



Mycetoma and Squamous Cell Carcinoma in Ethiopia: Case Report

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Abstract

Background: Mycetoma is a chronic suppurative infectious disease of the subcutaneous tissue affecting skin, muscle and bone, with high morbidity. It is characterized by nodules and sinus tracts that discharge watery fluid or pus containing grains, although, it is a common infection in the “mycetoma belt”, which includes Ethiopia, there are no published cases mycetoma in Ethiopia despite cases having been reported in Ethiopians living in Israel. Squamous cell carcinoma is a common skin cancer, at times arising from chronic wounds, but rarely reported to arise in tissue affected by deep fungal infection.

Case Report: A 43 year old male patient presented with bilateral lesions on the lower extremities. The lesions began 11 years earlier as a solitary, painless, firm sub-cutaneous lesion, draining hard pea-sized matter with pus. The patient was treated for mycetoma for 2 years with no marked improvement, until a biopsy finally confirmed squamous cells carcinoma.

Conclusion: Despite, mycetoma being present in Ethiopia, it is not reported. This could be due to under-diagnosed and poor /unsupported reporting formats of the health management information system. Squamous cell carcinoma should be suspect if there is no/poor response to treatment.

Keywords: Mycetoma; Squamous cell carcinoma; Ethiopia

Background

Mycetoma is a Neglected Tropical Disease (NTD) which causes chronic disfiguring and stigmatizing lesions primarily in poor populations of tropical and subtropical regions. It is a chronic suppurative infection of the skin and subcutaneous tissues that affects the lower limbs in more than 80% of cases [1]. The etiologic agents of mycetoma are aerobic and anaerobic bacteria (actinomycetoma) and fungi (eumycetoma). Mycetoma is characterized by a triad of tumor-like swelling, multiple sinus formation, and a grain-containing discharge [2-4]. Mycetoma was added to the World Health Organization list of NTDs in 2016 [5], but getting basic data on its epidemiology is still a big challenge. Sudan is the only country to have a national mycetoma control program worldwide [6,7], while for many other countries no strategies can be devised because of a lack of data. Part of the problem is that mycetoma is not a notifiable disease, and therefore no surveillance and routine data registration systems exist. Even though Ethiopia is in the so-called “mycetoma belt”), and patients of Ethiopian origin have been reported in Israeli and Sudanese case series [1,7], mycetoma is rarely reported from Ethiopia [7,8]. The Ethiopian NTD program desperately needs data to formulate a mycetoma control strategy. We would like to present a patient with poorly responsive mycetoma who was later diagnosed with a squamous cell carcinoma at Boru Meda General Hospital, Amhara region, Ethiopia.

Case Presentation

A 43 years old male patient self-presented at the dermatology out-patient department at Boru Meda General Hospital since three years before. He is a farmer living in a rural area called Worebabo where nearly 53 km away from Boru Meda Hospital, cultivating fruits and “Chat” in the vicinity. He presented with swollen bilateral lower legs covered with dry and crusted plaques, with severe scarring and pus draining areas. He reports that it had started, seven years before as a solitary, painless, firm nodule, which started draining pus and hard pea-sized matter. He had difficulty recalling the color of the lesion (pus, bloody tinged, black etc). Then through time multiple small lesions appeared with increasing pussy discharge, with some lesions coalescing into each other. There was swelling

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Received Date: 30 Jun 2021

Accepted Date: 22 Jul 2021

Published Date: 28 Jul 2021

Citation:

Zewdu FT, Lambert SM. Mycetoma and Squamous Cell Carcinoma in Ethiopia: Case Report. *Ann Clin Case Rep.* 2021; 6: 1958.

ISSN: 2474-1655

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Figure 1: Multiple globular granuloma with epithelial cells and inflammatory mixed squamous cells.

