Research Paper

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# Primary Tuberculous Osteomyelitis of Skull -A Rare Presentation

Medical Science

**KEYWORDS :** Tuberculous osteomyelitis, skeletal tuberculosis.

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## ABSTRACT

*In recent years, there has been a substantial increase in number of reports of tuberculosis from uncommon sites. Skeletal tuberculosis constitutes about 1% of all tuberculosis cases. Usually spine and limb bones are involved. However calvarial tuberculosis has been reported very rarely in world literature till now. We are reporting a case of primary tuberculous osteomyelitis of sphenoid and frontal bones in a 4year-old boy. With prompt as well as careful workup we were able to establish the diagnosis and with appropriate surgical intervention and anti-tubercular treatment, we were able to halt the disease progression and excellent response to treatment was observed on follow up.*

**Introduction**

The occurrence of tuberculous osteomyelitis is about 1-6% among all cases of extra pulmonary tuberculosis (1). Primary tuberculosis of skull vault is an extremely rare presentation among cases of primary extra-pulmonary tuberculosis (1). Only a few cases of primary tuberculous osteomyelitis of skull vault have been reported in the world till now.

Although precise aetiology behind rare incidence of calvarial tuberculosis (TB) is largely undefined, the peculiar pattern of blood supply in flat bones is sometimes implicated in causation of this rarity (2). We are reporting a case of a primary tuberculous osteomyelitis of sphenoid and frontal bones in a 4 year old boy.

**Case report**

A four-year-old boy presented with complains of painless swelling over right periorbital area. It had been growing slowly over a period of last 6 months. It was not associated with fever, headache, visual disturbance, any focal neurological deficit or ear discharge. There was no history of tuberculosis in the patient or of tuberculosis contact in past. There was no history of weight loss, bone pains, lump in the abdomen or over any part of the body, cough or breathlessness and the child was fully immunized for age.

On examination, the child was stable with a pulse rate of 86/min, and blood pressure of 90/60 mm Hg. There was no pallor, lymphadenopathy or bony tenderness. BCG scar was seen over the left arm. The abdomen revealed firm, non-tender hepatomegaly with a liver span of 10cm. The liver surface was smooth and had rounded margins. Local examination revealed the presence of an oval diffuse firm swelling (4cmx 3cm) in the right temporal region near the eye without any changes in the overlying skin or sinus formation. No other abnormalities were detected on the general or systemic examination.

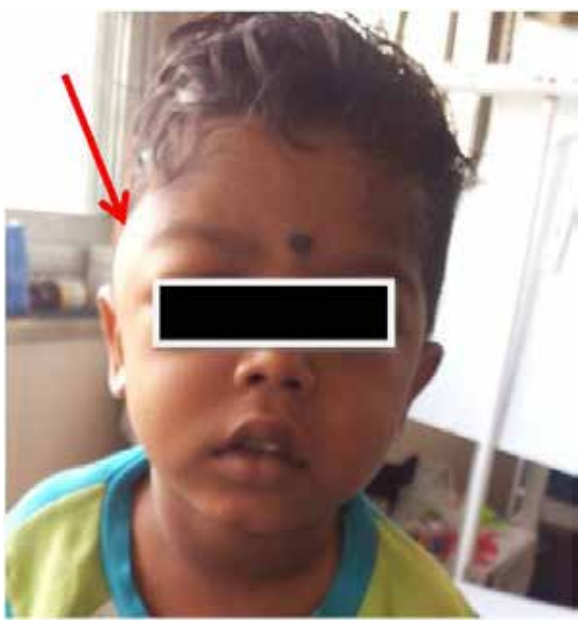

**Figure 1: showing location of the swelling in right temporal region at presentation (with arrows).**

**Figure 2: showing location of right sided temporal swelling in lateral view (with arrows)**

Investigations carried out in the child revealed anemia (Hb 6.4gm.dl) with normal leukocyte and platelet counts. No abnormal cells were seen on the peripheral smear. The ESR was 90 mm at the end of one hour. The serum levels of transaminases were normal (SGOT: 60 IU/ml; SGPT: 45IU/ml) and other liver function parameters [serum albumin 3.3gm/dl; Prothrombin Time: Patient: 13sec (Control: 14 sec); serum bilirubin: 0.8 mg/dl] did not reveal any abnormal levels. Mantoux test was negative (2mm X 2mm induration at 72 hours). The skull radiograph revealed multiple osteolytic lesions without sclerotic margins over frontal, sphenoidal and temporal skull bones. No abnormalities were detected on chest radiograph or ultrasonography of the abdomen. The gastric lavage did not demonstrate any acid fast bacilli on smear or culture. The serum levels of uric acid and lactate dehyrogenase (LDH) were normal.

