IMPRESSIVE OF PRECISION DIAGNOSIS IN MANAGEMENT OF GLUTEAL MYCETOMA: CASE REPORT

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ABSTRACT

BODY: A 48 year old civil servant who had four year copious discharge from multiple left gluteal sinuses is presented. He had had traditional herbal treatment self medication and empirical antituberculous treatment. Plain X-ray of pelvis, sinogram and detailed mycotic studies enabled diagnosis of actinomyces israeli. A course of co-trimoxazole was efficacious in drying up the discharge, healing of the sinuses and reversal of the patients early depressive overlay. Precise etiologic diagnosis in Mycetoma located at unusual site would enable direct efficacious antimicrobial treatment since the lesions are caused by different organisms.

BACKGROUND: Mycetoma lesions can constitute a major challenge in diagnosis and treatment.

Key Words: Mycetoma gluteal region, actinomyces Isreali, mycotic Studies, Sinuses.

INTRODUCTION

Mycetoma is a chronic cutaneous and subcutaneous infection caused by fungi and actinomycoses, and occasionally aspergillus species. The disease is characterized by progressive destruction of soft tissue and nearby structures. It was first recognized as a disease entity in 1842 by Gill in the province of

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Madura, India. Seventy percent of all mycetomas affect the foot, while other sites where it has been reported include the upper extremities, trunk, buttocks, eyelids, lacrimal glands, paranasal sinuses, mandible, scalp, neck, perineum and testes.

It is relatively common in parts of Africa, including Nigeria. The frequency appears to be directly related to heavy rain and hot climates. Predisposing factors include history of trauma, walking barefoot, agricultural work, poor personal hygiene, poor nutrition and wounds of multiple infection.

Forty percent of mycetoma are caused by fungi while other organisms account for sixty percent. The destruction may affect muscles, bone, blood, lymphatic vessels and nerves. When mycetoma affects any of the rare sites, including the gluteal region, high index of suspicion is required to arrive at a diagnosis. Since it is caused by a multiplicity of organisms requiring different medications, precise diagnosis is prerequisite for successful treatment.

The differential diagnosis includes tuberculosis, osteomyelitis, elephantiasis, malignancy, yaws and a host of other deep mycoses. This report is presented to highlight the symptomatology, difficulty in diagnosis and outcome of treatment of mycetoma in a relatively rare gluteal site.

**CASE REPORT**

A 48 year old male civil servant presented at the orthopaedic clinic of our hospital with complaint of multiple discharging sinuses of left buttock for four years. It was said to have started as a single painless boil with surrounding induration. The boil was said to have gradually increased in size with progressive pain, and finally ruptured after three weeks. The discharge from the ruptured boil was thick, milky and copious and terminally become mixed with blood. There were several episodes of recurrent ruptures, discharges and temporary healing of the swelling. Subsequently, other swellings and sinuses developed to occupy the entire left gluteal region. The discharges from the numerous sinuses were such that
the patient had to wear pads to avoid staining of his clothing. He had associated episodes of mild fever that preceded ruptures of each swelling. He also experienced pain that was exacerbated by sitting and relieved by lying supine. There was no associated history of trauma and patient did not observe any granules from the discharge. Patient showed signs of depression and was considerably worried about his state of hygiene. He resented the idea of having to wear absorbent cotton wool always. He had had an operation on the right gluteal region about eighteen years before. He also had exploration of the right foot for a two year old discharging, non-healing wound, said to be Athlete’s foot eight years before presentation and has a residual scar on the third and fourth metatarsal spaces. Patient had both oral and topical traditional herbal treatment and alternated these with self-medication using erythromycin, without much improvement.

On presenting at the hospital, the initial managing team had commenced empirical anti-tuberculous treatment following a finding of uniform sclerosis of the left iliac bone extending to the left hip joint, which showed cloudy opacification.

On examination, there was indurated swelling of left gluteal region down to the left gluteal fold.

The skin showed patchy areas of dark pigmentation, wet with discharges and punctuated by twelve discharging sinuses (fig I). A sinogram showed multiple interconnecting sinus tracks with four areas of pooling of contrast, but not connected to bone (fig. 2)
The discharge was subjected to mycotic study vide infra:

LABORATORY MATERIALS AND METHODS

Two samples were collected, the first was by means of a sterile syringe and needle, while the second was by means of a sterile swab stick.

The specimens were immediately transported to the laboratory within the hospital premises for laboratory work. A portion of the pus was gram-stained and also examined for sulphur granules. Another portion was then grown in enriched media of brain-heart infusion broth, both anaerobically (10% CO₂)
and aerobically at 37°C. A fixed portion was then subsequently cultured on sabouraud dextrose Agar (SDA) and both thioglycollate broth and thioglycollate agar and incubated at 37°C for one week. Further microscopical and biochemical tests were carried out on the growth from these two media.

Culture grown from the thioglycollate agar was subjected to gram and modified Ziel Neelson stain and club-shaped filamentous bacteria with molar teeth appearance. Zeil Neelson and catalase tests were negative, while the fermentation tests were positive with production of gas for glucose, lactose, and mannitol. This confirms the presence of non-acid fast Actinomyces Israelii. 7

TREATMENT
Following the diagnosis of mycetoma caused by Actinomyces Israelii, the patient was placed on co-trimoxazole 1g b.d. The discharges progressively diminished and much of the sinuses commenced healing.

After six weeks of treatment, patient asked for discharge, having progressively become dry without need to wear pads. He defaulted on follow using 1% volume by volume (v/v) acid alcohol, while catalase and fermentation tests of actose, glucose, mannitol with peptone water base and 10% sugar was also carried out.6,7.

RESULTS
Direct microscopic examination of the pus smear for sulphur granules was negative. However, examination of cultures grown on thioglycollate broth and agar revealed gram positive twined up, and therefore, not available for a final clinical picture to show healed lesion.

Figure 1: Multiple discharging gluteal sinuses with induration of the entire left gluteus.
DISCUSSION
The case presented in this report had a morbidity of four years to presentation. During this time, the patient had traditional herbal treatment and self-medication. The location of the mycetoma and the consequent unacceptable copious discharge in the gluteal region might have accounted for patient’s reluctance to seek appropriate medical treatment. Furthermore, the chronicity of the condition coupled with the natural life history of mycetoma accounted for the changes in the iliac bone and hip joint which further confused the picture and pointed towards tuberculous ileitis and tuberculosis of the hip joint.\(^1,3\).

However, the multiplicity of the sinuses should have pointed to the diagnosis\(^1-5\). The sinogram was invaluable in reaching a working diagnosis\(^1,2,3\). The detailed laboratory study to arrive at a diagnosis was an essential part of management of this patient, in that the use of antimycotic drugs such as ketoconazole, amphotericin – B and anti-tuberculous drugs could not have been effective and would have added to the patient’s frustration.\(^1\)

We note that simple therapy such as co-trimoxazole proved\(^1\) quite effective in the control of the infection. We did not have to adapt any surgical procedure, such as incision and drainage/debridement on this patient since the
symptoms responded to medical treatment.
The patient was lost to follow-up immediately he noticed signs of improvement, in spite of counselling to that effect. This is the usual attitude of our patients in this environment who detest the idea of being used for a study. We conclude that high index of suspicion, full scale laboratory investigation and precise drug treatment is required for successful treatment of mycetoma when it affects unusual sites.

REFERENCES: