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Isolated Spinal Neurocysticercosis with Clinical Pleomorphism

İzole Spinal Nörosistiserkosiz ve Klinik Çeşitliliği

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ABSTRACT

Neurocysticercosis, involvement of the central nervous system by taenia solium, is one of the most common parasitic diseases worldwide. However, isolated spinal involvement by neurocysticercosis, either intramedullary or extramedullary, is uncommon. The authors report a case of C1-C2 intradural extramedullary neurocysticercosis in a young male that presented initially with signs of raised intracranial pressure followed by high cervical myelopathy. Transventricular migration of the neurocysticercal cyst along the cerebrospinal pathways leading to clinical pleomorphism as described by the authors can perplex the best of clinicians leading to a missed diagnosis as in the present case. Diagnosis was confirmed after surgical excision and the patient is doing well at six months follow up with no neurological deficits. Spinal neurocysticercosis should be considered in the differential diagnosis in high-risk populations with new symptoms suggestive of a spinal mass lesion.

KEY WORDS: Clinical, Extramedullary, Neurocysticercosis, Spinal

ÖZ

Santral sinir sisteminin tenya solium ile infestasyonu (Nörokistoserkosis) dünyada en fazla görülen parazitik hastalıklardan birisidir. Nörokistoserkosizin izole spinal formu olarak nadir görülmektedir. Yazarlar genç bir kadın hastada C1-C2 yerleşimli intradural ekstramedullar yerleşimli nörokistoserkosis tanımlamışlardır. Bu hastada kafa içi basınç artışı bulgularının yanısıra üst servikal bölgede myelopati gelişmiştir. Nörokistoserkosis kistleri beyin- omurilik sıvısıyla serebrospinal sistem içerisinde değişik bölgelere transfer olabilir; bu nedenle taşındığı yerde yaptığı basınç etkisi ile ilişkili bulgular verir. Sunulan vakada olduğu üzere; bu da klinisyenlerin yanlış tanı koymalarına yol açabilir. Bu vakada kesin tanı cerrahi eksizyon sonrası konulmuştur. Ameliyat sonrası dönemde hastada bir komplikasyon gelişmemiştir.Yüksek riskli toplumlarda spinal kitle bulguları olan hastalarda Spinal nörokistoserkosiz ayırıcı tanıda düşünülmesi gereken bir patolojidir.

ANAHTAR SÖZCÜKLER: Clinical, Extramedullary, Neurocysticercosis, Spinal

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INTRODUCTION

Cysticercosis is the most common parasitic infection affecting the central nervous system (CNS) worldwide and is now frequently identified in the developed and other industrialized countries because of increased immigration and improved diagnostic methods (2,5,10). Purely spinal neurocysticercosis (NCC) is rare and requires timely diagnosis and appropriate management because of the natural confines of the spinal canal and its potential to cause irreversible neurological deficits (1,7,10). The authors report a rare case of the C1-C2 level intradural extramedullary NCC which had a bizarre presentation and was diagnosed only after histopathological examination.

CASE REPORT

A 34-year-old male was referred to us with complaints of headache/vomiting on and off for 3 years. Non-contrast computed tomography (CT) of the head performed one year previously had revealed hydrocephalus that was not investigated further as the headache and vomiting had subsided. He developed neck pain with restriction of neck movements and weakness of all four limbs for three months. Examination revealed restricted neck movements with spastic quadriparesis (4/5 MRC grade) along with 40 -50 % sensory loss to all modalities below the C2 level. A high cervical pathology, possibly craniovertebral junction (CVJ) anomaly was suspected. X-rays as well as magnetic resonance imaging (MRI) of the brain were normal but MRI of the spine revealed an intradural extramedullary pathology at the C1-C2 level with peripheral contrast enhancement. (Figure 1) Pre-operatively, a diagnosis of tuberculoma/ inflammed epidermoid was considered on neuroimaging. C1-C2 laminectomy with decompression of the lesion was done. The lesion (pink fleshy tissue) was densely stuck to the cord on posterior and lateral aspects with poor cleavage plane with cord tissue. It appeared like granulation tissue and a diagnosis of tuberculosis was considered. The lesion extended from the foramen magnum to the C2 level. Subtotal decompression was carried out in view of the intraoperative suspicion of tuberculosis and adherence of the lesion to cord tissue. The final diagnosis however turned out to be cysticercosis after histopathological examination. (Figure 2) The patient was given a course of albendazole with steroids for 8 weeks after surgery and recovered from his weakness and pain at six months follow-up with no residual lesion on follow-up MRI (Figure 3).

