Learning from errors

Quadriparalytic disseminated neurocysticercosis

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Summary

Cysticercosis is the most common parasitic infection of the central nervous system. Cysticercosis infrequently affects the spine, but when it does, it can present with symptoms similar to other more common spinal diseases. Here, the authors report a case of disseminated cysticercosis, with simultaneous involvement of brain and the spinal cord. Initially, the patient was misdiagnosed as tuberculoma on the basis of cerebrospinal fluid examination and CT scan of brain and was being treated with antitubercular therapy. Later on the patient developed quadriplegia which was investigated and diagnosed to be disseminated neurocysticercosis.

BACKGROUND

Worldwide, neurocysticercosis (NCC) is the most common parasitic infestation of central nervous system (CNS) caused by ingestion of eggs of *Taenia solium*.¹ The common presentations of NCC are headache, seizure and focal neurological deficit. NCC usually involves brain parenchyma, eye and subcutaneous tissues. The incidence of spinal NCC (SNCC) has been reported as 1.5%–3% by most authors.^{2–4} Both brain tuberculosis and NCC are common in countries like India and cause the difficulty in differentiating NCC and TB only on the basis of neuroimaging.⁵ Here, we report a case of disseminated NCC which was initially misdiagnosed as brain tuberculoma and was being treated for it. Later on, when he developed quadriplegia, he was investigated further and was found to have disseminated NCC.

CASE PRESENTATION

A 20-year-old young man presented with recurrent seizures from last 3 years, fever, headache and neck pain (1 month), weakness of both lower limbs (15 days), inability to hold objects properly with both hands (5 days) and urinary retention (2 days), history except for seizures was insignificant. On examination, vitals were normal. CNS examination showed, presence of neck rigidity, higher mental functions were normal. All the cranial nerves were intact. Motor system showed normal bulk with spastic limbs. Power was grade 3/5 in both lower limbs and grade 4/5 in both upper limbs. Handgrip was weak on both sides, right and left. Biceps and triceps reflexes were exaggerated on both sides with normal supinators. Knee and ankle jerks on both sides were exaggerated. Plantar was extensor on both sides. Rest of the examination was normal.

Initially, the patient was treated elsewhere, cerebrospinal fluid (CSF) examination (total cell count-5/mm³ with all lymphocytes and protein-356 mg/dl) and CT scan brain (ring enhancing lesions in left occipital lobe with perilesional oedema as shown in figure 1A) was done there, for which antitubercular treatment according to his body weight which consisted of rifampicin, isoniazid, pyrazinamide, ethambutol and injection streptomycin and

prednisolone 40 mg daily had been given, but the patient did not respond, rather, he developed weakness of lower limbs (15 days), inability to hold objects properly by either both hand (5 days) and urinary retention (2 days) for all these complaints he was referred to our centre.

INVESTIGATIONS

Routine investigations like complete blood profile, blood sugar, serum urea, creatinine and electrolytes were normal. Erythrocyte sedimentation rate was 25/1st h and purified protein derivative was negative. Chest x-ray and cervical spine x-ray were normal. MRI brain (figure 1B,C) and spinal cord (figure 2) were suggestive of NCC cysts. Serum anticysticercus IgG antibody was also positive.

DIFFERENTIAL DIAGNOSIS

A differential diagnosis spectrum should be considered when cystic, intramedullary lesions are detected in MRI images: tuberculosis, congenital lesions (simple or complex arachnoid cysts, dermoid cysts), hydatid cysts, sarcoidosis or subarachnoid metastatic neoplasm.^{5–9}

TREATMENT

The patient was treated with albendazole 400 mg twice daily for 28 days with oral prednisolone in tapering dose. Phenytoin 300 mg once daily was given for seizures.

OUTCOME AND FOLLOW-UP

In the next follow-up, after 1 month, weakness of upper limbs had progressed; power was 3/5 in upper limbs while lower limbs had similar power of 3/5. However, he did not have any episode of seizure in between. The patient was advised to repeat neuroimaging and to seek neurosurgical consultation for excision of the cyst. But unfortunately, the patient was lost to follow-up.

