

A TYPICAL PRIMARY EUMYCETOMA OF THE KNEE MIMICKING AN EPIDERMAL INCLUSION CYST: A RARE CASE REPORT

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ABSTRACT

Eumycetoma is a chronic granulomatous fungal infection of the skin and subcutaneous tissue that predominantly affects the foot following traumatic inoculation. Primary involvement of the knee is exceptionally uncommon and may present without the classical triad of painless swelling, multiple discharging sinuses, and extrusion of fungal grains, resulting in diagnostic uncertainty. Such atypical presentations can closely resemble benign soft tissue lesions, delaying definitive diagnosis and appropriate management. Primary eumycetoma involving the knee is exceedingly rare and poses a significant diagnostic challenge because of its nonspecific clinical presentation. The absence of characteristic sinus tracts and grain discharge may lead to misdiagnosis as benign cystic lesions, while cytological evaluation can be inconclusive due to failure to sample fungal grains. Histopathological examination with special fungal stains remains the gold standard for definitive diagnosis. Early recognition and complete surgical excision, combined with appropriate antifungal therapy, are essential to prevent disease progression, recurrence, and osteoarticular involvement. This case highlights the importance of considering eumycetoma in the differential diagnosis of chronic painless knee swellings in endemic regions, even in the absence of classical clinical features. Histopathological examination should be pursued when clinical and cytological findings are inconclusive, as early diagnosis facilitates timely treatment and favorable functional outcomes.

KEYWORDS: Eumycetoma, Mycetoma, Knee swelling, Epidermal inclusion cyst

INTRODUCTION

Eumycetoma is a chronic, progressively destructive subcutaneous fungal infection characterized by the classical triad of painless swelling, multiple draining sinuses, and the discharge of fungal grains. It results from traumatic inoculation of filamentous fungi into the skin and subcutaneous tissues and predominantly affects individuals living in tropical and subtropical regions. The disease most commonly involves the foot ("Madura foot"), although other anatomical sites such as the hand, leg, and trunk have occasionally been reported. Owing to its indolent course and nonspecific early clinical manifestations, diagnosis is frequently delayed, resulting in progressive soft tissue destruction and, in advanced cases, bone involvement and deformity.^{1–3}

Primary eumycetoma involving the knee is exceedingly rare, particularly in the absence of the characteristic discharging sinuses or grain extrusion. Such atypical presentations may clinically mimic benign soft tissue lesions including epidermal inclusion cysts, ganglion cysts, bursitis, or lipomas, thereby creating a significant diagnostic challenge. Imaging findings may also be nonspecific during the early stages, and definitive diagnosis generally relies on histopathological examination, microbiological culture, and increasingly, molecular diagnostic techniques.^{2–5}

The differential diagnosis of a long-standing, painless medial knee swelling encompasses a wide range of benign and malignant conditions. However, in endemic regions, chronic fungal infections such as eumycetoma should remain an important differential diagnosis, particularly when lesions demonstrate slow progression or recur after incomplete excision. Early recognition is crucial because delayed diagnosis may necessitate prolonged antifungal therapy, repeated surgical procedures, or even limb-sacrificing interventions in advanced disease.^{3,6}

We report a rare case of primary eumycetoma presenting as a long-standing medial knee swelling clinically suggestive of an epidermal inclusion cyst. The absence of classical clinical features contributed to the diagnostic dilemma, with the definitive diagnosis established only after surgical excision and histopathological examination. This case highlights the importance of maintaining a high index of suspicion for atypical presentations of eumycetoma in endemic areas and emphasizes the pivotal role of histopathology in establishing the diagnosis and guiding appropriate management.

Case Presentation

A 40-year-old male presented with a swelling over the medial aspect of the left knee that had been present since childhood. The swelling had remained static for several years. One month prior to presentation, the patient noticed a gradual increase

in the size of the swelling, associated with pain and tenderness. There was no history of trauma, fever, weight loss, or discharge of black or yellow granules. There was no history of previous surgical intervention at the site.

Clinical Examination

On local examination, a solitary, oval-shaped swelling measuring approximately 5×4 cm was identified over the medial aspect of the left knee. The overlying skin was stretched, and a small central punctum was noted. There was no evidence of sinus formation, active discharge, or surrounding cellulitis.

Diagnostic Investigations

Fine-Needle Aspiration Cytology

Ultrasound-guided fine-needle aspiration cytology (FNAC) was performed from the swelling over the medial aspect of the left knee. The aspirate was satisfactory for interpretation and showed no cytological evidence of malignancy.

Microscopic examination demonstrated numerous cyst macrophages arranged singly and in sheets, accompanied by abundant neutrophils and lymphocytes in a background of cellular debris, suggestive of a chronic inflammatory cystic lesion. Special staining for fungal elements was negative, and Ziehl–Neelsen staining for acid-fast bacilli (AFB) was also negative.

