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A RARE CASE OF INTRADURAL SPINAL HYDATID CYST IN A PAEDIATRIC PATIENT

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Intradural extramedullary type of spinal hydatid disease is a rare variety of hydatid disease, and is even rarer in paediatric age group. Spinal hydatid disease should be considered in the differential diagnosis of spinal cord compression syndrome in endemic countries and should be evaluated with imaging and serological investigations. Our case was a 9- year-old boy who presented with lower back pain lasting for 8 months and progressive bilateral lower extremities weakness lasting for 2 month. Neurological examination was suggestive of lower motor neuron type of paraperesis. Magnetic resonance images of the lumbar spine showed an intradural cystic lesion displacing and compressing the lower cord and cauda equina. The cystic mass was explored with L1-L4 laminectomy and after durotomy; it was separated from cord and dura mater by hydrodissection. It contained a clear fluid. The pathological diagnosis was hydatid disease.

Key-word: Echinococcosis.

Hydatid (which means 'watery cyst' in Greek) disease is an uncommon but clinically and radiologically challenging parasitic disease in endemic areas. Bone involvement is a rare complication of this disease and spinal involvement in a child being even rarer.

Despite the introduction of modern surgical and pharmacological therapy the disease still remains difficult to cure and highly prone to recurrence. The infestation persistently erodes the spinal column, eventually leading to its destruction and neurological deterioration.

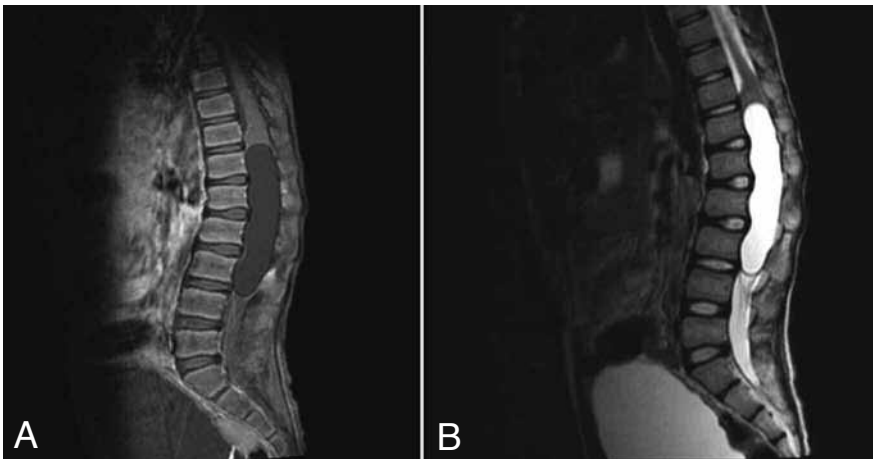

Fig. 1. — Sagittal T1 (A) and T2 (B) weighted MR sequence of the dorso-lumbar spine showing presence of a single smooth walled intradural cystic lesion extending from the level of the D11-D12 disc to L3-L4 intervertebral disc level widening of the spinal canal and posterior scalloping of the vertebral bodies. Lower spinal cord and cauda equina are compressed and displaced towards the right side.

Case report

A 9-year-old boy presented to the neurosurgical out-patients' department with complaints of back pain lasting for last 8 months as well as progressive weakness involving the lower limbs. He was also complaining of overflow urinary incontinence for a few days. On examination, there was wasting of muscles of both lower limbs (more severe on right side) with decreased power and absent tendon reflexes in bilateral knees and ankles. The chest radiograph was found to be normal. Abdominal ultrasound showed the presence of multiple, double walled cystic lesions of variable size within the right lobe of liver suggestive of hydatid cysts. MRI of the dorso-lumbar spine revealed the presence of a single, smooth walled, intradural cystic lesion extending from the level of D11-D12 intervertebral (IV) disc to L3-L4 IV disc causing widening of the involved spinal canal segment, lower spinal cord compression and displacement to the right (Fig. 1, 2). Immunologic studies were positive for Echinococcus granulosus. The erythrocyte sedimentation rate was 80 mm/h and the complete blood count showed an eosinophilia of 8%. On the basis of these radiographic, sonographic and immunological findings a diagnosis of hydatidosis was made, and treatment with albendazole was begun.

In order to relieve the spinal cord compression, laminectomy was performed between the levels of L1 and L4. The dura mater was distended between these levels; the spinal cord was compressed and displaced to the right by the cyst. The dimensions of the cyst were 7 x 2 x 2 cm (Fig. 3). The histopathologic findings confirmed the diagnosis. The patient showed steady improvement after surgery and was discharged with a healthy note.

