Cystic Tuberculomas of the Brain

—Two Case Reports—

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Abstract

A 19-year-old male presented with raised intracranial pressure and a peripherally enhanced multinodular lesion with a large subjacent cyst in the right medial frontal lobe. Surgery revealed a solid tuberculoma and an underlying cyst lined with normal brain tissue. A 22-year-old male with a history of tuberculous meningitis and hydrocephalus presented with an exophytic brain stem and a left temporal tuberculoma, which were excised in two stages. The temporal tuberculoma had a peripherally located cyst that was lined with tuberculous tissue. Intracranial tuberculomas are an important part of the differential diagnosis where tuberculosis is endemic and in immunocompromised patients. Cystic tuberculomas are uncommon. The cysts may be centrally or peripherally intralesional, or either subdural and extralesional, or occur between the lesion and the brain. The type of cyst is important to recognize preoperatively, so that adequate precautions can be taken during the excision of these lesions.

Key words: cyst, classification, tuberculoma, brain

Introduction

The incidence of cerebral tuberculomas and the incidence of tuberculomas requiring surgical intervention are both declining. However, the population of immunocompromised patients is increasing, and, together with easy transportation and population translocations, results in tuberculous infections of the central nervous system appearing in all parts of the world. Intracranial tuberculomas commonly present as well-circumscribed masses, which are nodular, firm to hard, and relatively avascular. The size varies from a less than 1 cm to more than 10 cm in diameter.⁷⁾ They are surrounded by varying degrees of edema and gliosis. Other than the classical presentation described above, there are certain atypical forms which may confuse the diagnosis. One of the rarer types of tuberculomas is that associated with cysts. We report two cases of intracranial tuberculoma associated with cyst, present a classification of cystic tuberculomas, and discuss the pathogenesis of these unusual lesions.

Case Reports

Case 1: A 19-year-old boy presented with a history of febrile illness persisting for one month for which all investigations were negative. He developed bifrontal headache associated with vomiting one week before admission to our department. There was no history of childhood tuberculosis, nor any family history of tuberculous infection. On admission, he was fully conscious and well oriented with normal speech. Fundus examination showed bilateral papilledema. There were no focal or lateralizing signs. There was no neck stiffness and Kernig's sign was negative.

Computed tomography (CT) of the brain showed an isodense multinodular lesion with peripheral enhancement in the right medial frontal region (Fig. 1). The lesion reached the brain surface just above the right cribriform plate. A large hypodense, non-enhanced circumscribed area was seen subjacent to the lesion and between the lesion and the brain, which caused pressure on the ipsilateral frontal horn of the lateral ventricle and shift of the midline to the left, with evidence of subfalcine herniation.

The lesion was approached by a right frontal craniotomy. The lesion reached the surface at the

Fig. 1 Case 1. Computed tomography scans of the brain with contrast medium showing a ring-enhanced lesion with a large hypodense cyst.

medial and inferior aspects of the frontal lobe. The lesion was tough and well circumscribed with minimal surface vascularity, and was easily separated from the surrounding white matter. A large cyst containing faintly xanthochromic clear fluid was located posterior and superior to the lesion. The wall of the cyst was normal brain tissue and was therefore left untouched. Two smaller cysts were located superior and medial to the lesion, separate from the large lateral cyst, and were also lined by normal brain tissue. The tough solid lesion was totally excised. Gross examination of the lesion revealed a thick-walled, multinodular lesion with thick, white caseous material in the center (Fig. 2). Histological examination of the specimen confirmed the diagnosis of tuberculoma, with the presence of Langhans giant cells, epithelioid cells surrounding areas of caseation, and lymphocytic infiltration (Fig. 3). The cyst fluid contained no cells, and no bacteria or fungal elements were detected by Gram's stain, acid fast bacilli stain, and fungal preparation. The fluid had a protein content of 1.3 g% and sugar content of 8 g%.

The patient was started on a three-drug anti-tuberculosis chemotherapy and made an uneventful recovery. He has been advised to continue the anti-tuberculosis medication for 18 months.

Case 2: A 22-year-old male presented with imbalance on walking and generalized seizures. He had received a full course of anti-tuberculosis

Fig. 2 Case 1. Photograph of the gross specimen of the tuberculoma cut open to show the thick shaggy walls and the caseous material content.

Fig. 3 Case 1. Photomicrographs of the pathological section of a tuberculoma showing multinucleated Langhans giant cells (arrow), epithelioid cells (double arrow), and lymphocytic infiltration in the surrounding fibrous tissue (thick arrow). HE stain, A: $\times 10$, B: $\times 40$.

chemotherapy 2 years before for tuberculous meningitis and had undergone ventriculoperitoneal shunting for hydrocephalus. CT showed an exophyt-

Fig. 4 Case 2. Computed tomography scan with contrast medium showing a left temporal tuberculoma with a peripherally located cyst.

ic brain stem and a left temporal tuberculoma (Fig. 4). The former was excised at the first stage surgery. However, he continued to have symptoms of raised intracranial pressure, right hemiparesis, and non-resolution of the temporal lesion despite a five-drug anti-tuberculosis therapy. Therefore, the left temporal lesion was treated surgically 3 weeks later. The solid intracerebral tuberculoma included a large cyst containing xanthochromic fluid in the posterior part of the lesion. The cyst was lined with tuberculous granulation tissue which was also excised. The patient improved in the postoperative period and now being followed up.

