

STREPTOMYCES SOMALIENSIS CAUSING MYCETOMAS IN SOUTH INDIA

V. V. TARALAKSHMI* V. V. PANKAJALAKSHMI † AND S. ARUMUGAM ‡

Summary

Biopsy specimens from 102 patients with mycetoma were examined histologically; *Streptomyces somaliensis* was found in 5 cases. The clinical features, radiology and histology of actinomycotic mycetoma due to *S. Somaliensis* are described.

The geographic distribution of the organism, its incidence and prevalence in India and the importance of histological examination in the diagnosis of the infection are discussed.

Introduction

Mycetomas caused by *Streptomyces somaliensis* are common in Africa, but reports are rare from other countries. The first published report of this disease caused by *S. Somaliensis* in India was by Murti and Padmavati¹ 1963, who isolated the organism in 3 cases of mixed infection from Guntur where they had encountered more than one species of organisms from the same case and in all the three cases, *S. Somaliensis* was isolated either with *Actinomadura madurae*, *Nocardia asteroides* or *Rubromadurella longeroni*. Klokke et al² reported 3 cases from Vellore and 2 cases each, from Madras and Madurai, based on histopathology. Desai et al³ from Bombay reported two cases based on histology and from one, the organism had been isolated in culture. Reddy et al⁴ from Vishakhapatnam and Grueber and Kumar⁵ from Ludhiana reported one and four cases respectively from the histologic appearance of the granules in sections.

Besides, Chugh and Arora⁶ reported 3 cases from Rohtak, all based on histology.

The histological appearance of the granules in mycetoma tissue sections is characteristic for most of the species of organisms. Mere histopathological examination of the biopsy material not only establishes the diagnosis of the disease but also allows specific identification of the causal agent in 94-95% of mycetomas^{2,7}. However, it has its limitations. In white-grain mycetoma no species differentiation can be made between *Allescheria boydii* and other white-grain producing species such as *Acremonium*, *Fusarium* etc., or between small grain mycetoma caused by *Nocardia asteroides*, *Nocardia brasiliensis* or *N. Caviae*. Similarly in black-grain mycetoma, it will not always be possible to identify the causal agent by histological examination alone⁸. But the distinctive appearance of the granule of *S. somaliensis* allows a specific etiological diagnosis to be made from histological sections⁸.

* Associate Professor of Pathology,

† Associate Professor of Microbiology,

‡ Professor of Pathology,

Madras Medical College, Madras-3

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Based on the histological description of the granules of *S. somaliensis* in

tissue sections^{9,10}, we have diagnosed 5 cases in our study from Madras. Since only few reports were available on *S. somaliensis* mycetomas from India, we thought it worthwhile to report these.

Materials and Methods

The pathological material used in this investigation was obtained from the collection of the Department of Pathology, Madras Medical College, Madras, for the years 1964-1976. Paraffin blocks were resectioned, stained with haematoxylin-eosin and reclassified according to the morphologic appearance of the granules. All sections of actinomycotic mycetomas were further stained by Brown and Brenn's modification of Gram and Kinyoun's acid-fast methods.

Observations

Of 102 cases of mycetoma examined histologically, 5 were diagnosed to be due to *S. somaliensis*, based upon the morphological appearance of the granules in tissue sections. The relevant details of those 5 cases are presented in Table I.

Histopathological study

Biopsy material from all 5 cases showed the presence of granules conforming to the description of *S. somaliensis*. The granules were of variable size, round or oval with smooth borders amorphous centre, staining pale pink with haematoxylin and eosin and without clubs. (Fig. 1 and 2). Often the granules were fractured by the microtome knife because of the hard cement material. The individual filaments in the cement were faintly basophilic and visible when stained by Gram's stain (Fig. 3). These were not acid-fast by Kinyoun's acid-fast method. The cellular reaction was also characteristic for this actinomycete in that the granules were surrounded by a narrow zone of polymorphonuclear

TABLE 1
Details of 5 patients with *S. somaliensis* mycetomas

Case No.	Sex/Age	Clinical syndrome	X-ray	Histo-pathology
1	M/25	Purulent discharge from umbilicus; Ulcer in the right inguinal region with sero-sanguinous discharge; 15 days. Had conservative treatment for appendicitis 6 months earlier. Bilateral inguinal lymphadenopathy present.	Abdomen Normal Chest Normal	<i>S. somaliensis</i> from 2 biopsy specimens. Ulcer; lymphnode
2	M/12	Swelling with sinuses of the right foot; serous discharge. History of injury with a stone an year earlier.	Right foot-No bony lesion	<i>S. somaliensis</i>
3	M/14	Swelling with multiple sinuses left foot; purulent discharge 6 months. No injury	Left foot: No bony lesion	<i>S. somaliensis</i>
4	M/40	Swelling right ankle and foot with multiple sinuses; purulent discharge 2 years following thorn prick.	Right foot: only soft tissue swelling.	<i>S. somaliensis</i>
5	M/50	Swelling with multiple sinuses left foot, serous discharge; History of injury with a wooden log 2 years earlier.	Left foot: No bony lesion.	<i>S. somaliensis</i>

