DOI: https://doi.org/10.3329/ewmcj.v12i1.77190

Case Report

TUBERCULAR OSTEOMYELITIS OF SKULL: CASE REPORT AND REVIEW OF THE LITERATURE

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Article History:
Received: June 2024
Accepted: July 2024

Keywords:
Tuberculosis, Osteomyelitis, Skull TB, Cranioplasty

Abstract:

Skeletal tuberculosis constitutes about 1j of all cases. Usually, spine and limb bones are involved. Tubercular osteomyelitis of the skull is a rare entity; therefore, diagnosis is rarely suspected. We report one such rare case of an atypical calvaria skull osteomyelitis in a14 years old female who presented with swelling on the right frontoparietal region for 3 months. CT scan of brain and bony window revealed a lytic bony lesion at the right parietal bone with associated epidural soft tissue component at the right parietal top causing compression over adjacent parenchyma- possibly eosinophilic granuloma or chronic osteomyelitis. Patient was treated surgically by excision of the lesion with cranioplasty by synthetic bone cement. After surgical treatment, the specimen was sent for histopathology which showed granulomatous inflammation histologically consistent with tuberculosis. She was treated with antitubercular therapy. Our Case highlights all these aspects of skull tuberculosis with review of the available literature relevant to skull tubercular osteomyelitis.

EWMCJ Vol. 12, No. 1&2, January 2024-July 2024: 111-119

Introduction:

Osteomyelitis is an inflammatory process associated with infection-induced bone destruction. Osteomyelitis is most common in the long bones like the tibia. But it can also involve the skull bone from other primary sources. Though tuberculosis (pulmonary) in Bangladesh is very common, tuberculosis in the skull bone (tubercular osteomyelitis) is a rare entity. Skull base osteomyelitis was first described and identified in 1959, by Meltzer and Keleman.[1] Skull Base Osteomyelitis is an infection of the temporal, sphenoid, or occipital bones that is caused as a complication of otogenic, sinonasal, odotogenic and rhinogenic infections, especially in immune-compromised conditions.[2] Tuberculosis of the skull is a very rare occurrence. It usually affects children, 50j being less than 10 years of age and 75-90j less than 20 years of age.[3] A high degree of suspicion and minimal investigations mainly radiological are necessary to exclude this uncommon diagnosis and more importantly, start prompt therapy after diagnosis is confirmed by biopsy and histopathology. Especially in a developing country like ours, where TB is a leading health problem, this rare clinical presentation of the infection should not be missed.

Given the rarity of the condition, only a few patients with Tubercular osteomyelitis of the skull have been reported in the medical literature. Here we present an intriguing case of Tubercular Osteomyelitis of Skull that was successfully treated by medical and surgical treatment. We also review the current literature and provide a summary of different factors including treatment and outcome.

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Case Report:

A 14-year-old female, normotensive, nondiabetic, and nonasthmatic presented to the neurosurgery department with complaints of swelling over the right side of the scalp for 3 months. Initially, the swelling was soft and nodular type which was growing rapidly and occasionally painful. She felt pain during the combing of her hair while taking a shower for 3 months. She also complained of discharge from swelling which was clear. There was no history of fever, loss of consciousness, convulsions, vomiting, blurring or loss of vision, or difficulties in hearing. She also did not report any history of weight loss, night sweats, or loss of appetite.

On examination, the swelling was present on the right frontoparietal region which was 5 cm in diameter, having a smooth surface, firm in consistency, margins in continuity with bone ridges, immobile but skin over it was free and without signs of local inflammation. Lungs were clear. Neurological examinations revealed no abnormality.

Routine investigation revealed Hemoglobin was 9.8 g/dl. ESR- 61 mm/hr. Total WBC count 5300/cumm. Neutrophils 59j, Lymphocyte 34j, Monocyte 4j, Eosinophil 3j. Chest X-ray showed normal study. CT scan of the Brain with Bony window with 3D Reconstruction revealed a lytic bony lesion at the right parietal bone with an associated epidural soft tissue component at right parietal top causing compression over adjacent parenchyma- possibly eosinophilic granuloma or chronic osteomyelitis [Fig-1].

