NOCARDIA PELLETIERI CAUSING MYCETOMA IN INDIA

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Mycetoma is a chronic inflammatory lesion caused by a variety of actinomycetes fungi. It is easily diagnosed by the characteristic clinical features of nodules and draining sinuses eventually leading to tumefaction of the affected parts. The causative organism in any particular case has to be determined by careful evaluation of several laboratory data.

Nocaridiosis is caused by several species of aerobic actinomycetes. Among these *N. asteroides* is of world wide distribution. Nocardial mycetomas are more prevalent in the tropics and subtropics. So far all cases of nocardial mycetomas seen in our clinic have been due to infection by *N. asteroides*. The purpose of this paper is to present our first case of mycetoma caused by *N. Pelletieri* and report a few hitherto undescribed features encountered in this case.

CASE REPORT

Patient was a 36 years old man He was a lifetime resident of south India and a palmist by profession. He reported to the dermatology clinic for the first time in December 1968 In 1962 patient injured his left insole with a thorn. Three months later, a nodule developed at that site. This broke down to form a discharging sinus. With the development of more nodules and sinuses, the lesion spread to involve most of the left insole and one or two adjacent areas on the dorsum. By this time patient began to notice small white granules in the discharge. About one year after onset of the complaint, a local excision was done in a nearby hospital. This however, failed to arrest progress of the disease. By 1967 the lesion had spread upto the ankle on its medial aspect. One year prior to patient's visit to hospital, he developed new lesions on the lateral border of foot, popliteal region and thigh. Nodules and sinuses at these sites also extruded small white granules with pus.

General physical examination revealed abnormality confined to the left lower extremity. The left foot was grossly deformed and swollen with multiple nodules and discharging sinuses. Small creamy white granules about 1 mm. in diameter could be expressed from these sinuses. Another ulcerated nodule about 1 cm. in size was seen on the lateral aspect of foot dorsally. Some ulceration and scarring were seen in the popliteal fossa and medial aspect of the thigh. In close proximity to the ulcers in these latter two areas, were present fusiform nodular swellings

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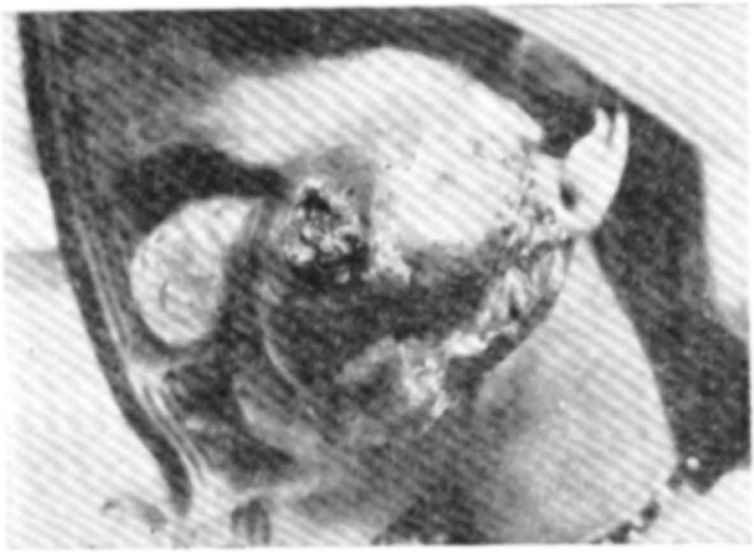
Received for publication on 12-8-1970.

extended from the side of nose to the tragus of ear. (fig. 2). It was not tender.

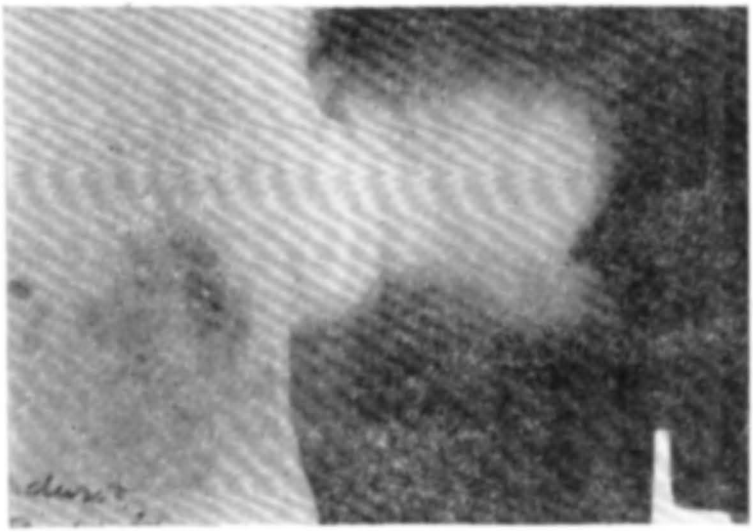
Fig. 2



The consistency varied, below the ulcerated area it was somewhat softer than rest of it, which was firm to hard. The overlying skin was red in colour and was adherent to the growth. The swelling was also fixed to the underlying muscles, clinching of teeth caused restriction in the mobility of the swelling. It was not adherent to the underlying bones. No abnormality was detected in abdomen, lungs and heart.

