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Extradural Hydatid Cyst of the Lumbo-Sacral Spine

Primary spinal hydatid cyst is a rare and uncommon entity but a significant manifestation of hydatid disease. Here, we report a case of primary extradural hydatid cyst of the sacral region causing cord compression. Pre-operative differential diagnosis was that of Tarlov cyst owing to the radiological appearance and location of the cyst. The diagnosis of hydatid cyst was established intra-operatively which was later confirmed by histopathology report. Hydatid cyst may not fall under differential diagnosis of extradural lesions of the spine due to its rarity but should be kept under high suspicion in endemic countries.

Key words: Extradural, Hydatid cyst, Spinal cord compression

Extradural hydatid cyst of the lumbo-sacral spine

Hydatid disease is caused by the parasite *Echinococcus granulosus*, a helminth belonging to the cestode group. The liver is the most common site of the hydatid cysts, followed by the lungs. Cysts in the spleen, kidneys, heart, bone and central nervous system are less common. Hydatidosis of the bone occurs in 0.5 - 3% of all cases; the vertebral column is involved in 50% of these. Most common site of vertebral hydatidosis is the thoracic vertebrae and cervical, lumbar vertebrae and sacrum are rare sites to be involved. The spread of the disease to the spine is either by direct extension from pulmonary or abdominal infestation. Primary spinal hydatid disease without any systemic foci is extremely rare. Most commonly presenting symptoms are radiculopathy, myelopathy and/or local pain owing to bony destructive lesions, pathological fracture and consequent cord compression.

It should be considered under pre-operative diagnosis in patients from endemic countries who present suspicious spine lesions. Pre-operative diagnosis can help prevent the rupture and dissemination of cyst, prevent anaphylaxis during surgery and prevent recurrence with appropriate treatment post-operatively.

Case Report

A 23-year-old male presented with complaints of progressively increasing bilateral lower limb pain, radiating up to the toes. Pain was described as pricking and burning type associated with tingling sensation and numbness. Numbness was more on the right and pain was more on the left lower limb. There was slight restriction in walking. Limb pain was often associated with back pain usually during activity like bending. General physical examination revealed no abnormality. On straight leg raising test both legs were found to be 80 degrees, extensor hallucis longus (EHL) and flexor hallucis longus (FHL) 5/5 and sensory examination showed reduced sensation on S1 dermatome on the right.

Magnetic resonance imaging (MRI) of the lumbo-sacral spine revealed an extradural cystic lesion in the postero-left lateral aspect of sacral spine causing erosion of the adjacent bone, extending outline through the neural foramen at S1-S2 showing fluid signal in all pulse sequences. Other investigations revealed no abnormality. Serum ELISA for hydatid cyst test was negative.

Patient underwent L5 laminectomy. Sacrum was exposed and a thick capsulated cystic lesion was found in the extradural space. Multiple cystic lesions were noted and removed. Posterior end plate of L5 vertebra was eroded by the lesion. Eroded bone was also removed. The operative field was then washed with normal saline and

hydrogen peroxide. Histology report confirmed it to be hydatid cyst.

Post operative period went well and patient's pre-operative symptoms subsided. Patient was started on albendazole therapy, which was continued for a month.

Discussion

Hydatid cyst of the lumbo-sacral spine is a rare form of parasitic infection, causing focal neurological signs. Cases of hydatid cyst of spine account for 1 % of all cases of hydatid disease¹. Hydatid cysts are usually located at the thoracic level (52 %), followed by lumbar (37 %), and then cervical and sacral levels¹. Braithwaite and Lees have classified this disease into 5 types²: 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.

Echinococcus affecting the spine was first described by Churrier in 1807³. The disease has a characteristic geographic distribution and is prevalent in most of the sheep-raising regions of the world^{4,5}. Echinococcosis in humans occurs as a result of infection by the larval stage of taenia cestodes of the genus *Echinococcus*. Six species have been recognized, but four are of public health concern: *Echinococcus granulosus* (which causes cystic echinococcosis), *Echinococcus multilocularis* (which causes alveolar echinococcosis), *Echinococcus vogeli* and *Echinococcus oligarthrus* (which cause polycystic echinococcosis)⁶. *Echinococcus granulosus* is a cestode whose life cycle involves dogs and other canines as definitive hosts for the intestinal tapeworm as well as domestic and wild ungulates as intermediate hosts for the tissue-invading metacestode (larval) stage. The metacestode (echinococcal cyst) is a fluid-filled, spherical, unilocular cyst that consists of an inner germinal layer of cells supported by a characteristic acidophilic-staining, acellular, laminated membrane of variable thickness⁷. The adult *Echinococcus granulosus* (3–6 mm long) resides in the small bowel of the definitive hosts, dogs or other canines. Gravid proglottids release eggs that are passed in the feces. After ingestion by a suitable intermediate host (under natural conditions: sheep, goat, swine, cattle, horses, camel), the egg hatches in the small bowel and releases an oncosphere that penetrates the intestinal wall and migrates through the circulatory system into various organs, especially the liver and lungs rarely spine. In these organs, the oncosphere develops into a cyst that enlarges gradually, producing protoscolices and daughter cysts that fill the cyst interior. The definitive host becomes infected by ingesting the cyst-containing organs

