Case Reports

Tuberculous osteitis of the skull in a child

Mohammed Benzagmout, MD, Saïd Boujraf, MD, PhD, Khalid Chakour, MD, Mohammed E. Chaoui, MD.

ABSTRACT

على الرغم من أن داء السل ما زال مرضا وبائيا منتشرا في الدول النامية ، فإن إصابة جمجمة الرأس تعتبر حالة شديدة الندرة. في هذا البحث ، نتحدث عن حالة طفل في العاشرة من عمره أدخل قسم جراحة الدماغ والأعصاب بسبب تورم العظم الجداري الأيسر لجمجمة الرأس . أظهر الكشف بالأشعة المقطعية تجويفا بالعظم الجداري الأيسر للجمجمة، مرتبطا بانتفاخ خارج الجافية .وبعد خضوع المريض لعملية جراحية، أظهر التشريح الدقيق التهابا حبيبيا سل درنيا نموذجيا. من خلال هذه الملاحظة، نناقش المظاهر السريرية وطرق تدبير علاج هذه الحالة النادرة لداء السل.

Tuberculosis is endemic in developing countries. However, skull tuberculosis is uncommon with few cases reported in the literature. We report a 10-year-old boy admitted for a left parietal painless swelling. A CT scan demonstrated a left parietal bony defect, destroying both inner and outer tables. This was associated with an enhanced epidural collection and scalp swelling. The patient was operated, and the microscopic examination revealed typical tuberculosis granuloma. The clinical presentation and management of this rare location of tuberculosis are discussed.

Neurosciences 2008; Vol. 13 (1): 70-72

From the Departments of Neurosurgery (Benzagmout, Chakour, Chaoui), and Biophysics and Clinical MRI Methods (Boujraf), Faculty of Medicine and Pharmacy, University of Fez, Morocco.

Received 1st April 2007. Accepted 9th June 2007.

Address correspondence and reprint request to: Prof. Saïd Boujraf, Department of Biophysics and Clinical MRI Methods, Faculty of Medicine and Pharmacy, University of Fez, BP. 1893, Km 2.200, Sidi Hrazem Road, Fez 30000, Morocco. Tel. +212 (67) 780442. Fax. +212 (35) 619321. E-mail: sboujraf@hotmail.com

Ithough rare, the incidence of tuberculosis of Λ the skull is on the rise in developing countries because of poor socioeconomic conditions and immunodeficiency syndromes.1 It usually presents as a painless scalp swelling, and most commonly involves the frontal and parietal bones. Therefore,

the goal of this paper is to report a new case of calvarial tuberculosis and to discuss its possible pathogenesis, clinical, radiological, and therapeutical features.

Case Report. A 10-year-old boy was hospitalized at the neurosurgery department for a scalp swelling, which appeared 4 days after a benign cranial trauma. There were no associated symptoms, especially signs of intracranial hypertension. The skull clinical examination revealed a painless left parietal fluctuant mass, of 3 x 4 cm in dimension. It had a regular edge, and was mobile according to the underlying bone, without modifying the overlying scalp. The patient was afebrile, and there were no palpable lymph nodes in the head and neck region. General and neurological examination did not show any abnormalities. Conventional radiographs of the skull showed a left parietal circumscribed lytic lesion (Figure 1). The complementary CT scan demonstrated a bony defect associated with an adjacent enhanced epidural collection (Figure 2). Biological assessment revealed an increasing erythrocyte sedimentation rate (90 mm at the first hour) and negative tuberculin reaction. Radiography of the chest was normal. The lesion was directly approached through an arciform incision; this allowed discovery of purulent material in the subgaleal space, bony sequestrum, and epidural collection with granulation tissue. A large craniectomy was performed until the healthy bone was seen, and the epidural collection was removed. The dura was infiltrated without being ruptured, therefore, it was not opened during surgery. Regarding the acid-fast bacilli and the bacterial infection, the cultures were negative. The histological examination was suggestive for caseous follicular tuberculosis. Postoperative antibacillar chemotherapy was then started; it consisted of rifampicin (10 mg/kg/day), isoniazid (5 mg/kg/day), streptomycin (15 mg/kg/day), and pyrazinamide (30 mg/kg/day). This was associated with corticosteroid therapy at a rate of 0.5 mg/ kg/day for 3 weeks. Streptomycin and pyrazinamide were withdrawn after 2 months of therapy while the remaining antituberculous medications were continued for 9 months. A post-treatment CT scan was performed and revealed an obvious decrease of the epidural collection size, which was well correlated with the clinical improvement (Figure 3). He remains very well with no recurrence of the infection after 2 years follow up.



Figure 1 - Lateral skull x-ray showing circumscribed lytic lesion.



Figure 3 - Post-treatment computerized tomography scan at one month showing complete regression of the epidural collection with right parietal bone defect.



Figure 2 - Computerized tomography scan in a) parenchymatous and b) bone windows showing bone erosion associated with soft tissue and epidural collections.

