ISOLATED CAUDA EQUINA CYSTICERCOSIS - A RARE CAUSE OF CAUDA EQUINA SYNDROME

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ABSTRACT

BACKGROUND

Cysticercosis is the most common parasitic infection of the nervous system caused by Taenia solium, with humans being the definitive hosts and pigs being the intermediate hosts. Cerebral involvement is more common than spinal involvement. Spinal involvement is commonly associated with the concomitant involvement of the brain, and spine is affected in 1-3% of the all cases of the neurocysticercosis. Isolated spinal cysticercosis is very rare without evidence of concomitant cranial disease and thoracic spine is commonly involved, followed by cervical, lumbar and sacral regions. Isolated cauda equina neurocysticercosis is extremely rare and only 15 cases of isolated cauda equina involvement were reported in the literature to the best of our knowledge. We present a case with isolated cauda equina involvement of cysticercosis with review of literature.

METHODS

In July 2016, we performed a literature search for isolated neurocysticercosis of the cauda equina with no limitation for language and publication date. The search was conducted through http://pubmed.com, a well-known worldwide internet medical address. To the best of our knowledge, we could find only 15 cases of isolated neurocysticercosis of cauda equina. We reviewed the literature with illustration of our case. We present a case of a 27-year-old female patient who presented with insidious onset of low back pain. MRI lumbosacral spine demonstrated an oblong, intradural CSF intensity cystic lesion extending from L1 to S1. With hemilaminectomy from L1 to L5, the cyst was excised totally. Histopathological diagnosis was cysticercosis.

RESULTS

Patient totally recovered from her pain and sensory symptoms within a week and bladder and bowel symptoms improved gradually over a period of one to two weeks.

CONCLUSION

Isolated cauda equina neurocysticercosis is extremely a rare cause of cauda equina syndrome and this should be considered in the differential diagnosis of cauda equina cystic lesions. These can be excised safely with good postoperative recovery.

KEYWORDS

Isolated Neurocysticercosis, Cauda Equina, Spinal Intradural Cyst.

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INTRODUCTION: Cysticercosis is the most common parasitic infection of the nervous system caused by Taenia solium, with humans being the definitive host and pigs being the intermediate host.[1] Cerebral involvement is more common than spinal involvement.[1,2,3,4,5,6] Spinal involvement is commonly associated with the concomitant involvement of the brain and spine is affected in 1-3% of the all cases of the neurocysticercosis.[1,2,4,6] Isolated spinal cysticercosis is very rare without evidence of concomitant cranial disease and thoracic spine is commonly involved by cysticercosis, followed by cervical, lumbar and sacral regions.[1,2,3,4 5,6]

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Isolated cauda equina neurocysticercosis is extremely rare and only 15 cases of isolated cauda equina involvement were reported in the literature to the best of our knowledge.[2,3,4,5,7,8]

REVIEW OF LITERATURE: In July 2016, we performed a literature search for isolated neurocysticercosis of the cauda equina with no limitation for language and publication date. The search was conducted through http://pubmed.com, a well-known worldwide internet medical address. To the best of our knowledge, we could find only 15 cases of isolated neurocysticercosis of cauda equine.[2,3,4,5,7,8] We present a case with isolated neurocysticercosis of the cauda equina was excised totally with good postoperative recovery with review of literature. A 27-year-old female presented with low backache radiating to left lower limb, difficulty in walking with tingling and numbness in both lower limbs of two months duration. Patient had difficulty in passing urine with history of constipation of one week duration.

Patient underwent caesarean section two months back under spinal anaesthesia after which patient became symptomatic. On neurological examination, patient had asymmetric weakness of both ankle dorsiflexion, more on left side with absent ankle jerk. Saddle anaesthesia was present. MRI Lumbosacral spine demonstrated an oblong, intradural, extramedullary CSF intensity mass lesion in the lumbar canal from lower border of L1 to the upper border of S1 level, displacing the roots of the cauda equina peripherally; possibility of arachnoid cyst was reported. We thought the possibility of arachnoid cyst because of typical MRI findings and arachnoid cysts were reported following spinal anaesthesia.[9] Patient was already having bladder and bowel disturbances of recent onset with progression of symptoms. We operated the patient without any further delay and we did not do any serological investigations in view of progressive neurological symptoms of the patient. Left-sided hemilaminectomy was done from L1 to L5 with total excision of the cystic lesion done.

Intraoperatively, lesion was bulging out after opening the dura and it was a light bluish coloured, thin-walled cyst, containing clear fluid with all the roots displaced posteriorly and it was excised in toto without any difficulty. Histopathologically, cystic lesion with complex infoldings with characteristic three layers, inner chitinous layer, middle nuclear layer and outer muscular layer, without evidence of inflammation, consistent with cysticercosis. Patient totally recovered from her pain and sensory symptoms within a week and bladder and bowel symptoms improved gradually over a period of one to two weeks. Patient was discharged with the advice of albendazole for four weeks along with steroids. Followup period, patient did not have any further complaints.