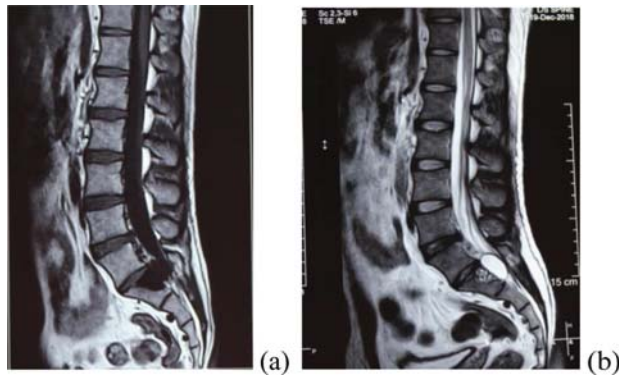


Figure 1 (a) and (b) showing the sagittal MRI of lumbo-sacral spine with extradural cystic lesion in the postero-left lateral aspect of sacral spine causing erosion of the adjacent bone



Figure 3: After removal of the cystic lesion/ hydatid cyst

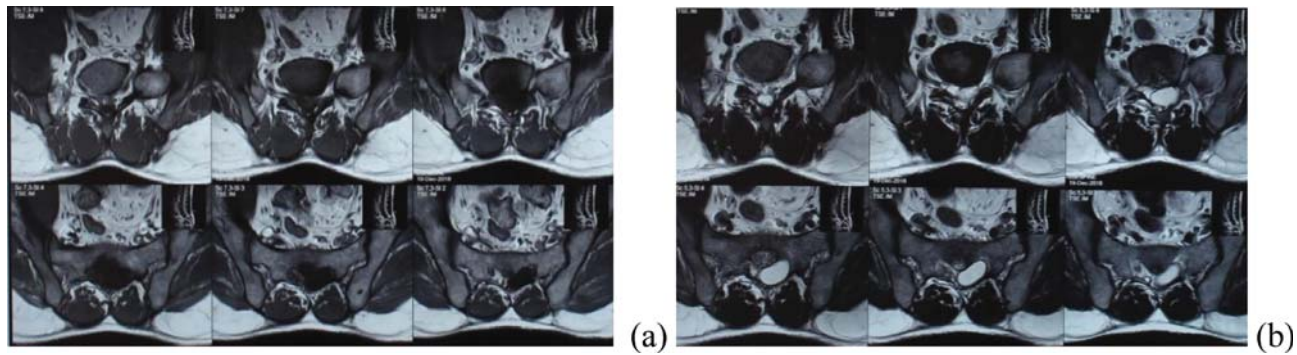


Figure 2 (a) and (b) showing axial view of cystic lesion in the sacral region, compressing the nerve root

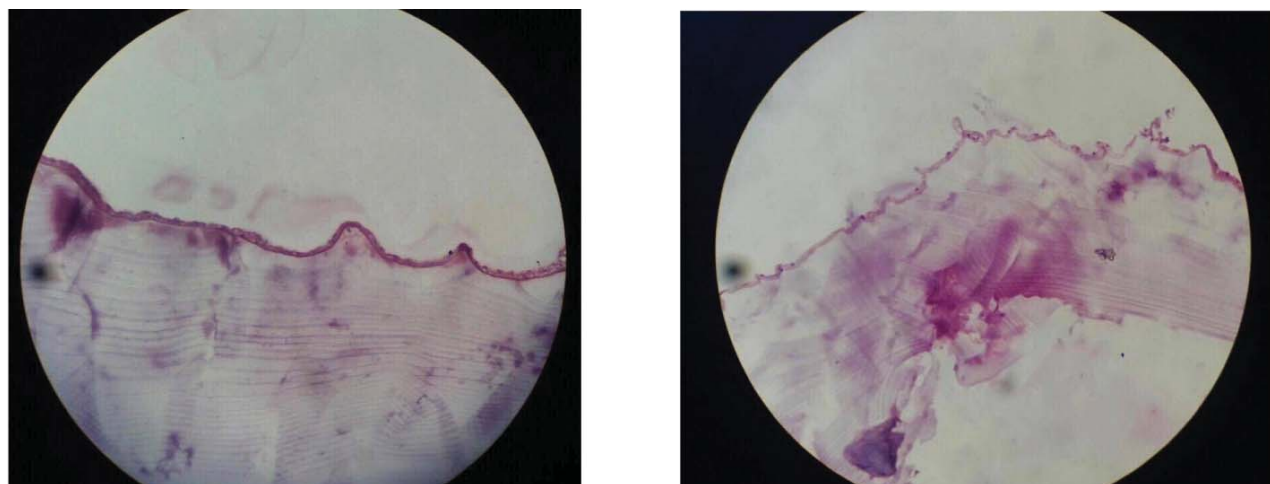


Figure 4: Histopathology of hydatid cyst showing inner germinal layer with daughter cysts and laminated chitinous layer

of the infected intermediate host. After ingestion, the protoscolices evaginate, attach to the intestinal mucosa and develop into adult stages in 32 to 80 days⁹.