Invesigation: Haemoglobin- 10 gm%, total and differential leucocyte counts were within normal limits. Screening of the chest did not reveal any abnormality. X-ray of the left side of the face revealed a soft tissue shadow without involvement of the underlying bone (fig. 3). A provisional diagnosis of fibrosarcoma was made.

Fig. 3



At operation: a wide excision of the swelling along with the involved muscle fibres was done. Created raw area was covered by split skin graft. The post-operative period was uneventful except for lower eyelid oedema which developed on 2nd day. The patient was discharged on 10th post operative day. Some oedema of the left lower eyelid persisted. Zygomatico-temporal facial palsy was present.

Pathology report: Gross appearance: It was a soft irregular tumour mass surmounted by ulcerated skin, the size of which was 10 x 8 5 cms. It cut soft with the knife. The cut surface was smooth, pink in colour, here and there blotched by known coloured arees. The streaks and whorls of fibrous tissue traversing the whole mass could be seen.

Microscopic appearance showed proliferated and interlacing bundles of fibroustissue in the sub-cutaneous resion. Bundles of spindle and oval cells intersecting and at places forming whorls were also seen. The cytoplasm was scanty and the nuclei were large and vesicular with hyper-chromatism. Skin corium was also having ce ls with hyperchomatic and mitotic figures (Fig. 4 and 5).

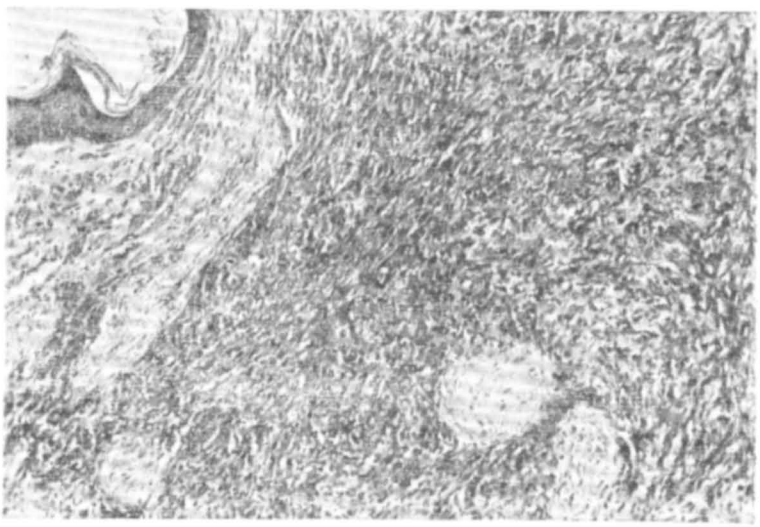


Fig. 4

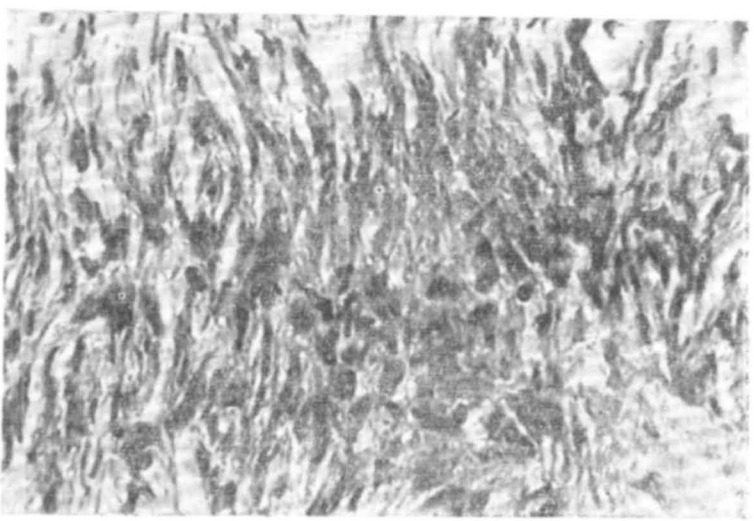


Fig. 5

It is not possible to give the exact results of the treatment since the patient did not again report after one year, till then there was no evidence of recurrence.

