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RESEARCH ARTICLE

MYCETOMA OF THE FOOT: A RARE CASE OF MADURAMYCOSIS IN A SUDANESE MALE LIVING IN SAUDI ARABIA

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Abstract

Mycetoma, a rare tropical disease, manifests as chronic subcutaneous granulomatous lesions predominantly found in the mycetoma belt, encompassing Sudan, Mexico, and India. This study offers a comprehensive discussion and literature review, delving into the historical evolution of mycetoma since its initial description as "Madura foot" in 1832. Clinical presentation typically involves painless subcutaneous masses, sinus tracts, and discharges, with the foot being the primary site of affliction. In the context of this broader discussion, we present the case of a 52-year-old Sudanese male residing in Saudi Arabia, who presented with a chronic, painful swelling in the right big toe. The subsequent diagnosis revealed Maduramycosis, providing a nuanced perspective within the clinical landscape. Diagnostic methodologies rely on clinical features, imaging, and cultures to identify causative microorganisms. Actinomycetoma, induced by gram-positive filamentous actinomycetes, demonstrates responsiveness to antibiotics like trimethoprim-sulfamethoxazole or high-dose penicillin G. Conversely, eumycetoma, associated with Mudrellamycetomatis, exhibits poor responses to antifungals, necessitating surgical interventions ranging from excision to amputation. This study underscores the dynamic epidemiology of mycetoma, with emerging cases noted outside traditional endemic regions. By unraveling the clinical enigma surrounding mycetoma, this research contributes to an enhanced understanding of this complex disease, paving the way for improved diagnostic and therapeutic modalities. Ongoing research remains pivotal to addressing the challenges posed by mycetoma and advancing global efforts to manage and prevent its impact on affected populations.

Introduction:-

Mycetoma, a chronic granulomatous infection characterized by the formation of localized subcutaneous masses, poses a diagnostic challenge in clinical practice [1,2]. Mycetoma consider one of that rare tropical disease, characterized by swelling , discharge and sinus [3]. This condition is caused by either bacteria (actinomycetoma) or

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fungi (eumycetoma) and typically affects individuals residing in tropical and subtropical regions [4]. Mycetoma primarily involves the extremities, with the feet being a common site of manifestation [5]. The infection progresses slowly over the years, leading to the development of painless swelling and discharging sinuses, often accompanied by the formation of grains [3]. These grains, composed of aggregates of microorganisms and host tissue, are pathognomonic for mycetoma [3]. Despite the localized nature of the disease, mycetoma can result in severe deformities and disabilities if left untreated [6].

Due to the indolent nature of mycetoma, delayed diagnosis is not uncommon, leading to the persistence of the infection and increased morbidity [7,8]. Diagnosis is typically confirmed through a combination of clinical evaluation, radiological imaging, and histopathological examination of the excised lesions [2]. The treatment options of Mycetoma is consider difficult as it is dependent on the causative organism [9]. Treatment involves a combination of surgical excision and prolonged antifungal or antibacterial therapy, tailored based on the identified causative agent [9].

In this context, we present a case of a 52-year-old Sudanese male residing in Saudi Arabia, who presented with a chronic, painful swelling in the right big toe, ultimately diagnosed as Madura-mycosis.

Case presentation:

A 52-year-old Sudanese male, currently residing in Saudi Arabia, presented to the surgery clinic with a longstanding complaint of painful swelling in his right big toe, persisting for approximately 20 years. The patient reported a gradual increase in the size of the swelling over time and denied any history of discharge or trauma related to the affected toe. Notably, he had a previous medical history of right hand mycetoma, which had been excised but recurred, leading to re-excision.

Upon examination, the patient appeared vitally stable, and a small swelling was noted in the left big toe. There were no signs of discharge during the clinical assessment. To further investigate the condition, X-ray imaging of the right big toe was performed, revealing a small soft tissue lesion without apparent bony involvement (Figure 1). Additionally, ultrasound imaging showed a well-defined hypoechoic oval-shaped lesion measuring 4x8mm (Figure 2). Importantly, this lesion exhibited no vascularity, calcification, or connection to the bone.

Subsequently, the patient underwent surgical en-block resection of the mass, during which granules were detected. The histopathological examination of the excised mass, measuring 2x1.5x0.5cm, revealed gross features of multiple yellowish-brown foci (Figure 3). Microscopically, the specimen exhibited suppurative granulomas surrounding characteristic grains in the subcutaneous tissue. A neutrophilic infiltration was observed, surrounded by palisading histocytes. Beyond this, a mixed inflammatory infiltrate was identified, compromising lymphocytes, plasma cells, eosinophils, macrophages, and fibrosis. Occasional multinucleated giant cells were also noted. Importantly, no malignancy was detected.