Based on these findings, the lesion was initially reported as a cystic lesion with associated inflammation, and clinical correlation was advised.

The FNAC findings were nonspecific and favored a benign inflammatory cystic process, thereby supporting the initial clinical diagnosis of an epidermal inclusion cyst. However, cytology alone was insufficient to establish the definitive diagnosis.

Histopathological Examination

Following complete surgical excision of the lesion, the specimen was submitted for histopathological evaluation.

Gross Examination

Gross examination revealed three grey-white to grey-brown soft-tissue fragments measuring collectively $5 \times 3.5 \times 3$ cm. On cut section, the lesion appeared cystic with a relatively firm consistency.

Microscopic Examination

Microscopic examination of hematoxylin and eosin-stained sections revealed skin with underlying dermis and subcutaneous tissue showing dense chronic inflammatory cell infiltrates composed predominantly of lymphocytes, plasma cells, and histiocytes.

Multiple microabscesses and areas of granulation tissue formation were identified. Embedded within these inflammatory foci were colonies of filamentous organisms morphologically consistent with mycetoma grains.

Special histochemical staining with Periodic acid–Schiff (PAS) and Gomori methenamine silver (GMS) demonstrated fungal hyphae within the inflammatory foci, confirming the fungal nature of the organisms. No evidence of malignancy was identified.

The overall histopathological features were diagnostic of eumycetoma (fungal mycetoma).

Clinicopathological Review

The lesion had initially been clinically suspected to be an epidermal inclusion cyst and was reported cytologically as an inflamed cystic lesion. In view of the unusual presentation and clinicopathological discrepancy, the histopathological slides and paraffin blocks were reviewed at a tertiary referral centre.

On review, fungal grains composed of branching fungal filaments were identified and highlighted by PAS and GMS staining. No evidence of malignancy was observed. The reviewed findings established the final diagnosis of eumycetoma (fungal mycetoma).

Final Diagnosis

Primary eumycetoma (fungal mycetoma) of the medial aspect of the left knee.

Surgical Management

In view of the patient's long-standing swelling, recent increase in size, and associated tenderness, along with inconclusive preoperative cytological findings suggestive of an inflamed cystic lesion, the patient was planned for excision biopsy for definitive diagnosis and treatment.

Under regional anaesthesia and strict aseptic precautions, an elliptical incision incorporating the punctum and adherent overlying skin was made over the medial aspect of the left knee. Skin flaps were carefully elevated, and meticulous sharp and blunt dissection was performed in the subcutaneous plane.

The lesion was identified as a well-circumscribed cystic mass with dense fibrotic adhesions to the surrounding soft tissue. Circumferential dissection was carried out outside the pseudocapsule, and the lesion was excised en bloc along with a cuff of surrounding fibro-inflammatory tissue to minimise the risk of residual disease and local recurrence. Care was taken to avoid rupture of the lesion and spillage of its contents during dissection.

The operative field was thoroughly irrigated with normal saline, meticulous haemostasis was achieved, and the wound was closed in layers. The excised specimen was sent for histopathological examination.

Following histopathological confirmation of eumycetoma, the patient was planned for appropriate systemic antifungal therapy and regular follow-up to monitor wound healing and detect any evidence of recurrence.

Final Diagnosis

Cystic lesion with inflammation suggestive of eumycetoma (fungal mycetoma).

Surgical Management

In view of the patient's long-standing swelling with a recent increase in size and associated tenderness, along with inconclusive preoperative cytological findings suggestive of an inflamed cystic lesion, the patient was planned for excision biopsy for definitive diagnosis and treatment.

Under regional anaesthesia and strict aseptic precautions, an elliptical incision incorporating the punctum and adherent overlying skin was made over the medial aspect of the left knee. Skin flaps were carefully elevated, and meticulous sharp and blunt dissection was performed in the subcutaneous plane. The lesion was identified as a well-circumscribed cystic mass with dense fibrotic adhesions to the surrounding soft tissue. Circumferential dissection was carried out outside the pseudocapsule, and the lesion was excised en bloc along with a cuff of surrounding fibro-inflammatory tissue to minimise the risk of residual disease and local recurrence. Care was taken to avoid rupture of the lesion and spillage of its contents during dissection.

The operative field was thoroughly irrigated with normal saline, meticulous haemostasis was achieved, and the wound was closed in layers. The excised specimen was sent for histopathological examination.

The initial histopathological examination demonstrated dense chronic inflammatory infiltrate with giant cells and microabscess formation, with organisms morphologically consistent with actinomycosis and PAS-positive staining. Owing to the unusual histopathological findings and clinicopathological discrepancy, the slides and paraffin blocks were reviewed at a tertiary referral centre. Review microscopy demonstrated fungal grains composed of branching fungal filaments highlighted by PAS and GMS stains, with no evidence of malignancy, establishing the final diagnosis of eumycetoma (fungal mycetoma). The patient was subsequently planned for appropriate systemic antifungal therapy and regular follow-up to monitor wound healing and detect any evidence of recurrence.



