AFRICAN JOURNAL OF MEDICINE
and medical sciences

VOLUME 21, NUMBER 2, DECEMBER 1992

EDITOR: B.O. ONADEKO
ASSISTANT EDITORS:
B.O. OSOTIMEHIN and A.O. UWAIFO

SPECTRUM BOOKS LIMITED
Ibadan • Owerri • Kaduna • Lagos

ISSN 1116–4077

Afr. J. Med. med. Sci. (1992) **21**, 19-21

African histoplasmosis (Duboisii) of the skull with neurological complication — *A case report and review of literature*

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Summary

A case of African histoplasmosis of the skull associated with neurological deficit has been reported. There was complete recovery of neurological features after excision of the lesion followed by a course of co-trimoxazole. A review of the available literature indicates the rarity of this particular mode of presentation. The reversibility of the neurological complications makes it important that clinicians increase their awareness of this treatable condition.

Résumé

Un cas de l'histoplasmosis du crâne Africain associé a la déficit neurologique a été signale il ya en un recourement complete des traits néurologiques après l'exersion de la lasion suivi par une course de co- trimoxazole. Une revue de la littérature accessible indique la rarité de cette mode de présentation particuliere.

La réversibilite des complications neurologiques rend important que les chiciens augementent leur savoir de cette condition traitable.

Introduction

The clinicopathological changes which result from infection of *Histoplasma capsulatum* are well recognized and have been documented in many articles[1,2,3,4,5,6]. Infection by *Histoplasma duboisii* leads to a wide spectrum of clinical conditions. The lungs are not often involved although pulmonary lesions have been described[7]. It is the skin, subcutaneous tissues, bones and joints that are predominantly affected [6]. Dissemination to the lymph nodes, spleen, liver and bowel has been described[8,9,11]. When the disease is disseminated, fever, anaemia and weight loss are common. The course is rapidly progressive and the outcome is generally fatal [8]. When the lesion is localised, it tends to affect only one tissue although local spread by contiguity may occur. The disease may be represented by a circumscribed skin lesion or a localised lesion in the bone. In such cases, there are no signs of systemic disturbances such as fever, anaemia and weight loss. The course of the disease is chronic with phases of progression and regression but throughout there is a strong tendency to spontaneous healing[11].

In the skeletal system, *H. duboisii* forms a granulomatous lesion which destroys bone trabeculae and with expansion erodes the cortical bone. If the periosteum becomes affected, it is lifted off the surface with the production of extracortical new bone. The majority of patients with skeletal manifestation show multiple foci; flat bones are more frequently affected but long bones and the spine can also be involved[6,8,12]. Radiologically-detectable skull granulomata and the sequential development of the lesion from a small diploic destruction extending to the inner and outer tables, have been described. Multiple skull lesions are normally seen[8]. This paper reports a case of African histoplasmosis of the skull with reversible neurological signs; a condition which had hitherto not been reported.

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Case report

BA was a 12 years old male student who was referred to the neurosurgical unit with history of a swelling over the left frontal area of over 6 months duration. The mass, which had been progressively increasing in size, had not been tender and there was no antecedent history of trauma. He had a history of frontal headache which precipitated his presentation at the general out-patients department two weeks prior to being seen in the neurosurgery unit. A diagnosis of subgaleal cyst was then initially entertained and the patient was treated symptomatically with analgesics and ampiclox 500mg QID for one week on out-patient basis. He was reviewed by the paediatric surgery unit one week later and after reviewing the

skull-x-ray, a diagnosis of osteomyelitis of the skull was made. His antibiotics were then changed to chloramphenicol and lincomycin.

Within one week the patient had developed loss of appetite, fever, vomiting and severe headache with increase in size of the swelling. On examination, he was drowsy, febrile and moderately pale with equivocal evidence of meningism. There was a left fronto-orbital swelling about 5cm x 1.5cm in dimension. The mass was warm and fluctuant to touch. It was tender with hyper- pigmentation of the skin on the lateral aspect. The mass was non- pulsatile and had no bruit. The entire scalp had evidence of subgaleal collection of fluid, which turned out to be pus on aspiration. There was opthalmoplegia of the left eye with paralysis of the 6th, 4th and partial paresis of the 3rd cranial nerves. He also had a right hemiparesis, more prominent in the upper extremity.

Investigations

Haemograms showed packed cell volume of 32% and a white blood cell count of 14,380 with 44% neutrophils, 48% lymphocytes, 6% Eosinophils and 2% Monocytes. The roentgenographic studies (Fig.1) included a composite frontal and lateral skull x-ray and chest x-ray. The skull x-ray showed a circular bone lesion with sclerotic edges suggestive of osteomyelitis. The chest x-ray was essentially normal. Fluid aspirated from the subgaleal space was negative on gram staining and showed no growth after 48 hours.