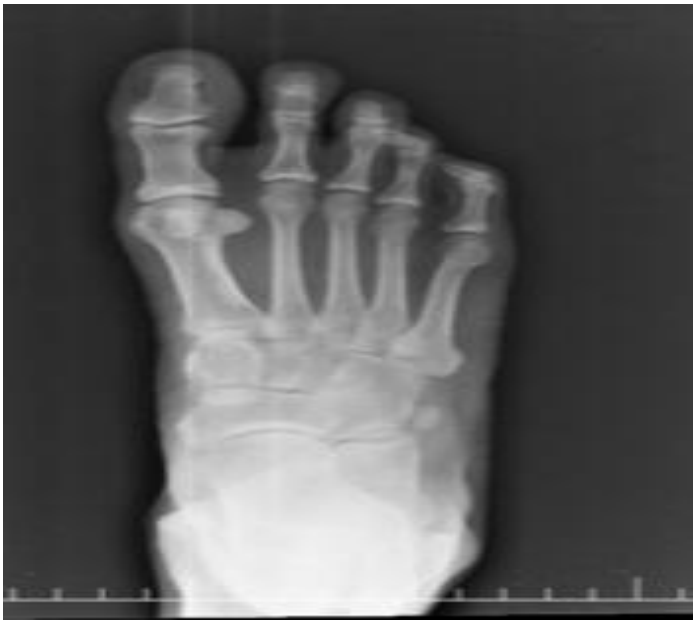


Figure 1:- The X-ray results of the caseshowing a small soft tissue lesion without apparent bony involvement (Black arrow).

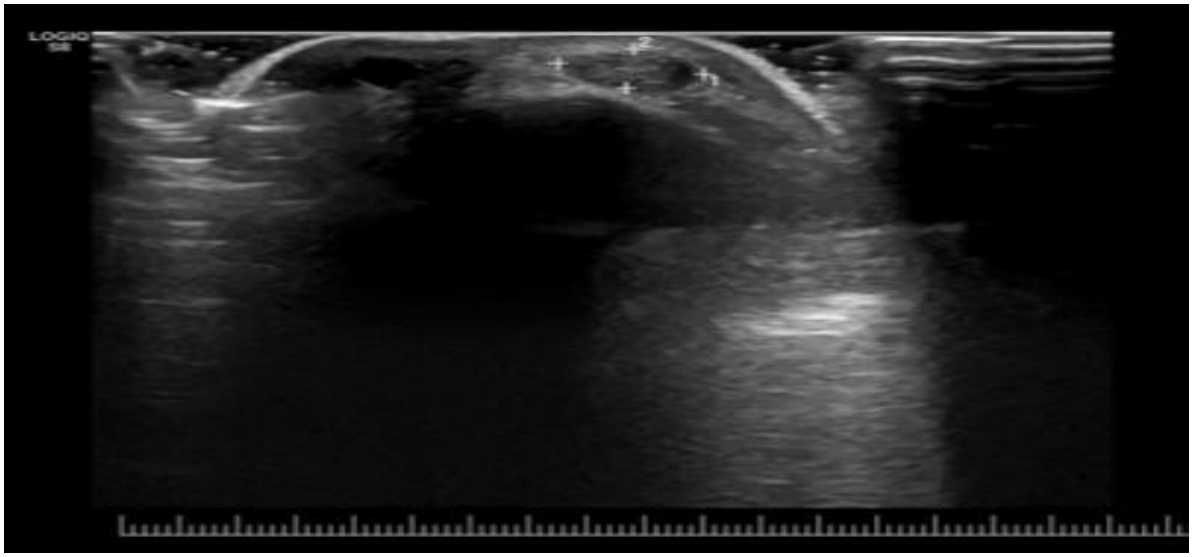
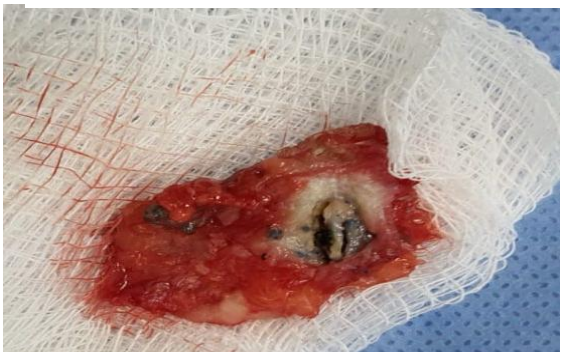


Figure 2:- Ultrasound imaging showing a well-defined hypoechoic oval-shaped lesion measuring 4x8mm (White arrow).



