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Suprasellar cistern tuberculoma presenting as unilateral ocular motility disorder and ptosis: A case report and literature review

Bi-Bo Zhao *et al*. Suprasellar cistern tuberculoma

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Abstract

BACKGROUND

Intracranial tuberculoma is a rare and serious type of tuberculosis, which mostly occurs in the frontoparietal and cerebellar hemispheres, with a predominance of the gray-white matter junction area, while tuberculomas only in the cistern are extremely rare with only few reported cases in the literature. We describe a unique case of isolated tuberculoma in the suprasellar cistern, with only right ocular motility disorder and upper eyelid ptosis.

CASE SUMMARY

A 5-year-old boy without any medical history presented with right ocular motility disorder and upper eyelid ptosis one month ago. He had no history of fever, headache, vomiting, convulsions, and limb weakness. Neurological examination showed the right third cranial nerve palsy with restrictions of eye movements and ptosis, pupil dilation and negative light reflex. Imaging suggested a space-occupying lesion in the suprasellar cistern with calcification and ring-enhancement. Moreover, no Mycobacterium tuberculosis was found in cerebrospinal fluid by PCR. The lesion was initially diagnosed as a tumor, while postoperative pathology combined with PCR indicated tuberculoma. The patient continued postoperative anti-tuberculosis treatment. At present, the patient's condition is stable and the symptoms are partially relieved compared with those before operation.

CONCLUSION

This case confirmed that isolated intracranial tuberculoma can occur in the suprasellar cistern. Therefore, for space-occupying lesions in the suprasellar cistern, tuberculoma should be included in the differential diagnosis even if there is no history and indication of tuberculosis infection.

Key Words: Tuberculosis; Intracranial tuberculoma; Suprasellar cistern; Ocular motility disorder; Ptosis; Case report

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Core Tip: Intracranial tuberculoma is a rare and serious type of tuberculosis, which mostly occurs in the frontoparietal and cerebellar hemispheres, with a predominance of the gray-white matter junction area, while tuberculomas only in the cistern are extremely rare. Isolated intracranial tuberculoma in the suprasellar cistern mimicking a tumor with only ocular symptoms has not been reported before. This case confirmed that isolated intracranial tuberculoma can occur in the suprasellar cistern. Therefore, for space-occupying lesions in the suprasellar cistern, tuberculoma should be included in the differential diagnosis even if there is no history and indication of tuberculosis infection.

INTRODUCTION

Tuberculosis remains one of the most serious infectious diseases in the world currently, with Mycobacterium tuberculosis (MTB) infecting more than 2 billion people worldwide, and 10% of whom develop active tuberculosis[1]. Tuberculosis is a significant cause of morbidity and mortality globally, especially in developing countries, and has tended to increase in recent years with the increase in HIV infection and the emergence of multi-drug resistant MTB. Tuberculosis can involve all parts of the body, predominantly the lungs, and tuberculosis in central nervous system is the rarest but most serious type, accounting for 5-10% of extrapulmonary tuberculosis and about 1% of all tuberculosis[1].

Central nervous system tuberculosis mainly includes tuberculous meningitis, tuberculoma, tuberculous abscess and spinal cord tuberculosis[2]. Intracranial

tuberculoma is a granulomatous lesion formed by the hematogenous spread of MTB. Lacking specificity in the clinical presentation as well as imaging features of intracranial tuberculoma makes its diagnosis difficult in patients without the evidence of MTB infection or the history of extracranial tuberculosis. Intracranial tuberculomas only in the cistern are extremely rare with only few reported cases in the literature, and we reported a unique case of isolated tuberculoma in the suprasellar cistern with right ocular motility disorder and upper eyelid ptosis as the main symptom and reviewed the relevant literature.

CASE PRESENTATION

Chief complaints

A 5-year-old boy was admitted to the Neurosurgery Department of Tianjin Huanhu Hospital on February 2021 due to right ocular motility disorder and right upper eyelid ptosis.

History of present illness

The patient suffered from right ocular motility disorder, accompanied by right upper eyelid ptosis and diplopia in absence of apparent cause for one month, which had recently worsened. He had no fever, and denied symptoms of headache, vomiting, convulsions, and limb weakness. In addition, he denied fatigue, night sweats, wasting and other tuberculosis symptoms.

History of past illness

The patient had no history of previous disease, including tuberculosis, and he was not taking any medication.