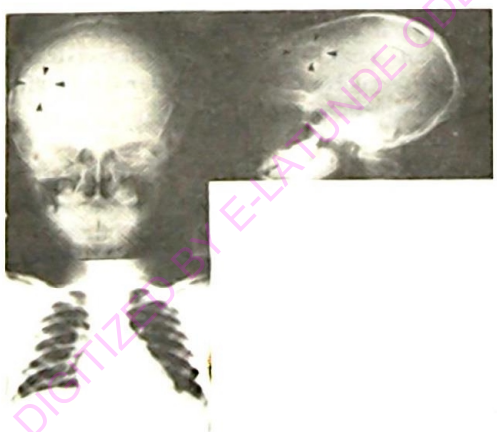

Fig.1: Composite frontal and lateral skull x-rays. Chest x-ray. Note arrows by skull defect.

Surgery and follow up

In view of the above presentation and the result of the investigation, surgery was undertaken. The mass was incised through a crescent-shaped scalp incision over the left fronto-parieto temporal area. After raising the skin flap; a fungating necrotic mass was noted to be projecting from the calvarium through the bony defect in the skull. The capsule of the mass was opened; the content appeared cheesy. The capsule and surrounding exuberant subgaleal granulation tissue and the sclerotic edge of bone were excised. The immediate underlying *dura mater* was noted to be thickened with deposits of the cheesy material. This too was excised and moderately oedematous brain was noted below. The dural defect was closed. The entire specimen including the dural tissue was sent for histopathology.

Microscopy of the cheesy material revealed yeasts which was confirmed histopathologically by special staining – (Fig. 2a and b). Post-operatively the patient was treated with co- trimoxazole, two tablets twice daily for a duration of two weeks. He improved with resolution of neurological deficits. He was discharged to follow up as out-patient. There has been no recurrence in the past nine months.

Discussion

A review of the literature has revealed no cases of African histoplasmosis in which there is involvement of the brain and its coverings. Cockshott and Lucas[8] reported that the skull was involved in twelve of their cases and described the radiological features of the gradual destruction of the diploic tables in the development of a calvarial granuloma. Williams *et al*[11], reported that the skull was affected in 18 of the 101 cases of African histoplasmosis which included the 56 cases reported by Cockshott and Lucas earlier. There was no mention of intracranial abscess formation in any of the above cases. Dubois and Janssen[9] reporting from the Congo in 1952 described a case of "head abscess" from *Histoplasma duboisii* although no mention was made of neurological deficits as a sequelae of the disease. Cockshott and Lucas[8] mentioned as private communication, osteolytic lesion of the skull in a case in Northern Nigeria. Ours is perhaps the only case in which neurological changes have been observed to be associated with the skull lesion. That the neurological deficits resolved following wide excision of the granulomatous lesion and a course of co-trimoxazole confirms the experience of other reporters that in localised lesions, complete excision is curative[6,8,11].

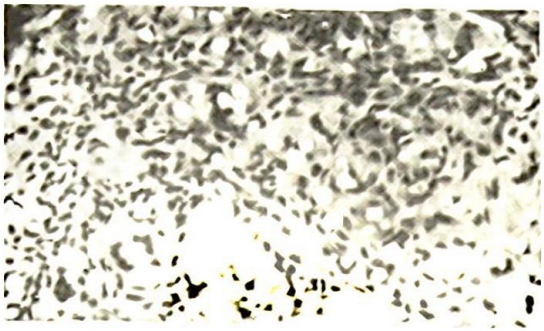

Fig.2(a): (HE stain) Histoplasma are seen in the cytoplasm of histiocytes and giant cells. Some are found extracellularly.

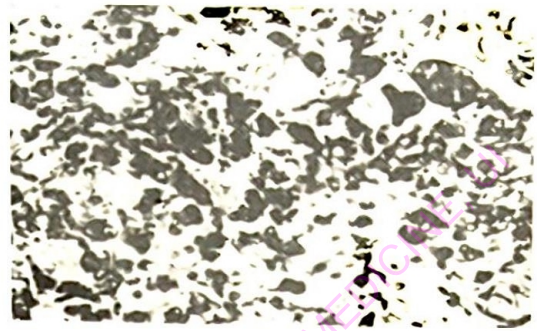

Fig.2(b): Methenamine-Silver stain demonstrating the fungi. They appear as black oval bodies.

Although Amphotericin B has proved efficacious in the management of disseminated disease[4], it is of interest that where this drug was not available the substitution of co-trimoxazole in our case following excision has not been associated with recurrence.

Such neurological deficits as were noted may be due to inflammation of the *dura mater* and its contiguity with the underlying brain tissue and subsequent focal oedema. This may be confused with cavernous sinus thrombophlebitis but the neurological picture of the latter is quite distinct from the presentation of our patient. Complete resolution of the neurological deficit in this case is explained by the removal of the inflammatory tissue adjacent to the brain.

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(Accepted 11 June, 1990)