DISCUSSION

SNCC has been identified in vertebral, extradural, intradural and intramedullary regions.¹⁰ It should be noted that intramedullary NCC in the cord is very rare with most

Figure 1 (A) Axial contrast enhanced CT shows two ring enhancing lesions in left occipital lobe, perilesional edema (arrow). (B) Gadolinium enhanced axial T1 image shows multiple ring and nodular enhancing lesions suggestive of neurocysticercosis cysts. (C) Gadolinium enhanced axial T1 image shows two ring enhancing lesions in cerebellum and one in cerebrum-left parieto-occipital region (arrow).

lesions being subarachnoid.¹¹ Intramedullary spinal cysticercosis often presents in the patients between 20 and 45 years old.¹⁰ Human beings are the definite host where adult worm stays in intestine and lays eggs which are taken by pigs, the intermediate host. The embryo crosses the intestine and resides in CNS, muscles, eyes and skin as cysticerci. Human beings acquire infestation through ingestion of the eggs, just as pigs get infected. The eggs hatch in the intestine and develop into cysticerci in the various organs including CNS.

The cysticerci larvae dissemination into the intradural extramedullary spinal space has been explained by the accumulation of a great volume of blood originating from valve-less epidural venous plexus, with extremely thin-walled veins, this may conduct the blood in any direction, under the influence of intra-abdominal and intrathoracic pressure variations. Cases have been reported which have corroborated the hypothesis of retrograde flow of those larvae through veins epidural venous plexus, dissemination of primary cysticerci larvae into the intradural-extramedullary spinal canal without cerebral involvement. In contrast, Isamat de La Riva's postulated on the parasite's descending migration from the cerebral subarachnoid space into

the spinal space.¹² SNCC is rare compared with intracranial as cysticerci do not pass through subarachnoid space at cervical level due to its size (sieve effect). Besides CSF reflux at craniovertebral junction propels floating cysts back into intracranial space rather than the spinal canal. Brain blood flow (100-fold greater than spinal) is also an important determining factor which plays a role in having more intraventricular lesions. SNCC has predilection for thoracic region attributed to more blood flow in this region. The location of cysticerci is cervical-34%, thoracic-44.5%, lumbar-15.5% and sacral-6%. SNCC in the absence of previous history or subcutaneous nodule poses great diagnostic challenge. The unusual finding in our case is the intramedullary NCC which is extremely uncommon. The location of mass lesion, its size, the inflammatory response generated by cyst breakdown are important factors for management.

In our case, CT scan brain misled to the initial diagnosis of tuberculoma and the patient was receiving antitubercular treatment, but he was not improving, rather, new symptoms were appearing. CSF findings in both NCC and tuberculoma are similar; both may have pleocytosis with predominance of lymphocytes. The protein level in CSF

Figure 2 Sagittal T2 weighted imaging of cervical spinal cord shows well-defined intramedullary lesion with hypointense rim (open arrow) and a central dot like hypointensity (suggestive of scolex) at level of second cervical vertebra with hyperintensity of spinal cord extending from cervicomedullary junction to C7 vertebral level -suggestive of oedema.

may be elevated in both as well.¹³ We did not go for repeat CSF examination because it was not helpful and the space occupying lesion contraindicated it too. The patient was lost to follow-up therefore; histopathology could not be done which is an absolute criterion for the diagnosis of cysticercosis. The patient did not respond to albendazole therapy, if he would have responded, it would have been another consistent finding for diagnosis of cysticercosis. The possibility of CNS tuberculosis in this patient was not ruled out with adequate evidence because in the CSF neither adenosine deaminase level was determined, nor PCR for tuberculosis performed. Nevertheless, here diagnosis of cysticercosis was considered because the patient fulfilled the criteria proposed by Del Brutto *et al* for diagnosis.¹⁴ The patient fulfilled two major criteria that is, neuroradiological lesion was suggestive of NCC (cystic lesion, one or more nodular calcification), and second major criteria

fulfilled was antibodies to cysticerci by enzyme linked immunoelectrotransfer blot was present in serum, plus the patient fulfilled one minor criteria (seizure as clinical presentation) and the patient belonged to an area endemic for cysticercosis fulfilled the epidemiological criteria.

Learning points

- ▶ Disseminated NCC is a rare presentation where concurrent involvement of brain parenchyma and spinal cord can occur. It is noteworthy that intramedullary NCC in the cord is very rare with most lesions being subarachnoid.
- ▶ Spinal NCC should also be considered in the differential diagnosis in patients coming from endemic areas presenting with symptoms suggestive of a spinal mass lesion.
- ▶ This report highlights that NCC may have diagnostic difficulty in being differentiated from brain tuberculoma in countries where both the diseases are endemic.

Competing interests None.

Patient consent Obtained.

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