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Case report

Apropos of an unusual and menacing presentation of neurosarcoidosis: The space-occupying trapped temporal horn

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ABSTRACT

The trapped ventricular horn syndrome is characterized by the exclusion of a compartment of the ventricular tract from the rest of the CSF pathways. This syndrome has been reported in a variety of diseases including meningitis, intraventricular haemorrhages and tumours. Only two previous cases of neurosarcoidosis-related trapped temporal horn syndromes have been reported. The authors add a third case of a trapped temporal horn revealing neurosarcoidosis, presenting as a space-occupying process requiring urgent temporal tip lobectomy.

1. Introduction

Sarcoidosis is a systemic granulomatosis of unknown aetiology originally described by Hutchinson in 1878 [1]. Whereas lungs, lymph nodes and skin are frequently involved, the central nervous system (CNS) invasion remains rare and subdiagnosed [1,2]. When isolated, this latter form of the disease known as neurosarcoidosis (NS) is very problematic to diagnose. Many neurological presentations may reveal neurosarcoidosis, including cranial nerve palsies, parenchymal granulomas, leptomeningitis, vasculitis, endocrinological symptoms and hydrocephalus [3]. A particular form of compartmentalized hydrocephalus consisting in an exclusion of a ventricular horn from the rest of the CSF pathways exceptionally reveals NS. To the best of our knowledge, only two cases of trapped temporal horn revealing NS were previously published [4,5]. The authors present such a case in a woman with a looming presentation, requiring urgent temporal tip lobectomy permitting surgical decompression and histological diagnosis.

2. Case report

A 42-year-old woman, without any medical relevant history, was admitted in our emergency department after two brief generalized convulsive seizures and intense cephalalgia beginning 16 h before her arrival. The post-ictal anamnesis revealed a 3-month history of headaches and vomiting, but also recent memory disturbances concerning the short-term memory. At examination, she was somnolent, with a left hemiparesis and a slight right pupillary dilatation. There was evidence of bilateral papilloedema. The rest of the somatic examination including lung, lymph nodes and skin was normal. The routine laboratory tests were within normal range. The chest X-ray was also standard. Cerebral MRI showed a cystic space-occupying process within the right temporal fossa with herniation signs (Figs. 1–3). The cyst's magnetic patterns were those of normal CSF with evidence of transependymal resorption, corresponding to a right temporal trapped horn. In addition, MRI depicted several signal anomalies and granular gadolinium enhancements involving essentially the left temporomesial lobe, the brainstem, the infundibulum and the optic chiasm. Spinal MRI was normal. Cerebral granulomatosis secondary to tuberculosis and sarcoidosis were discussed. An endoscopy-assisted internal ventriculo-cisternal shunt with biopsy was considered, but the unavailability of the endoscopic device for maintenance reasons at the moment of this case led us to propose an urgent open surgical procedure. This latter consisted in a right temporal tip lobectomy through a pterional approach. This lobectomy opened largely the right temporal trapped horn

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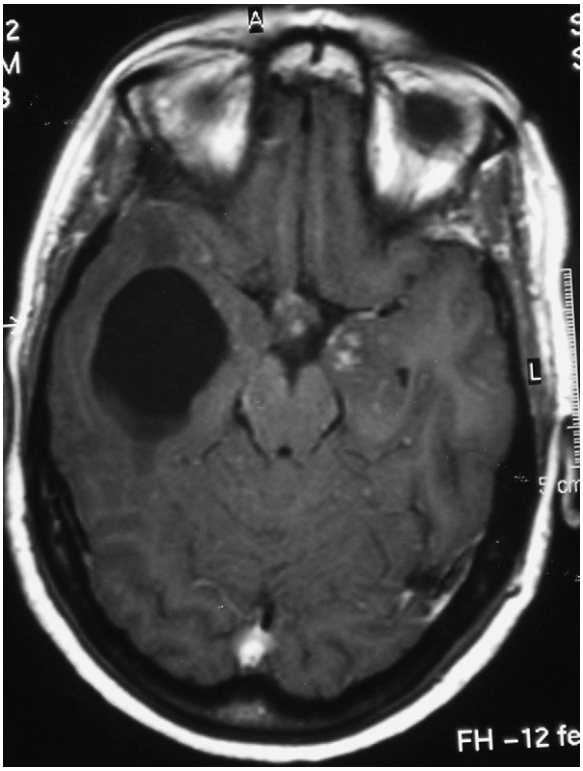

Fig. 1. Axial MRI (T1 sequence with gadolinium). Cystic space-occupying process within the right temporal lobe corresponding to a trapped temporal horn. Note the granular enhancements in the left temporal lobe and the optic chiasm.