Discussion

The first report of a cystic tuberculoma was in a 4-year-old boy with hemiparesis in 1962.¹⁾ The cyst contained xanthochromic fluid and was located within the tuberculoma. A localized frontal tuberculoma with a cyst was described in 1963.⁵⁾ Acid fast bacilli have also been demonstrated in the fluid of a cyst within a tuberculoma.⁴⁾ Other cases of similar cystic tuberculomas have been reported.^{2,3)} A description of several unusual forms of tuberculomas included tuberculomas with cysts.⁶⁾ One patient in the series had a subdural cyst lying superficial to a subcortical tuberculoma, and three other cases, called cystic tuberculomas, had cysts lined by tissue that was recognizable as tubercular.

Fig. 5 Classification of cystic tuberculomas. Type I: intralesional, cyst within the center of the tuberculoma; Type II: intralesional, cyst at the periphery of the tuberculoma; Type III: extralesional, cyst is subdural in location; Type IV: extralesional, cyst lies between the tuberculoma and the brain.

The cysts associated with tuberculomas can be classified into four different types, depending on their relationship with the tuberculoma (Fig. 5): Type I, completely intralesional and centrally located⁶⁾; Type II, intralesional but peripherally located⁶⁾ (our Case 2); Type III, extralesional and subdural⁶⁾; or Type IV, extralesional and between the lesion and the brain (our Case 1). Types I and II are intralesional whereas types III and IV are extralesional. Intralesional cysts are more common than extralesional cysts and probably represent the end result of liquefaction necrosis of the central caseous material of tuberculomas. Whether anti-tubercular chemotherapy is involved in the formation of these cysts is not known. The cyst fluid in these cases is high in protein content and may contain lymphocytes and large mononuclear cells. No acid fast bacilli are found in the fluid. Type II cysts are peripherally located in the lesion and may pose problems in diagnosis and surgery. Unless the surgeon is aware of the possibility of peripherally located cyst (Type II), the lesion may not be completely excised, leaving behind the pathological cyst wall. A previous case appeared to be a large multiloculated cyst at initial surgery, which was excised but the solid component of the tuberculoma was missed and required a second surgery.⁶⁾

Extralesional cysts associated with tuberculomas are extremely rare, with only our case and one other case reported.⁶⁾ These cysts are totally outside the

tuberculoma. In the previous case, the cyst was large and subdural (Type III), and was separate from the dura, the underlying cortex, and the subjacent tuberculoma. In our case, the cyst was completely extralesional and was the major cause of the mass effect. However, the cyst was not subdural, but lay between the tuberculoma and the brain. The lining of the cyst was gliotic brain tissue and did not incorporate tuberculous tissue.

The pathogenetic mechanism of the formation of extralesional or perilesional cysts is unclear. Type III cyst may possibly result from a localized inflammatory response of the meninges to the adjacent tuberculous infection, with a resultant thickening of the meninges and formation of a cyst. Type IV cyst is probably due to other causes. The possible pathogenetic mechanisms may be similar to those seen in peritumoral cysts associated with meningiomas and vestibular schwannomas, or a result of loculation of cerebrospinal fluid in an adjacent subarachnoid space. Other possible mechanisms include ongoing inflammatory response to the presence of the infection, exudation caused by inflammatory vasculitis, development of an inflammatory arachnoid cyst, or a combination of all these factors. The rarity of these lesions makes determining the exact causative mechanism very difficult.

Preoperative diagnosis of the type of cysts is possible to a certain extent. Magnetic resonance imaging with contrast medium will outline the pathological tuberculous lining of the cysts, allowing the surgeon to differentiate intralesional from extralesional cyst. Magnetic resonance imaging with contrast medium will also differentiate peripheral intralesional cyst (Type II) from extralesional cyst (Type IV). However, identifying the pathology of the main solid lesion as tuberculous is still difficult, as these lesions can mimic many other intracranial pathologies.

Type I and II cysts require total removal of the

cyst wall along with the solid portion of the tuberculoma to ensure complete excision of the lesion. Types III and IV only require excision of the solid lesion. However, wide opening of the cyst cavity and establishment of communication to the normal subarachnoid spaces is optimum to prevent persistence or reaccumulation of fluid within the cyst.

References

- 1) Dastur HM, Desai AD, Dastur DK: A cystic cerebral tuberculoma treated surgically. *J Neurol Neurosurg Psychiatry* 25: 370, 1962
- 2) Kamiya K, Yamashita N, Nagai H, Mizawa I: [Intracranial tuberculoma with difficult preoperative diagnosis. Case report]. *Neurol Med Chir (Tokyo)* 31: 991-994, 1991 (Jpn, with Eng abstract)
- 3) Kunishio K, Sunami N, Yamamoto Y, Asari S, Akagi T, Ohtsuki Y: [Cystic cerebral tuberculoma—a case report]. *No Shinkei Geka* 14(3 Suppl): 429-434, 1986 (Jpn, with Eng abstract)
- 4) Lieberman A, Dart L, Bennett R: Intracerebral tuberculoma. Case report. *J Neurosurg* 33: 331-333, 1970
- 5) Rao BD, Subrahmanyam MV, Sathe NM: Cerebral tuberculoma simulating cystic glioma—a case report. *J Neurosurg* 20: 172, 1963
- 6) Sinh G, Pandya SK, Dastur DK: Pathogenesis of unusual intracranial tuberculomas and tuberculous space-occupying lesions. *J Neurosurg* 29: 149-159, 1968
- 7) Tandon PN: *Tuberculosis of the central nervous system*, in Ramamurthi B, Tandon PN (eds): *Textbook of Neurosurgery*. New Delhi, Churchill Livingstone, 1996, pp 473-488

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