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Case report

Comprehensive management of actinomycetoma in a young male: A case report from Somalia

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ABSTRACT

Mycetoma is a neglected tropical disease that predominantly affects individuals in low socioeconomic strata, primarily in tropical and subtropical regions. This case report describes a 20-year-old male student from Bahdo City, Somalia, who presented with a persistent cervical mass following a history of trauma. The patient exhibited vital signs within normal limits, and imaging studies, including ultrasound and computed tomography, revealed well-defined cystic masses. A Fine Needle Aspiration cytology confirmed the diagnosis of actinomycetoma by revealing sheets of neutrophils, multinucleated giant cells, and branching filamentous bacteria structures. The patient was treated with co-trimoxazole and amikacin, resulting in significant improvement after three months, highlighting the critical role of early diagnosis and appropriate medical management in enhancing patient outcomes. This report emphasizes the importance of thorough clinical evaluation and the use of cytological methods, particularly in resource-limited settings, to expedite the diagnosis and treatment of actinomycetoma.

Introduction

Mycetoma, a neglected tropical disease, is a significant public health concern primarily affecting individuals from low socioeconomic backgrounds in tropical and subtropical regions [1–3]. The condition is categorized into two main types: actinomycetoma, caused by various species of filamentous bacteria, and eumycetoma, resulting from fungal infections [3]. Both forms are characterized by a chronic, progressive infection leading to severe morbidity and stigma associated with visible deformities [3]. Actinomycetoma, in particular, is caused by pathogens belonging to the *Actinomyces* and *Nocardia* genera, which can elude prompt diagnosis and treatment in resource-limited settings [3,4].

Clinically, actinomycetoma presents as painless subcutaneous nodules or masses that can develop into abscesses with or without sinus tract formation, potentially involving extensive soft tissue [3,4]. The condition's insidious nature and overlapping features with other neoplastic and infectious processes, such as lymphomas and tuberculous lymphadenitis, and botryomycosis [5–10], further complicate diagnostic efforts. In regions endemic to mycetoma, the failure to recognize the disease can lead to delays in treatment, resulting in increased morbidity and negative impacts on quality of life [3].

This case report details the clinical journey of a 20-year-old male student from Bahdo City, Somalia, who presented with a persistent cervical mass following a history of trauma. Through this case, we aim to illustrate the challenges in diagnosing and managing actinomycetoma by including a comprehensive review of clinical presentations, imaging studies, cytological findings, and the therapeutic approach. The report serves as an important reminder of the necessity for timely and appropriate medical interventions, which are crucial for improving patient outcomes and addressing the burden of this neglected tropical disease.

Case presentation

A 20-year-old male student from Bahdo city, Galgaduud region, Galmudug state, Somalia, presented to the surgical department at Shaafi Hospital with persistent swelling on the right side of his neck. The patient reported that the condition began five months prior with a small, painless swelling localized to the right side of his neck (Fig. 1). He has a

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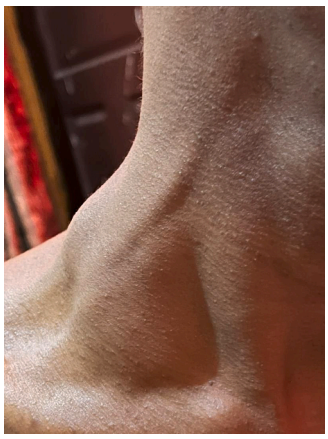

Fig. 1. The image illustrates the prominent cervical mass observed during the clinical examination.

notable history of trauma to the area two years ago.

Upon examination, the patient exhibited vital signs within normal limits: pulse rate of 78 beats per minute, respiratory rate of 18 breaths per minute, blood pressure of 123/70 mmHg, and a temperature of 36.7 °C. Systemic evaluations—including cardiovascular, central nervous system, endocrine, and gastrointestinal assessments—were all normal. Locally, the examination revealed a non-compressible, non-pulsatile cystic mass located on the right side of the neck, measuring 5 cm × 4 cm. The overlying skin appeared normal, with no hypo- or hyperpigmentation, and there were no observable sinuses.

Laboratory tests indicated normal complete blood count and liver function, with alkaline phosphatase recorded at 102.9 U/L. Renal function tests yielded a blood urea of 27 mg/dL and a serum creatinine level of 0.76 mg/dL. Random blood glucose measurement was 106.2 mg/dL. Screening for *Mycobacterium tuberculosis* and viral infections, including human immunodeficiency virus (HIV), Hepatitis B, and Hepatitis C, returned negative results.