Then, the patient and her guardians were counseled about the disease and the possible need of an operation to confirm the diagnosis, as bone erodes and this lytic bony lesion was associated with epidural soft tissue component that may compress brain parenchyma causing irreversible neurological deficit. With adequate counseling and written consent, surgical management was done.

A linear incision was given over the swelling and excision of scalp swelling was done. As there was erosion of bone found, we made a 2 c.m margin clearance. The lesion was extradurally so we tacked up the dura with 4-0 vicryl round body. The defect area of the bone was covered by cranioplasty using synthetic bone cement - polymethyl methacrylate (PMMA). After ensuring hemostasis, the wound was closed in layers keeping a drain tube beneath the

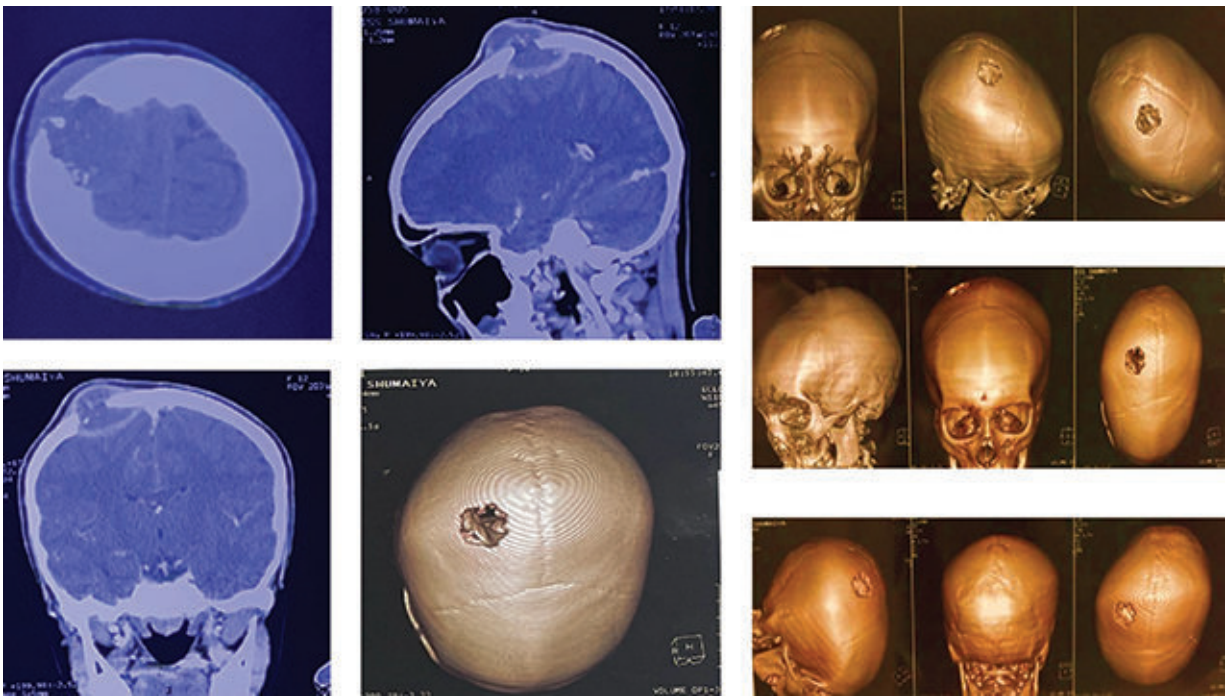

Figure 1 : *CT Scan of Brain with Bony window with 3D Reconstruction revealed lytic bony lesion at right parietal bone with associated epidural soft tissue component at right parietal top causing compression over adjacent parenchyma.*

