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CASE REPORT

Calvarial Tuberculosis in a Preschool-Aged Child

An uncommon entity

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سل قبي في طفل ما قبل المدرسة

ظاهرة غير شائعة

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ABSTRACT: Calvarial tuberculosis is a rare manifestation of tuberculosis, especially in children under five years of age. We report a two-and-a-half-year-old male child who presented to the Nilratan Sircar Medical College, Kolkata, India, in 2015 with frontoparietal fluctuant swelling of three months' duration. He had also had chronic sinus discharge from the left lower eyelid over the previous six months. Computed tomography of the head revealed a frontal swelling along with erosion of both the outer and inner plates of the left frontal bone. Fine needle aspiration of the pus indicated the presence of acid-fast *bacilli*. Unfortunately, no primary focus of tuberculosis could be established. The patient improved after one year of antitubercular therapy without requiring any surgical intervention and with no sign of subsequent disease recurrence.

Keywords: Tuberculosis; Skull; Preschool Child; Antitubercular Agents; Case Report; India.

الملخص: السل القبي هو ظاهرة نادرة للسل، وبالأخص في الأطفال تحت عمر الخمس سنوات. نعرض هنا حالة طفل ذكر عمره سنتان ونصف تم عرضه لكلية نلراتان سركار الطبية، كالكتا، الهند، عام 2015 وكان مصاب بورم جبهي جداري متموج لمدة ثلاثة أشهر. وكان أيضا مصاب بخراج جيب مزمن من الجهة اليسرى للجفن السفلي خلال الستة أشهر الماضية. الأشعة المقطعية للرأس أظهرت وجود ورم جبهي يرافقه تآكل في اللوح الخارجي والداخلي من جهة اليسار للعظم الجبهي. الأبرة الشافطة الدقيقة للخراج أظهرت وجود عصيات صادمة للحمض. لسوء الحظ لم يتم التعرف على البؤرة الأولى للسل. تحسن المريض بعد مرور سنة على أستخدام علاج مضادات السل بدون الحاجة للتدخل الجراحي مع عدم وجود ظواهر لعودة المرض.

الكلمات المفتاحية: السل؛ الجمجمة؛ طفل ما قبل المدرسة؛ مضادات السل؛ تقرير حالة؛ الهند.

Calvarial tuberculosis (TB) is a rare form of extrapulmonary TB arising in the calvarial bones of the skull; despite the high prevalence of TB in India, this form of TB is encountered infrequently.[1] Nevertheless, the incidence of calvarial TB is on the rise due to malnutrition, poor socioeconomic conditions and an increase in the prevalence of HIV/AIDS and other immunodeficiency conditions.[2] This report describes a rare case of calvarial TB occurring in a preschool-aged child who presented with a left frontoparietal swelling.

Case Report

A two-and-a-half-year-old male child presented to the neurology outpatient department of the Nilratan Sircar Medical College in Kolkata, India, in 2015 with a gradually progressive yet painless swelling on the left side of the head of three months' duration associated with a low-grade fever. There was no history of severe head trauma, coughing, loss of appetite or failure to thrive. Six months previously, the child had experienced trivial trauma to the lateral aspect of the left eye and head; this was followed by swelling of the lateral part of the lower left eyelid. An ophthalmologist had diagnosed this as an abscess and incision and drainage of the area was performed. However, intermittent serous discharge from the eyelid continued to occur despite repeated courses of antibiotics. The child's father had a history of TB lymphadenitis, for which he had completed six months of directly observed treatment short course (DOTS) antitubercular therapy (ATT) one year previously. All of the child's immunisations were up-to-date, including the *bacillus* Calmette-Guerin (BCG) vaccine which he had received when he was three days old.

