CASE REPORT

SPINAL HYDATID CYST: CASE REPORT WITH REVIEW OF LITERATURE

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ABSTRACT: Spinal hydatid cyst is a rare and a serious manifestation of the parasite Echinococcus, involving less than 1% patients with hydatid disease. Intraduralhydatid cysts are extremely rare compared to other types of spinal hydatid cysts. We report a case of a 28-yr-old female who presented with history of dorsal backache and stiffness since last 3 months with progressive increasing in severity. Clinical examination revealed tenderness of upper dorsal spine on palpation, with mild lower motor neuron type weakness of both lower limbs. A plain radiograph and MRI of the dorsal spine was advised and a provisional diagnosis of spinal hydatid cyst was given which was confirmed per-operatively. A better understanding of this rare but clinically challenging disease is intended by reporting this case.

INTRODUCTION: Hydatid disease is caused by infestation of the larval stage of the parasitic tapeworm Echinococcus granulosus, a helminth belonging to the cestode group. The most common sites of infection are liver (75%), lung (15%), brain (2–4%), and genitourinary tract (2–3%). Bone involvement is very rare (1%) and in most of these cases involves the spine.

CASE REPORT: A 28-yr-old female presented with history of dorsal backache and stiffness since last 3 months, progressively increasing in severity. Recent onset of lower limb weakness was also reported. There was no history of fever, weight loss or chest pain.

Clinical examination revealed tenderness of upper dorsal spine on palpation with mild lower motor neuron type weakness of both lower limbs. No significant incontinence of bowel or bladder function was observed. Plain radiograph and MRI of the dorsal spine was advised.

Plain radiograph of dorsal spine AP view revealed well-defined homogeneous opacity in left paraspinal region at D5/D6 level obliterating the left paraspinal line and forming obtuse angle with the left lung parenchyma. Mild erosions of left lateral margin of D5 vertebral body were also observed to suggest paraspinal infective pathology. No significant rib erosions were appreciated (Figure 1).

MRI of the dorsal spine revealed multiseptated predominantly cystic lesion on sagittal STIR images involving D5 vertebral body and their posterior elements (Figure 2). Partial collapse of the involved vertebral body observed on sagittal T1W images (Figure 3) with intermediate to low signal intensity lesion replacing the normal osseous marrow with extramedullary spinal extension at D5 level (Figure 4) causing compressive myelopathy. MR myelography revealed multicystic lesion in left paraspinal and intraspinal location causing severe thecal stenosis (Figure 5). A provisional diagnosis of spinal hydatid cyst was given.

Patient underwent surgical exploration and there were multiple daughter cysts seen in D5 vertebral body and with right paraspinal and intraspinal extension (Figure 6). Postoperative recovery was uneventful.

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DISCUSSION: Hydatid is a Greek word meaning “watery cyst”¹. Hydatid disease is caused by infestation of the larval stage of the parasitic tapeworm *Echinococcus granulosus*, a helminth belonging to the cestode group. Far less commonly infestation by *Echinococcus Multilocularis* or *Oligoarthus* may be responsible for the disease.²

Although rare in northern Europe and the United States, it is a considerable cause of morbidity in large geographic regions. The disease is especially prevalent in areas where livestock is raised in association with dogs: the Middle East, the Mediterranean, South America, Australia, India and Northwest China, with an incidence that ranges from 3 to 50 cases per 100,000 inhabitants.³ Adult worms mature in the intestine of the dog (definitive host), and the eggs are released in the faeces. Intermediate hosts such as sheep and cattle ingest the eggs. Humans contract the disease either by direct ingestion of parasitic eggs from contact with dogs or indirectly from contaminated water or food. The cysts are transported from the intestinal wall via the blood stream to different organs.

The most common sites of infection are liver (75%), lung (15%), brain (2–4%), and genitourinary tract (2–3%). Bone involvement is very rare (1%) and in most of these cases involves the spine. Disease usually spreads over the spine by the direct extension from pulmonary, abdominal, or pelvic infestation and most commonly affects the dorsal region of the spine.⁴

Hydatid cysts of the spine constitute 1% of all cases of hydatidosis and are most commonly located in the thoracic spine and less so in the lumbar, sacral and cervical regions.¹

According to Islekel et al⁵ spinal hydatidosis was first described by Churrier in 1807 and the first surgical intervention was reported by Reydellet in 1819.

