

NOCARDIA PELLETIERI CAUSING MYCETOMA IN INDIA

By

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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.

Nocardiosis 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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simulating lymph glands (fig. 1). Left inguinal group of glands were markedly



Fig. 1.

enlarged. On few occasions, it was possible to demonstrate presence of creamy white granules in the discharge from the ulcers on the thigh and popliteal region. Clinical diagnosis of mycetoma was made. A probable tuberculous infection, superimposed on mycetoma was suspected, because of the presence of metastatic lesions along the course of ascending lymphatics.

LABORATORY STUDIES

Direct examination of granules obtained from the sinuses and biopsy material failed to reveal chlamydo spores or fungal filaments.

Gram's stain showed delicate gram positive branching filaments (fig. 2).

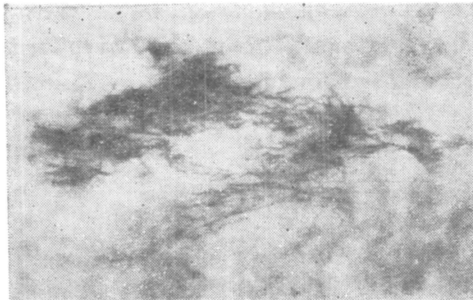


Fig 2.

These were non-acid fast by the Kinyon's method of staining.

Biopsy nodules on the foot showed presence of multiple granules within abscesses deep in the dermis. Gram staining of the section revealed gram positive filaments (fig 3).

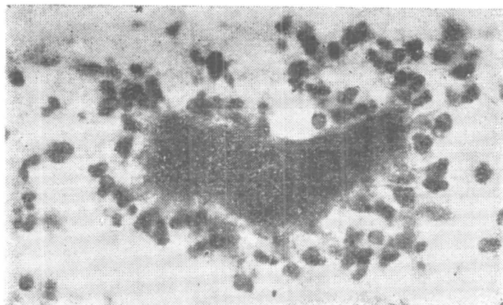


Fig. 3.

Multiple granules obtained from biopsy material were cultured on Sabourauds Dextrose Agar (SDA). SDA showed a slow growing fungus which grew to an appreciable size only in 4–5 weeks. However, subcultures from colonies grew in 3 weeks' time. To start with, the colonies were folded and pinkish. They became deep coral red in colour in course of time. The full grown colonies were heaped, glabrous, cerebriform and coral red with no aerial mycelium or diffusible pigment. Growth was best at 37° F. Smears of the growth also showed delicate branching non-sporulating filament which were non acid fast. The growth was identified as *N. Pelletieri*. Granules recovered subsequently from nodules on the thigh also grew the same organism. Subcultures of two of the patient sent to Dr. Libero Ajello, National Communicable Disease Centre (NCDC) Atlanta, Georgia, confirmed their identity as *N. Pelletieri*.

X-ray of left foot showed extensive bony destruction. X-ray chest revealed opacity in right 3rd intercostal space and left 4th space. These were thought to be patches of pneumonitis.

Biopsy of a lymph gland removed from left thigh in close proximity to a nodule and sinus showed only chronic nonspecific lymphadenitis.

No evidence of tuberculosis seen in the sections from the nodules or lymph gland.

Acid fast stain of the pus from nodule was negative for T. B.

E. S. R.—1st hour 25mm. 2nd hour 56mm

Other routine blood, urine and stool examinations revealed no abnormalities.

COURSE IN HOSPITAL

On January 22, 1969, below knee amputation was done although the diagnosis of Nocardiosis was made, because the orthopaedists were of the opinion that the bone changes in the foot were severe enough to justify such a procedure. Amputation stump healed except for three areas of discharging sinuses. Creamy white granules could be expressed from these sinuses also. On direct smear these granules showed Gram positive filaments. On February 27, 1969, 5 weeks after the amputation, patient was operated again. The 'lump' on the thigh was dissected out and was traced proximally to its termination along a lymphatic vessel. This tissue on histology and other laboratory investigations showed same features as that of earlier specimen from the foot. An adjacent lymph gland which was also removed showed no evidence of fungus or tuberculous infection.

From March 5, 1969 patient was given sulphamezathine 4 gms. per day. He was discharged from hospital in good condition. Patient continued to take Sulphamezathine for 5 months and he stopped treatment on his own. Patient was seen again in January 1970, ten months after initiation of sulphamezathine treatment. At this time his amputation wound and other areas of ulceration had healed. There was no evidence of any existing sinus.

DISCUSSION

Large majority of *N. Pelletieri* infections have been reported from Africa and South America. Cases have been reported from Yemen. One case of *N. Pelletieri* has been reported from the United States. To the best of our knowledge, ours is the first case of *N. Pelletieri* to be reported from the Asian-Continent

N. Pelletieri is known to produce red coloured granules as one of its specific characteristics. This had been so in all previously reported cases. However, in an article by Pardo-Castello¹ and Trespalacios¹, casual mention was made of 2 Cuban cases where the colour of the granules was not red. Because the granule did not show red colour, the diagnosis on these cases was questioned by Bergeron et al². Further details on these cases were not available to us.

