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Case Report

Coexistence of eumycetoma and tuberculosis in a same lesion in right thigh in an adult: a rare case report

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ABSTRACT

Eumycetoma is an uncommon infection in adults, especially as a co-infection with extra pulmonary tuberculosis. We report a unique case of Co-existence of Eumycetoma and tuberculosis in a same lesion in an adult from a known endemic area of Tamilnadu, India which posed a diagnostic and therapeutic challenge.

Keywords: Eumycetoma, Tuberculosis, Co-existence

INTRODUCTION

Eumycetoma is an uncommon infection, especially as a Co-infection with tuberculosis in a same lesion in an adult. Eumycetoma is a chronic cutaneous and subcutaneous infection caused by various genera of fungi, leading to progressive destruction of soft tissue and the nearby anatomical structures.[1] Eumycetoma is mainly a disease of the tropical and subtropical zones, seen in adults, belonging to low socioeconomic status or rural background who are manual workers like agriculturalist and who walk with barefoot in dry, dusty environment.

It is characterized by a triad of tumefaction, draining sinuses and presence of colonial grains in the exudates.

A major cause of morbidity and mortality in developing countries like India is tuberculosis. We present a unique case of Co-existence of Eumycetoma and tuberculosis in a same lesion in the Right thigh of a 35 year old woman.

CASE REPORT

A 35 Year-old woman from a rural area of thiruvallur district, Tamilnadu (India) presented with a lesion on the posterolateral aspect of right thigh with multiple discharging sinuses since last 3 years.

On physical examination, mild fever and moderate anemia was observed. The lesion measured 18 x 15cm in size on the posterolateral aspect of right thigh which was mobile, tender and firm with hyperpigmented overlying skin (Figure 1).

Figure 1: Right thigh lesion with multiple sinuses with hyperpigmentation.

Touch smear from back of right thigh lesion was taken and it showed Blastospore, thick septate hyphae, features suggestive of Eumycetoma.

Figure 2 shows, high power view showing peripheral radially arranged hyphae with central irregular hyphal arrangement surrounded by splendor hopple phenomenon.

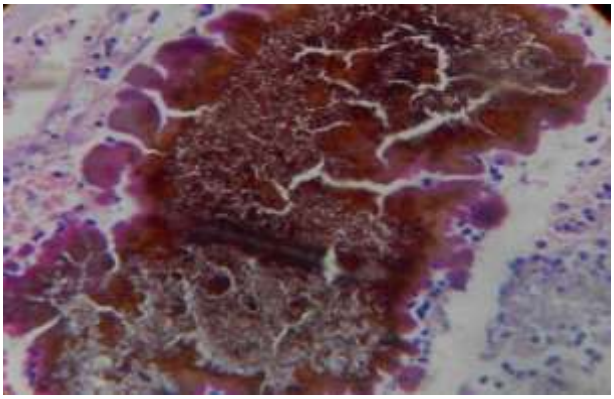

Figure 2: High power view of hyphae surrounded by splendor hopple phenomenon

X-ray Right thigh was normal. There was no extension of the sinus tract to the bone. Abnormal laboratory Parameters were microcytic hypochromic anemia with hemoglobin level of 10 gms/dl, urine and blood cultures were negative.

Based on the clinical and laboratory parameters, diagnosis of Eumyceoma right thigh was made. Patient was started on Antifungal medication in the form of Terbinafine 250mg twice daily, Itraconazole 200mg once daily.

Monthly follow up of the patient showed signs of improvement like being afebrile and pain free has. However, there was no improvement in the healing of the sinus on the right thigh even after 12 months.

There were discharging sinuses from the right thigh, although its size remained the same.

Wide excision and split skin grafting was done. Specimen were examined Histopathologically, section showed stratified Squamous epithelium with acanthosis and hyperkeratosis with focal ulceration, underlying dermis and subcutaneous tissue showed neutrophilic abscesscess, multiple granulomas composed of Langhans type of muttinucleated gaint cells, epithelioid histiocytes and lymphocytes and congested blood vessels. No fungal organisms were seen.

Hematoxylin and eosin (H and E) stained sections showed no evidence of grains finally, features diagnostic of Tuberculoid Granuloma. These smears were further subjected to ziehl - Neelsen (ZN) staining for acid fast bacilli, which showed negative for acid fast bacilli (AFB).

Mantoux's test was carried out which was positive, with a wheal of 26mm, X-ray chest was normal, Erythrocyte sedimentation rate was 60 cumm/hr. No significant lymphadenopathy was noted.

In view of this histopathological finding, a diagnosis of Tuberculosis was made. The patient was put on Antitubercular treatment (ATT) in the form of category - I, Rifampicin, Isoniazid, Ethambutol and Pyrazinamide, Monthly follow-up of the patient showed signs of improvement and complete healing of the wound was observed.

