CASE REPORT

NON-ULCERATIVE SOFT TISSUE MYCOSIS OF LONG DURATION MIMICKING SOFT TISSUE SARCOMA– A CASE REPORT

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Abstract. Soft tissue mycosis usually presents with a triad of tumefaction, suppuration and ulceration. We report an unusual case of soft tissue mycosis in a 42 year old male teacher who presented with painless swelling over the anterolateral aspect of the right shin for 4 years duration.

INTRODUCTION

Mycetoma is a chronic granulomatous inflammatory disease that usually involves the subcutaneous tissue after a traumatic inoculation of the causative organism (Gonzalez-Ochoa, 1975). It may be caused by true fungi (eumycetes) or higher bacteria (actinomycetes). Thus, it is classified into Eumycetoma or Actinomycetoma (Gonzalez-Ochoa, 1975; Fahal, 2004). This condition has been known since ancient times. It was mentioned in a Sanskrit writing "*Atharva Veda"* from India as "pada valmikam", which means ant-hill foot (Kwon-Chung and Bennet, 1992). French missionaries first recorded it in Pondhicherry India in the 18th century (Kwon-Chung and Bennet, 1992). The fungal origin of the disease was discovered by Van Dyke Carter in 1860, by isolation of the organism. He coined the term 'Mycetoma' (Fahal, 2004).

There are not less than 18 different organisms believed to cause the condition, reported from different regions of the world. These organisms are saprophytic in nature residing on thorns or in soil, accidentally introduced into the body by penetrating injuries or soil contamination of wounds (Fahal, 2004).

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Upon infection, patients usually present with a triad of tumefaction, suppuration and ulceration. We report an interesting case presenting only with swelling of long duration, without the other two signs, leading to an incorrect diagnosis of a soft tissue tumor.

CASE PRESENTATION

A 42 year old male secondary school teacher, presented with two painless swellings over the anterolateral aspect of his right shin for 4 years. They were insidious in onset without affecting the limb function. There was neither history of trauma nor any other constitutional symptoms. On examination, two swellings were seen over the anterolateral aspect of the right shin, firm, non-tender and fixed to the underlying fascia (Fig 1). The overlying skin was normal. No regional lymphadenopathy was detected.

His routine blood counts, electrolytes, blood sugar and chest X ray were normal.

A plain radiograph revealed soft tissue swellings with no involvement of the underlying periosteum or bone. Small, linear flakes of calcification were noted in the centers of these swellings (Fig 2). MRI noted three discreet vertically arranged swellings over the right leg in the anterolateral compartment, arising from the fascia without infiltration into deeper structures. They were homogeneous and showed a differ-

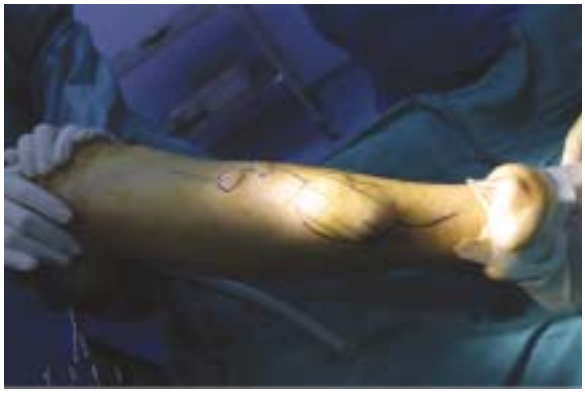

Fig 1–A preoperative photograph of the lesion showing three discrete lobulated swellings over the anterolateral aspect of the right shin.

Fig 2–Plain radiograph of the lesion. Note the soft tissue swelling and the linear flecks of calcifications (arrow).

Fig 3–Gross specimen showing two nodules within the soft tissue strip composed of well defined capsulated lesions with central cavitation containing yellowish friable material.