On examination, the child appeared healthy, well-nourished and weighed 13 kg. His vital parameters were all within normal limits. A fluctuant non-tender swelling of 4 x 3 cm in size was noted over the left frontoparietal area without any evidence of overlying inflammation [Figure 1]. In terms of visual acuity, the patient had a 6/6 Snellen equivalent fraction as measured using a crowded acuity test (Kay Pictures

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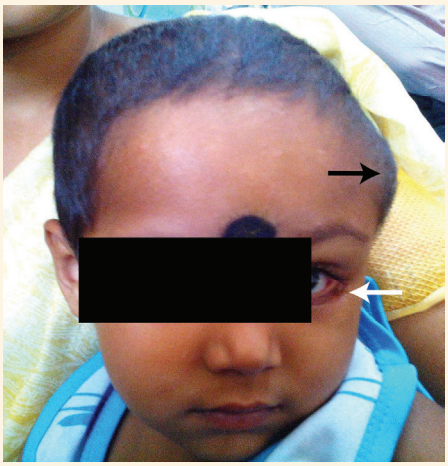

Figure 1: Photograph of a two-and-a-half-year-old male child with a swelling in the left frontoparietal area (black arrow). A small discharging sinus can be seen in the lateral part of the left lower *palpebra* (white arrow).

Ltd., Tring, Hertfordshire, UK). An ocular examination was normal, except for the small discharging sinus present over the lateral part of the lower left *palpebra*. A characteristic BCG scar was present on the lateral aspect of the patient's left arm and a single small non-tender left submandibular lymph node was palpable. The rest of the physical examination was essentially normal. A complete blood count revealed mild anaemia with a haemoglobin level of 10.6 g/dL. The total leucocyte count was 11,000/mm^3, with 48% neutrophils and 46% lymphocytes. The results of renal function, liver function and serum electrolyte tests were all within normal ranges.

Subsequently, a non-contrast computed tomography (CT) scan of the brain showed a well-defined hypodense lesion of 3 x 2.5 cm over the left frontal region of the scalp, along with erosion of the underlying left frontal bone [Figure 2]. Fine needle aspiration (FNA) of the scalp swelling yielded purulent material and a Ziehl-Neelsen stain indicated the presence of acid-fast

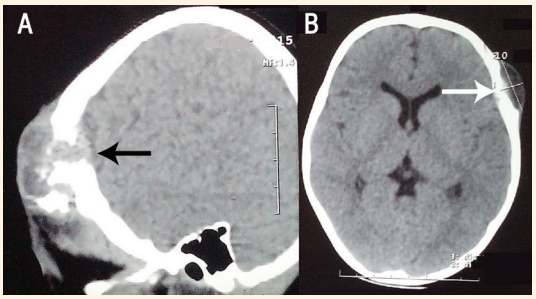

Figure 2: Computed tomography scans of the brain of a two-and-a-half-year-old male child from the (**A**) reconstructed sagittal and (**B**) axial views showing a hypodense lesion in the left frontal region of the scalp (white arrow). Note the erosion of the underlying left frontal bone (black arrow).

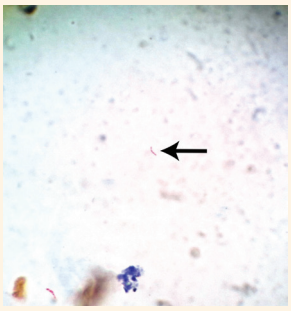

Figure 3: Ziehl-Neelsen stain at x1,000 magnification confirming the presence of acid-fast *bacilli* (arrow) against a caseonecrotic background.

bacilli (AFB) [Figure 3]. The FNA cytology panel also indicated the presence of an epithelioid granuloma and Langhans giant cells on a caseonecrotic background. A cartridge-based nucleic acid amplification test also confirmed the presence of a rifampicin-sensitive *Mycobacterium tuberculosis* complex. However, both a chest X-ray and ultrasonography of the abdomen did not reveal any abnormalities. Due to its small size, FNA cytology of the left submandibular lymph node was not attempted. An AFB smear of the serous discharge from the left lower eyelid sinus was negative. However, a Mantoux test with two tuberculin units was positive with an induration of 14 mm.