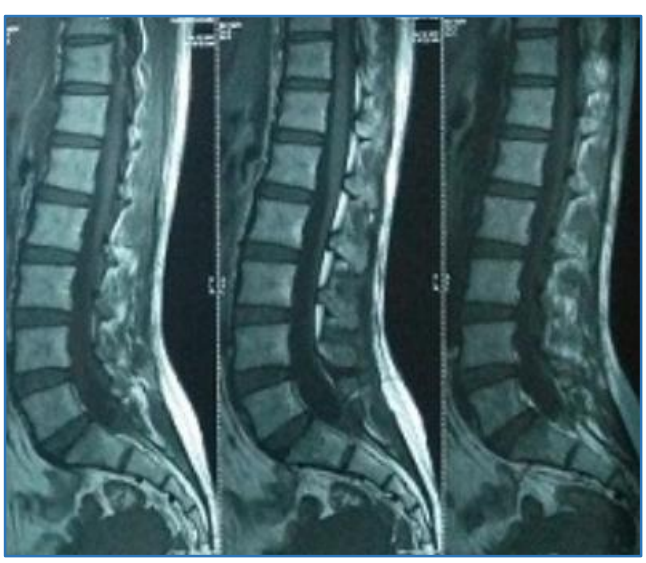

Fig. 1: T1 Weighted Images Show CSF Intensity, an Oblong, Cystic Mass Extending From L1 to S1, Anterior to the Cauda Equina

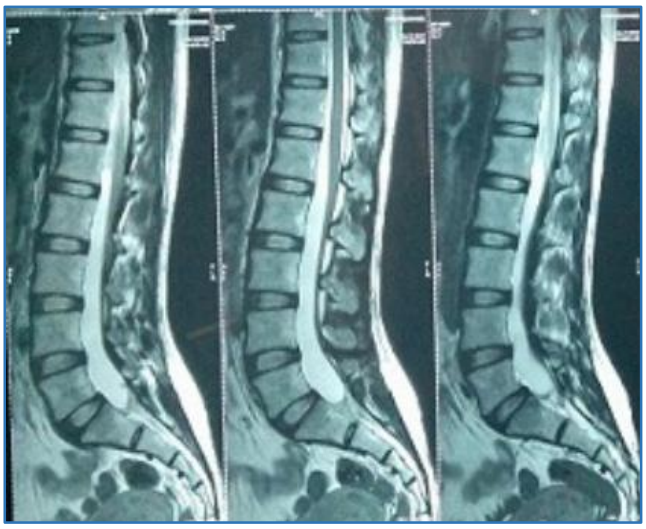

Fig. 2: T2 Weighted Images Showing an Oblong, Well-Defined CSF Intensity Intradural Lesion, Abutting the Posterior Vertebral Margin, Extending from L1 to the Upper Border of S1

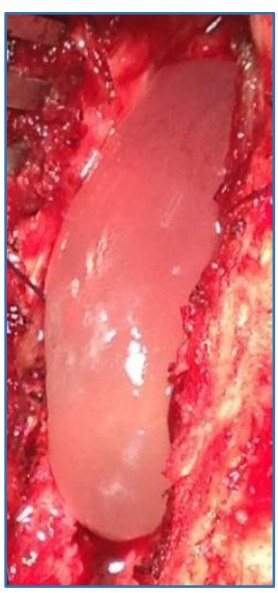

Fig. 3: Intraoperative Photo Showing Thin-Walled Cystic Lesion Bulging Out from the Dural Sac

DISCUSSION: Cysticercosis is the most common parasitic infection of the nervous system caused by Taenia solium, with humans the only known definitive hosts and pigs the natural intermediate hosts and humans may become accidental intermediate hosts for the larval form or cysticercus.[1] Neurocysticercosis is endemic in low socioeconomic countries, mainly affecting Africa, Latin America and Asia.[2] Cysticercosis, commonly involves the brain and spinal involvement is uncommon and spinal involvement is reported in 1-3% of the all neurocysticercosis cases.[1,2,4,6,8] Usually, spinal neurocysticercosis is commonly associated with brain involvement. But isolated spinal cysticercosis without cranial involvement is very rare.[1,2,3,4,5,6,7,8] Thoracic region is commonly involved by the isolated neurocysticercosis because of high blood supply to this region, followed by cervical, lumbar and sacral regions. Isolated cauda equina neurocysticercosis is very rare and

only 15 cases were reported in the literature to the best of our knowledge.[2,3,4,5,7,8] Spinal neurocysticercosis can be extraspinal or intraspinal.[3,4] Intraspinal can be intradural, extramedullary or intramedullary.[3,4] Most commonly, these involve the thoracic region because of preferential blood supply to this region, even though it can occur anywhere in the spine.[2,3] Usually, these are located in subarachnoid space (80%), result of CSF dissemination directly from the brain.[2,3,4] but isolated spinal cysticercosis cases were reported and these isolated cases may be because of haematogenous spread.[2,3] Most of the reported cases were multiple cysts.[2,3,4] and isolated single cyst was very rare.[5]