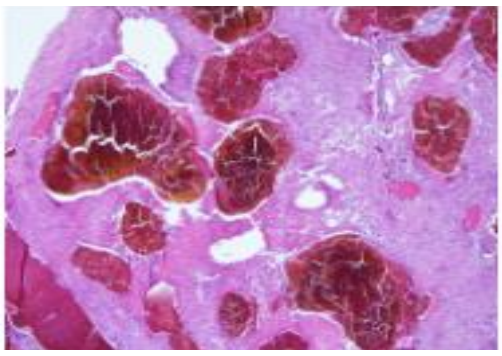


Figure 3:- The histopathological examination of the excised mass, measuring 2x1.5x0.5cm, revealed gross features of multiple yellowish-brown foci.

The final diagnosis based on the histopathological findings was Maduramycosis. Postoperatively, the patient recovered well and received fluconazole as part of the management plan. Regular follow-up will be crucial to monitor the patient's progress and address any potential recurrence or complications associated with Maduramycosis.

Discussion:-

Mycetoma, often referred to as a "fungal tumor," is a rare and neglected tropical disease, as recognized by the World Health Organization (WHO) following the Geneva meeting in May 2016 [10,11] (Figure 4). Initially described as "Madura foot" by Gill in 1832 in the South Indian city of Madurai, mycetoma presents as an unusual nodular appearance of the leg, particularly among field workers in the district [11]. Over the years, its prevalence in the mycetoma belt, encompassing Sudan, Mexico, and India, has been acknowledged [12,13]. Recent literature estimates suggest varying prevalence rates, emphasizing the need for comprehensive epidemiological studies. The disease primarily affects adults, with a notable male predominance [14,15]. Its clinical presentation as a devastating chronic subcutaneous granulomatous disease underscores the importance of early diagnosis and intervention [15,16].

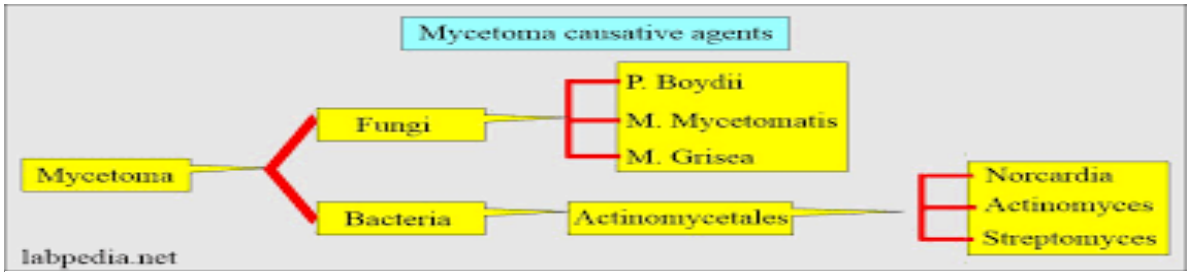


Figure 4:- The different types of mycetoma and its causative agents (Source: https://labpedia.net/wp-content/uploads/2020/01/Mycetoma-causative-agents2.jpg).

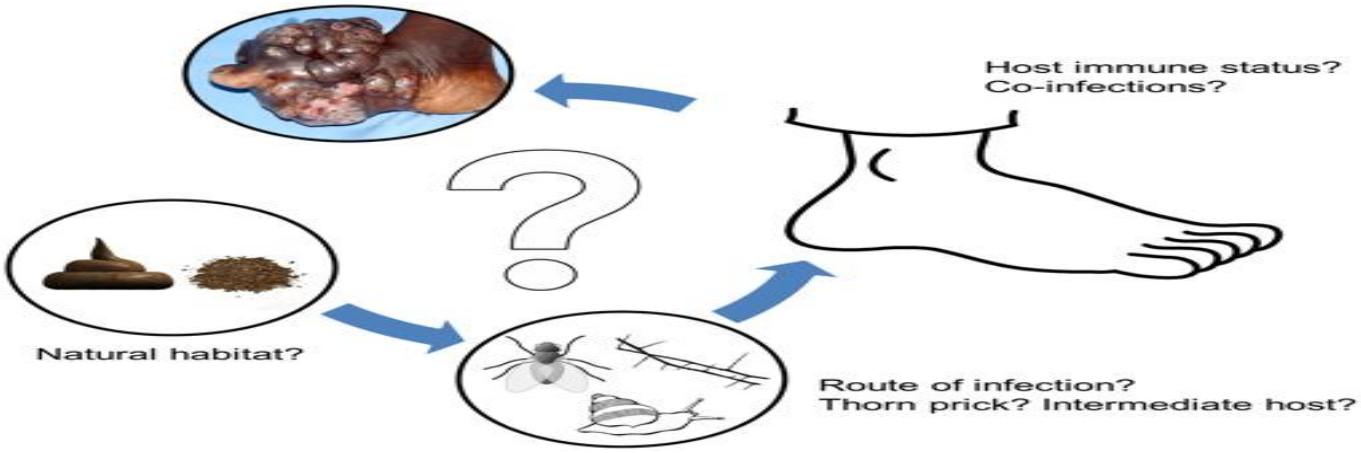


Figure 5:- The mode of transmissionofMaduramycosis
(Source:https://journals.plos.org/plosntds/article?id=10.1371/journal.pntd.0002667)