A final diagnosis of microbiologically-confirmed calvarial TB was made, although a primary focus could not be determined. Category I DOTS ATT was initiated as per the Revised National Tuberculosis Control Programme (RNTCP) guidelines.[3] This consisted of a two-month intensive phase of treatment with the thrice weekly administration of 150 mg of rifampicin, 150 mg of isoniazid, 400 mg of ethambutol and 500 mg of pyrazinamide, followed by a 10-month continuation phase involving the thrice weekly administration of 150 mg of rifampicin and 150 mg of isoniazid. While the patient was receiving ATT, the scalp swelling and left submandibular lymphadenopathy gradually reduced in size and the lower left eyelid lesion healed completely. No surgical intervention was required. At the time of writing, the patient had been under observation for one year following the completion of ATT, with no sign of disease recurrence.

Discussion

Among all active TB cases, approximately 1–2% have involvement of the skeletal system of which only 0.2–1.3% constitute calvarial TB.[1] Due to its rarity, a high degree of clinical suspicion is essential to enable early identification of this entity as it can cause extensive bony destruction and significant neurological impairment if left untreated.[4] In particular, younger

individuals are at higher risk of developing TB of the skull; Raut *et al*. reported 42 cases of calvarial TB over a 10-year period, with an age range of 5–48 years old and a mean age of 16 years.[5] In another study, the mean age at presentation was 15.09 years among 11 patients with calvarial TB.[1] However, in the current case, the patient presented at a much younger age. Both previous studies also documented a male predominance.[1,5] Scalp swelling is the most common presenting feature of calvarial TB, followed by sinus discharge, localised pain, seizures and meningitis.[1] The duration of such symptoms is usually six months or less, as noted in the current case. A similar case was reported from Mumbai, India, describing a 10-year-old boy who presented with a left frontal swelling of two months' duration.[6]

Although ocular TB is quite common in endemic countries like India, TB of the eyelid is another extremely rare condition that often presents in the form of chronic sinus discharge.[7,8] In the present case, the patient had a non-healing discharging sinus over the lower left *palpebra* and, although the exact origin of the TB could not be determined, the site healed completely during the course of the ATT. Trauma has been mentioned as a predisposing factor to skeletal TB among 42.8% of patients.[5] Following injury, increased vascularity, decreased resistance and the discovery of a latent infection might be contributing factors, although this hypothesis has been challenged by other researchers.[9,10] Among common sites of skull TB, involvement of the parietal or frontal bones is most frequent, as these both have greater diploic space and more cancellous bone.[1,5] It is believed that skull TB occurs as a consequence of the haematogenous seeding of *bacilli* to the diploë from a primary TB infection elsewhere in the body.[5] However, it was impossible to establish a primary TB focus in the current patient.

Often, the microbiological confirmation of skull TB is not possible due to necrosis.[1] In a previous case series, only one out of six patients with calvarial TB was found to have an AFB-positive stain.[11] An appropriate clinicoradiological presentation combined with histopathological evidence of a caseating granuloma is often sufficient for diagnosing TB, while a positive Mantoux test or raised erythrocyte sedimentation rate is only an ancillary aid.[1,5] A CT scan usually demonstrates soft tissue swelling with the accompanying destruction of one or both of the inner and outer tables of the *calvaria* while highly specific magnetic resonance imaging characteristics often allow for a conclusive diagnosis.[5] Under the RNTCP protocol, a 12-month ATT course is recommended for cases of central nervous system TB, consisting of a two-month intensive phase followed by a 10-month continuation phase.[3] However, ATT may be administered for up to 24 months depending on the clinicoradiological response. Surgical intervention may be required, especially in cases with large extradural collections causing neurodeficits or large scalp swellings with sinus formation, with some evidence to suggest that combined surgery and ATT may yield better outcomes than ATT alone.[12]

Conclusion

Calvarial TB is an extremely rare form of skeletal TB which needs to be diagnosed early. Among young patients, TB of the skull should be considered as a differential diagnosis, particularly among those presenting with a frontoparietal swelling within the first two decades of life, even if there is no evidence of active TB in other parts of the body. With or without surgical intervention, ATT remains the cornerstone of therapy for this condition.

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