Ultrasound of the neck revealed a well-defined ovoid-shaped cystic mass localized in the right posterior cervical region with mobile internal debris measuring 4.6 cm × 2.1 cm × 3.2 cm. Additionally, a second cystic mass with a thick wall was observed in the supraclavicular region, measuring 3.6 cm × 1.4 cm × 2.3 cm. A subsequent computed tomography (CT) scan highlighted a well-defined, fluid-like, partially lobulated collection with thin enhancing walls located in the right lower aspect of the neck, beneath the sternomastoid muscle and anterior to the scalene muscles, measuring approximately 5.1 cm × 2.6 cm × 4.4 cm. This mass was noted to be in close proximity to the right carotid sheath, with no evidence of calcifications, internal air densities, or intramuscular abscess formation.

Several differential diagnoses must be considered in this case. The primary candidate, is actinomycetoma, particularly linked to the *Actinomyces spp.,* which can manifest as a cervical mass following a history of trauma. Other considerations include tuberculous lymphadenitis, which would typically present with cervical lymphadenopathy, often accompanied by systemic symptoms such as fever and night sweats. Additionally, lymphomas usually display significant systemic effects and may involve lymph nodes. Other possibility is *Nocardia spp*.,as it can also involve the face and neck mass. Therefore we requested a Fine needle aspiration cytology from the swelling. Fine needle aspiration of the neck mass revealed sheets of neutrophils, multinucleated giant cells, and areas surrounding variably sized branching, radiating filamentous threads reminiscent of bacteria, consistent with actinomycetoma. Ziehl-Neelsen (ZN) stain was performed on the aspirated material, and the results were negative. This finding further consolidates the final diagnosis of actinomycetoma, as the absence of acid-fast bacilli helps differentiate it from other mycobacterial infections and *Nocardia spp*The patient was initiated on a treatment regimen that included cotrimoxazole at a dosage of 960 mg administered twice daily for five weeks, along with amikacin at a dosage of 15 mg/kg administered twice daily for three weeks. Additionally, the patient received folic acid supplementation at a dosage of 5 mg once daily for five weeks, administered in cycles. After three months of treatment, the patient showed significant improvement and was considered cured. Throughout the treatment course, the patient was closely monitored for any potential side effects associated with the medications. This diligent oversight ensured timely intervention in the event of adverse reactions, contributing to the overall positive outcome of the treatment.

Discussion

Actinomycetoma, while a relatively uncommon condition in Africa, poses significant diagnostic challenges due to its overlapping clinical presentations with other infectious, inflammatory, and neoplastic processes [5–10], particularly in areas where healthcare resources are limited, such as Somalia [1]. This case of a 20-year-old immunocompetent male presenting with a persistent cervical mass following trauma exemplifies the nuances involved in diagnosing actinomycetoma, especially when differential diagnoses must be meticulously considered to guide appropriate management.

In Somalia despite it considered one of the country that is located within the mycetoma belt, there is scarce literature regarding this infection and there are very few published cases of mycetoma in Somalia in the medical literature [11,12]. The condition is often underdiagnosed due to a lack of awareness and limited access to healthcare facilities. The typical diagnostic pathway involves clinical evaluation and history taking, but resources for advanced imaging and laboratory diagnosis are often lacking. Consequently, many cases are initially misclassified or treated for other conditions before the correct diagnosis of mycetoma is made. In rural settings, healthcare workers may rely heavily on clinical signs, with the characteristic appearance of the lesion often leading to a presumptive diagnosis. However, the integration of techniques such as culture, fine needle aspiration cytology (FNAC) and Histopathology has begun to help in accurately identifying the causative organisms and confirming the diagnosis [11,12].

The differential diagnosis for a cervical mass in a young male, particularly in the context of a previous trauma, is broad and necessitates a high index of suspicion for both infectious and neoplastic conditions [5–10]. In this case, the main considerations included: Actinomycetoma as the patient's had a history of trauma and the cystic nature of the mass, actinomycetoma, resulting from infection with filamentous bacteria such as *Actinomyces spp.*, was a prime candidate. Its typical presentation includes painless masses, often with a chronic course, aligning closely with this patient's sign and symptoms. Another differential diagnosis was tuberculous lymphadenitis since this condition is common in endemic regions and often presents with cervical lymphadenopathy. It is characterized by systemic symptoms such as fever and night sweats, which were absent in this case. The chronicity of symptoms and history of trauma could have complicated the diagnosis, yet the cytological findings and imaging studies favored actinomycetoma. Moreover, other differential diagnosis can include Lymphoma as the significant mass effect along with cervical swelling raises concern for lymphoproliferative disorders including lymphomas [13]. However, lymphomas typically present with systemic manifestations and may lead to significant changes in blood counts, which were not observed in this case. Furthermore, other Infectious processes such as abscesses from other bacterial infections or deep fungal infections could also present similarly but often come with acute systemic signs or localized pain. The absence of such features moves actinomycetoma to the forefront.