DISCUSSION

The tumour is neither rare nor common. The incidence is slightly less than 0.1 per cent (Pack and Tabah, 1951). The condition is more common in females (Hoffmann, 1925; Pack and Tabah, 1951) and occurs between the ages of 20–45 years, though it has been described at the age of 3 years (Penner, 1951; Basto and Martins, 1964). The average age reported is 44.3 years (Pack and Tabah, 1951).

The etiology of the tumour is controversial. Many theories have been advocated in an attempt to explain the origin and histogenesis of these tumours: parasitic (Darier and Ferrand, 1924); traumatic (Hoffmann, 1925); angiomatous (Hoffmann, 1925); mammogenic (Binkley, 1939) and the fibroblastic theory (Pack and Tabah, 1951). There is no evidence of familial occurrence of this neoplasm. The influence of pregnancy on these tumours has been mentioned (Pack and Tabah, 1951). McGregor (1960) described the epidermal layer normal except for the alteration and loss of rete pegs. The corium is occupied by a uniform layer of connective tissue with lacunar spaces. Beneath this layer is a stratum composed of bundles of spindle cells with a few mitotic figures, which penetrate the normal adipose tissue underlying the deepest portion of the growth.

The tumour has a greater predilection for the trunk than for the extremities (Hoffmann, 1925; Pack and Tabah. 1951). The head, scalp, back, limbs, buttocks and genitalia, however, may also be involved (Binkley, 1939).

It starts as a firm cutaneous or subcutaneous nodules covered with normal epidermis, the early or so called "plaque stage". These nodulels may coalesce as the lesions advance, develop red to blue colouration and grow to be very large tomours, pedunculated or broad based, the latter so called as "tumour stage". As a rule, these tumours are hemispherical and sessile growths. Ulceration may occur at this stage with formation of a soft, fungating granulomatos mass. The underlying plaque spreads laterally and much more slowly into the deeper tissues, which accounts for the difficulty encountered in complete removal and in the local recurrence. Fixation to the overlying skin is always present. Invasion of muscle and fascia is comparatively unusual (Pack and Tabah, 1951).

The condition is practically asymptomatic and does not affect the general helth. Discomfort owing to size and/or lacation of the tumour is perhaps the most frequent complaint. Pain and bleeding are rare symptoms. The bleeding is never gross haemorrhage and in some cases, it is a sero-sanguinous discharge. The lesion in its early stages may be mistaken for a keloid, wen, desmoid tumour, true fibro-sarcoma, neurogenic sarcoma, nonpigmented malignant melanoma. sclerosing haemangioma and sweat gland carcinoma.

The rate of growth is imperceptibly slow at first. It may take many years before the tumour strarts to enlarge with increasing rapidity in weeks or months'

The history of long duration of the tumour emphasizes its relative slowness of growth. Pack and Tabah (1951) reported four patients, in which the lesions were present at birth.

There is a lot of controversy regarding the nature of the tumor. Darier and Ferrand (1924) considered it to be locally malignant, while Binkley (1939) thought it of low grade malignacy. Metastases are very rare, but these have been reported when either the tumour has been present for a long time, 38 years (Bezency, 1933) and 50 years (Binkley, 1939) or after many inadequate local excisions. There is not a single report of metastatases to the lymph nodes (Basto and Martins, 1964); though there are proved instances of metastases at other sites (Bezecny, 1933, Penner, 1951). Recurrences of the tuumour are known (Srivastava et al. ,1964; Razdan and Seth, 1969).

Wide surgical excision gives good risults (Borrie, 1952). The removal of fascia overlying the muscles along with the specimen is advocated routinely (Pack and Tabah, 1951). The subjacent muscles may be excised, if the neoplasm infiltrates through the fascia. Amputation for this tumour is never indicated. These tumours are radio-resistant. The therapy has been recommended post-operatively, when treating recurrences (Basto and martin, 1994). Radioactive isotopes and chemotherapy have not proved of value in the treatment of these tumours.

The prognosis as regards life is good. Pack (1954) reported a 5- year survival rate of 69.2 per cent after surgical treatment.

SUMMARY

A case of dermatofibrosarcoma protuberans of the face, a rare neoplasm of the skin, rs reported The relevant literature on the subject is briefly discussed.

ACKNOWLEDGEMENTS

We are thankful to Dr. D. G. Ojha, M., S. F. A. C. S., Principal and Controller, S. P. Medical Colleget and Associated Group of Hospitals, Bikaner for permitting to publish this case.

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