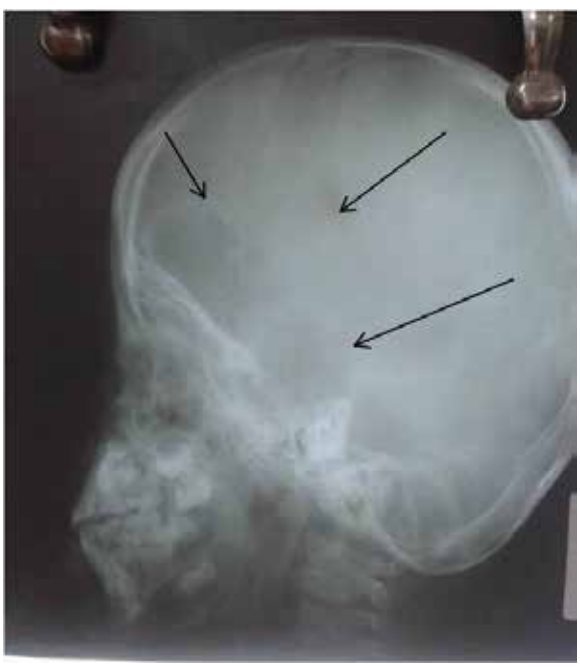

**Figure 3: Shows non-sclerotic osteolytic lesions in skull (showed with arrows).**

CT scan of local part showed diffuse irregular osteolytic lesion involving right frontal, squamous part of temporal and greater and lesser wings of sphenoid with thinning of lateral margins of right orbit.

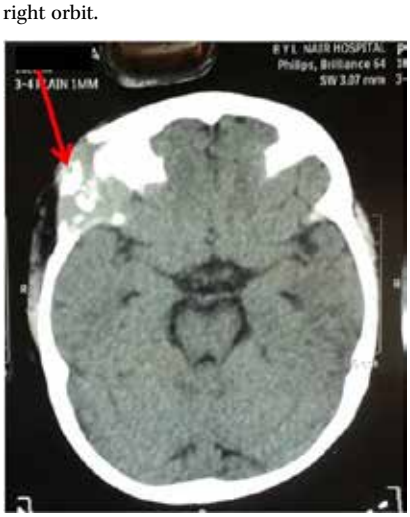


**Figure 4 (a): CT scan of brain with temporal cuts showing osteolytic lesions.**

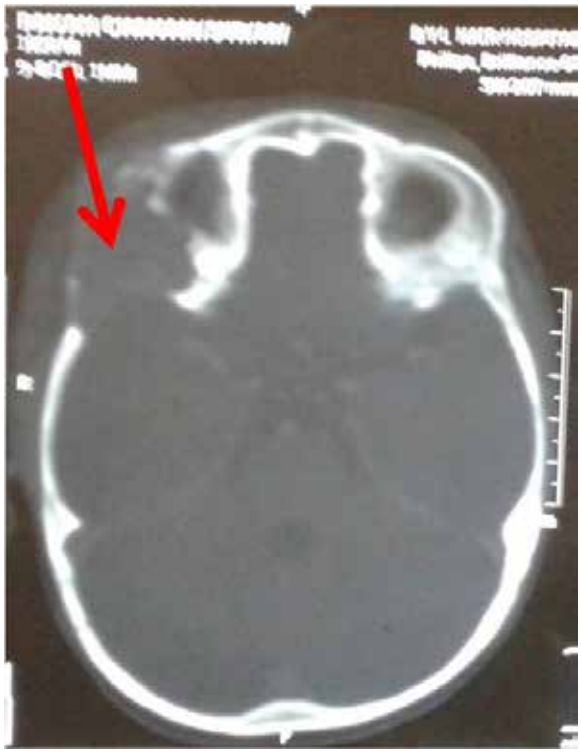

**Figure 4(b): CT scan brain with temporal cuts showing right sided osteolytic lesion with thinning of lateral margin of right orbit.**

Bone scan-did not reveal osteolytic lesions in any other part of body.

**Further management**- USG of local part showed hypoechoic lesion in right temporal fossa with coarse, immoble internal echoes within with extensive erosive changes in the underlying bone. USG guided biopsy was done which showed multiple mononuclear cell and macrophage infiltration with necrosis. No evidence of acid fast bacilli in the biopsy specimen; fungal stains were also negative. Bone marrow biopsy was done to rule out malignancy, Langerhan cell histiocytosis. It was normal except for increase in histiocytes however CD 1a, S100 stains were negative.

Excision of the right sided granulomatous lesion involving up to right sphenoid bone was done. Histopathology of the excised tissue revealed tuberculous osteomyelitis of right sphenoid and frontal bones and and with characteristic granulomas and caseous necrosis. No evidence of neoplasm or fungal growth. BACTEC culture of excised tissue for tuberculosis came out to be positive.

Child was started on anti-tubercular treatment with isoniazid, rifampicin, ethambutol and pyrazinamide for 6 months. Child is doing well without any complications or recurrence of swelling over last 3 months and is having adequate weight gain.