The diagnostic imaging studies, including ultrasound and computed tomography (CT), played a crucial role in identifying the characteristics of the mass and its fluid-containing structure, helping to narrow down

the differential diagnoses. The definitive diagnosis was ultimately achieved through fine needle aspiration cytology (FNAC), revealing distinct cytological features characteristic of actinomycetoma [14,15].

Our final diagnosis was established through a comprehensive integration of clinical history, imaging findings, and cytological analysis, which proved crucial in confirming the presence of actinomycetoma. Fine needle aspiration cytology (FNAC) is a minimally invasive procedure that facilitates rapid cytological examination, making it particularly valuable in low-resource settings. This technique not only helps to differentiate actinomycetoma from eumycetoma effectively but also allows for the timely initiation of appropriate treatment. By expediting the diagnostic process in environments where access to histopathological services is limited, FNAC plays a vital role in enhancing patient care and ensuring that individuals receive the necessary treatment without unnecessary delays [16]. The presence of neutrophils, multinucleated giant cells, and characteristic radiating filamentous structures on cytology strongly suggested an actinomycetoma diagnosis [14,15].

The therapeutic approach taken in this case reflects an understanding of the disease's etiology and the pharmacological management available. The combination of co-trimoxazole and amikacin is well-supported in the literature for treating actinomycetoma caused by *Actinomyces* species, highlighting the importance of selecting an empiric treatment regimen tailored to the likely pathogen while also considering the potential for resistance patterns specific to the region [17].

In resource-limited countries, such as Somalia, health policy initiatives must prioritize the development and enhancement of diagnostic and treatment capacities for conditions like actinomycetoma. Building healthcare infrastructure is essential to improve access to essential diagnostic tools and imaging technologies that can aid in the timely identification of such complex conditions. Training healthcare professionals in the recognition and management of actinomycetoma and other mycetomas, along with fostering partnerships between local health authorities and international organizations, can facilitate knowledge transfer and resource sharing. Furthermore, implementing community health education programs can raise awareness of mycetoma symptoms and encourage early medical consultation. Strengthening laboratory capabilities, including the provision of training in cytological techniques such as fine needle aspiration, can enhance diagnostic accuracy. Additionally, promoting research into local pathogen profiles and resistance patterns can guide targeted treatment plans, ultimately leading to better patient outcomes. A comprehensive approach that encompasses education, training, and resource allocation is vital for improving the management of actinomycetoma, ultimately addressing health disparities and enhancing the overall efficacy of healthcare systems in endemic regions.

In conclusion, this case underscores the complexities involved in diagnosing actinomycetoma, particularly in resource-restrained settings. It illustrates the vital importance of comprehensive clinical evaluation, thorough consideration of differential diagnoses, and the utility of cytological techniques in achieving a definitive diagnosis. Prompt recognition and appropriate management of actinomycetoma are crucial to ameliorating patient outcomes, particularly in regions where the disease burden remains significant. Further research and education regarding the epidemiology and clinical presentation of mycetoma may improve diagnostic accuracy and enhance treatment strategies in endemic areas.

CRediT authorship contribution statement

Emmanuel Siddig: Writing – review & editing, Writing – original draft, Visualization, Validation, Supervision, Resources, Project administration, Methodology, Investigation, Formal analysis, Data curation, Conceptualization. **Ayman Ahmed:** Writing – review & editing, Writing – original draft, Visualization, Validation, Supervision, Methodology, Investigation, Formal analysis, Data curation, Conceptualization. **Claude Muvunyi:** Writing – review & editing, Writing – original draft, Visualization, Validation, Supervision, Methodology. **Mohamed Mahamud:** Writing – review & editing, Writing – original draft, Visualization, Validation, Methodology, Formal analysis, Data curation, Conceptualization.

Authors' contributions

MAM, AA, and EES conceived and designed the study; AA, MAM, and EES analyzed the data; AA, MAM, CMM, and EES wrote the manuscript. AA, MAM, CMM, and EES revised the manuscript. All authors read and approved the final manuscript.

Authors' statement

Mohamed Adam Mahamud: Conceived, design, analyzed, wrote the first draft, revised and approved the final draft. **Claude M. Muvunyi**: Analyzed, wrote the first draft, revised and approved the final draft. **Ayman Ahmed:** Conceived, design, analyzed, wrote the first draft, revised and approved the final draft. **Emmanuel Edwar Siddig**: Conceived, design, analyzed, wrote the first draft, revised and approved the final draft.

Consent

Written informed consent was obtained from the patients for publication of this case report and any accompanying images.

Ethical approval

Written, informed consent to publish history, findings, and images for educational purposes were obtained from the patients.

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Competing interests

The authors declare that they have no competing interests.

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Availability of Data and Materials

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

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