Personal and family history

The patient had no related personal and family history, and he denied any tuberculosis contact. Bacillus Calmette-Guerin (BCG) vaccination was performed after birth.

Physical examination

The patient's vital signs were normal. Physical examination showed no remarkable findings of positive signs, including rales in both lungs, tenderness in the abdomen and palpable lymphadenopathy. Neurological examination showed the right third cranial nerve palsy with restrictions of eye movements and ptosis, pupil dilation and negative light reflex. There were no meningeal signs. Muscle tone and tendon reflexes were normal. The patient was not found to have the Marcus Gunn syndrome in the eye examination.

Laboratory examinations

Routine laboratory tests, including complete blood count, hepatic and renal function and electrolytes were all in the normal ranges. MTB and HIV serologies were negative.

Imaging examinations

The patient's brain CT scan showed a mixed density mass in the suprasellar cistern with poorly defined borders, and multiple patchy calcifications were observed within it, with intact adjacent bone structure (Figure 1). The brain magnetic resonance imaging (MRI) scan showed that the lesion was about 1.6×2.2×1.5cm, downward involving the prepontine cistern, with clear borders, and the lesion appeared isointense in T1-weighted images (T1WI) and hypointense to isointense in T2-weighted images (T2WI), with irregular ring-enhancement after injection of gadolinium (Figure 1). No abnormality was observed on a plain CT scan of the patient's chest, therefore active pulmonary tuberculosis was not considered.

Further diagnostic work-up

To determine the presence of MTB in the cerebrospinal fluid (CSF) , which suggested tuberculosis, the patient subsequently underwent lumbar puncture. The CSF was clear with normal pressure, and negative PCR for MTB.

FINAL DIAGNOSIS

The patient's clinical manifestations, physical signs, and imaging characteristics were combined to make a preliminary diagnosis of tumor, so surgery was performed through an inferior temporal approach. The mass was observed above the petrous apex, with a small part protruding into the perimesencephalic cistern. The P2 segment of the posterior cerebral artery (PCA) and oculomotor nerve were compressed and closely adhered to the mass. To avoid damaging to blood vessels and nerves, the mass was not forcibly removed from the adhesion with PCA and the oculomotor nerve. The final diagnosis was made after surgery, based on a histopathological examination and PCR of the resected tumour. Grossly, the mass was slightly firm with white caseating contents. Histopathology of the mass showed epithelioid cell hyperplasia nodules without tumor cell proliferation. The center of the nodules was caseous necrosis, surrounded by numerous lymphocyte infiltration and Langerhans giant cells, which suggested tuberculosis (Figure 2). MTB was found in the operative specimen by PCR, thus confirming the diagnosis of tuberculoma.

TREATMENT

After surgery, the patient was advised to go to a specialist hospital to start anti-tuberculosis treatment. The patient received anti-tuberculosis treatment with rifampicin (225mg QD), isoniazid (200mg QD) and pyrazinamide (500mg QD) for 3 mo.

OUTCOME AND FOLLOW-UP

After the surgery and 3 mo of anti-tuberculosis treatment, the symptoms of ocular motility disorder and ptosis were partially relieved. The right light reflect was normal, and pupil dilation disappeared completely. Follow-up brain contrast-enhanced MRI showed a significant decreased in size of the residual lesion (Figure 3). Anti-tuberculosis treatment will continue to be administered and we will continue to follow up on his condition.

DISCUSSION

Intracranial tuberculoma is a focal tuberculosis disease that develops from the hematogenous dissemination from other parts of the body and disperses in the parenchyma, meninges or adjacent tissues, forming multiple small granulomatous lesions of different sizes. As the tuberculosis develops, these foci continue to proliferate and aggregate, resulting in larger mature granulomatous lesions, or tuberculomas, with solid caseous necrosis in the center containing small amounts of MTB, surrounded by an envelope of epithelioid cells, multinucleated giant cells, lymphocytes, and proliferating glial fibers[3]. Etiopathogenically, combining with the study of Rich and McCordock in 1933[4], the patient was infected by inhaling particles containing MTB. Many bacilli were killed but some survived, which were disseminated hematogenously to the central nervous system. Subsequently, mediated by the complex cellular immune, the small lesions called "Rich foci" were formed in the suprasellar cistern. As these lesions did not rupture into the subarachnoid space, there was an absence of tuberculous meningitis.

