

CASE REPORT

Tuberculosis of the skull mimicking a bony tumor

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ABSTRACT

We present a rare case of calvarial tuberculosis mimicking a solitary bone tumor, which was surgically removed. A 52-year-old female presented with a right forehead swelling, which gradually enlarged over the course of 2 years, with no symptoms or raised intracranial pressure or neurological deficits. Plain and contrast-enhanced brain computed tomography scans were done, revealing a punched-out lesion of the right frontal bone, with a nonenhancing lytic mass. With an initial diagnosis of an intraosseous meningioma, and later on intraoperatively thought to be a metastatic tumor, the mass was excised along with a rim of bone. Histopathological examination results came back as caseous necrosis, highly suggestive of tuberculosis. The patient was then treated with a 1 year regimen of anti-tuberculous medications. Tuberculosis of the cranium is a rare entity, and can mimic tumors or multiple myeloma. A high index of suspicion and knowledge is required for an early diagnosis. A combined surgical and medical therapy is curative.

Key words: Calvarial tuberculosis, cranium, mimic, skull

Introduction

Tuberculosis remains endemic and is among the biggest health problems in developing countries. Tuberculosis had the second highest incidence and the highest mortality rate among all communicable diseases in Malaysia in the year 2013 with 78.28 and 5.37 each per 100,000 population according to the health facts year 2014 by the Ministry of Health Malaysia.^[1] In Africa and developed Western Countries, the increase is mainly due to the migration of people from developing countries, and to HIV infection. The incidence of skeletal tuberculosis is rare, accounting to 1–2% of cases. Bones generally involved are the dorso-lumbar spine, skull, shoulder girdle, and hip bones. Multifocal involvement is more common than unifocal ones.^[2]

Case Report

A 52-year-old female with a background history of hypertension was referred to our center by a physician at

a private clinic for a large right frontal swelling. According to the patient, she first noticed the swelling 2 years back, and it was initially the size of a marble (1 cm). The swelling gradually increased in size. It would occasionally cause itchiness on the overlying skin, and caused some discomfort as it grew bigger, but otherwise the swelling was painless. She denied experiencing a change of color or temperature over the swelling, and she denied ulceration and/or discharge from the swelling. The patient did not have any headaches, blurring of vision, nausea, and seizure. As the swelling had never caused her any problems, she only agreed for further workup after her physician coaxed her during her follow-up for hypertension.

On examination, there was an obvious single lump involving most of the right side of the patient's forehead, measuring 8 cm × 8 cm. The skin overlying it was slightly stretched, but otherwise normal. We could not see any sinuses, ulcerations, nodules, or even sinuses. It had the same temperature as the rest of her face. The swelling had diffuse borders and was firm-to-hard to the touch. It was not compressible or indentable, and was nontender. The swelling was not attached to the skin, but it was not mobile; it was most likely attached

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to the bone. Neurological examination did not yield any abnormality.

Our initial impression was a dermoid cyst, a lipoma, or an osteoma. In view of her age, we were also considering a malignant bony tumor. The patient was admitted for further workup. Blood investigations did not show any abnormality, with normal full blood count, renal function test, and serum calcium. The erythrocyte sedimentation rate (ESR) was not raised, at 15 mm in the 1st h. Plain and contrasted computed tomography (CT) imaging was done, revealing that the swelling was a lytic bone lesion, with a punched-out appearance of the affected bone. On the outer surface of the cranium, the lesion extended beyond the bony defect and sloped off at the edges. The purely extra-axial lesion was isodense with brain tissue, and only the inner surface took up contrast, likely due to the involvement of the dura [Figure 1].

From the CT imaging, our impression was a right frontal intraosseous meningioma. We counseled the patient for surgery, with the aim for cytoreduction and histological diagnosis. We designed a question mark flap to encompass the right fronto-temporo-parietal regions, and as one of our differential diagnoses was an intraosseous meningioma, we also dissected the patient's neck in a curvilinear manner at the level of C4/C5 to expose the internal and external right carotid arteries in the event that we needed a proximal control for hemorrhage.

