

Letter to the Editor

Empirical antitubercular therapy even in endemic area should be started with caution: Missed case of calvarial epidermoid presenting with discharging sinus

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Sir,

Spontaneous discharging sinus due to a bony lesion is not an unknown entity. When encountered in endemic areas or developing countries, tubercular osteomyelitis is found to be a leading cause. Occasionally, some bony lesions may mimic tubercular osteomyelitis.^[2] We present an unusual case of intradiploic epidermoid of sphenoid ridge that was initially misdiagnosed and treated as tuberculosis.

An 11-year-old child presented with complaint of intermittent whitish discharge from right anterior temporal area [Figure 1a] for 4 years without any history of trauma or fever. The pus culture was sterile. Cytology of discharge showed only inflammatory cells but no granuloma. Computed tomography (CT) scan showed

a lytic lesion in the sphenoid ridge [Figure 1b and c]. Patient was empirically treated with antitubercular therapy (ATT) drugs as calvarial tuberculosis was suspected clinically. The patient did not respond to ATT and the discharge persisted even after 6 weeks.

Magnetic resonance image (MRI) acquired at this time showed sharp well-defined margin and uniform diffusion restriction [Figure 2a-c] that raised suspicion of other lesion. Sinus track was excised completely through a frontotemporal flap. The track was in continuity with the well-defined bony lesion and underneath the bone extradurally [Figure 3a and b].

The initial histopathology showed inflammatory cells with some stratified squamous epithelium. On careful evaluation, keratin was seen confirming the diagnosis to be epidermoid. ATT was stopped. The child improved,

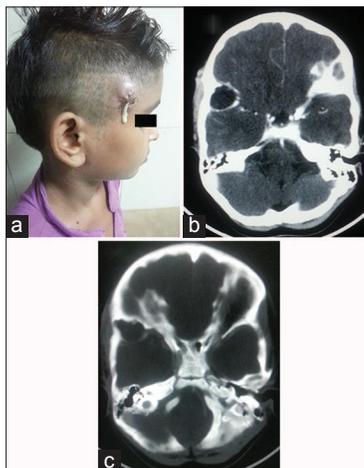


Figure 1: (a) Whitish discharge from the wound. (b) Computed tomography scan showing hypodense lesion in the right sphenoid ridge intra diploic space. (c) Bone window computed tomography shows expanded lytic lesion of the sphenoid ridge

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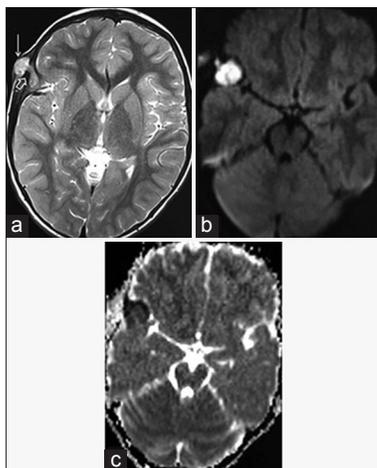


Figure 2: (a) Magnetic resonance axial T2-weighted images depict widening of the intradiploic space with hyperintense soft tissue lesion within it. Small similar signal intensity lesion is seen in the overlying subgaleal tissue, adjacent to the bony lesion which is communicating with the intradiploic lesion. (b and c) Axial diffusion-weighted images and the corresponding apparent diffusion coefficient maps demonstrate the hyperintense signal in the intradiploic and the subgaleal lesions with uniform hypointense signal on respective apparent diffusion coefficient maps, consistent with restricted diffusion within the lesion

wound healed completely [Figure 3c] and there is no recurrence at 6 months follow-up.

Spontaneous discharging sinus from the calvaria has been reported in infections such as tuberculosis, mycetomas, or posttraumatic osteomyelitis and occasionally in epidermoid.^[1-3] It is important to establish the diagnosis as management differs in each case. The diagnostic dilemma increases as all of them appear as lytic lesion on radiology with subtle differences.