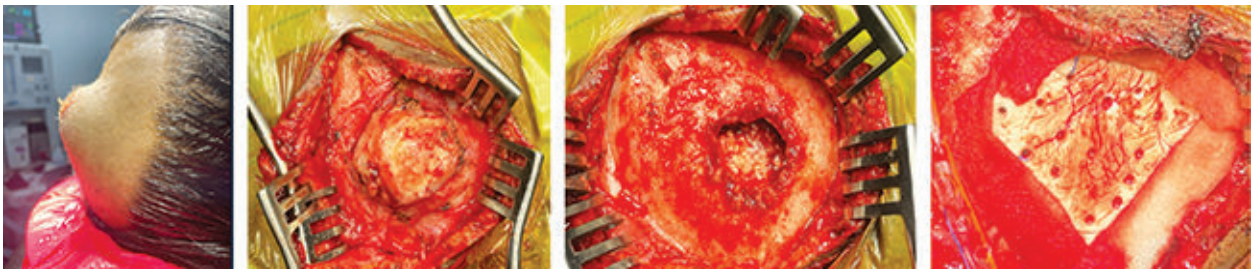

Figure 2 : *Per operative picture from left to right: a) Skull Swelling before incision; b) Exposure of lesion involving parietal bone; c) after excision of lesion and 2 cm bone margin clearance; d) cranioplasty done by bone cement and fixed with non absorbable prolene 2-0 suture materia*l.

scalp which was removed on 1st postoperative day. [Fig-2]. Scalp swelling and Bone margin were sent for histopathology. A post-operative Ct scan of the Brain was performed on 1st post-operative day to see any residual and status of cranioplasty which revealed no residual lesion at site and synthetic bone was placed in place. [Fig-3].

Histopathological examination showed granulomatous inflammation histologically consistent with tuberculosis [Fig-3].On the basis of the histopathology

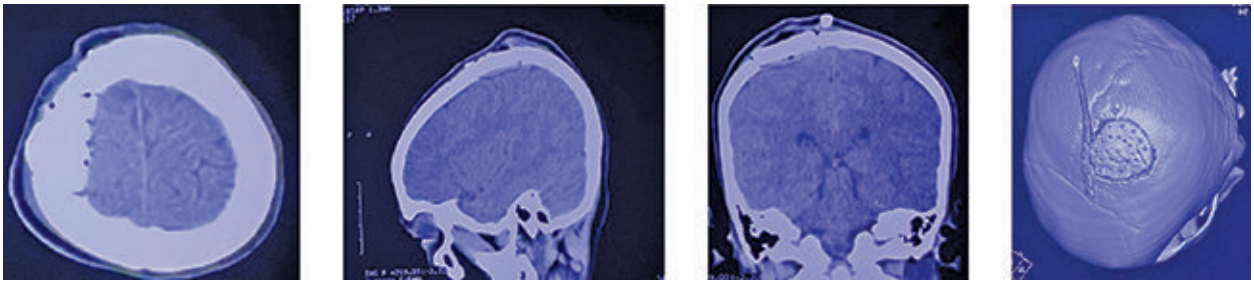

Figure 3 : *Craniotomy and excision of scalp swelling and cranioplasty by bone cement - polymethyl methacrylate (PMMA).*

