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NEUROLOGICAL PICTURE

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Torcular Erdheim–Chester disease

Erdheim–Chester disease (ECD) is a rare non-Langerhans histiocytosis associated with multiple organ systems. Typical features include osteosclerotic lesions in the metaphysis of long tubular bones, exophthalmia, pulmonary and retroperitoneal fibrosis,¹ which highlight the diagnostic difficulties that ECD presents. Intracranial association is exceptional and its treatment is controversial.²

We present the case of a 52-year-old man with a history of progressive visual deficit, pseudotumour cerebri and diabetes insipidus, who presented with a tumour in the torcular region; the lesion included the falx, transverse sinuses and straight and superior sagittal sinuses and had the radiological features of a meningioma (fig 1). Angiography of the cerebrum showed thrombosis of both transverse sinuses, with a narrowed anterior superior sagittal sinus but no hypervascular tumour stain. The lesion infiltrated the posterior falx and tentorium with a nodule indenting, but not infiltrating, the left cerebellar hemisphere. After subtotal resection of the mass, histological examination showed numerous lipid-laden histiocytes and scattered multinuclear Touton-type giant cells (fig 2) and rare eosinophils. The diagnosis was a xanthogranuloma of the Erdheim–Chester type. Further imaging showed multiple lesions in both femurs, pleural and retroperitoneal fibrosis with the kidneys being affected. The patient had a progressive worsening of respiratory and renal functions and died 6 months after diagnosis.

Most reported cases of intracranial ECD are dural granulomas affecting the brain stem, cerebellum and vertebral and basilar arteries.^{1–5} Diabetes insipidus and cerebellar signs are the most common neurological symptoms.^{3–4} A characteristic feature on MRI is the prolonged retention, even after several weeks, of gadolinium; this may be because of the abnormal content of histiocytes in the lesions.⁵ In this unusual case, ECD was diagnosed after resection of the intracranial mass. The progression of neurological symptoms further distinguishes this case from previous reports.

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Figure 1 Brain MRI axial T1-weighted postcontrast image shows abnormal homogeneous infrasupratentorial tumour at the level of the torcula.

Figure 2 (A) Mixed, chronic inflammatory infiltrate of lymphocytes and foamy histiocytes; some of the histiocytes are multinucleated giant cells, original magnification $\times 250$. (B) High-power view of (A). Touton-like multinucleated giant cell is readily identified, original magnification $\times 400$.

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