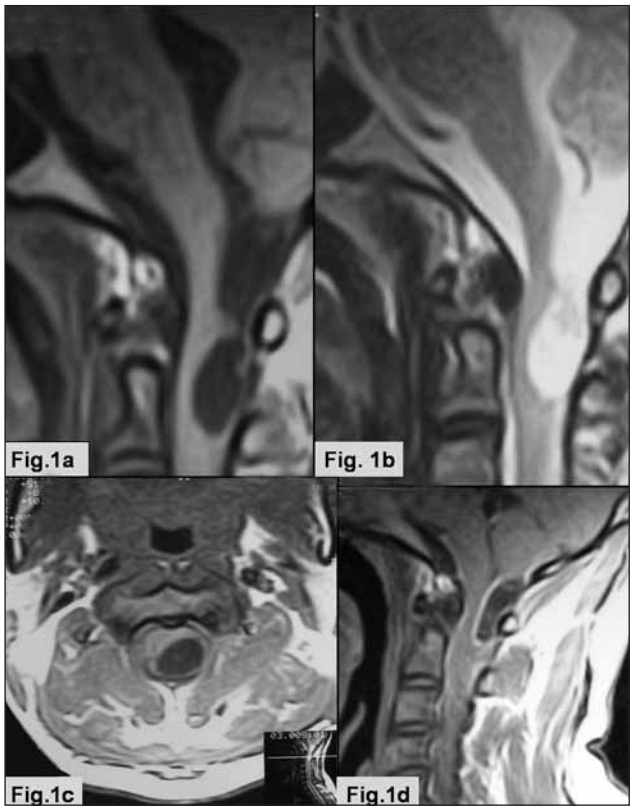

Figure 1: *Sagittal T1W (Fig. 1a), Sagittal T2W (Fig. 1b) and axial T1W (Fig. 1c) MR image showing a relatively well defined intradural extramedullary lesion hypointense on TIWI and hyperintense on T2WI with peripheral enhancement on contrast administration (Fig. 1d).*

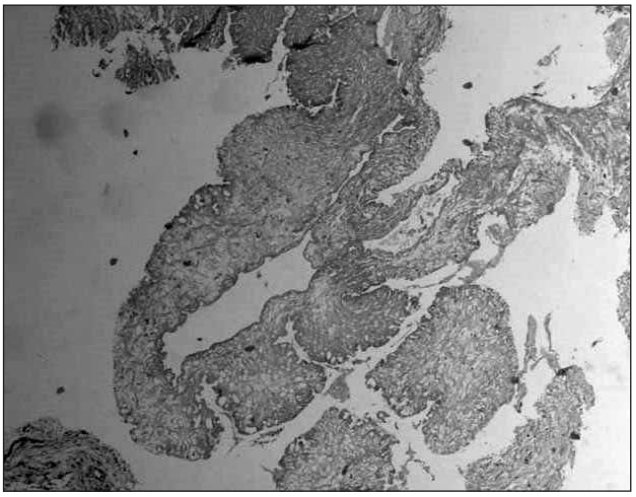

Figure 2: *Photomicrographs of the histopathological specimen showing scolex with a portion of the cyst wall (H & E X 100).*

DISCUSSION

Cysticercal involvement of the central nervous system by taenia solium characteristically involves

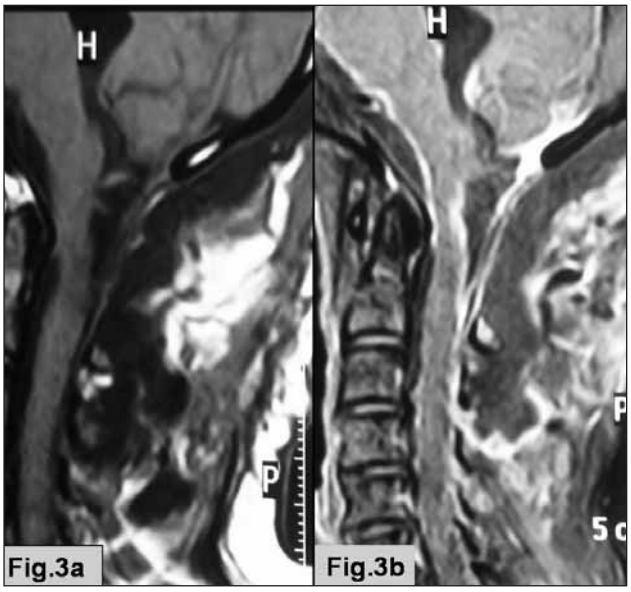

Figure 3: *Postoperative sagittal T1W (Fig. 3a) and post-contrast sagittal MR image (Fig.*3b*) showing no residual lesion.*