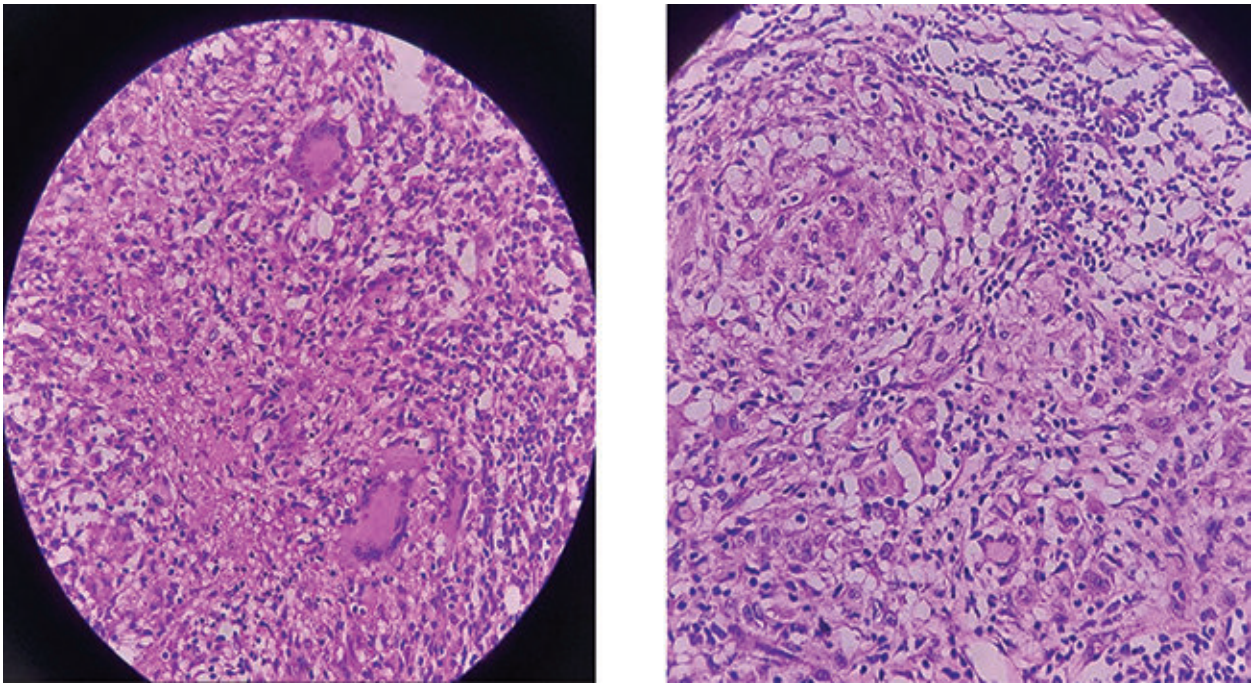

Figure 4: *Histopathology which shows granulomatous inflammation histologically consistent with tuberculosis*

report, the patient was diagnosed case of Tubercular osteomyelitis of the skull. So the patient was treated with anti-tubercular drugs as per the National Guideline For The Management of Tuberculosis.

During OPD follow-up on the 7th POD, 15th POD, and at 1 month, the patient improved significantly without any neurological deficit. She was advised to continue anti-tubercular therapy for 12 months. Physical and occupational therapy noted the patient to progress well, returning well, returning very near baseline upon discharge.

Literature Review:

Details of the literature search strategy using PRISMA guideline, we found 54 search results on Pubmed and MedLine via 3 search terms: "Tubercular osteomyelitis of skull", "Skull osteomyelitis", "TB Skull" from a period between 2000 to 2024 with full text and reviewed article. 38 results remain after the exclusion of inconsistent material and duplicated articles. After a full-text review, 7 articles remain for further review which was included in qualitative synthesis summarized in Table I.