Calvarial tuberculosis usually appears as a cone-shaped lytic bony lesion, wider at the inner table or diffuse hypodensity with peripheral enhancement on CT imaging. Enhancing extradural inflammatory collection may be found in some cases.^[2] In posttraumatic osteomyelitis, the involved bone is sclerotic with sequestrum. Fungal lesions such as mycetoma appear as punched out lytic lesion with intradural enhancing mass.^[1] Calvarial epidermoid commonly appears as a lytic lesion with sclerotic margin on CT scan often eroding the inner table.^[3] In our case, the diagnosis was clinched on MRI which showed uniform hyperintense signal on diffusion-weighted images and hypointense on apparent diffusion coefficient map corresponding to restricted diffusion [Figure 2b and c]. Tubercular osteomyelitis shows patchy restriction and lacks uniformity.

The modality of treatment differs in each case. In our case, the initial cytological evaluation suggested infection

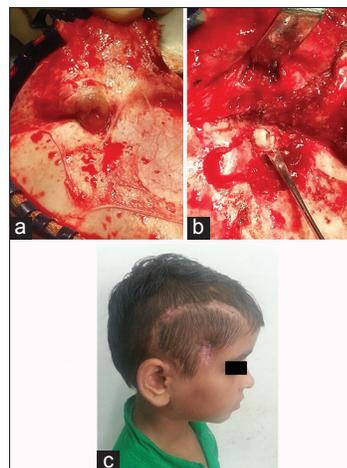


Figure 3: (a) Intraoperative image showing subgaleal sinus track and whitish caseous material seen inside the lesion (b). (c) Completely healed lesion at 6 month follow-up

and empirical ATT was started. As the lesion did not respond to ATT, radiology was reevaluated including MRI and complete surgical excision was planned. After thorough discussion with the pathologist, a careful evaluation of the slides showed stratified squamous epithelium with keratin.

Therefore, discharging sinus from the calvaria may not necessarily be tubercular even in an endemic area and should be properly evaluated. Cautious evaluation of radiology and histopathology helps establish the correct diagnosis, else the patient may receive unnecessary ATT.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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Commentary

In general, I do not believe in the Western World this article would be contributing much to neurosurgery. In fact, I was considering to reject it. However, the article underscores in some regions of the world universal standards and requirements as to assessing a given medical problem may not commonly exist. To establish the exact diagnosis is critical in patient care. Hence, failure to obtain appropriate data can render the incorrect treatment. It is not just standard as to a neurosurgical approach to obtain bacterial, fungal, and mycobacterium microbiological data (if clinically indicated), but securing instructive imaging studies (computed tomography scans or magnetic resonance image) is mandatory. I was struck by the author(s) conclusion that imaging and histopathology as being critical. It would seem obvious that it is essential. This is very informative that in different regions of the world a more relaxed approach may exist. I suspect this may occur secondary to immense shortages as to medical facilities, overextended physicians and the lack or paucity of nursing care, not to mention limited educational advantages, financial and region pragmatic issues. In some regions of the world, this article to me endorses “modern medicine” is not part of the algorithm of assessment. Doctors seemingly because of circumstances conclude diagnoses based on endemic probability. I suspect the evidence-based approach may be dangerously unrealistically and impractical for some populations.

The imaging studies demonstrated an expanding sphenoid greater wing lesion that was with relatively defined margins, not with erosive and irregular boundaries. Albeit, typical or atypical mycobacterium can be variable and a “great imitators” of pathological processes. However, the images did not support an infectious process with high likelihood. The authors were correct, ultimately, in their efforts concluded the lesion was not infectious.

As a reviewer, I was more impressed that in today's medicine a classical approach was not employed. That is to conduct reasonable imaging correlations with the clinical findings and then confirming with histopathological studies. This profoundly aroused my own appreciation of the disparity of the most basic 20th century, not 21st century, expected and required approaches to this given situation. Why should there be such regional variations of the practice of medicine? The approach and investigative tools selected by the author(s) were correct and should be acknowledged as such. This article in a tangent but in a real way exemplifies a socioeconomical-educational medical conundrum that must be solved.

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