the brain parenchyme, intracranial subarachnoid space or ventricular system (1,5). Intraspinal involvement by cysticercosis is reported in only 1 to 5% of patients with NCC, most of these cases being associated with concomitant cerebral involvement (2-4,6-8,13). Isolated spinal involvement by cysticercosis, either intramedullary or extramedullary is rare (7,10).

Clinical symptomatology due to spinal NCC depends on several factors such as location, spinal level, lesion size and presence (or absence) of inflammation/arachnoid scarring due to cyst degeneration (1,2,7,15). Our case presented with features of high cervical myelopathy preceded by symptoms of raised intracranial pressure. Cerebral cysticercosis is more common than spinal forms and could have been responsible for the initial raised pressure symptoms caused by hydrocephalus as was noticed on CT which however may have missed the neurocysticercal cyst. Further tranventricular migration of the cyst with impaction at craniovertebral junction leading to features of high cervical myelopathy had been thought of by the authors as a plausible explanation for this clinical pleomorphism. Clinical suspicion of spinal cysticercosis is difficult especially in the absence of a previous history of NCC or associated intracranial NCC. Though high eosinophil count and calcification of soft tissues in the plain radiogram may be suggestive, such findings are not common (9,14). Cerebrospinal fluid (CSF) findings typically include moderate lymphocytic pleocytosis, variable eosinophilic pleocytosis, elevated protein and low/normal glucose levels (2,5). Very high sensitivity and specificity has reported with enzyme-linked immunosorbent assay in CSF for diagnosis of neurocysticercosis (12). CSF studies were not performed in this case as the diagnosis was not considered preoperatively but if performed could have provided the diagnosis.

Current MR imaging can help diagnose these lesions on which the cysticercal cysts appear hypointense on T1W MRI and hyperintense on T2WI with surrounding edema and ring enhancement on post contrast T1W MRI (6,10,11). A scolex, if present, appears as hyperintensity on T1WI and hypointensity on T2WI and can be of diagnostic importance (6,10). In the absence of a characteristic scolex, which is not uncommon in spinal NCC, and of synchronous cerebral involvement as in our case, the diagnosis is difficult to arrive at considering the rarity of this lesion in spine (4,6,8).

Though various therapeutic options exist for spinal NCC, the rarity of spinal involvement has precluded the evolvement of definite guidelines as compared to cerebral NCC (1-9). The efficacy of antihelminthic therapy in cases of spinal cysticercosis is well described in a number of cases by various authors with resolution of neurological deficits (1-4,15). Surgery does not provide an unequivocal cure without morbidity in all cases of spinal cysticercosis, especially intramedullary cysts (9,14); due to the multiple mechanisms responsible for neurological symptoms in spinal cysticercosis (1,2,5,9,10,14,15). However, in patients presenting with acute onset of symptoms and in those where the diagnosis is in doubt, surgical excision should be done to eliminate the compressive element as histopathology not only confirms the diagnosis but early surgery also provides maximal chances of recovery before any irreversible cord changes take place (1,2,9,14). This should be followed with albendazole therapy to address the other factors implicated in the pathogenesis.

CONCLUSIONS

Spinal NCC should be considered in the differential diagnosis in high-risk populations with new symptoms suggestive of a spinal mass lesion.

Current MR imaging may help to diagnose this condition but this may not be feasible in a significant number of cases, especially in isolated spinal involvement. This compounded by the clinical pleomorphism as described in the present case can perplex the best of clinicians. Though both surgical as well as medical therapy has a role in the management of spinal cysticercosis, clinical suspicion and laboratory confirmation of the diagnosis in an appropriate setting with timely institution of medical therapy can lead to a successful outcome in patients with spinal cysticercosis.

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