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**Figure 5: post-operative photograph of patient showing extent of incision made during surgery.**

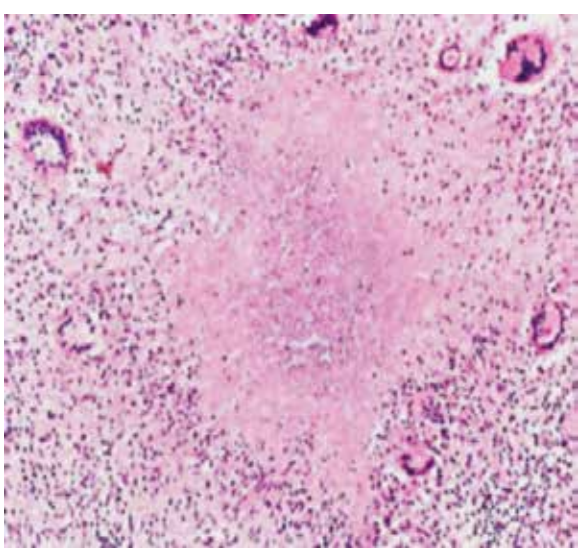

**Figure 6: Histopathology from excised specimen (focused under high power) showing classical caseous necrosis suggestive of tuberculosis.**

**Discussion:**

Tuberculosis of skull is a very rare occurrence and accounts for approximately 1% of skeletal tuberculosis (3,4,5,6). Skull tuberculosis occurs in only 0.01% of patients with mycobacterial infections (3). Most cases occur in the first two decades; however infants are rarely affected, probably because of the paucity of cancellous bone in the skull (7). It usually affects children, 50% being less than 10 years of age and 75-90% less than 20 years of age (3,8,9). The frontal and the parietal bones are usually involved and have been attributed to relatively more cancellous bone elements in these bones as compared to other bones of skull vault (8). A solitary discrete round or oval punched out osteolytic defect with minimal surrounding sclerosis in the fronto-parietal bones is the commonest presentation of skull tuberculosis. Occipital and sphenoid bones are rarely affected (10, 11). However in our case, sphenoid bone was affected along with frontal bone. The type of clinical presentation depends perhaps on the immunity of the individual (12). Once the marrow of the diploe is seeded with the inoculum, the infection spreads towards the inner and outer table, causing bone destruction and formation of granulation tissue. The extension of the infection through the diploe is resisted by proliferation of an encircling layer of concentrically placed fibroblasts and if the process is not arrested, extension then takes place through either table (7). If the process is rapid, sequestration may occur; this appears as "bone sand" on radiography. Involvement of the outer table is usually associated with scalp swelling or a discharging sinus while involvement of the inner table results in extradural granulation tissue. The dura acts as a barrier to further spread, however intradural involvement is occasionally seen. A good immunity will cause slow and restricted evolution of the lesion, while decreased resistance will rapidly lead to subgaleal or extradural collections. Our patient initially presented with painless swelling of right sided temporal region without any discharging sinus. It is emphasized that proper radiological examination, and a fine needle aspiration cytology (FNAC) /local biopsy can be very helpful. It is important to subject every surgical specimen for histo-pathological examination, as caseous material of tuberculous origin and cheesy material of sebaceous cyst may look alike on gross examination. Though dura acts as strong barrier against intracranial spread, such spread is rarely known to occur (3,9,13,14). A raised ESR and positive tuberculin skin test are usually found. However in our case though ESR was high but tuberculin test was negative. Calvarial tuberculosis is known to have three types of radiological findings, the common circumscribed lytic type or perforating tuberculosis of the cranium with small circumscribed punched out lesions (our case showed radiological features of this type); the diffuse type, in which there is widespread involvement of diploe with destruction of the inner table and epidural granulations; and circumscribed sclerotic type (8,10). CT scan though non- specific, may reveal an irregular bony defect which is wider at inner table, a diffuse hypodense lesion with enhancing margins and a well circumscribed enhancing lesion. Other pathologies mimicking such picture include pyogenic osteomyelitis, eosinophilic granulomas, metastases, haemangiomas, aneurismal bone cyst, meningioma, neuroblastoma and syphilis (6, 16, 17). Ziehl – Neelsen stain and culture for mycobacterium are diagnostic (14,16) but may not be positive in 50 percent of cases. FNAC/excision biopsy should be done in lesions with overlying intact skin (17). Histopathology revealing epitheloid granulomas with Langhans type giant cells and caseation necrosis is diagnostic. Surgery is indicated for removal of large extradural collections causing neurological deficit, large pockets of caseating material, associated sinus formation or fulminant secondary infection and when diagnosis remains uncertain (3,8,18). In our case the main indication for surgery was uncertain diagnosis and it also facilitated removal of large amount of caseous material. This case represents primary tuberculous osteomyelitis of skull as no other primary focus of tuberculosis could be obtained even after extensive investigations. Anti-tubercular therapy has been recommended for 18-24 months (14, 16, 17) by some authorities but one year course has also been found adequate [19]. As per latest Revised National Tuberculosis Control Program (RNTCP) of India which follows WHO guidelines, 6 months treatment is recommended.

Reports of involvement of rarer sites by tuberculosis are continuously pouring in, particularly after the rise in number of HIV

cases all over the world. Tuberculosis of skull vault must be considered in the differential diagnosis of swellings of skull when a chronic infective pathology is suspected as timely intervention can save the patient from impending serious neurological complications or death. Successful treatment in this case, further establishes the usefulness of WHO recommended DOTS treatment in rare extrapulmonary forms of tuberculosis.

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