ACTINOMYCETOMA OF HAND AND FOOT

Lt Col MAN MOHAN HARJAI*,
Maj AK SHARMA⁺, Lt Col JS PANAYCH[#],
Wg Cdr PK MENON^{**}, Col BM NAGPAL^{VSM⁺⁺},
Col Y SINGH^{VSM^{##}}

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Introduction

Mycetoma refers to the chronic suppurative pathological process in which exogenous etiological agents generate pus and sulphur like granules. These agents belong to two groups : true fungi and the actinomycetes. Eumycetoma (caused by fungi) and actinomycetoma (caused by actinomycetes) must be distinguished as their treatment is different. These causative agents are introduced into the skin by minor trauma. Most cases of mycetoma occur in tropical regions such as Asia, Africa and Central and South America but this disease is endemic in India. *Nocardia brasiliensis* is the most common isolate found in India [1]. The pathologic process is characterized by tumefaction, subcutaneous nodules and in most cases discharging sinuses that drain exudate containing granules. It gradually invades the tissues and bones causing a functional disability. Bone involvement depends on the duration of the disease and the causative agent. Here we present two cases of actinomycetoma, one affecting the hand and the other the foot. The pathogenesis, radiological features and the therapeutic management of this entity are reviewed.

Case Report

A 32-year old individual, tailor by profession reported with a gradually increasing painless swelling of the left hand associated with progressive development of multiple discharging sinuses, of 8 years duration. He did not give any positive history of direct prick or any injury. Local examination revealed a diffuse swelling of the left hand involving mainly the palm and the dorsum, sparing the fingers and thumb. The feel of the swelling was woody and there were multiple discharging sinuses present all over the dorsum and palmar aspect of the hand (Fig 1). The granules contained in the discharge were yellowish in colour. 1-2 mm in size, multiple and discharging intermittently. The surrounding skin was thickened, unhealthy and hyperpigmented. Systemic examination was normal. There was no evidence of any distant spread. Gram stain of the granules showed gram positive branching bacillary filaments (1 μ in diameter) having a distinct sunray appearance,

diagnostic of actinomycetoma. Repeated culture both under aerobic and anaerobic conditons did not grow any organism.(the patient gave history of prior antibiotic treatment). X-ray of the left hand showed a patchy resorption of all the metacarpal bones with lucent filling defects and thickened cortex with partial destruction of the carpal bones (Fig 2). There was no involvement of the small joints. He was managed with high dosage of penicillin along with dapsone (DDS) and rifampicin to which he responded well. The chemotherapy was then switched over to dapsone (DDS) and trimethoprim-sulfamethoxazole combination at the time of discharge, for the patient's convenience. He is under close follow up.

Case Report 2

A 70-year old woman from rural back ground and a farmer by occupation, presented with a progressive painless swelling of the left foot for the last 20 years, with multiple sinuses discharging yellow granules. The patient stated that the swelling started after a thorn prick to the foot, sustained while working in the fields. Local examination revealed a diffuse swelling over lateral half of the dorsal aspect of the left foot. The swelling was nodular, indurated, nontender and having multiple sinuses discharging yellowish granules. X-ray of the left foot showed a soft tissue shadow over the lateral three metatarsals with no bony involvement. Aspiration cytology gave the impression of lesion being an actinomycetoma.

Fig. 1 : Palmar view of the left hand showing massive soft tissue swelling, subcutaneous nodules and multiple discharging sinuses.

*Reader and Pediatric Surgeon, ⁺Trainee in Surgery, [#]Reader in Pathology, ^{**}Reader in Microbiology, ⁺⁺Associate Professor Surgery, ^{##}Professor and HOD Surgery, Armed Forces Medical College, Pune - 40.

Fig. 2 : X-ray photograph of the left hand showing patchy resorption of all the metacarpal bones with lucent filling defect and thickened cortex with partial destruction of the carpal bones. Small joints of hand are normal

Local excision of the granulomatous tissue was carried out extending down upto the bone. Histopathological examination of the excised tissue revealed a background of vacularised fibrous stroma with multiple pus filled abscesses, containing varying sized matted colonies of intertwined branching, haematoxyphilic filamentous bacteria forming a granule or grain. Splendore-Hoepli phenomenon was seen surrounding the colonies of the bacteria (Fig 3). Special stains revealed that the organisms were gram positive and weakly acid fast. Presently, the patient is on dapsone (DDS) and doxycycline and responding well. She is under regular follow up.

Discussion

The term actinomycetoma refers to a slowly progressive subcutaneous infection caused by actinomycetes of the genera, *Nocardia*, *Nocardiosis*, *Streptomyces* and *Actinomadura*. Eumycetoma on the other hand is caused by the true fungi (with thick septate hyphae) of many different genera such as *Petridium boydii* (*Allescheria boydii*), *Madurella grisea* and *Madurella mycetomatis* (*mycetomis*). The discharged granules are colourless in the case of former, and black in the case of the latter two. Sixty percent of the mycetomas are caused by actinomycetes whereas the remaining are eumycetomas. The infection usually remains localised and involves the cutaneous tissue, subcutaneous tissue, fascia and even bone. The organisms are inoculated into the hands or feet either directly following minor trauma, or from infected vegetation. It predominates among farm workers in tropical, subtropical and adjacent zones. The disease is characterised by painless swelling, woody induration, nodule formation, multiple sinuses discharging granules or grains of the causal agent and serious bone involvement. The bone changes include sclerosis, erosions, periosteal thickening and osteoporosis [2]. Sometimes the ultrasonography delineates the extent

Fig. 3 : High power view : edge of colony showing tangled mass of filamentous haematoxyphilic bacteria. Splendore-Hoepli phenomenon and more peripherally placed pus cells can be appreciated at this magnification.

of mycetoma more accurately than clinical examination. In eumycetoma lesions, the grains produce numerous, sharp hyper-reflective echoes and there are single or multiple thick-walled cavities with no acoustic enhancement. In actinomycetoma, the findings are similar but the hyper-reflective echoes are fine closely aggregated and commonly settle at the bottom of the cavities [3]. The colour of granules, their size, consistency and the histopathology contribute to their identification. In actinomycetoma the colour of grains is usually yellow, but in literature the red grains mycetoma caused by *Actinomadura pelletieri* from Rajasthan [4] and pale grain mycetoma from Madras [5] are also reported. In endemic regions this entity should be differentiated from tubercular infection, chronic osteitis and botryomycosis (in which pyogenic bacteria, usually staphylococcus aureus, form granules similar to those seen in mycetoma). The diagnosis of the disease is confirmed by the microscopic identification and by isolating and identifying the infective agent. It can be fairly accurately diagnosed by the fine needle aspiration cytology [6]. All types of actinomycetoma infections should be treated with early limited surgical debridement. The excised tissue should be sent for gross, microscopic, and histopathologic evaluation as well as for culture. In addition to the surgical management, these patients should also be managed with a prolonged course of adjunctive chemotherapy. Streptomycin with dapsone or trimethoprim-sulfamethoxazole is the treatment of choice for actinomycetoma. Prolonged doxycycline chemotherapy, is also successful in some cases [7]. Amikacin is used for severe and resistant cases [8]. Amphotericin B, Ketoconazole, Itraconazole : these drugs combined with surgery are

the best possible therapeutic options for true fungi eumycetomas [9,10].

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