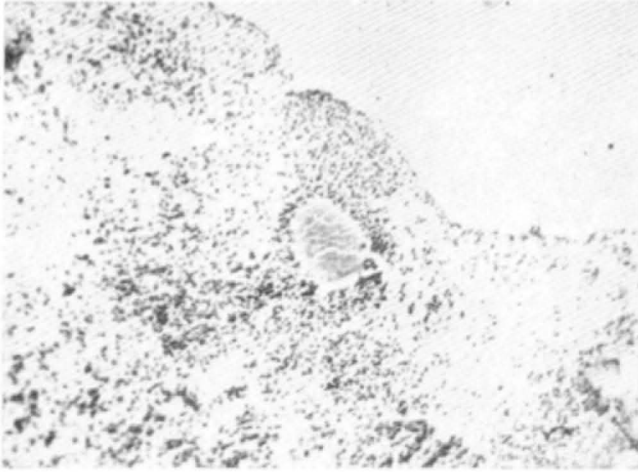


Fig. 1

Biopsy specimen of lesion showing the weakly stained, amorphous granule of *S. somaliensis*. haematoxylin and eosin x 100.

leucocytes and then by more chronic cells including plasma cells and rare giant cells.

Therapy

With streptomycin, crystalline penicillin and isoniazid, case 1 showed marked improvement in 4 weeks' time. The discharge from the umbilicus ceased and the inguinal ulcer healed. In the other 4 cases, excision of the lesion with skin grafting was done. They were treated with a combination of drugs which included crystalline penicillin, potassium iodide and dap-

son. Since there was no bony involvement, the lesions healed well and all patients were discharged with the advice to continue the therapy.

Discussion

The species causing mycetoma change from country to country. *S. somaliensis* as a causative agent of mycetoma has mainly been reported from the African countries—Sudan, Somalia, Senegal, Ethiopia, Nigeria, Republic of South Africa and Tanzania and the Latin American countries—Brazil and Mexico 7, 11, 12. In Sudan, it accounts

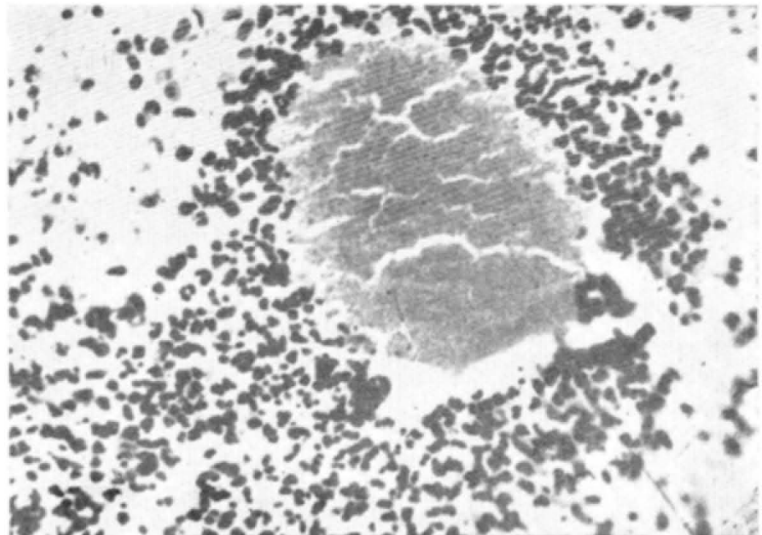
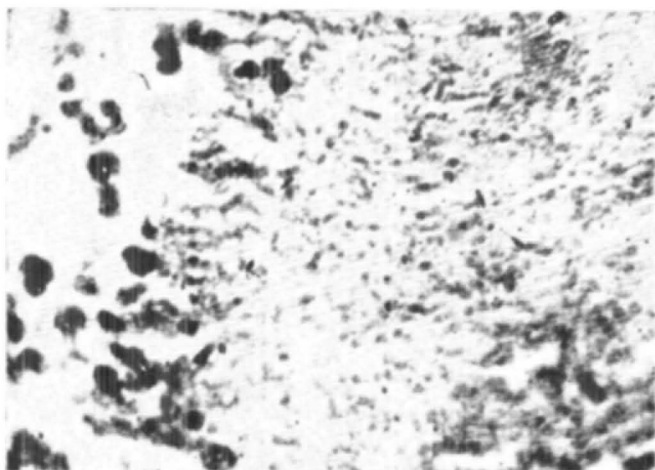


Fig. 2

Same Section x 450

Fig. 3

Section showing the delicate, gram positive, branched non fragmenting filaments. Gram x 100



for 20% of mycetoma cases¹³. Symmers⁵ diagnosed one case of *S. somaliensis* mycetoma in England in a student from Sudan in 1966.

