CASE REPORT

Intraspinal hydatidosis with retroperitoneal extension: an uncommon location

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SUMMARY

Hydatidosis is a ubiquitous disease that is endemic in India. It most commonly involves the liver (75%) and lungs (15%) with only 10% occurring in the rest of the body. Primary hydatid cyst in the spinal canal is extremely rare. Intraspinal hydatid accounts for 0.5–1% of the cases and carries a poor prognosis. It presents as a diagnostic and therapeutic challenge. We present one such case of a 64-year-old man with associated radiculopathy and myelomalacia.

BACKGROUND

Hydatidosis is a zoonotic infection and an important health problem in some countries including India. It is caused by the larval stages of *Echinococcus granulosus*.¹ Intraspinal involvement is seen only in 0.5–2% of cases.² Furthermore, sacral and lumbar vertebral involvement is extremely rare with commonest occurrence at dorsal level.³

According to Braithwaite and Lees,⁴ spinal hydatid cysts are classified into five types:

1. Primary cysts of the spinal cord (intramedullary),
2. Primary intradural cysts (intradural extramedullary),
3. Primary extradural cysts,
4. Hydatid cyst of the vertebrae,
5. Paravertebral lesions extending to the spinal column.

The first three groups of hydatid disease are rare, and only sporadic cases have been reported. The present case showed no evidence of pulmonary or chest wall hydatidosis on CT scan and was classified as type 5 spinal hydatid cyst.

CASE PRESENTATION

A 64-year-old man presented with a six-month history of progressive low back pain and difficulty in walking. He also reported urinary incontinence suggesting late onset bladder involvement. General physical examination showed abdominal distension. Neurological examination revealed mild weakness in bilateral lower limbs with power of grade 4/5 in knees and ankle. Chest X-ray, CT of the thorax and abdomen showed no evidence of pulmonary or hepatic hydatid disease. MRI of the lumbosacral spine, a non-enhancing heterogenous cystic lesion, hypointense on T1 and hyperintense on T2 was noted within the spinal canal. The cystic lesion demonstrated clumped membranes with peripheral enhancement in the spinal canal extending from L3 to S2 along with a huge left paraspinal and retroperitoneal component extending from L5 to S1.

Bilateral sciatic and L3–L4 root inflammation was also present. The lesion displaced the left psoas medially encasing the middle third of the left ureter ([figure 1](#)). In view of clinicoradiological evidence a diagnosis of primary type 5 intraspinal hydatidosis was made. Surgical decompression was done by combined anterior and posterior approach.

INVESTIGATIONS

MRI of the lumbosacral spine, a non-enhancing heterogenous cystic lesion, hypointense on T1 and hyperintense on T2 was noted within the spinal canal. The cystic lesion demonstrated clumped membranes with peripheral enhancement in the spinal canal extending from L3 to S2 along with a huge left paraspinal and retroperitoneal component extending from L5 to S1. Bilateral sciatic and L3–L4 root inflammation was also present. The lesion displaced the left psoas medially encasing the middle third of the left ureter.

TREATMENT

Patient was treated with anti-helminthics preoperatively and a double-J (DJ) stent was inserted in the left ureter. The retroperitoneal component was addressed through a left paramedian incision and extraperitoneal approach after identifying the spermatic cord, ureter and gonadal vessels ([figure 2](#)). The lesion was confirmed via palpation and hypertonic saline was instilled into it prior to excision. The intraspinal component was excised via a posterior approach (prone) with L3–L5 laminectomy and excision of the intradural extramedullary lesion via an adequate dural opening and watertight duroplasty thereafter.

Postoperatively, the patient did well neurologically and at the time of discharge was able to walk with minimal support. DJ stent was removed after 1 month and the patient was continued on albendazole (400 mg once daily) for 3 months. He has been asymptomatic at 6-month follow-up ([figure 3](#)).

OUTCOME AND FOLLOW-UP

Intraspinal hydatidosis is an extremely rare entity associated with high morbidity. Integrated therapeutic approach in the form of surgical decompression and medical treatment with mebendazole is the preferred management of these patients.

DISCUSSION

Paraplegia caused by compression of spinal cord is the most serious complication of intraspinal hydatid cyst.⁵ Cysts located intraspinally have a

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Figure 1 Preoperative MRI of the L-S spine (axial, coronal and sagittal sections) showing the intraspinal cysts with retroperitoneal extension.

tendency to rupture spontaneously. For this reason the high recurrence rate (30–40%) is still a major problem in management.² Surgical excision after decompressive laminectomy and additional medical treatment is still the critical mainstay and most effective treatment.^{6–7} The present case of intraspinal hydatid cyst classified as a combination of intraspinal extradural, vertebral and paravertebral forms according to the Braithwaite

and Lees' classification. The patient was treated by surgical decompression. Long-term results after surgical decompression are generally poor.^{3–8} However, our case showed remarkable improvement following the surgery. Further, it has been suggested that irrigation with hypertonic saline solution during the operation might be beneficial to prevent recurrence following spontaneous or intraoperative rupture.

Figure 2 Intraoperative image showing excision of the hydatid cyst.

Learning points

- ▶ Intraspinal hydatid accounts for 0.5–1% of the cases and carries a poor prognosis.
- ▶ Radiological investigations usually clinch the diagnosis since serology for amoebiasis is usually negative or equivocal.
- ▶ Integrated therapeutic approach in the form of surgical decompression and medical treatment with mebendazole is the preferred management of these patients.
- ▶ Irrigation with hypertonic saline solution during the operation might be beneficial to prevent recurrence following spontaneous or intraoperative rupture.

Competing interests None.

Figure 3 Postoperative MRI of the L-S spine (axial, sagittal and coronal sections) showing near total excision of the cyst with postoperative changes and fibrosis.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed..

REFERENCES

- 1 Tiseo D, Borrelli F, Gentile I, *et al.* Cystic echinococcosis in humans: our clinic experience. *Parassitologia* 2004;46:45–51.
- 2 Adilay U, Tuğcu B, Gunes M, *et al.* Cauda equina syndrome caused by primary lumbosacral and pelvic hydatid cyst: a case report. *Minim Invasive Neurosurg* 2007;50:292–5.
- 3 Charles RW, Govender S, Naidoo S. Echinococcal infection of the spine with neural involvement. *Spine* 1988;13:47–9.
- 4 Braithwaite PA, Lees RF. Vertebral hydatid disease. Radiological assessment. *Radiology* 1981;140:763–6.
- 5 Kotil K, Tari R, Savas Y. Medical treatment of primary extradural solitary lumbar hydatid disease. *J Clin Neurosci* 2010;17:793–5.
- 6 Güneş M, Akdemir H, Tuğcu B, *et al.* Multiple intradural spinal hydatid disease: a case report and review of literature. *Spine (Phila Pa 1976)* 2009;34: E346–50.
- 7 Lakhdar F, Arkha Y, Rifi L, *et al.* Spinal intradural extramedullary hydatidosis: report of three cases. *Neurosurgery* 2009;65:372–6; discussion 376–7.
- 8 Pamir MN, Akalan N, Özgen T, *et al.* Spinal hydatid cysts. *Surg Neurol* 1984;21:53–7.

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