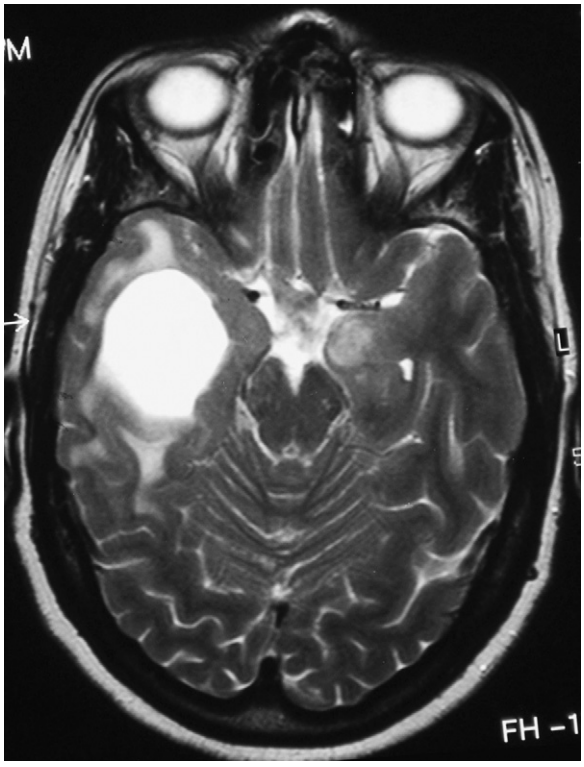

Fig. 3. Axial MRI (T2 sequence). Peri-cystic hyperintensity corresponding to transependymal resorption. Note the brainstem and left hippocampus signal anomalies. Right temporal herniation.

and permitted a wide communication with the Bichat's fissure. A thickened arachnoiditis involving the optic nerves and the right carotid artery was noted, but no evident granuloma was encountered. The post-operative course was uneventful and the neurological examination normalized rapidly. The neuropsychological tests were normal. An angiotensin-converting enzyme level in the peroperative sampled CSF was normal. The pathological examination of the temporal lobectomy piece found numerous non-caseating epithelo-gigantocellular granulomas, confirming the suspected diagnosis of neurosarcoidosis (Fig. 4). Oral prednisone (1 mg/(kg day)) was given during 8 weeks associated with phenytoin (300 mg/day), with a disappearance of the cerebral lesions in an asymptomatic and seizure-free patient after a 2 years follow-up without relapses or systemic signs.

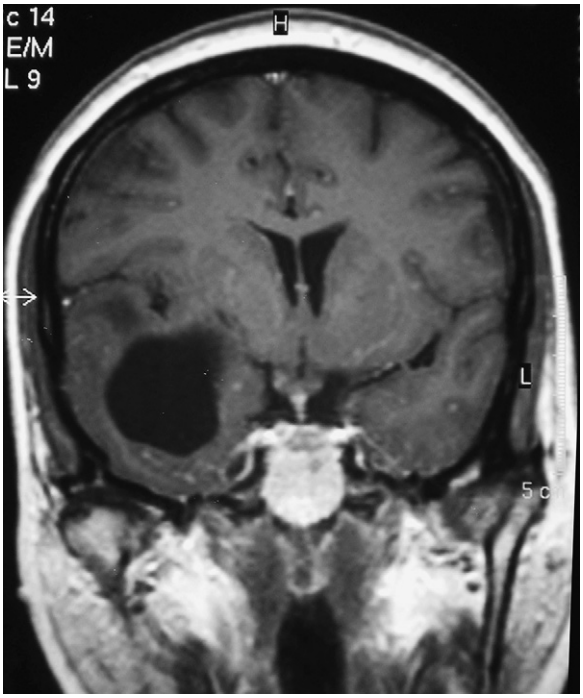

Fig. 2. Coronal MRI (T1 sequence with gadolinium). Pituitary stalk involvement.

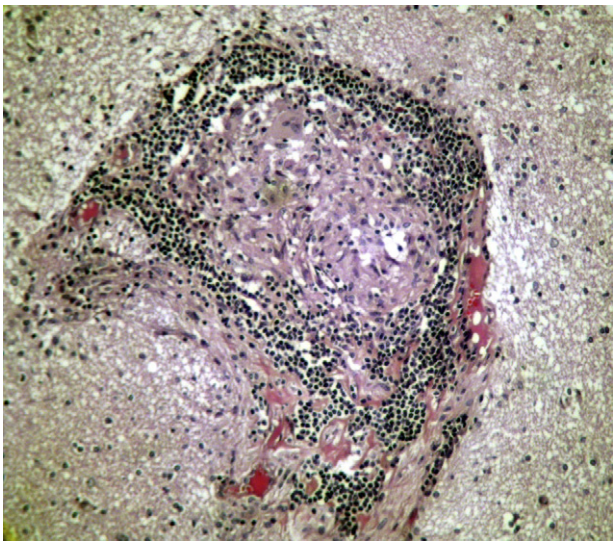

Fig. 4. Right temporal tip lobectomy piece. High-power view of small non-caseating giganto-cellular granuloma typical for sarcoidosis. No micro-organisms were demonstrated (hematoxylin and eosin, 40× original magnification).