Intracranial tuberculoma can be solitary or multiple, but most of them are isolated, and multiple only accounts for 15-34% of cases[5]. Tuberculomas can occur anywhere in the brain, with the frontoparietal lobe predominating supratentorially and the cerebellar hemispheres predominating subtentorially, commonly in the gray-white matter junctional area and around the ventricles, mainly because these areas have rich blood supply and are the destination for hematogenous spread[6]. However, tuberculoma in the cistern alone is extremely rare, so that we only found five published tuberculoma cases through PubMed literature search. Three of them were multiple, namely, multiple small tuberculomas mimicking aneurysms in the suprasellar cistern reported by Gücüyener K *et al*[7], diffuse multiple tuberculomas in the suprasellar cistern and lateral fissure reported by Pinto DS *et al*[8], and Fujii T *et al*[9] reported multiple old calcified nodular lesions within the suprasellar cistern. Two cases were solitary, as in our case, one was mediastinal tuberculosis with right prepontine cistern tuberculoma reported by

Agu CC *et al*[10], while another was suprasellar cistern tuberculoma involving the pituitary gland reported by Sundar US *et al*[11]. The clinical manifestations of intracranial tuberculoma mainly depend on its location, size and number. Most tuberculomas present epilepsy, headache, and intracranial hypertension as the first symptoms, which may or may not be accompanied by systemic symptoms such as fever, night sweats and wasting. In our case, only ocular symptoms were the main clinical manifestation, which was relatively rare and mainly due to the compression of the right oculomotor nerve.

Intracranial tuberculoma is diagnosed mainly based on clinical and relevant imaging evidence. It often occurs in patients with the signs of pulmonary tuberculosis infiltration or immunodeficiency[12]. However, the immune function of the patient was normal, HIV was negative and his chest CT indicated no pulmonary tuberculosis infection. Therefore, the patient did not have favorable condition for intracranial tuberculoma. In addition, the patient was negative for MTB, which might be attributed to the fact that MTB enters the CSF circulation only when the tuberculoma envelope ruptured. As a result, the patient had no direct clinical evidence of tuberculosis infection.

Tuberculoma may appear as iso- or mixed density on CT scan, and annular or irregular enhancement after contrast injection. Typical tuberculoma can be manifested as peripheral annular enhancement with calcification and a central hypodense zone, which is called "target sign"[6]. The manifestation of tuberculoma on MRI depend largely on the presence of caseous necrosis. Non-caseating granulomas show hypointense on T1WI and hyperintense on T2WI with uniform enhancement, while caseating granulomas show hypo- or isointense on T1WI and hypointense on T2WI with annular enhancement. Caseous granuloma has high iron and manganese content, resulting in hypointensity on T2WI[5]. The CT and MR findings of the patient were consistent with those of caseous granuloma. The brain MRI showed that the mass was adjacent to the right internal carotid artery and cavernous sinus, but the compression was not obvious. In addition, the radiation dose of CT angiography was high relative to the young age of the patient, therefore the CT angiography was not performed.

Magnetic resonance spectroscopy is of adjunctive value in differentiating tuberculoma from other intracranial diseases, which typically shows elevated lipid and lactate peak, more choline and less N-acetylaspartate and creatine[13]. In short, the imaging findings of the patient were not specific and there was no clinically relevant indications of tuberculosis infection, so the diagnosis of tuberculoma becomes extremely difficult, which was also the reason for the initial diagnosis of tumor.

The "gold standard" for the diagnosis of intracranial tuberculoma remains pathological diagnosis, relying primarily on stereotactic biopsy or surgery. Stereotactic biopsy was limited by the deep location of the lesion and the risk of neurovascular damage. Finally, the patient received surgery and was diagnosed with tuberculoma by the combination of histopathology and PCR. As the tuberculoma was tightly adherent to the P2 segment of PCA and oculomotor nerve, partial residual existed postoperatively, so the patient continued postoperative anti-tuberculosis treatment. And now the patient is stable with partial relief of symptoms compared to before.

CONCLUSION

Intracranial tuberculoma is difficult to diagnose owing to the uncertainty and non-specificity of its clinical manifestations and imaging features. This case confirmed once again that isolated intracranial tuberculoma can occur in the suprasellar cistern. Therefore, for space-occupying lesions in the suprasellar cistern, tuberculoma should be included in the differential diagnosis even if there is no history and indication of tuberculosis infection.

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