



BASIDIOBOLOMYCOSIS - A REPORT OF TWO CASES.

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Abstract : Basidiobolomycosis a rare chronic granulomatous subcutaneous fungal infection caused by *Basidiobolus haptosporus* is seen predominantly in children. Here we report two cases of basidiobolomycosis who had been diagnosed elsewhere as chronic abscess, cutaneous tuberculosis and soft tissue tumour. Both of them presented with well circumscribed subcutaneous plaque with India rubber consistency. Fingers could be insinuated beneath the margins of swelling. A clinical diagnosis of basidiobolomycosis was made. Histopathology showed granulomatous infiltrate consisting mainly of eosinophils and few giant cells with fungal elements seen as unstained tubes and haloes surrounded by splender hoeppli material which confirmed the diagnosis of basidiobolomycosis. Both the children were started on mixture of potassium iodide to which they showed good clinical response. This case report is being reported to highlight the importance of early diagnosis and prompt treatment of the infection which has a characteristic clinical morphology and histopathology, so as to avoid unnecessary surgical intervention.

Keyword : Basidiobolus haptosporus, granulomatous, potassium iodide.



FIGURE 1 - Subcutaneous plaque over gluteal region

INTRODUCTION :

Basidiobolomycosis is a rare chronic granulomatous subcutaneous fungal infection seen mainly in children. The causative organism is *Basidiobolus haptosporus*. Most of them are misdiagnosed as chronic abscess, paniculitis, morphoea and subsequently subjected to surgical intervention. But this infection which is rare has characteristic clinical and histopathological feature and is well amenable to treatment with potassium iodide.

CASE REPORT :

CASE 1 : A six year old female presented with painless subcutaneous swelling over right gluteal region of six months duration.

The child was diagnosed elsewhere as chronic abscess and was subjected to incision and drainage and subsequently biopsy was done and a diagnosis of cutaneous tuberculosis was made and was started on antituberculous therapy was started. Since there was no response to ATT, the child was referred from Tiruvannamalai to ICH&HC, Egmore where dermatology opinion was sought. General and systemic examination of the child was normal. Dermatological examination revealed a subcutaneous plaque of size 10x8 cm over right gluteal region with India rubber consistency and finger insinuation was possible beneath the margins. Three scars of size 2x1 cm, 1.5x1 cm and 1x0.5 cm were present.[Figure 1] **CASE 2 :** A three old male child presented with painless swelling over right side of trunk of one year duration. The child was diagnosed elsewhere as soft tissue tumour and was referred to dermatology op. General and systemic examination of the child was normal. Dermatological examination showed subcutaneous plaque of size 15x10 cm over right side of trunk with India rubber consistency and fingers could be insinuated beneath the margin of the swelling.[Figure 2]



FIGURE 2 - subcutaneous plaque over rt. side of trunk

Histopathological study of both patients showed granulomatous infiltrate consisting mainly of eosinophils and few giant cells with fungal elements seen as unstained tubea and haloes surrounded by splender hoeppli material [Figure 3,4] which was consistent with basidiobolomycosis.

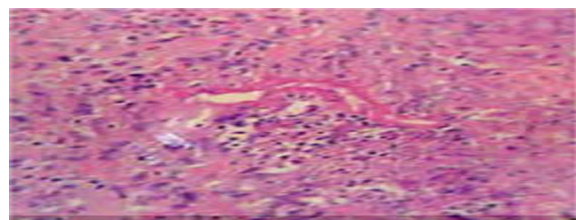


FIGURE 3 - Histopathology of case 1

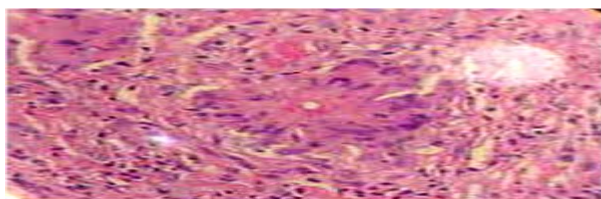


Figure 4 Histopathology of case 2

Gomori methenamine stain showed coenocytic hyphae with black wall and unstained centre which further confirmed the diagnosis. [Figure 5,6]. Fungal culture was not done as the patient was referred with slide and blocks and biopsy specimen was not available.



FIGURE 5 - GMS stain of case 1

Both the patients were started on mixture of potassium iodide in low dose and observed for signs of iodism and subsequently dose titrated to a dosage of 40 mg/kg/day. Good clinical response was seen over a period of 2 months and complete resolution was seen in 6 months and they are on follow up. [Figure 7,8]



FIGURE 8 - case 2 - post treatment

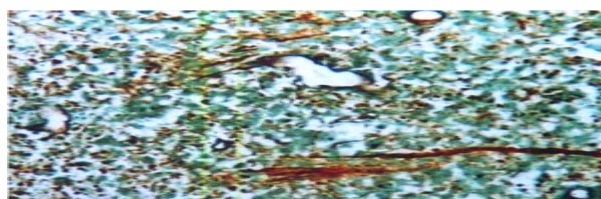


FIGURE 6 - GMS stain of case 2

DISCUSSION :

Entomophthoromycosis is a rare subcutaneous fungal infection. It manifests in two clinically distinct forms namely, basidiobolomycosis and conidiobolomycosis. Basidiobolomycosis is caused by *Basidiobolus haptosporus*.¹ Conidiobolomycosis is caused by *Conidiobolus coronatus* and *C. incongruus*. *Basidiobolus haptosporus* is a saprophytic fungus present in soil, decaying vegetable matter and gut of amphibians and reptiles. First case of basidiobolomycosis was described by Lei

Kion Joe in 1956 in Indonesia. It is seen in tropical areas of Asia and Africa mainly in children and adolescents. Males are affected more than females. Mode of infection is by inoculation, ingestion and inhalation.² Basidiobolomycosis manifests clinically as well demarcated painless subcutaneous plaque or nodule with a firm India rubber consistency and finger insinuation was possible beneath the margins of the swelling.³ The surface skin may be pigmented. Regional lymphadenopathy may or may not be present. Complications are persistent elephantiasis, muscle invasion with resulting tenderness and hydronephrosis due to pressure effect on ureters.⁴ Differential diagnosis include chronic abscess, soft tissue tumour, paniculitis, localised morphea, sarcoidosis.

Investigations include wet mount of the biopsy tissue in 10% KOH showing broad non septate or infrequently septate coenocytic hyphae. Culture in Sabouraud's dextrose agar with acidition shows grey to yellow glabrous colonies with central raised growth and radial folds. Zygospores are characteristic. Histopathological examination is characterised by granulomatous inflammatory infiltrate consisting of eosinophils, giant cells, histiocytes plasma cells in deeper dermis and subcutis. Fungal elements are seen as poorly stained or unstained tubes and haloes surrounded by splender hoepli material. Vessels are not involved. The fungal elements are better stained with PAS (purple) and GMS (black).⁵

Potassium iodide is the gold standard drug given in a dose of 40-60 mg/kg/day.⁶ Other drugs found to be effective are itraconazole in a dosage of 3-6 mg/kg and ketoconazole 4 mg/kg. These drugs are given upto 1 month after resolution. In widespread infection IV amphotericin B 0.6 mg/kg is used.

CONCLUSION :

Though a rare subcutaneous fungal infection, this disease has characteristic clinical and histopathological features. So a early correct diagnosis and prompt treatment will help in avoiding unnecessary surgical intervention.

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