Discussion. Tuberculosis of skull is very rare, representing 0.2-1.3% of all cases of skeletal tuberculosis.² Since the first description by Reid et al in 1842,3 numerous cases reports and short series of calvarial tuberculosis have been reported in the literature.^{1,4,5} The disease is mostly seen in infants.⁶ Approximately 50% of the cases reported in the literature were in patients younger than 10 years, and 70-90% were younger than 20 years.^{2,7} The pathogenesis of this pathology remains unclear. Many authors have hypothesized that trauma is a predisposing factor in the formation of bony lesions.² Increased vascularity, decreased resistance, and unmasking of a latent infection secondary to trauma are thought to be the predisposing factors.¹ In addition, inflammatory cells are attracted to the site of trauma and act as vectors.8 Moreover, apparently calvarial tuberculosis occurs by hematogenous spread of bacilli from a primary active or latent focus, which is typically located in the lungs. However, lymphatic dissemination of tuberculosis, common in other bones, is not thought to occur in the skull.9 The common sites of involvement are frontal and parietal, followed by the occipital, and sphenoid bones. This is probably accounted for by a greater amount of cancellous bone with diploeic channels at these sites.¹⁰ Generally, concentrically placed proliferating fibroblasts encircle the tuberculous granulation tissue and prevent its further extension through the diploe.⁹ If the process is not contained, further extension can occur through the inner or outer table of the skull. Cranial sutures do not prevent the spread of granulation tissue, and hence extensive destruction can occur before a sinus or swelling manifests.¹¹ Clinically, skull tuberculosis may presents as a painless subgaleal swelling (Pott's

puffy tumor)12 with a discharging sinus when the outer table is involved. Involvement of the inner table is associated with formation of underlying extradural granulation tissue. Initial presentation with seizures,² motor deficit, or other manifestations of meningitis⁸ is uncommon. The erythrocyte sedimentation rate was often raised, and the tuberculin test was found positive in most studies reported so far.¹³ Depending on the nature of calvarial destruction, 3 types of the lesions of tuberculosis osteitis are described at conventional radiography: "circumscribed lytic," "diffuse spreading," and "circumscribed sclerotic" lesions. They depend on the virulence of the organism and the immune response of the host. The presence of sclerosis is thought to represent secondary infection.¹⁴ These conventional radiographic features are inconclusive. Nevertheless, CT scan demonstrates soft tissue swelling with accompanying destruction of one or both skull tables. A bony sequestrum may also be seen. It also shows spread of the disease process to the extradural space, the surrounding meninges, and brain parenchyma. The skull tuberculosis in children may mimic other diseases such as histiocytosis and neuroblastoma.¹⁵ Therefore, the microbiologic or histological confirmation is essential before starting chemotherapy. Treatment involves surgical excision and antituberculous chemotherapy. It is mandatory to excise all diseased bones and the sinus tract, however, total excision of granulation tissue is not necessary. Indeed, surgery is performed in cases with large extradural collections causing neurologic deficits or large scalp swellings with sinus formation leading to fulminant secondary infections.8 Surgery would also be indicated if there is doubt about the diagnosis.⁵ Finally, cranioplasty is not recommended at an immediate stage because ossification of bony defects could occur spontaneously in children.⁴

In conclusion, tuberculosis continues to be among the greatest health problems in developing countries and has an enormous social and economic impact. The involvement of the skull is highly exceptional with very good prognosis after treatment.

References

- 1. Raut AA, Nagar AM, Muzumdar D, Chawla AJ, Narlawar RS, Fattepurkar S, et al. Imaging Features of Calvarial Tuberculosis: a Study of 42 Cases. *AJNR Am J Neuroradiol* 2004; 25: 409-414.
- 2. Strauss DC. Tuberculosis of the flat bones of the vault of skull. *Surg Gynecol Obstet* 1933; 57: 384-398.
- 3. Reid E. Medizinisches Correspondenzblatt Bayerischer Arzte. Erlangen: 1842. p. 33.
- Gazzaz M, Bouyaakoub FA, Chaoui FM, EL Khamlichi A. Tuberculosis osteitis of the skull. *Pan Arab Journal of Neurosurgery* 1998; 2: 43-45.
- Tyagi AK, Kirollos RW, Kang NV. Tuberculous osteitis of the skull: a case report and review of the literature. *Br J Neurosurg* 1996; 10: 339-401.
- 6. Meng CM, Wu YK. Tuberculosis of the flat bones of the vault of the skull. *J Bone Joint Surg* 1942; 24: 341-353.
- Mohanty S, Rao CJ, Mukherjee KC. Tuberculosis of the skull. *Int Surg* 1981; 66: 81-83.
- Jadhav RN, Palande DA. Calvarial tuberculosis. *Neurosurgery* 1999; 45: 1345-1349.
- 9. Davidson PT, Horowitz I. Skeletal tuberculosis. A review with patient presentations and discussion. *Am J Med* 1970; 48: 77-84.
- Patankar T, Varma R, Krishnan A, Prasad S, Desai K, Castillo M. Radiographic findings in tuberculosis of the calvarium. *Neuroradiology* 2000; 42: 518-521.
- Gupta PK, Kolluri VR, Chandramouli BA, Venkataramana NK, Das BS. Calvarial tuberculosis: a report of 2 cases. *Neurosurgery* 1989; 25: 830-833.
- 12. Brown TS, Franklyn PP, Marikkar MS. Tuberculosis of the skull vault. *Clin Radiol* 1980; 31: 313-315.
- 13. Bhandari B, Mandowara SL, Joshi H. Tubercular osteomyelitis of the skull. *Indian J Paediatr* 1981; 48: 113-115.
- Mukherjee KK, Kaushik R, Nada R, Khosla VK, Khandelwal N, Kak VK. Calvarial tuberculosis. *Surg Neurol* 2002; 57: 195-202.
- 15. Wessels G, Hesseling PB, Beyers N. Skeletal tuberculosis: dactylitis and involvement of the skull. *Pediatr Radiol* 1998; 28: 234-236.

COPYRIGHT

Whenever a manuscript contains material (tables, figures, etc.) which is protected by copyright (previously published), it is the obligation of the author to obtain written permission from the holder of the copyright (usually the publisher) to reproduce the material in Neurosciences. This also applies if the material is the authors own work. Please submit copies of the material from the source in which it was first published.