DISCUSSION

Eumycetoma is mainly a disease of the tropical and subtropical zones, especially between the tropic of cancer and the tropic of Capricorn, that is between latitudes 15* S and 30* N.

The disease is commonly seen in adult men. It usually presents between 20 and 50 years of age with male to female ratio 3.5: 1.[2] It is commonly seen in people who live in rural areas in endemic region. Agents that cause eumycetoma are Primarily Saprophytic microorganisms that are found in the soil and on plant matter.

Healthy persons become inoculated with these agents as a result of the traumatic implantation of thorns, Splinters and other Plant matter. Therefore, the incidence of mycetoma is more particularly occupation dependent as people like farmers, herdsmen and fieldworkers are more likely to come in contact with the causative agents.[3]

The most common site is foot as 70% of all mycetomas affect the foot hence the name madura foot.[4] However, extrapedal involvement also occurs and has been detected in hand, leg, head and neck, abdominal wall, buttock and perineum.[2]

The rural area to which the patient belongs has similar environmental condition, i.e. dry and dusty with thorny vegetation all around. All these factors might have favoured the development of Eumycetoma right thigh in this case.

We also postulate that the Tuberculosis was most likely acquired by contact with an infected individual in the community as tuberculosis is endemic in southern tropical region, due to the low- Socioeconomic status of the people, malnutrition, low immunity, which predisposes the population to this overwhelming disease. Eumycetoma may present as a small localized tumor like mass, with or without sinuses, or can be associated with significant morbidity interms of gradual enlargement, destruction and deformity of the affected site. The

diagnosis of Eumycetoma is made tentatively clinically, when discharging grains are visible to the naked eye.[2]

The grains vary in colour, size and consistency depending on the causative agent and can be confirmed by culture method.

Granules of eumycetoma are firm 0.2 - 5mm aggregates of organized, vegetative septate hyphae, which often are embedded in a matrix cement substance of the eumycetoma, producing black granules. M. Mycetomatis accounts for most cases worldwide. Pseudoallescheria boydii is the common aetiologic agent in the United States, while madurella grisea is a common aetiologic agent in south America.[4]

In general, the geographical distribution of the various mycetoma agents is related to the amount of rainfall and other climatic conditions, and thus, each geographical region has a different list of most common agents.

The smears from eumycetoma lesions have a distinct cytological appearance, characterized by brown to black colonies of branching, septate hyphae embedded in a matrix which stain positively with PAS or Gomori's methenamine silver stains, both demonstrating large sized hyphae of eumycetoma.[5,6]

A diagnosis of eumycetoma should be suspected in case of discharging sinuses, especially those exhibiting black granules. The cytological diagnosis of eumycetoma can be as accurate as histological diagnosis, and techniques such FNAC as well as imprint smears can definitely be taken in to consideration before planning any medical or surgical treatment as these are simple, inexpensive and fairly reliable techniques without any obvious disadvantages. Special fungal stains can also be well applied to cytological specimens for further confirmation, whereas culture studies are helpful in confirmation of diagnosis and species identification.

In literature, we found no reports of Eumycetoma patients coinfected with Tuberculosis in a same lesion, so the Present case highlights that in a known endemic areas, predisposing environmental factors may further aggravate the disease by Co-existing together.

Present case posed a diagnostic dilemma as the patient was a 35-year-old female having multiple discharging sinus lesion on right thigh and Tuberculosis which was not responding to Anti-fungals alone. Hence, it is suggested that physicians/ surgeons should be alert to any discharging/ non - discharging, single/ multiple sinus in an adult from a known endemic area of tuberculosis/ mycetoma. The basis of management includes: clinical examination, high index of suspicion, histopathology, isolation and culture of organism.

Eumycetoma are only partially responsive to anti-fungal therapy but can be treated by surgery due to their normally well circumscribed nature. Surgery in combination with azole treatment is the recommended regime for small eumycetoma lesions in extremities.[7]

This patient has been on a regular follow-up for 4 months and has shown complete healing of the wound.

CONCLUSION

Eumycetoma is mostly found in areas with low living standards, the disease is often neglected in the initial stage. Due to this, there is a high incidence of secondary bacterial infection, deformities and recurrences. So, increased awareness and emphasis on early and definite diagnosis after meticulous clinical examination assisted by histological and microbiological studies is required. [8]

In the setting of full susceptibility, other entities, such as Co-infection must be suspected, irrespective of age. The tuberculosis and Eumycetoma occurring simultaneously in a patient in a same lesion is rare and therefore presents a diagnostic challenge and dilemma in clinical practice, especially in developing countries where tuberculosis still has a very strong foot hold.

Early identification of both these entities prevents not only cost and prolonged diagnostic and therapeutic interventions but also the complication.

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