Mycetoma is endemic in specific regions forming the so-called "mycetoma belt," with notable prevalence in Sudan, Mexico, and India [17,18]. Recent literature estimates the prevalence to be around 1.8 to 3.49 cases per 100,000 people in Sudan and Mauritania, with potentially higher rates in Senegal due to its location in the mycetoma belt [10]. Although traditionally associated with the mycetoma belt, cases are now emerging in other countries, likely due to increased travel [19]. The disease is reported to affect individuals between 20-40 years of age, predominantly males with a ratio of 2.2:1 [18].

Mycetoma manifests as a chronic subcutaneous granulomatous disease, characterized by a triad of painless subcutaneous masses, multiple sinus tracts, and discharge [16–18]. The most common site of occurrence is the foot, followed by the hands [20]. It is classified based on the causative microorganisms into fungal (eumycetoma) or bacterial (actinomycetoma) origin [17,18]. The disease often arises after minor trauma to the foot, making it more prevalent in populations that walk barefoot, leading to the implantation of the causative agent in subcutaneous tissues [10,18]. The exact mode of transmission remains unclear, with some literature suggesting soil as a potential source as the organisms is cultured from the soil and for N.asteroides has been isolated from cow dung in India then minor prick for the skin and subcutaneous skin causse grains form[21] (Figure 5).

The diagnosis of mycetoma is based on clinical characteristics such as tumor-like swelling, sinus tracts, and the discharge of pus and/or grains. Imaging studies and cultures are crucial for identifying the causative organisms [11]. Bacterial causative agents include gram-positive filamentous actinomycetes, with Streptomycessomaliensis and Nocardiaspp. being common culprits. On the other hand, Mudrellamycetomatis, a gram-negative organism, is a frequent agent in eumycetoma[10].

Mycetoma is classified based on lesion size, with small lesions measuring less than 5 cm in diameter, moderate lesions ranging from 5-10 cm, and massive lesions exceeding 10 cm. A significant portion of patients (37%) may present with massive lesions at initial diagnosis [22].

No self-cure has been reported for mycetoma, necessitating accurate diagnosis through examination of grain morphology, which varies in size and color based on the causative organisms [17]. Early diagnosis and management are crucial in preventing disease progression, disability, and potential amputation [10]. Treatment strategies depend on whether the causative agent is bacterial or fungal. Actinomycetoma is typically treated with antibiotics, commonly employing a regimen of trimethoprim-sulfamethoxazole (TMP-SMX) for several months or high-dose penicillin G. Conversely, eumycetoma exhibits poor response to antifungal medications (ketoconazole or itraconazole), necessitating surgical intervention ranging from excision to amputation [10,21].

In conclusion, the intricate nature of mycetoma, as explored through this case and literature review, underscores the imperative for continual research and vigilance in addressing this challenging tropical disease. The historical roots of mycetoma, coupled with its dynamic epidemiology, emphasize the need for adaptive diagnostic and therapeutic approaches. As cases extend beyond traditional endemic regions, a global perspective and collaborative efforts become increasingly essential in managing and mitigating the impact of mycetoma. The multidisciplinary approach and advancements in understanding this complex disease will pave the way for more effective interventions and improved outcomes for affected individuals worldwide.

Summary points:

1. Mycetoma, a neglected tropical disease, remains challenging to diagnose and treat, often leading to misdiagnosis, especially in non-tropical regions.
2. This case report details a 52-year-old Sudanese male living in Saudi Arabia with a 20-year history of mycetoma, emphasizing the chronic nature and diagnostic complexities associated with the disease.
3. Accurate diagnosis relies on clinical characteristics, imaging, and cultures, with histopathology revealing suppurative granulomas and characteristic grains in subcutaneous tissue.
4. Treatment options vary based on the causative agent, with actinomycetoma responding to antibiotics and eumycetoma requiring surgical intervention due to limited antifungal efficacy.
5. The case underscores the need for increased awareness, early detection, and a multidisciplinary approach to effectively manage mycetoma and prevent disability or amputation.

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