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Case Report An atypical manifestation of mycetoma around knee joint - A case report

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| ARTICLE INFO | A B S T R A C T |
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| Article history: Received 19-08-2024 Accepted 03-10-2024 Available online 24-12-2024 | A chronic granulomatous disease called mycetoma affects subcutaneous tissue and in more advanced stages, destroys bone. Large, painless swellings that resemble tumours, nodules and draining sinuses and a discharge that contains grains are its distinguishing features. A 40-year-old man arrived at our outpatient clinic complaining of diffuse edema and pain surrounding his left knee joint that had been present for the previous two years, along with sinus and granular discharge. The |
| Keywords: Knee mycetoma Eumycetoma Wide excision Antifungal agents Knee infection Knee infective arthritis Joint infection | patient claims to have had a thorn prick thirty years ago. One differential diagnosis for the current situation was septic arthritis. Mycetomas are usually classified using a classification called "staging-classification of mycetoma," and the current case was classified as Stage-C. Based on the "Staging classification of mycetoma," the patient's condition was identified as Stage-C mycetoma due to pain and generalized oedema around the left knee. The isolated organism was Madurella mycetomatis. A comprehensive curettage involving multiple stages, debridement and excision of damaged tissue from the anterior and posterior aspects of the knee joint using various techniques was carried out. Anti-fungal medication was added for six months prior to and six months following the procedure and the results at the 6-month mark indicated a favourable outcome. |
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1. Introduction

A localized, persistent infection known as mycetoma is brought on by a variety of fungal or actinomycete species. It is identified by the accumulation of the causing organisms (grains) within abscesses. Draining sinuses allow grains to be released to the surface.¹

A chronic granulomatous illness called mycetoma affects subcutaneous tissue and in more advanced stages, destroys bone.² Sinus creation, granule production and bulk in the subcutaneous and intramuscular regions are the defining characteristics.

Actinomycetoma (bacteria) and Eumycetoma (fungi) are the two forms of mycetomas that are most frequently observed.²

Although mycetoma can affect any body part with a small male predominance (3-4:1), the foot is the most frequently affected part.³

The care of mycetoma necessitates a thorough clinical history, as well as examinations such as ultrasonography, magnetic resonance imaging, fine-needle aspiration and histological testing. For the purpose of surgical planning and result prediction, the causative agent, the extent of the disease and the location are critical factors. Wide surgical excision combined with medicinal care is the preferred course of treatment in these cases.^{4,5}

It can cause serious tissue loss, subsequent bacterial infections, limb abnormalities and other terrible problems if it is not addressed appropriately.

Despite receiving medicinal and surgical care, 25-50% of individuals experience recurrences. There are currently relatively few published works on this uncommon

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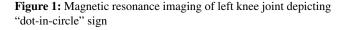
mycetoma appearance around the knee joint.

2. Case Report

A 40-year-old man arrived at our outpatient department complaining of two years of pain and widespread oedema around his left knee joint, along with sinus and granular discharge. The patient claims to have had a thorn prick thirty years ago. One differential diagnosis for the current situation was septic arthritis.

A thorough history and clinical examination revealed pain, gross swelling with previous scar mark, multiple sinuses with granular discharge and restriction in the left knee's movement. Swelling was in medial aspect of left knee joint, surface was nodular, margins were indistinct, hard in consistency, not reducible, not compressible, fixed to overlying skin with involvement of underlying muscle. Haematological investigations and digital radiographs were taken (Figure 3).

Consequently, thickened and inflammatory synovium along with numerous variable-sized hyperintense lesions resulting in a central "dot-in-circle" sign were seen in the subcutaneous, intramuscular and synovial regions of the left knee joint during magnetic resonance imaging (Figures 1 and 2).



"Staging-classification of mycetoma" is usually used for classifying mycetoma and as per classification, the current case was classified as Stage-C (Figure 4).

Stage description

A – No sinuses only swelling

 $B\ -\ Formation$ of sinuses and pustules with woody inducation

C – Digital radiograph showing bony involvement

D – Multiple lesions/spreading to distant sites.

Before the patient was taken for the surgical treatment, their informed consent was obtained. As a result, a multistage, comprehensive curettage involving debridement and the removal of damaged tissue from the anterior and

Figure 3: Thickened and inflammatory synovium with destroyed joint's articular surface

Figure 4: Stage C of "staging classification of mycetoma" with bony involvement







Figure 2: Magnetic resonance imaging of left knee joint depicting

variable sized hyperintense lesions

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Figure 5: At previous incision site midline skin incision was given in the left knee

posterior aspects of the knee joint using various approaches was carried out (Figures 9, 10, 11 and 12).

Neuraxial anaesthesia was administered to the patient. The patient was placed on the operating table in a supine position. A 320 mmHg tourniquet was applied and an 15–20 cm midline skin incision was made on the left knee's previous incision site (Figure 5). The pathology was found in the knee joint's subcutaneous plane, intramuscular area and it's articular surface during surgery (Figures 6, 7 and 8).

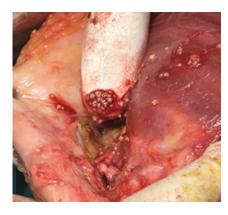


Figure 6: Granules in subcutaneous plane

Thorough curettage that included debridement and the excision of damaged tissue from the anterior and posterior aspects of the left knee joint and arthrotomy were performed (Figures 9, 10 and 11) and a thorough wash was administered. Closure was performed in layers.



