

Mediastinal Hydatid Disease with Spinal Involvement—A Rare Cause of Spinal Cord Compression

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ABSTRACT

Spinal hydatid disease accounts for less than 1% of human hydatid disease. It can very rarely lead to paraplegia. We report a rare case of spinal hydatid disease and review the pertinent literature. A young man presented with gradually progressive paraparesis and urinary incontinence. Imaging showed with thoracic extension, pathologically confirmed as hydatid cyst following surgical excision. He showed remarkable postoperative improvement and is now able to ambulate without support at 1-year follow-up. This case stands distinct in highlighting two aspects, the rare spinal location of a hydatid cyst and complete recovery of paraplegia following surgical excision.

Keywords: Echinococcus, Hydatid, Mediastinum, Spine.

How to cite this article: Chawda M, Salman ST, Chandan MB, Chandrashekhar DE. Mediastinal Hydatid Disease with Spinal Involvement—A Rare Cause of Spinal Cord Compression. *J Spinal Surg* 2019;6(1):19-21.

Source of support: Nil

Conflict of interest: None

INTRODUCTION

Cystic echinococcosis is caused by tapeworm cestode *Echinococcus granulosus*. Humans are accidental intermediate hosts. It is common in areas where cattle rearing is a prevalent occupation. While liver and lungs are commonly involved, bone involvement is seen in only 0.5–2% of cases. Amongst patients with bone involvement, vertebral involvement is seen in approximately 45% of cases.¹ It is associated with significant morbidity, disability, and mortality. Prognosis is often compared to malignancy due to the propensity for recurrence.² Here, we report case of primary hydatid cyst of the dorsal spine with an intrathoracic extension which was successfully managed with a combined posterior spinal and thoracic approach.

CASE HISTORY

A 32-year-old male with a history of playing regularly with street canines, presented with complaints of paresthesia of both lower limbs (right more than left) for 1 month followed by difficulty in walking for 15 days and urinary incontinence with constipation for 10 days. On examination, he was wheelchair-bound and unable to stand without support. He had spastic paraparesis with proximal power of grade 1/5 and distally power was 0/5. Deep tendon reflexes were exaggerated. Hypoesthesia was present below the T8 level. Post-void ultrasonography of the bladder revealed residual urinary volume of 160 mL. Clinically, features were suggestive of progressive thoracic myelopathy. Magnetic resonance imaging (MRI) showed a multi-loculated cystic lesion in the posterior mediastinal space extending into the spinal canal at T7–10 level, causing compression of the spinal cord (Figs 1 and 2). These features were suggestive of hydatid disease and, surgical excision of the lesion was planned. Forty-eight hours before surgery, oral albendazole 400 mg was started once a day. T7–T10 laminectomy and excision of the intra-spinal cystic lesion were performed followed by right postero-lateral thoracotomy, and complete excision of extraspinal hydatid cyst was achieved in a piecemeal fashion. Intra-operatively shiny pearl-like, grape-like (Fig. 3), confirmed as hydatid cysts on histopathological examination (Fig. 4). Povidone-iodine soaked gauze was used around the margins to prevent spillage and 3% hypertonic saline was used as a lytic



Fig. 1: T2 weighted axial section magnetic resonance imaging (MRI) showing a multi-loculated cystic lesion at T8 causing cord compression

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Fig. 2: T2 weighted sagittal section of MRI showing the multi-loculated cystic lesion

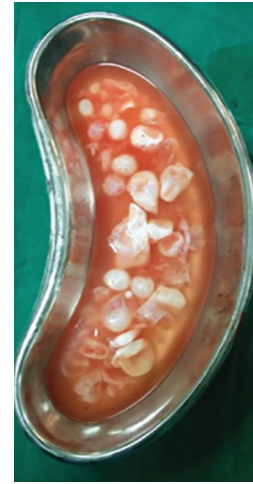


Fig. 3: Intraoperative image showing shiny pearl-like, grape like hydatid cysts after removal

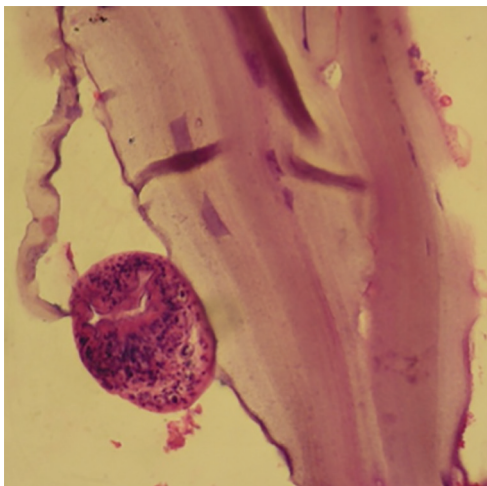


Fig. 4: Histology image from right to left shows fibros, laminar and germinal layer with brood capsule