In India 3 to 10% of mycetoma cases are due to *S. somaliensis* and the agent is thought to be more common in the Southern region⁵.

Of the 37 cases of *S. somaliensis* mycetoma recorded so far from India, 18 were from Southern India and 12 of them were from Tamilnadu. The organism is said to frequent areas with an annual rainfall of 50–250 mm⁷. The average rainfall in most areas of India being above 600 mm per year may account for the rarity of the infection here.

Enlargement of the regional lymph node is often attributed to secondary bacterial infection. Rarely it is caused by actual spread of fungus to the related lymph node and *S. somaliensis* was the predominant cause of lymphadenitis associated with mycetoma, being found in 9 out of 13 instances in the series reported by Hassen and Mahgoub¹⁴ from Sudan. We had also encountered one case (Case 1) in our series.

Although involvement of bone is an important feature of mycetoma, its

degree and extent varies with the species of infecting agent, site of lesion, stage of development and intensity of infection. According to Delahaye et al¹⁵, bone lesions appear during the 2nd year after infection and the affinity of the organisms for the bones increases in progressive order from *Streptomyces madurae*, *Madurella mycetomii*, *S. somaliensis* and *Streptomyces pelletierii*. Of the 4 cases of *S. somaliensis* mycetoma reported by Grueber and Kumar⁶, only one had bone lesions and the duration of the infection was 6 years. In our series, since the duration of the disease was not more than 2 years, bone involvement had probably not taken place.

The importance of histopathological examination in cases of mycetoma is evident by the fact that in all the 90 cases reported from Madras, where the granules were demonstrated, presumptive species identification was possible¹⁶. Several workers have repeatedly stressed the reliability of the histological appearance of the granules in tissue sections for the diagnosis of species responsible for mycetoma^{17,21}.

Comparative studies made during the last few years have shown that there is often complete agreement between identification by examination

of the grains in tissue section stained by haematoxylin and eosin and identification by culture^{17,19,22,23,26}. However, histological investigation has its limitations. With a little experience, specific diagnosis of the etiologic agent in mycetomas due to *Madurella mycetomi*, *Neotestudina rosatii*, *Actinomadurella madurae*, *A. pelletieri* and *Streptomyces somaliensis* is possible by examination of the granules in sections. In other cases, the identification is more difficult^{20,23,26}. It is only possible in such cases to recognize a group of organisms which produces morphologically similar granules: *Leptosphaeria senegalensis*-*L. tompkinsii*²⁷⁻²⁹; *Nocardia asteroides*-*N. brasiliensis* - *N. caviae*^{20, 30}; *Allescheria boydii* - *Acremonium* spp *Fusarium* spp³¹. In some rarer mycetomas e.g., due to *Aspergillus nidulans*^{32,34} *Phialophora jeanselmei*^{33,35} *Acremonium* spp.,^{36,38} some dermatophytes^{39, 40} and some particular actinomycetes^{41,42}, the comparative morphology of the grains and their etiology have not yet been sufficiently studied.

Though the ideal method for the specific diagnosis of mycetoma is by isolation of the causal agent in culture, mere histological examination of the granule is sufficient to establish the diagnosis in many cases. This is very important since specific identification by culture is very slow and not even always possible⁷.

Early diagnosis of mycetoma before any bony involvement and identification of the species of fungi causing it is very important, as the ideal specific therapy depends upon it. Clinical cure was obtained in all our 5 patients with a combination of antibiotics which included streptomycin together with excision and skin grafting since the bones were not involved in them.

In mycetomas caused by *A. madurae*, *A. pelletieri* and *S. somaliensis*, Streptomycin has been used most successfully⁴³. Clinical improvement was

noticed with an initial dose of 3g of Streptomycin daily for 3 weeks, decreasing to 2g daily and then 1g for similar interval by Abbot⁴³ and Lynch⁹. Other broad spectrum antibiotics either singly or in combination have been used with some success in these infections.

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