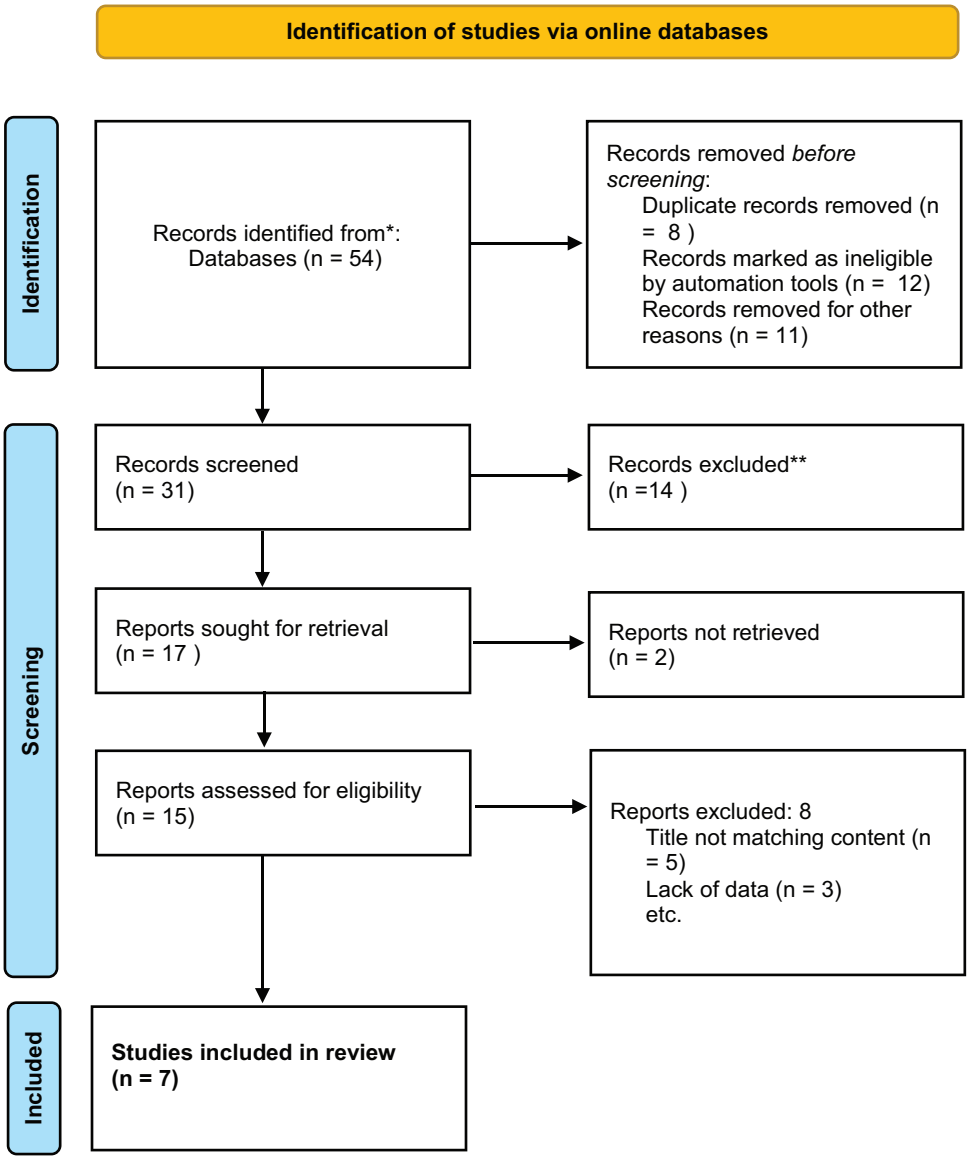

Figure 5: *Literature search strategy using Prisma guideline 2020*

Author	Year	Age/ Sex	Radiological findings	Hematological/ Microbiological Findings	Neurological deficit	Organisms	Types of Treatment	Medications	Outcome
B.Bhandari, S.L.Mandowara, Harish Joshi [6]	1981	3Y/F	1. Skiagram of skull- Osteolytic lesion in frontal bone 2. CXR- Patch of pneumonitis on the left side with hilar adenopathy	1.Hb% 7gm/dl 2.ESR – 20 mm/1st hr 3. Total leucocyte count- 10000/cumm 4. Histopathology- Tuberculus granulomatous tissue	None	*Mycobacterium Tuberculosis*	Biopsy of swelling	Anti-TB	Improved
Singh et al [12]	2005	F, 12	1.CT Scan of brain-Multiple osteolytic lesions in the skull vault and in addition showed extradural granulations in contrast enhanced CT scan 2. CXR- Normal	1.Hb% 11 gm/dl 2.ESR – 40 mm/1st hr 3. Total leucocyte count- 8400/cumm 4. Tuberculin test- Positive	None	*Mycobacterium Tuberculosis*	Excision & Biopsy of ulcers	Anti-TB	At 3 months follow up, her wounds had completely healed
Col P Singh *et al* [4]	2006	10y/ F	X-ray: Multiple osteolytic lesion in skull vault. CT: Small Extradural granulations	Tuberculin Test: Positive	None	*Mycobacterium Tuberculosis*	Excisional Biopsy of the ulcers	Anti Tubercular Drugs	Completely healing of wound