We had initially planned to raise the scalp as two layers, and to harvest the pericranium in the event that we needed to do a fascia duraplasty. However, the pericranium had eroded. The lesion was noted to be smooth and had a firm consistency, with a well-demarcated margin along the punched-out bone edge. We could not see any vessels feeding into the swelling. A craniotomy was made 1.5 cm away from the margin, but the dura, tightly adherent to the inner cranial surface, was circumferentially torn during bone cutting. There was not much bleeding during the craniectomy. We noted a single pedicle under the inner surface of the bone: The enlarged right middle meningeal artery (MMA). After ligation using sutures, the MMA was severed, and the lesion, along with a rim of

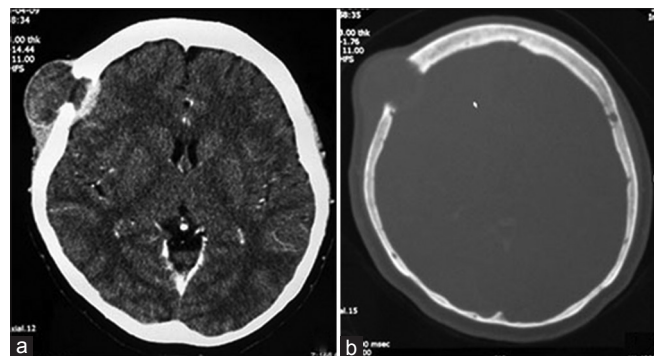


Figure 1: Axial view of the patient's computed tomography scan. (a) Contrast-enhanced computed tomography; (b) bone window

bone, was excised en bloc. The brain surface was indented, but otherwise the arachnoid layer was intact, and we did not see any abnormality on the brain surface. The dura defect was closed using an artificial tissue dura, and we proceeded with primary closure of the scalp [Figure 2].

The lesion was cut in half, and we noted that under the firm, fibrous capsule, the lesion was whitish and soft-to-firm inconsistency. In view of the scalp tissue and dura involvement, we suspected a calvarial metastatic tumor.

The patient recovered well-postoperatively. She did not have any neurological deficit, and she did not develop any seizures. We planned for a full workup for a primary source; however, the histopathological report came back as extensive areas of caseous necrosis surrounded by a collection of epithelioid histiocytes, lymphocytes, and several Langerhans-type giant cells forming granulomas. Ziehl–Nielsen stain for acid-fast bacilli was negative. The granuloma was also seen within the bone. The histopathological diagnosis was a caseating granulomatous inflammation, highly suggestive of tuberculosis.

The patient was then subjected to anti-tuberculosis treatment, planned to complete a 1 year therapy. She was also planned for a titanium mesh cranioplasty during subsequent follow-ups. The patient was well at the time of writing, and contact screening for tuberculosis was negative.

Discussion

Before the advent of chemotherapy, calvarial tuberculosis was estimated to represent 0.2–1.3% of all cases of skeletal tuberculosis.^[3] 50% of the cases reported in literature were in patients younger than 10 years of age, and 70–90% were younger than 20 years, but rarely seen among infants [Table 1].^[4]

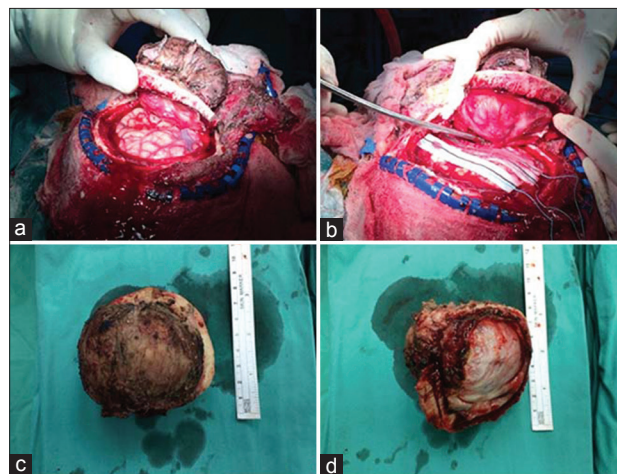


Figure 2: Intraoperative findings of the lesion. (a) The tumor was noted to have a well-demarcated fibrous outer capsule, with erosion of the pericranium, and tight dura adherence; (b) the lesion received its main blood supply from the engorged middle meningeal artery; (c) superior view of the lesion; (d) inferior view of the lesion

