Case Report

# Spinal intradural hydatid cyst causing arachnoiditis: A rare etiology of cauda equina syndrome


**ABSTRACT**

This study aims to focus on a rare presentation of spinal hydatid cyst as cauda equine syndrome and misdiagnosed as intradural extramedullary (IDEM) benign lesion on magnetic resonance imaging. In this article, we report a case of spinal hydatid cyst masquerading as IDEM tumor, and intraoperatively, we accidently find clumped granuloma with severe arachnoiditis and hydatid cyst in lumber region, which was present as bilateral S1 radiculopathy with cauda equina syndrome. An 11-year-old boy who presented with symptoms and signs of cauda equina syndrome and planned for surgical excision. His radiological impression was IDEM possibly neurofibroma. To our surprise, we found multiple intradural cystic lesions with arachnoiditis. Dissecting in plane cyst was flushed out, and surgical cavity was irrigated with 3% saline. Postoperatively histopathology and serum tests confirmed the diagnosis of hydatid cyst. Hydatid disease is rare cause of cauda equine syndrome which can be miss diagnosed on radiological investigations. A high index of suspicion should be kept especially in a young patient from the Indian subcontinent.



**Key words:** Arachnoiditis; cauda equina; hydatid cyst.


## Introduction

Low back ache *per se* has a wide spectrum of etiologies. Spinal hydatid cyst is one of the rare causes with poorly understood pathogenesis and often misdiagnosed. Man is both an intermediate and accidental host in the life cycle of *Echinococcus*. Almost all organs of the human body have been reported to harbor *Echinococcus*. The presence of parasitic cyst causes compression and patients present either with compressive myelopathy or radiculopathy. Spinal hydatids have classical radiological features on magnetic resonance imaging (MRI), but the diagnoses are often missed, especially in the lack of diffusion weighted images. When the cysts do not show characteristic features on MRI, the differential diagnosis is quite difficult as the lesion can imitate various other pathologies. Missing the diagnosis of spinal hydatid can be hazardous, especially if the cyst ruptures intraoperatively. In this article, we report a case of spinal hydatid cyst masquerading as posterior disc bulge with features of radiculopathy.


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## Case Report

An 11-year-old boy presented with low back ache and bilateral lower limb radiculopathy in S-1 and S-2 dermatome. There were early bladder and bowel involvement. On examination, the patient had moderate Oswestry disability with power 4/5 Medical research council (MRC) grade in both lower limbs and 5/5


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in both upper limbs. Deep tendon reflexes were 2+ all four limbs, and bulbocavernosus and superficial anal reflexes were normal. There was no sensory loss. Other hematological investigations were within normal limits. The prospective radiology shows a contrast enhancing mass on right side at level of L4-S1 spine with extension along nerve roots. The cord intensity changes were present, there were no bony changes. Diffusion images were not available. A possibility of neurofibroma was kept in mind. Decompression laminectomy was planned, but intraoperatively, there were clumped nerve roots without any cleavage. There was thick arachnoiditis and roots were adhered and displaced which made extraction of cyst very difficult. The radiology was reassessed, and the laminectomy was further extended cranially. To our surprise, we found an intradural single hydatid cyst. Cyst was removed in to [Figure 1] and the surgical field was irrigated with 3% saline. Postoperative period patient had improvement in pain, but weakness persisted. The patient was kept on albendazole for 3 months in follow-up. Repeat MRI after 3 months showed no evidence of cyst. Histopathology was consistent with hydatid cyst.

## Discussion

Our case report emphasizes the difference between radiologic "misses" and "surprise." We are reporting a case of radiological "surprise" with a well-established radiological diagnosis of prolapsed intervertebral disc which was reviewed by more than one specialist. In our case, there was discrepancy between radiological diagnosis and intraoperative findings. Pathology established the diagnosis of hydatid disease (HD) which was preoperatively thought to be herniated disc. Misdiagnosis or error is often under reported. A recent review found error rate among radiologists in their day-to-day practices averages 3%–5%, comparing clinical diagnoses with postmortem