in both lower limbs and some limitation involvement. When the lesions become aggressive and multiple, the patient had gone to the local health institution and received an unspecified oral medication, with no improvement. He then visited multiple-local traditional healers where he received unknown topical mixtures to applied and liquor to be drunken, with no improvement in the improvement of the lesions, discharge, and size. But as the time gone and the lesion progresses to the shin, it become extensive and the patient felt difficulty of walking due to pain. He presented at Boru Meda in 2018, where he started to be diagnosed using Fine Needle Aspiration (FNA) with Potassium Hydroxide (KOH) examination, X-ray and clinical evaluation. Then the result was reviled inconclusive cytological examination and recommends biopsy (FNAC), no fungal elements seen (KOH), chronic osteomyelitis (X-ray) around the ankle area. He was diagnosed as having mycetoma. He started on the ketoconazole (200 mg) and cotrimoxazole (960 mg po BID) after doing baseline investigations. On review after nine months of treatment he was found to have no improvement and maybe possibly some worsening in his condition. Treatment was changed from ketoconazole to itraconazole (200 mg daily) with co-trimoxazole for the next 9 months (3 cycles). At 18 months post-initiation of unsuccessful treatment, the patient wanted for referral to get a second opinion at another hospital, where he was diagnosed with mycetoma and referred back to Boru Meda Hospital. With the poor response to anti-fungal and anti-bacterial treatment, a biopsy and FNA was requested. The pathologist reported "Multiple globular granuloma with epithelial cells and inflammatory mixed squamous cells" (Figure 1) multiple grains and squamous cell carcinoma. The patient has now been transferred for better treatment like chemotherapy and/or radiotherapy at the national referral centre to ALERT.

Discussion

Mycetoma are chronic granulomatous infections of the dermal and subcutaneous tissues caused by filamentous aerobic and anaerobic bacteria (actinomycetoma), or true fungi (eumycetoma), and which can be diagnosed using a combination of different diagnostic methods [9]. But in poor resource countries like Ethiopia, mycetoma can be diagnosed clinically with the presence of a symptomatic triad: A subcutaneous mass (tumefaction), draining sinuses and the presence of grain containing discharge. The most specific diagnostic tool is the examination of the grains discharged from sinuses. Both spontaneous drainage and manually expressed material from sinus should be carefully examined macro and microscopically to visualize

the grains. The samples can be obtained from any open sinus or by deep surgical biopsy. Mycological and bacteriological examinations are very important for the correct diagnosis. Over 30 species have been identified as causative agents of mycetoma, but the grains of many of these agents have overlapping morphological characteristics, therefore, cultures are needed to properly identify the causative agent. Mycetoma is mainly found in young adults, with cases having been report in children and elderly. In most countries, males are more often infected than females. One of the risk factor for mycetoma is thorn injuries due to bare foot and being farmers whose daily activities are associated with cultivating vegetable. In countries like Ethiopia, 85% of the population is a farmer who used bare foot for their daily practices [10]. Treatment of mycetoma depends on a correct diagnosis. It is important to define the fungal or bacterial etiology because the treatments are different. Mycetoma cannot be cured without active treatment. It requires antibiotics (for actinomycetoma) or oral anti-fungals (for eumycetomas) for weeks, months or years. Damage to subcutaneous tissues (muscle, bones, joints or tendons) often persists, so local surgery (including amputation) and physiotherapy may be required [11]. Moreover, mycetoma with the cure rate which takes at least 6 months even using a combination of anti-fungal and systemic antibiotics. (Antibiotics and antifungals effective in the treatment of mycetoma are included in the WHO model list of essential medicines, but the long duration of treatment [8] and monitoring means they may be prohibitively expensive). Even though mycetoma is present, diagnosed, treated and had a regular follow up in the department of dermatovenereology unit in Ethiopia like ALERT, and other respective regional dermatology clinics, they are not reported to the respective NTD departments due to inconclusiveness of deep fungal infections in the Health Management Information System (HMIS). With few dermatologists in Ethiopia, deep fungal infections are under diagnosed and un-reported to the Ministry of Health and/or the World Health Organization, even though it is listed as a NTD. Many mis-diagnosed mycetoma patient develop sever chronic wounds which put them at risk of carcinomas, such as squamous cell carcinoma. Mycetoma is quite common in the lower extremities where wound healing is slow. With the long delay in diagnosis or the unavailability of treatment, mycetoma infections may develop into a chronic wound with osteomyelitis. Thus, chronic ulcer is one of the sources of squamous cell carcinoma. Any chronic wound which may be of fungal, bacterial or viral etiology may be treated with the respective medication and routine nursing care. A non-healing ulcer should warrant further investigation to determine the presence of an underlying or super-imposed condition is unable to have marked improvement for long standing period, one should suspect the presence and absence of overlying disease condition like cancer, superimposed undiagnosed disease condition and other. Therefore, presence and absence of clinical improvement could be one of the sign of cancer imposed mycetoma.

Conclusion

Mycetoma is present in Ethiopia but due to many reasons it is under-diagnosed and not reported and when diagnosed it is often diagnosed late making treatment complicated and increasing the risk of disability, loss of income and social stigma. In all chronic ulcers and wounds not responding to treatment, malignant changes should be suspected. This case emphasizes the importance of biopsy and cultures as fungal infection and squamous cell carcinoma is often have similar, non-specific clinical appearances.

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