Table 1: Previous studies on calvarial tuberculosis

Author (year)	Study type	Number of patients
Strauss (1933)	Case report	1
Meng and Wu (1942)	Case report	1
Barton (1961)	Case series	3
Prinsloo and Kirsten (1977)	Case report	1
Gupta <i>et al.</i> (1989)	Case report	2
LeRoux <i>et al.</i> (1990)	Case report	1
Nergizoglu <i>et al.</i> (1999)	Case report	1
Sharma <i>et al.</i> (2000)	Case report	1
Raut <i>et al.</i> (2004)	Retrospective study	42
Diyora <i>et al.</i> (2009)	Retrospective study	11
Pan and Zulkarnaen (2014)	Case report	1

In a case report by an author in the neighboring state of Sarawak, the patient was 34 years of age.^[5] Our patient is much older, at 52 years of age.

Many authors hypothesized that the bony lesions is predisposed by trauma due to increased vascularity, decreased resistance, and the unmasking of a latent infection secondary to trauma. Several authors such as Meng and Wu, and Barton, had challenged the significance of trauma, as they found no patients with a history of head injury.^[6,7] There are two schools of thoughts regarding the seeding of calvarial tuberculosis: Hematogenous seeding of bacilli to the diploe without lymphatic transmission; and the other favoring the likelihood of lymphatic transmission, which explains the rarity of calvarial tuberculosis in view of poor lymphatic supply to the skull. In many cases, the primary infection lies elsewhere, with the lungs being the most common site. However, it has been reported that primary focus of infection could not be established, as in our case.^[4,5]

Concentrically placed proliferating fibroblasts encircle the tuberculous granulation tissue, preventing further extension through the diploe. Cranial sutures do not prevent the spread of granulation tissue; extensive bony destruction may occur before a swelling is seen. The most common sites of involvement are the frontal and parietal bones, followed by the occipital and sphenoid bones.

Patients presented with a painless scalp swelling, which can be multiple, and at times, a discharging sinus in the scalp. Initial presentation with seizures, neurological deficit, or other manifestations of meningitis is uncommon. Two types of lesions are generally recognized: The circumscribed/perforating type, and the progressive infiltrating type. The perforating type is more common.^[2]

Plain X-ray of the skull can show areas of rarefaction early in the disease, which later on develop into punched-out defects with a central sequestrum. Both osteolytic and osteoblastic areas are seen; sclerosis, rarely seen, indicates a secondary infection.^[8] CT

imaging helps in the assessment of the extent of bone destruction, scalp swelling, and degree of intracranial involvement.

As the tuberculosis lesions are osteolytic and can be multiple, they need to be differentiated from multiple myeloma, secondary metastasis, lymphangioma, Ewing's sarcoma, and bacterial osteomyelitis. A positive Mantoux test and raised ESR are important diagnostic clues for tuberculosis, but can be negative in 10% of patients. Demonstration of acid-fast bacilli in smear by Ziehl-Nielsen staining or the isolation of mycobacterium by culture is diagnostic but rarely seen. Histopathological findings give a definitive diagnosis.

A high index of suspicion and knowledge is required for early diagnosis of calvarial tuberculosis. Treatment for calvarial tuberculosis includes surgery and anti-tuberculous therapy. Although there are reports that favor anti-tuberculous therapy alone, studies have indicated that combined treatment is better, as extensive areas of diseased bone may become foci of tuberculous bacilli unless surgically removed.^[8,9] Surgery is indicated to establish the diagnosis, to remove thick extradural granulation tissue and necrotic bone, as well as to relieve mass effect. However, surgery is not indicated in small lesions, even in the presence of a sequestrum, provided there is a favorable response to chemotherapy.

Anti-tuberculous therapy has to be continued for 9 months to 1 year, and the response to drugs should be monitored by clinical examinations, ESR and CT scanning.^[8]

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Conflicts of interest

There are no conflicts of interest.

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