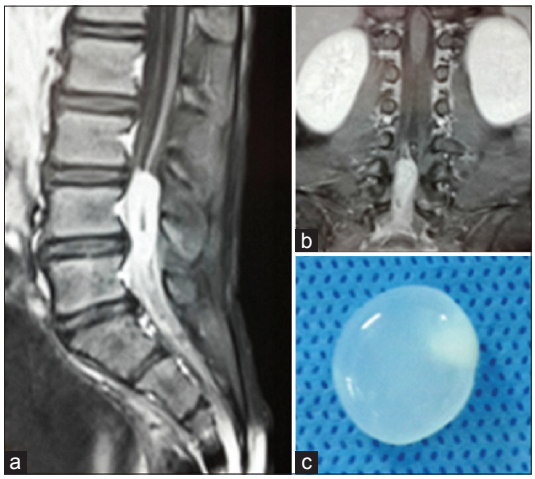


**Figure 1: (a and b) Sagittal and coronal section of magnetic resonance imaging lumbosacral spine showing hyperintense intradural extramedullary lesion in L5–S1 region, (c) unruptured hydatid cyst**

diagnoses. The same study found that in 26% of cases, a major diagnosis was missed clinically.[1] HD is caused by larval stage of *Echinococcus* with two forms involved in spinal disease including *Echinococcus granulosus* and *Echinococcus multilocularis*. HD involves almost all organs in the body. Spine is one of the rare locations for echinococcosis with incidence varying from 0.2% to 1% in vertebral column 40%–50% of which involves spine.[2] The disease most commonly involves the thoracic spine, followed by the lumbar, sacral, and cervical spine.[3] With such low incidence and rarity even in endemic regions, the above disease is often missed as a cause of low back ache.[4] Few case reports suggest that missing a diagnosis of hydatid could be devastating and should be kept as a differential diagnosis if one encounters cystic lesion in spine. Spillage of cystic fluid in surrounding may cause grave inflammatory reaction.[5] There are many case reports suggesting spectrum of signs and symptoms by which spinal HD can present ranging from simple low backache to paraplegia.[6] MRI is the preferred imaging modality in the diagnosis of hydatid cysts.[7] Hydatid cyst has no special features on MRI. The presence of a markedly hypointense cyst wall on T1- and T2-weighted (T1W/T2W) images is characteristic of HD.[8] In endemic countries, some MRI features may suggest an infection; these features consist of Dense calcification rim around a lytic lesion, a water like signal for daughter cysts, a muscle like one for the parent cyst in T1W with multiloculation, and hyperintensity of parent and daughter cysts on a T2W with the appearance of rose or wheel.[9] Turgut *et al*. have described many typical and atypical signs suggesting HD such as perifocal edema, nonhomogeneous contrast enhancement, multiplicity or septations and calcification, various unusual manifestations due to rupture or infection of the cyst. Familiarity with these signs is critical for diagnosis of spinal *Echinococcus*.[10] Braithwaite and Lees had classified hydatid spine disease in five types:

1. Primary intramedullary hydatid cyst
2. Intradural extramedullary hydatid cyst
3. Extradural intraspinal hydatid cyst
4. HD of the vertebrae; and
5. Paravertebral HD.[11]

The mainstay management of HD is surgery followed by chemotherapy for 1–3 months. The surgical procedure varies from simple decompression to excision of cyst depending on spread of disease. Strict follow-up is critical in the management of these patients, and regular MRI scans should be done during the postoperative period to ensure that any recurrence is detected early as despite optimal, and medical therapy, recurrence and thus reoperations are generally needed. The recurrence rate of 30%–40% is described in literature.[12] Pamir *et al*. have a recurrence rate of 18% in his

case series with neurological improvement in 63% of cases.[13] Few case reports suggest recurrence rate as high as 100%[14] but with improved diagnostic imaging quality and microsurgery options recurrence rates are in decreasing trend.

## Conclusion

HD is one of rare causes of radiculopathy and myelopathy and should be kept in differential diagnosis especially in thoracic segment involvement in patients of the Indian subcontinent. The atypical features should be reviewed by radiologist while reporting MRI of spine.

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### Conflicts of interest
There are no conflicts of interest.

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