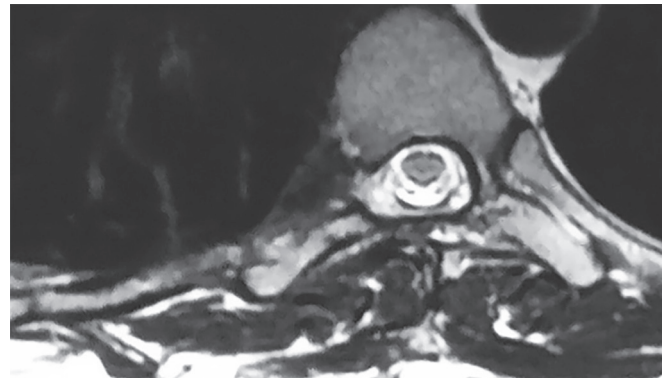


Fig. 5: Postoperative T2 weighed axial image at T8 level at 1 year

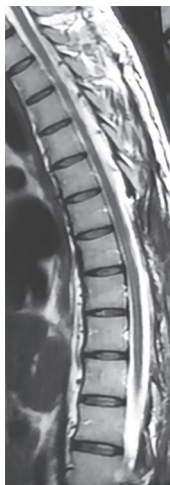


Fig. 6: Postoperative T2 weighted sagittal section 1 year after surgery at T8 level

solution. Thorough and extensive lavage of the surgical cavity with hypertonic saline was performed after the complete excision. Power gradually improved to 4/5 in both the lower limbs by the 10th postoperative day and the patient was able to walk with support by 12th

postoperative day, also being the day of discharge. He was advised to continue albendazole 400 mg daily for 24 months. There was a significant improvement in the bladder control at the time of 1st follow-up at 3 months. Follow-up was done at 3 monthly intervals. Presently, a year after surgery, he can do his routine chores, walk without support and has a normal bowel-bladder function. Follow-up magnetic resonance imaging (MRI) of the spine and high-resolution computed tomography (HRCT) chest has shown no recurrence (Figs 5 and 6).

DISCUSSION

Hydatid disease is one of the oldest diseases known to mankind, mentioned in the Talmud as “Bladders full of water”. Bremser reported the first surgically treated case of hydatid in a human patient in the year 1821.³ The larval stage of cestode *Echinococcus granulosus* leads to infestation. It is endemic in temperate climate zones. Definite hosts include sheep, goats, and cattle, while humans are accidental hosts. Primary hydatid disease is common in liver, spleen, and lungs. Extra visceral spread evolves from arterial dissemination of oncospheres. The other proposed route of spread is venous, through the port-vertebral shunts and retrograde passage of the

parasite from inferior vena cava to retroperitoneal and epidural venous plexus.⁴ Spinal involvement accounts for less than 1% of cases.

Mediastinal hydatid cysts are very rare, accounting for less than 0.1% of all cases and less than 1% of them have thoracic involvement. In a review of 74 cases; erosion of ribs, vertebra, and compression of spinal cord occurred in 57% of cases with posterior mediastinal cysts.⁵ Mediastinal Echinococcosis with rib and vertebral destruction with intraspinal extension can raise the possibility of aggressive malignant lesions like round cell tumors, lymphoma, metastasis and neurogenic tumors.⁶ Mediastinal hydatid should always be considered as a differential diagnosis of cystic lesions with intraspinal extension. Serology can often be negative, especially if the cyst is intact.⁷

Gold standard treatment for mediastinal hydatid is radical removal of germinative membrane and pericyst.⁸ The preoperative course of benzimidazoles has been reported to soften the cysts to reduce intracystic pressure, enabling surgeons to remove the endocyst easily. Postoperative chemotherapy with albendazole (albendazole 10–15 mg/kg, 1 month) or mebendazole (40–50 mg/kg, 3 months) is advised in cases where per-operative spillage of protoscolices may have occurred.⁹ If total excision is limited by the invasion of vital structures, partial peri-cystectomy following removal of the germinative membrane can be performed. Better results are obtained by combining surgical evacuation with anti-helminthic therapy. World Health Organization (WHO) recommends adjuvant chemotherapy for 2 years after surgery.

Recurrence is a rule because even after extensive surgery, as up to 40% recurrence within 2 years has been reported. Spinal stability should be the goal and has been achieved successfully in the past as shown in the case reported by Kellar et al.,¹⁰ wherein the patient was operated six times with preserved neurological status and spinal stability.

For recurrence, several authors recommend reinstating anti-helminthic pharmacotherapy. Most such patients have

a poor neurological status with up to 45% incidence of paraplegia.⁴ Recurrence is a bad prognostic marker having a reported operative mortality rate of 14.4%.⁴

CONCLUSION

Mediastinal hydatid cyst with spinal extension is a rare occurrence with a high propensity for recurrence despite complete surgical excision and adjuvant anti-helminthic treatment. This warrants a close clinical follow-up and radiological evaluation periodically.

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