

## INTRASPINAL HYDATID CYST- RARE CASE REPORT

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### PRESENTATION OF CASE

A 20-year-old male presented to us with complaints of urinary incontinence and right lower limb weakness. On examination the patient had continuous urinary dribbling with lower motor neuron type of bladder involvement. The lower limb weakness was limited to the dorsi-flexion of the ankle and the toes with extensor hallucis longus (EHL) weakness. The deep tendon ankle reflex was depressed on the right side and the plantar reflex was mute. Power and reflexes on the left side were normal.

### CLINICAL DIAGNOSIS

Diagnosis of a cauda equina compression was made which was confirmed on Magnetic Resonance Imaging (MRI), which showed a T2 Hyper-intense and T1 Hypo-intense, irregular walled cystic lesion in the spinal canal extradural space at the L5 to S2 vertebral levels extending more to the right side. The lesion was non-enhancing on contrast and had a hyper-intense thin rim. (Figure 1, 2, 3, 4)

### DIFFERENTIAL DIAGNOSIS

On the MRI a provisional diagnosis of a benign cyst more likely a Tarlov's cyst was made. Other differentials which were thought of were spinal tumours like cystic ependymoma or arachnoid cyst. Hydatid cyst of the spine was not thought of until surgical findings were suggestive of the same.

### PATHOLOGICAL DISCUSSION

Hydatid disease is the most widespread zoonosis caused by *Echinococcus granulosus*. Liver and lungs are the most common sites. Bone involvement is rare and reported in 0.5%–4% with spinal involvement reported in 50% of these cases.<sup>1</sup> In the vertebral column it affects the lumbar, thoracic and cervical region, in decreasing order of

frequency.<sup>2</sup> Hydatid Disease (*Echinococcus granulosus*) is endemic in the Middle East as well as other parts of the world, including India, Africa, South America, New Zealand, Australia, Turkey and Southern Europe.<sup>3</sup> Infestation by hydatid disease in humans most commonly occurs in the liver (55-70%) followed by the lung (18-35%); the two organs can be simultaneously affected in about 5-13% of cases.<sup>3</sup> Other less commonly involved organs are the Brain (1-2%), the bony skeleton (0.5 -4%) and cardiac (0.02 to 2%). (1, 3) Hydatidosis of the spine was first described by Churrier in 1807.<sup>4</sup> Primary vertebral hydatid disease without any other systemic involvement can occur with direct porto-vertebral venous shunts.<sup>2</sup> The cyst can be epidural and may be single or multiple. Intradural and extramedullary involvement is rare.<sup>5</sup> 45% of such patients are under 30 years of age. Generally, spinal hydatid cyst disease presents with radicular symptoms or symptoms of cord compression.<sup>6</sup> Immunodiagnostic tests are helpful with a diagnostic sensitivity of 90%.<sup>7</sup> Different serological tests are being carried out for the diagnosis, screening and post-operative follow up for recurrence. These include the hydatid immune-electrophoresis, enzyme-linked immunosorbent assay (ELISA), latex agglutination and indirect haemagglutination (IHA) test.<sup>3</sup>



**Figure 1. T2 Weighted MRI Sagittal Image Showing the Hyperintense Cystic Lesion in the L5-S Region**

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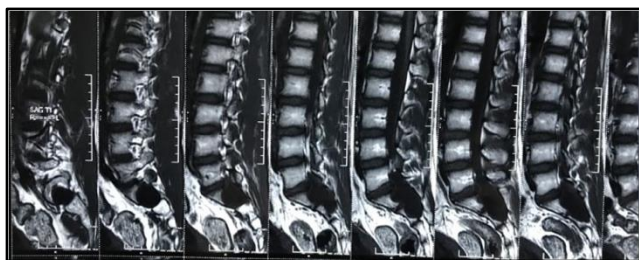
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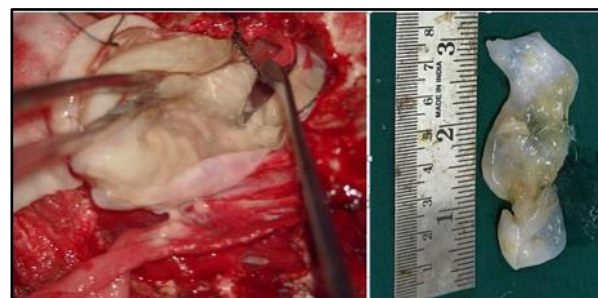
**Figure 2. T1 Weighted Pre-contrast MRI Sagittal Image showing the T2 Hypointense Cyst**