Figure 7: Granules in intramuscular area



Figure 8: Granules in articular surface of joint.



Figure 9: Removal of damaged tissue



Figure 10: Removal of thickened synovium



Figure 11: Arthrotomy and debridement was done

A number of $2 \times 1 \times 1$ mm granules were removed (Figures 12 and 13) and sent for gram's staining, fungal culture, fungal staining, ZN staining, TB-PCR and histopathological examination. The diagnosis of mycetoma (Madurella mycetomatis) was confirmed by histopathological investigation, which revealed several fungal colonies encircled by inflammatory infiltrates (Figure 14). Fungal culture likewise produced conclusive result. Therefore, tablet Itraconazole (200 mg, twice a day) was given for 6 months preoperative and 6 months postoperative. The patient was followed for 6 months postoperatively and the period was uneventful.

3. Discussion

Mycetoma is a chronic disease that affects the skin and subcutaneous tissue. In its severe stages, it can cause gross destruction of muscles, bone damage and if left untreated, can reoccur.^{2,6} This disease is typically observed at latitudes of 30° north and 15° south, in tropical locations like Yemen,



Figure 12: Excised granules



Figure 13: Excised thickened synovium with granules

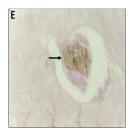


Figure 14: Histo-pathological examination of mycetoma

Mexico and India.

The case study originates from the arid region of western Madhya Pradesh, where a madurella infection of the knee joint accounts for the unusual nature of the illness. Because this condition presented in the outpatient department, we cannot comment on the incidence due to insufficient data.

Mycetomas typically show up with standard symptoms, however in 10-15% of cases, an unexpected presentation is noticed as in this example. The diagnosis of such presentations is mostly dependent on the clinical history, local examination, magnetic resonance imaging, and histological analysis.

The gold standard treatment for people with eumycetoma consists of broad local excision along with antifungal medications like itraconazole or ketoconazole. Including an anti-fungal medication in the treatment has the benefit of lowering the disease burden and fibrous tissue growth surrounding the lesion, resulting in sufficient lesion excision.⁷ There are a few different literatures that suggest 0.5–2cm as sufficient margins for local excision, but nothing is clear as of yet.^{8,9}

In the current instance, a multi-phased comprehensive curettage involving debridement and the excision of damaged tissue from the anterior and posterior aspects of the knee joint using various techniques was carried out. Itraconazole tablets (200 mg, twice daily) were administered for six months prior to surgery and six months following it.¹⁰ Because it is less hepatotoxic than ketoconazole, it was selected.

4. Conclusion

Based on the "Staging classification of mycetoma," the patient's condition was identified as Stage-C mycetoma and patient presented with pain and generalized edema around the left knee.¹¹ The isolated organism was Madurella mycetomatis. After a multi-staged, thorough curettage that included debridement and the excision of damaged tissue from the anterior and posterior aspects of the left knee joint using various approaches. Itraconazole (200mg, twice daily) was prescribed for 6 months preoperative and 6 months postoperative. The patient had a good outcome at final follow-up of 6 months.

5. Clinical Message

For accurate diagnosis and therapy in patients with unique features, radiographic inspection (MRI) and histopathological evaluation of the excised specimen are crucial.

6. Conflict of Interest

None.

7. Source of Funding

None.

8. Patient Consent

The patient provided informed consent, which the authors attest to, in order for this case report to be published.

References

- Suneet S, Anurag K. Surgical Diseases in Tropical Countries. 1st ed. New Delhi, India: Jaypee Brothers; 1996.
- Baki R, Mathur DR. Incidence and changing pattern of Mycetoma in Western Rajasthan. *Indian J Pathol Microbiol*. 2008;51(1):154–5.
- Panosian C. Tropical Infectious Diseases: Principles, Pathogens, and Practice: Edited by Richard L. Guerrant, David H. Walker, and Peter F. Weller. Philadelphia: Churchill Livingstone, 1999. 1644 pp., illustrated. \$295 . *Clin Infect Dis.* 2000;30(5):816.
- 4. Fahal AH. Management of mycetoma. *Expert Rev Dermatol.* 2010;5(1):1–7.
- 5. Fahal AH. Mycetoma: A thorn in the flesh. *Trans R Soc Trop Med Hyg.* 2004;98(1):3–11.
- Amaravati RS, Angamuthu N. Multiple foot sinues due to schizomycetes. *Indian J Orthop*. 2005;39(1):125.
- Suleiman SH, Wadella ES, Fahal AH. The surgical treatment of mycetoma. *PLoS Negl Trop Dis.* 2016;10(6):e0004690.
- Tamir G, Adler A, Hagler J, Hauben D, Goldberg I. Mycetoma of the foot-surgical treatment with free flap reconstruction. *Eur J Plast Surg.* 1995;18:124–6.
- Gismalla MDA, Abdulla GM, Ali MM, Mohamed SM. Wide surgical and reconstruction of eumycetoma in Gezira Mycetoma Centre. *Glob J Med Res*. 2016;16(2):14–21.
- 10. Scolding P, Fahal A, Yotsu RR. Drug therapy for Mycetoma. *Cochrane Database Syst Rev.* 2018;2018(7):CD013082.
- Singh B, Gehlot R, Saxena M, Bharwani N, Raichandani K, Bhati M. An unusual presentation of mycetoma around knee joint as a subcutaneous mass - A case report. J Orthop Case Rep. 2022;12(10):22–5.

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