In our patient, the granules expressed from all the sinuses at different times showed creamy white colour. Since the cultural characteristics were identical with those of *N. Pelletieri* and all the available tests confirmed the same both in our own laboratory and by the National Communicable Disease Centre, USA, we believe that, occasionally cream coloured granules can also be seen in *N. Pelletieri* infections.

Another unusual clinical feature in this case was the sporotrichoid picture. When first seen we suspected that this patient had mycetoma with tuberculosis, the latter accounting for an ascending lymphatic spread and scrofuloderma in the popliteal region and thigh. All investigations done to detect presence of tuberculous infection in these lesions were negative. No foci of tuberculosis were found anywhere in this patient. On the otherhand, fungal granules proved to contain *N. Pelletieri* were obtained from the lesion. We therefore, believe that this is a case of *N. Pelletieri* with sporotrichoid picture. In our cases of *N. asteroides* infection sporotrichoid feature has never been seen although such a picture has been reported rarely in *N. braziliensis* infections³.

Rey⁴ believed that *N. Pelletieri* is the most virulent of all the actinomycotic mycetoma agents. He therefore, advocates radical surgery wherever possible. Vanbreuseghan⁵ on the other hand claims good results with sulfones and sulfonamides. In our patient the response to sulfonamide was satisfactory. Ten months follow up on our patient showed that the sinuses in the popliteal region and thigh as well as those which formed at the site of amputation wound, had all healed well with medical treatment. An attempt at further follow up on this patient was unsuccessful.

SUMMARY

A case of mycetoma caused by *N. Pelletieri* with sporotrichoid clinical picture is presented

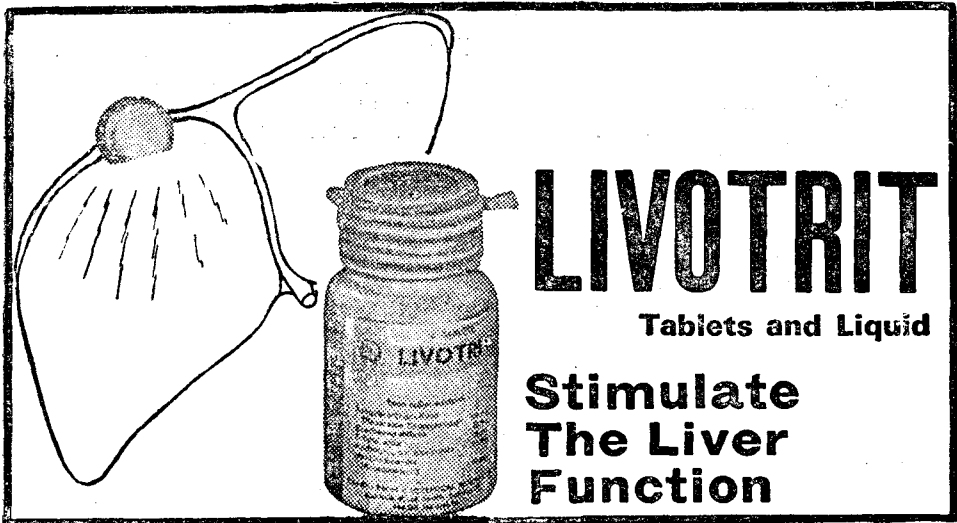
It is believed that this is the first published case of *N. Pelletieri* from Asia.

Although granules of *N. Pelletieri* are believed to be always red in colour, occasionally they could also be white.

We are grateful to Dr. Libero Ajello of the National Communicable Diseases Centre, Atlanta, Georgia for his help in confirming our identification of the fungal isolate from the patient, as *N. Pelletieri*.

REFERENCES

1. Pardo-Castello' V. and Trespalacios, F. 1959. Superficial and Deep Mycoses in Cuba - A report based on 1,174 cases. SOUTHERN MED. J. 52, 7-15.
2. Bergeron J. R. et al. 1969. Mycetoma caused by Nocardia Pelletieri in the United States of America. ARCH. DERM. 99, 564-566.
3. Moore, M. and Conrad A. H. 1967. Sporotrichoid Nocardiosis caused by Nocardia Brasiliensis. ARCH DERM, 99, 390-?
4. Rey, M. 1961 Les Mycetomas, dans l'ouest africain, Paris: Foulon & Cie.
5. Vanbreuseghem, R. 1967. The early diagnosis of Mycetoma. DERM. INT. 6, 123-140.

An advertisement for Livotrit. On the left is a detailed line drawing of a human liver. To its right is a glass bottle with a stopper, labeled 'LIVOTRIT'. The bottle has a textured surface and a small label with the product name. To the right of the bottle, the word 'LIVOTRIT' is printed in large, bold, black letters. Below it, 'Tablets and Liquid' is written in a smaller font. Further down, the text 'Stimulate The Liver Function' is written in a large, bold, sans-serif font.

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