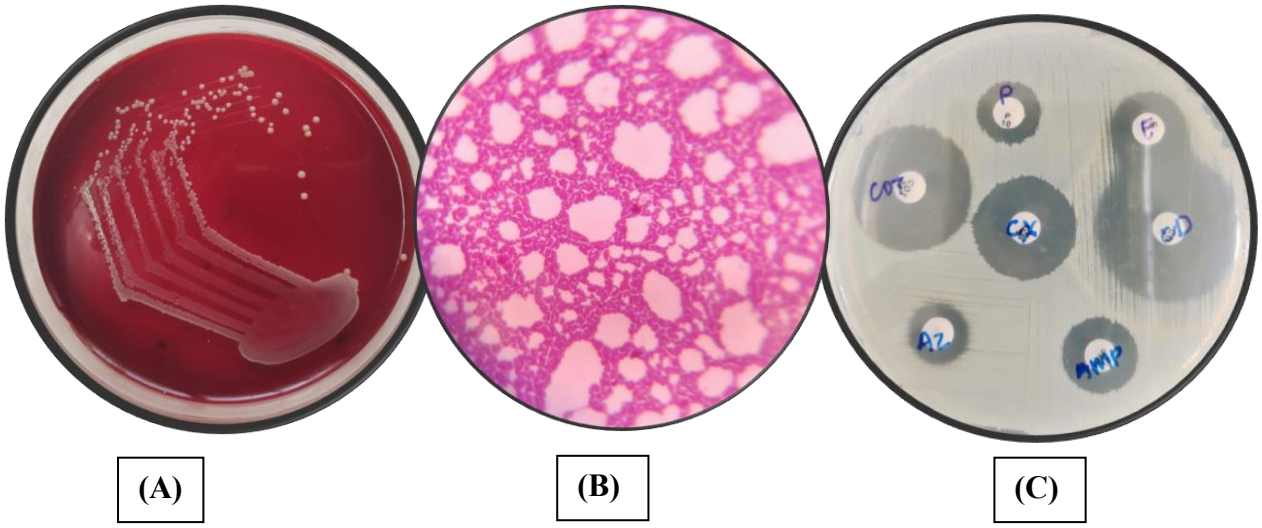


**Figure 2: Golden yellow beta-haemolytic colonies seen in blood agar (A); Gram positive cocci in cluster seen on Gram staining (B); Disk diffusion method showing Methicillin resistant Staphylococcus aureus (C)**

## DISCUSSION:

Botryomycosis, a rare bacterial infection, often mimics mycetoma and actinomycosis, leading to misdiagnosis. In this study, *Staphylococcus aureus* was identified as the causative organism, consistent with prior cases (1-3). Histopathology confirmed the diagnosis by demonstrating the Splendore-Hoeppli reaction, a hallmark feature of botryomycosis (4).

A review of the referenced literature highlights similar cases in diverse anatomical locations. Sirka et al. (2019) (1) and John et al. (2018) (2) reported cutaneous botryomycosis in immunocompetent individuals, challenging the notion that it primarily affects immunosuppressed patients. Chintaginjala et al. (2016) (3) described a rare case, underscoring the need for heightened clinical awareness. Ortiz et al. (2024)

(4) expanded the bacterial spectrum by identifying *Staphylococcus simulans* as a causative agent.

The findings align with previous reports that MRSA is an emerging pathogen in botryomycosis. Siddig et al. (2022) (5) documented *S. aureus* as the causative agent in foot botryomycosis, mimicking actinomycetoma. Similarly, Singh et al. (2020) (6) and Masuka et al. (2024) (7) emphasized prolonged antibiotic therapy as the key to treatment. Our study demonstrated a successful response to clindamycin and amikacin, reinforcing their efficacy against MRSA-related infections (8).

## New Findings from This Study:

While previous studies primarily focused on immunosuppressed or immunocompetent patients (1-3) this study presents a case of botryomycosis in a patient with multidrug-resistant tuberculosis (MDR-TB) and alcohol dependence an association not previously well-documented. The immunomodulatory impact of MDR-TB and chronic alcohol use could play a role in the pathogenesis of botryomycosis, warranting further research.

Additionally, while most studies report surgical intervention in cases of botryomycosis (4,6), our case showed complete resolution with prolonged antibiotic therapy alone, avoiding unnecessary surgical debridement. This highlights the potential for conservative management in select cases.

## CONCLUSION:

This study adds to the growing evidence of *Staphylococcus aureus*, particularly MRSA, as a significant etiological agent of botryomycosis. The case underscores the importance of microbiological culture, histopathological confirmation, and antibiotic susceptibility testing for accurate diagnosis and effective management. While previous reports have mainly focused on immunocompromised individuals, this case contributes new insights into botryomycosis in patients with MDR-TB and alcohol dependence. The favourable response to clindamycin and amikacin further supports their use in MRSA-related soft tissue infections. Early recognition and tailored antimicrobial therapy remain critical in preventing complications and improving patient outcomes.


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## CONFLICT OF INTEREST STATEMENT:

All authors declare that they have no competing interests.

## DATA AVAILABILITY STATEMENT:

The data that support the findings of this study are available from the corresponding author.

## CONSENT:

Verbal informed consent was obtained from the patient

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