The disease usually begins in the vertebral body preferentially in the center of the vertebra with a predilection for involvement of the pedicle. The intervertebral disc is usually spared. Perforation of the cortex and periosteum results in extraosseous extension which may be extraspinal or intraspinal. Primary extradural hydatid cyst is rare and primary intramedullary disease extremely rare. Parasites are considered to reach the highly vascularised centre of the vertebral body through portal vertebral venous shunts.⁶

Braithwaite and Lees⁷ have classified this disease into 5 type: 1) primary intramedullary hydatid cyst; 2) intradural extramedullary hydatid cyst; 3) extradural intraspinal hydatid cyst; 4) hydatid disease of the vertebrae; and 5) paraspinal hydatid disease.

Spinal hydatidosis is predominantly seen in adults, usually men of age 21–40 years. Because of the slow growth of the parasite, it may take several years before the disease becomes clinically evident. Patients most commonly present with a long history of back or radicular pain, depending on the exact localization of the cysts.⁸ Lower extremity weakness, paraplegia, sensory deficits, altered deep tendon reflexes, sphincteric dysfunction and cauda equina syndrome are frequently seen during the course of the disease.⁹ In 20% of the cases, symptoms are elicited by trauma.⁵

Suspicion should arise in cases of persistent back pain or progressive neurologic signs suggestive of spinal cord compression in endemic countries

Definitive diagnosis of hydatid disease remains a histopathological one. Casoni and other serodiagnostic tests are inconclusive, especially in extrahepatic disease, with sensitivity being as low as 25 to 56%.¹ Plain x-rays may reveal the characteristic vertebral moth eaten lesions with surrounding sclerosis and calcification of the adjacent soft tissues, which is indicative but not diagnostic of the disease.^{1,9}

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CT scan and MRI are the current diagnostic modalities of choice. Irregular bony erosions along with multilocular, nonenhancing flattened sausage shaped lesions with very thin, nonseptated walls are the hallmark of vertebral hydatid disease.¹⁰

Though CT can efficiently demonstrate bony erosions and the extent of the lesion, MR can demonstrate any cord compression throughout the length of the spinal cord and thus is the investigation of choice.¹

On MRI, hydatid cysts appear as well-circumscribed, cystic lesions, with CSF-like signal intensities. The cyst wall is usually thin and regular with no septations.¹¹ The cysts appear hypointense on T1W images. On T2W images they appear hyperintense with sharply defined, hypointense cyst wall which shows mild enhancement following intravenous gadolinium, reflecting the vascularity of the pericyst. No contrast enhancement is seen after intravenous gadolinium-enhanced MRI either in extradural or intradural hydatid cysts.¹²

"Bunch of grapes" appearance can be seen due to extradural spread of hydatid cysts through widened neural foramina into the muscle planes.¹¹

Jena et al¹³ pointed out that the intensity differences in T2W sequences of MRI can also determine the viability of the cyst. Decrease in hyperintensity and an increase in the intensity of the cystic wall signal are indicative of a succumbed cyst.

MRI is superior to CT scan when there is involvement of soft tissues or neural involvement due to its higher soft tissue resolution and the better delineation of CSF and the spinal cord.¹⁴

A number of pathologic conditions may mimic spinal hydatid cyst and pose difficulties in the diagnosis. The most important amongst them is Pott's disease, or tuberculosis of the spine, which closely resembles vertebral hydatid disease. The risk of false diagnosis is particularly high in regions where both tuberculosis and hydatidosis are endemic. Evidence of involvement of intervertebral disk with increased bony reaction and sparing of the pedicles and the contiguous rib are suggestive of Pott disease.¹⁵

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Figure-1

Plain radiograph of dorsal spine – AP view: reveals homogeneous opacity mass lesion in left paraspinal region with erosions and partial collapse of d5 vertebral body.

Figure-2

STIR MRI – Sagittal sequence: reveals fluid signal intensity multicystic lesion.

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Figure-3

Figure-4

Figure 3 and 4 – T1W MRI – Sagittal and Axial sequences, respectively: reveals intermediate to low signal intensity mass lesion with septations in paravertebral region with intraspinal extension causing compressive myelopathy. Partial collapse of D5 vertebral body is also observed.

Figure-5

MR Myelography: reveals multicystic extramedullary mass causing compressing myelopathy.

Figure-6

Peroperative images reveals multiple white colored daughter cysts in operative field.

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