Sanjay et al [7]	2021	M, 32	Extensive osteomyelitis of occipital bone & clivus extending bilaterally upto C1/C2 level with intramuscular abscess	Gram & ZN staining of pus shows no growth.	None	*Pseudomonas Aeruginosa*	Debridement of abscess with drainage of pus	Anti Tubercular drugs for 18 months	Complete resolution of osteomyelitis
A.S.Iyer, P.V. Patil, D. Pandey et al [8]	2021	12y/ F	CT Scan-Extensive erosion in anterior and posterior clinoid process and occipital protuberances with adjacent ill defined peripherally enhancing soft tissue in clinoid region	1.CBC-Normal limits 2.ESR-22mm/1st hr 3. CSF study-Raised proteins-95 mg/dl, low sugar- 47mg/dl and complete predominance of lymphocytic cells. 4. Histopathology-Chronic granulomatous inflammation with necrosis 5. Genexpert & culture-presence of MTB	Complete hemiparesis of the right side of the body	*Mycobacterium Tuberculosis*	USG guided FNAC biopsy of the largest involved cervical lymph node	1.Anti-TB 2.Physiotherapy for neurological deficit	In a span of 4 weeks showed remarkable improvement in her symptoms
Budiarti & Setyoningrum [5]	2022	9y/ M	Skull X-ray- Multiple punch out lesion in right frontal and parietal region	1. CRP-38.19 mg/L	None	*Mycobacterium Tuberculosis*	Fine needle aspiration biopsy	Anti-TB	After 2 weeks, fairly good clinical condition

Budiarti & Setyoningrum [5]	2022	9y/M	Skull X-ray- Multiple punch out lesion in right frontal and parietal region	1. CRP-38.19 mg/L 2.Histopathology-Suppurative granuloma inflammation, positive AFB	None	*Mycobacterium Tuberculosis*	Fine needle aspiration biopsy from nodule in forehead	Anti-TB	After 2 weeks, fairly good clinical condition
Sankalp Yadav, Gautam Rawal, Jeyaraman [11]	2024	14y/M	1. Xray skull- Soft tissue swelling in left parietal region 2.Non Contrast CT of head- left high parietal, subgaleal and peripherally enhancing extradural abscess with bony destruction	1. ESR- 63mm/1st hr 2. Histopathology- Necrotizing granulomatous inflammation with a few Langhan's giant cells suggestive of tuberculosis	None	*Mycobacterium Tuberculosis*	USG guided biopsy of parietal bone lesion	Anti-TB	At 8 months of treatment, the swelling decreased and complete resolution of scalp lesion observed

Discussion:

Tuberculosis of the skull is sporadic and accounts for approximately 1% of skeletal tuberculosis. [4,5] Although this had been a known entity during the first half of the 20th century, it became a rare clinical condition after the availability of anti-TB drugs. [6] Also, it has been more common in children rather than adults. Usually, the incidence rate is 50% between 10 years of age and 75-90% in 20 years of age. [7] Painless swelling is the common presentation with or without discharging sinuses and with or without any motor deficit. [5]

The Parietal and Frontal bones are most commonly involved as relatively more cancellous bone elements compared to other bones of skull vault. The ratio is 5:1. [6] The skull base osteomyelitis is broadly classified as typical & atypical. Typical osteomyelitis occurs secondary to uncontrolled infection of the temporal bone region, most often from necrotizing otitis externa. Atypical Skull Base Osteomyelitis occurs in the absence of obvious temporal bone or external auditory canal infection involving the sphenoid and occipital bone. In developing countries like Bangladesh, most cases are related to Paranasal sinus infections, direct head injuries, and scalp infections. In developed countries, post-operative craniotomy infections are the main source of skull base osteomyelitis.

Pseudomonas aeruginosa is the most common pathogen implicated in osteomyelitis secondary to malignant otitis externa, although other organisms have been reported including *Aspergillus, Mycobacterium, and Candida*. [7,8]

Presenting symptoms of skull base osteomyelitis (headache, fever, nasal congestion/ discharge, ear pain/ discharge) are non-specific. Diagnosis is often made when the disease is advanced and neurological deficits have occurred. [8]

Identification of acid-fast bacilli of smear or biopsy specimens is the definite diagnosis for tuberculosis with histopathological findings suggesting a granulomatous lesion. Although 75% of AFB staining often shows negative results. [5]

Antibiotic treatment for skull base osteomyelitis is often required for prolonged periods as culture sensitivities. The length of time antibiotics are administered is variable.[8] However, some patients respond well to anti-tubercular therapy only.

Conclusion:

A high degree of clinical suspicion is required to diagnose atypical Tubercular Osteomyelitis of Skull, as the condition may mimic other skull disease and non-specific radiological presentations. Early diagnosis, medical management, and surgical approach are important, as delays could prove fatal.

Compliance with ethical standards

Conflict of interest: The author declares no conflict of interest.

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Peer-Review History:
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