Discussion

Hydatid disease is a zoonotic disease caused by the larval stage of the tapeworm Echinococcus sp., and is known to be endemic in Middle Eastern, Mediterranean, and Australian regions (1). The first description of spinal hydatid disease was made by Churrier in 1807 and the first surgical intervention was

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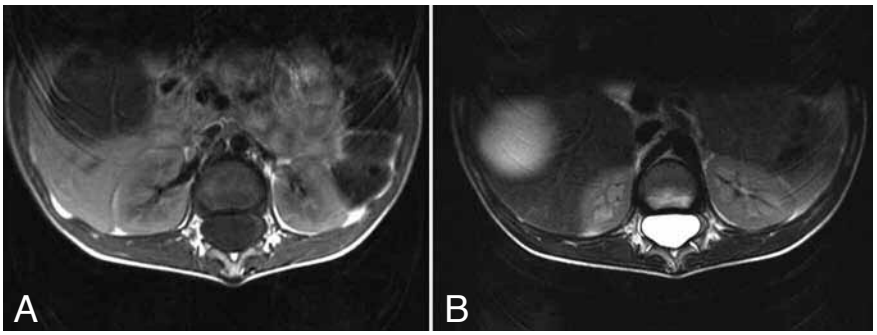

Fig. 2. — Axial T1 (A) and T2 (B) weighted MR sequence of the dorso-lumbar spine at L2 level showing the presence of a single smooth walled intradural cystic lesion causing widening of the spinal canal and compressing and displacing the lower spinal cord and cauda equina towards right side.

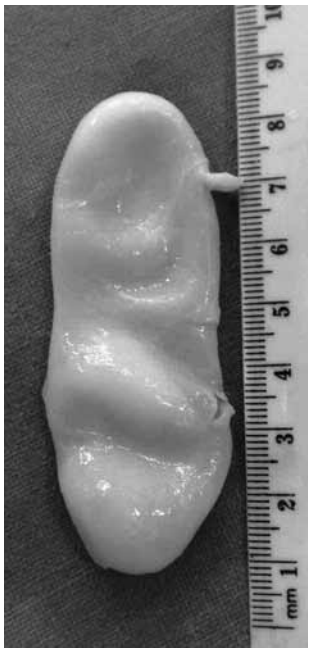

Fig. 3. — Post- operative specimen of the excised cyst showing a glistening white membrane and containing clear fluid.

reported by Reydellet in 1819.The liver is most commonly involved, followed by the lungs. Bone involvement is rare and in most of these cases concerns the spine. Spinal hydatid cyst represents less than 1% of all hydatid cyst cases (2). The distribution of the disease in the spinal area is as follows: 10% in cervical area, 50% in thoracic area, 20% in lumbar area, and 20% in sacral area. Cervical spine involvement is least common (3). Spinal hydatid cysts are categorized morphologically into 5 groups (classification of Braithwaite and Lees (4)): 1- intramedullary HC, 2- intradural- extramedullary HC, 3- extradural- intraspinal HC, 4- vertebral HC, and 5- paravertebral HC. In 90% of the cases the disease is confined to the bone and the epidural space. The first three groups are extremely rare and they are usually reported as sporadic cases. In spinal hydatid cysts, presentation differs according to the site of involvement of the spine and may vary from paraparesis (62%) or paraplegia (26%), backpain or radicular pain (55%), numbness or sensory loss (36%) and sphincter disturbance (30%) (5). On MRI, hydatid cysts are seen as spherical lesions with well defined margins and fine walls (4). The fluid content of HC is isointense with CSF on T1 and T2 WI, but the cyst wall is seen as low signal intensity rim on both T1 WI and T2 WI. After IV contrast injection, the rim generally does not enhance, and calcification of the wall of the cyst is rare. Enhanced ring-shaped wall can be seen if the cyst is infected. In our case, no contrast enhancement or calcification was present.

In the differential diagnosis of HC, archnoid cyst and epidermoid cyst of spine should always be considered, especially in children.

As a conclusion, spinal hydatidosis is a rare entity. Among these, isolated intradural- extramedullary thoracolumbar hydatid cyst cases are extremely rare. In the pre-operative period, MRI is a useful imaging modality in diagnosis of spinal HD and can play a crucial role in the management.

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