Figure 1. Intraoperative findings during excision of primary knee eumycetoma.

(A) Elliptical incision over the medial aspect of the left knee with exposure of the lesion. (B) Dissection of the lesion in the subcutaneous plane. (C) Mobilization and excision of the well-circumscribed lesion. (D) Surgical wound following removal of the lesion. (E) Gross appearance of the excised tissue fragments.

(B)



Figure 2. Gross photograph of the excised tissue fragments showing the cystic lesion and associated tissue material.

DISCUSSION

Mycetoma is a chronic granulomatous infection of the skin and subcutaneous tissue caused either by true fungi (eumycetoma) or filamentous aerobic bacteria (actinomycetoma). The disease is endemic in tropical and subtropical regions, particularly within the "mycetoma belt," which includes Sudan, India, Mexico, Senegal, and parts of South America. Despite being recognized as a neglected tropical disease by the World Health Organization, it continues to be underdiagnosed because of its slow progression, variable clinical manifestations, and limited awareness among healthcare professionals.^{2,4,7}

Eumycetoma most commonly affects the foot following traumatic implantation of fungal organisms from contaminated soil, accounting for nearly 70% of reported cases. Knee involvement is exceptionally uncommon, with only isolated cases reported in the literature. Lesions occurring at unusual anatomical sites frequently lack the characteristic triad of painless swelling, multiple discharging sinuses, and extrusion of fungal grains, making diagnosis particularly difficult.^{2,5,8}

Our patient presented with a chronic swelling over the medial aspect of the knee without sinus formation, grain discharge, or surrounding cellulitis. A small punctum was the only external cutaneous finding, and the lesion clinically resembled an epidermal inclusion cyst. Similar atypical presentations have been described in previous reports where eumycetoma masqueraded as benign soft tissue tumors, chronic bursitis, ganglion cysts, or lipomas, resulting in delayed diagnosis and treatment.⁷⁻⁹

Fine-needle aspiration cytology was initially suggestive of an inflamed cystic lesion, demonstrating cyst macrophages, neutrophils, lymphocytes, and cellular debris without evidence of fungal elements. Fungal and acid-fast bacilli stains were negative. These findings illustrate the limitations of FNAC in diagnosing eumycetoma because representative fungal grains may not be sampled, especially in localized lesions. Consequently, cytological findings are often nonspecific and may lead to an erroneous diagnosis of a benign inflammatory cyst.^{9,10}

Histopathological examination remains the gold standard for diagnosis in atypical cases. In the present case, excisional biopsy revealed chronic granulomatous inflammation with microabscess formation surrounding fungal colonies. The fungal hyphae were clearly demonstrated by Periodic acid-Schiff (PAS) and Gomori methenamine silver (GMS) staining, confirming the diagnosis of eumycetoma. Histologically, eumycetoma is characterized by fungal grains embedded within suppurative granulomatous inflammation accompanied by neutrophils, macrophages, multinucleated giant cells, fibrosis, and chronic inflammatory infiltrates.¹¹⁻¹³

Radiological imaging is useful in evaluating the extent of disease and detecting osseous involvement. Magnetic resonance imaging may demonstrate the characteristic "dot-in-circle" sign, which has been reported to be highly specific for mycetoma. However, this feature is generally observed in advanced disease and may not be present in small localized lesions, such as in the present case. The absence of bone involvement permitted complete surgical excision while preserving knee function.¹⁴

Current evidence supports combined surgical excision and prolonged systemic antifungal therapy for localized eumycetoma. Early diagnosis and complete excision significantly reduce recurrence and prevent progression to osteoarticular involvement, whereas delayed diagnosis may require multiple surgeries or even amputation in advanced disease.^{2,4,15}

The present case emphasizes that eumycetoma should be considered in the differential diagnosis of chronic painless swellings occurring at unusual anatomical sites in endemic regions, even in the absence of the classical clinical triad. Furthermore, FNAC may be nondiagnostic in localized disease, making histopathological examination with special fungal stains indispensable for establishing the diagnosis. Increased awareness of these atypical presentations may facilitate earlier recognition and improve patient outcomes.

CONCLUSION

Eumycetoma should be included in the differential diagnosis of chronic subcutaneous swellings of the extremities, particularly in endemic regions. Absence of classical sinuses or grain discharge may lead to misdiagnosis. Histopathological evaluation with fungal special stains and clinicopathological correlation remain the cornerstone for accurate diagnosis. Early recognition facilitates appropriate surgical management and timely initiation of antifungal therapy, thereby reducing recurrence and morbidity.

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