In our case, it is intradural, located in the subarachnoid space and is a single large cystic lesion involving the cauda equina without any other lesion elsewhere in the spine or brain, may be because of direct haematogenous spread of cysticercosis larva to the spine.[3,5] Clinical symptoms are not specific to the NCC and may not be distinguishable from other lesions.[2] The symptoms depend on the location, size, number of cysts and the presence or absence of any inflammation.[1,2,3,4,5] Most often the symptoms are due to the pressure effect by the cyst and symptoms may worsen in presence of inflammation. Inflammatory reaction may also produce symptoms by the parasite metabolism or by cyst degeneration. Choi KB reported a case of spinal neurocysticercosis in herniated disc patient, in which insidious onset symptoms may be because of inflammatory reaction produced by the cyst instead of mass effect. Symptoms may range from vague pain to cauda equina syndrome. Patient may present with radicular pain, paraesthesias, and sphincter disturbances or with typical cauda equina syndrome. Park YS, reported a case mimicking as spinal subarachnoid tumour. Mohapatra RN reported a silent neurocysticercosis case with pseudotumour cerebri. Arachnoid cysts were reported in relation to the trauma.[9,10] spinal anaesthesia, but none of the neurocysticercosis was reported following spinal anaesthesia except in our case, this patient underwent caesarean section under spinal anaesthesia after which patient became symptomatic. Trauma is having a role in development of arachnoid cyst by producing a tear in the arachnoid or by triggering a silent cyst into a symptomatic one.[9,10]

The same mechanism may be responsible for our case by triggering a silent cyst into a symptomatic one. This is the first case in the literature we are presenting, which has become symptomatic after spinal anaesthesia. Most of the reported cases were diagnosed only after the histopathological examination.[5] So in all cases of cystic lesions of the spine, we should always include the neurocysticercosis in the differential diagnosis. Plain radiographs are of less value except in cases of calcified cysticercosis.[3,4] Raised eosinophilic and white blood cell counts may be seen but not often. CSF shows low or normal glucose levels, increased proteins, lymphocytic pleocytosis and eosinophilia.[3] Serum immunomarkers may be high in cases associated with inflammation. Serum and CSF ELISA tests may help in the diagnosis of neurocysticercosis.[3,4]

MRI is the investigation of choice for detecting the cyst, because it gives non-invasive images of whole spinal cord, but it is difficult to differentiate from other intradural cystic lesions of the spine.[2,3,4,5] Cysts appear as hypointense on T1 images and hyperintense on T2 weighted images, typical of any cystic lesion and it may mimic other cystic lesions of the spine like arachnoid cyst, dermoid cyst, hydatid cyst. Contrast-enhanced MRI is mandatory for evaluating NCC, because wall of the cysticercosis is enhanced as compared to arachnoid cyst.[5] CT scan is not that useful unless in cases of calcified cysts.[4,5] Treatment of spinal neurocysticercosis is both medical and surgical according to the available literature even though still it is controversial.[1,2,3] Medical treatment is with anticysticercal drugs like albendazole or praziquantel.[2,3,4,5] Albendazole is preferred over praziquantel in view of high CSF penetration and possible high drug serum concentration in combined administration of steroids. Steroids are usually given along with anticysticercal drugs to counteract the inflammatory reactions following parasite death.[2,3,4,5] Medical treatment is useful in medically stable patients and even after surgery because neurocysticercosis is a systemic disease with focal manifestations.[2,4]

Spinal neurocysticercosis treatment is primarily surgical, especially in the presence of progressive neurological dysfunction and when the diagnosis is doubtful.[2,3,4,5] Other indications for surgery are very severe symptoms, symptom aggravation, medical treatment failure and acute aggravation of symptoms during treatment.[4] Surgical excision gives definitive diagnosis and removes the mass effect by the cyst.[2,3,4] Main aim of the surgery is complete excision of the cyst if possible without breaking the capsule.[2,3] But adhesions between cyst and nervous tissue may make total resection of cyst difficult in some cases. Prefer to have a latest MRI, because migration of cyst can occur posing difficulty in finding the cyst. Postoperative results were good in most of the cases.[1,2,3] Han SB.[4] reported bilateral motor weakness in both lower limbs which improved slowly with steroids but mild sensory impairment persisted, may be due at adhesions between the roots and cystic lesions. Patients may develop symptoms in the followup period because of adhesions between the cauda equina and dura because of chronic arachnoidal scarring.[6]

Prognosis depends on many factors such as location of the cyst, adhesions to the nervous tissue, arachnoiditis, duration of symptoms at the time of presentation.[2,3,4,5] Recurrent symptoms of pain is reported in patients with arachnoid adhesions.[4,6]

CONCLUSION: Isolated cauda equina neurocysticercosis is extremely rare cause of cauda equina syndrome, and this should be considered in the differential diagnosis of cauda equina cystic lesions. Surgical excision with histopathological examination only will provide a definitive diagnosis of these cysts in spite of so many investigations.

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