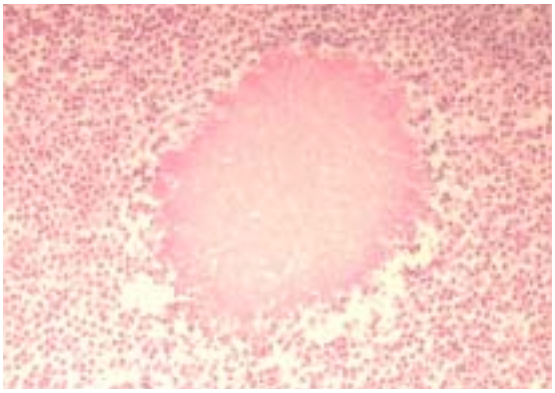

Fig 4–Microscopically, many colonies of organisms having a tangle of real hyphae are seen, surrounded by neutrophils and eosinophils in "Splendore-Hoeppli" phenomenon.

ent signal intensity from the surrounding fat. Associated calcifications were seen.

Clinical diagnosis

At that time, given the age, clinical presentation and imaging studies, the clinical impression was either one of a slow growing soft tissue tumor, possibly synovial sarcoma, or a well-differentiated malignant fibrous histiocytoma. The patient then underwent tumor excision.

Gross pathology examination revealed two nodular lesions, attached to the strip of removed fascia. On cut section there were central cavitations surrounded by friable soft yellowish tissue with a well circumscribed capsule (Fig 3).

Microscopic examination showed a suppurative granulomatous lesion with the presence of small non-pigmented grains in the center. These grains were made of numerous fungal hyphae (Fig 4) surrounded by neutrophils and eosinophils.

The final diagnosis was deep (soft tissue) mycosis, consistent with eumycetoma.

Unfortunately as all the tissue had already been fixed in formalin, culture to identify the organism was not possible. Formalin fixed tissue sent for PCR examination for fungal organisms was inconclusive, since DNA could not be extracted from the tissue due to prolonged fixation. The patient was treated with a short course

of systemic antifungal, and he remained well with no evidence of disease 23 months post surgery.

DISCUSSION

Mycetoma is prevalent in the tropics and subtropics (Kwon-Chung and Bennet, 1992). The highest incidence is in Sudan, followed by Mexico (Kwon-Chung and Bennet, 1992). It is not known to be endemic in Malaysia.

It is a chronic suppurative disease of the soft tissue and bone. The most common site is the foot (Lynch, 1964). Extra-petal mycetoma makes up approximately 30 to 40% of cases (Lynch, 1964). Initially, the lesion presents as a small subcutaneous swelling of a few millimeters in diameter, is painless and rubbery (Kwon-Chung and Bennet, 1992). The overlying skin is usually normal in the early stages. The infection then spreads along the subcutaneous tissue planes forming small abscesses connected by sinus tracts. These sinuses frequently connect to the skin surface and appear as soft vesiculated areas (Kwon-Chung and Bennet, 1992) discharging sero-sanguinous fluid. The progression from soft tissue swelling to draining sinuses occurs in almost all cases within a one year period (Lynch, 1964). The patient usually presents with advanced disease when treatment is difficult.

Our patient was unusual in that first, he did not have a profession which put him at risk for such an infection, and second, the lesion remain as localized swelling for a few years. Pre-operative diagnosis of mycetoma as opposed to a soft tissue tumor, as in this case, may be difficult. Radiological investigation is useful in late stages of mycetoma to demonstrate the presence and asses the extent of bony or organ involvement, not so much as a diagnostic tool (Weedon, 1992). In early disease, the appearance may not be discriminatory from other soft tissue lesions (Weedon, 1992).

The management of a mycetoma depends on the etiological agent and the extent of disease. Amputation of the affected part or debilitating surgery is often the treatment of choice for advanced disease (Fahal, 2004).

Medical treatment, such as the administration of ketoconazole 200 mg twice daily may not be effective against certain organisms (Fahal, 2004). Other agents, such as procaine penicillin, griseofulvin, dapsone, amikacin, streptomycin, rifampicin, co-trimoxazole and even Fansidar have been used with some success (Maghoub, 1994).

In this case, since the disease was confined to the soft tissue, surgical excision was indicated (Fahal, 2004). Since the capsule was not ruptured during excision and a rim of healthy tissue was removed with the lesion, this treatment can be curative for the patient. However, follow-up is still desirable, in case of the presence of any occult grains left behind which could lead to a future recurrence.

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