**Figure 3. T2 Axial Image of the Cyst Extending to the Extradural Space on the Right Side**



**Figure 4. Post Contrast MRI Showing Non-Enhancing Cyst with a Rim on Hyperintense Wall**



**Figure 5. Intraoperative Image of the Cyst with Post-op Image Showing the Size of the Capsule Excised as a Whole**

Sl. No.	Authors	Year	No. of Cases	Location	Treatment
1.	Pluchino and Lodrini	1981	1	T10-L2	Surgery
2.	Wani et al.	1989	1	T9-T10	Surgery
3.	Kars et al.	1990	1	C5-6	Surgery
4.	Bavbek et al.	1992	1	T5-T9	Surgery
5.	Tekkök and Benli	1993	1	L2-L5	Surgery
6.	Baysefer et al	1996	2	T5-T6, T6	Surgery
7.	Pandey and Chaudhari	1997	1	S1-S2	Surgery
8.	Bayar et al.	1997	1	L5-S1	Surgery
9.	Berk et al.	1998	1	T7-T9	Surgery
10.	Bouklata et al.	2000	1	T8-T11	Surgery
11.	Karadereler et al	2002	1	L2-L5	Surgery
12.	N. K. Sharma et al.	2003	1	L1-L2	Surgery
13.	Layadi F et al	2005	1	Sacral	Surgery
14.	Adilay et al.	2009	1	Lumbo-Sacral	Surgery
15.	Present case	2018	1	Lumbo-Sacral	Surgery

**Table 1. Review of Literature of All Reported Cases of Primary Spinal Extradural Hydatid Cysts**

## DISCUSSION OF MANAGEMENT

Patient was posted for surgery and a L5 to S2 right sided laminotomy was performed. Intra-operatively an extradural well encapsulated cyst measuring 3cm x 2 cm with multiple internal septae was found with whitish mucinous fluid in it. A classical friable whitish wall was seen (Figure 5).

The cyst was excised with its wall with minimal spillage of contents. Hypertonic saline soaked cottonoids were used during and after the procedure as there was suspicion of hydatid cyst (Figure 6).





**Figure 6. Intraoperative Image Showing the Field Soaked in Hypertonic Saline Cottonoids**

There was no dural involvement and the exiting nerve root was freed as soon as the cyst was excised. Histopathology confirmed the classical findings with multiple cystic structures with laminated membrane with few scolices. Patient was started on a course of Albendazole 400 mg twice a day for a period of 2 months. There were no other systemic hydatid cysts which were evaluated with a HRCT of the thorax and a Computerised tomography (CT) of the abdomen and brain. The urinary complaints of the patient improved post operatively with minimal weakness in the right ankle dorsiflexion at 2 months follow up. The treatment of hydatid cysts is principally surgical. The operative procedure of choice is laminectomy with excision of the cyst. However, decompression with anterior approaches and fusion also gives good results.<sup>5</sup> In the series published by Necmettin Pamir M et al,<sup>5</sup> neurological improvement was seen in 63% of the cases and recurrence in 18%. Overall, a recurrence rate of 30-40% is described in liver and lung cysts but recurrence is rare in spinal hydatid.<sup>8</sup> There is a correlation between cyst localization and recurrence. In the epidural osseous type of cysts, there are microvesicles which are spread diffusely inside the bone ("cystic disease of the vertebrae") and these cysts usually rupture during the surgical procedure causing recurrence.<sup>8,9</sup> Pre- and post-operative one month courses of Albendazole and two weeks of Praziquantel should be considered in order to sterilize the cyst, decrease the chance of anaphylaxis, decrease the tension in the cyst wall (thus reducing the chances of spillage during surgery) and to reduce recurrence after surgery. Intra-operatively the use of hypertonic saline helps prevent spread and anaphylactic reactions.<sup>3</sup>

## FINAL DIAGNOSIS

Primary Spinal Extradural Lumbo-Sacral Hydatid Cyst.

On review of literature only 15 cases have been reported of the same (Table 1).<sup>2,10,11,12</sup> Of the reported cases only two cases have involved the lumbar and sacral spine together. The rare nature of this disease, especially in non-endemic areas warrants careful reporting of these cases to highlight their management strategies.

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