The hexacanth embryo is believed to reach the bone along a tortuous and complicated systemic circulatory route passing through the liver and lungs. Primary extradural

hydatid disease of spine can be explained through direct porto-vertebral venous shunt³.

Patient may present with symptoms and signs related to spinal cord compression. In this case, the patient had presented with features of lumbar radiculopathy. There are no pathognomic signs and symptoms of this disease. This often leads to misdiagnosis of the disease. Radiological studies such as CT and MRI⁸ scan can help in recognizing the occurrence of the disease. MRI can demonstrate any cord compression throughout the length of the spinal cord and thus is the investigation of choice⁹. On MRI, hydatid cysts appear as well-circumscribed, cystic lesions, with cerebro-spinal fluid-like signal intensities. The cyst wall is usually thin and regular with no septations. The cysts are hypointense on T1W images¹⁰. On T2W images they appear hyperintense with sharply defined, hypointense cyst wall¹¹.

The differential diagnosis of cystic lesion of sacrum includes developmental cysts epidermoid, dermoid, teratoma, neurenteric and retrorectal cystic hamartoma (tail gut cyst), anterior sacral meningocele, necrotic sacral chordoma, schwannoma, arachnoid cyst, Tarlov cyst and aneurysmal bone cyst.

Surgery remains the preferred treatment for extradural hydatid cysts, where spinal cord decompression is the main purpose of the surgery¹². The treatment is essentially surgical and decompressive laminectomy with total excision of cyst, whenever possible, represents the treatment of choice⁹. Ideally, treatment involves total excision of all affected tissue, but this is very difficult in most cases. Due to the tight adhesion of the lesion with the surrounding tissues and multiloculation of cyst, chance of rupturing is high. If it ruptures during the procedure, the use of intra-operative hypertonic saline or 0.5% silver nitrate solution before opening the cavities kills the daughter cysts and prevents further spread or anaphylactic reaction. Many authors^{2,12} believe that after surgery, antihelminthic treatment is indicated even when the cyst remains intact during surgery.

Conclusion

Hydatid cyst of the spine is a rare presentation and should be considered as a differential diagnosis in endemic countries. Only radiological studies are not enough to confirm their occurrence but also require intraoperative and histopathological findings.

Conflict of interest

None of the authors have potential conflicts of interest to be disclosed.

References

1. Lakhdar F, Arkha Y, Rifi L, Derraz S, El Ouahabi A, El Khamlichi A. Spinal intradural extramedullary hydatidosis: Report of three cases. **Neurosurgery**. 2009;**65**(2):372–6. DOI: 10.1227/01.NEU.0000349751.39742.E3
2. Braithwaite PA, Lees RF. Vertebral hydatid disease: radiological assessment. **Radiology**. 1981;**140**(3):763–6. DOI: 10.1148/radiology.140.3.7280247
3. Fiennes AG, Thomas DG. Combined medical and surgical treatment of spinal hydatid disease: a case report. **J Neurol Neurosurg Psychiatry**. 1982;**45**(10):927–30. DOI: 10.1136/jnnp.45.10.927
4. Rayport M, Wisoff HS, Zaiman H. Vertebral echinococcosis: report of case of surgical and biological therapy with review of the literature. **J Neurosurg**. 1964;**21**:647–59. DOI: 10.3171/jns.1964.21.8.0647
5. Pamir MN, Akalan N, Özgen T, Erbenli A. Spinal hydatid cysts. **Surg Neurol**. 1984;**21**(1):58–60. DOI: 10.1016/0090-3019(84)90401-4
6. Moro P, Schantz PM. Echinococcosis: a review. **International Journal of Infectious Diseases**. 2009;**(13)**:125–33. DOI: 10.1016/j.ijid.2008.03.037
7. Thompson RCA. Biology and Systematics of Echinococcus. **Adv Parasitol**. 2017;**95**:65–109. DOI: 10.1016/bs.apar.2016.07.001
8. Moharamzad Y, Kharazi HH, Shobeiri E, Farzanegan G, Hashemi F, Namavari A. Disseminated intraspinal hydatid disease. **J Neurosurg Spine**. 2008;**8**(5):490–3. DOI: 10.3171/SPI/2008/8/5/490
9. Abbassioun K, Amirjamshidi A. Diagnosis and management of hydatid cyst of the central nervous system: Part 2: Hydatid cysts of the skull, orbit, and spine. **Neurosurgery Quarterly**. 2001;**(11)**:10–6. DOI: 10.1097/00013414-200103000-00002
10. Tekkök IH, Benli K. Primary spinal extradural hydatid disease: Report of a case with magnetic resonance characteristics and pathological correlation. **Neurosurgery**. 1993;**33**(2):320–3. DOI: 10.1097/00006123-199308000-00023
11. Gupta S, Rathi V, Bhargava S. Unilocular primary spinal extradural hydatid cyst - MR appearance. **Indian J Radiol Imaging**. 2002;**12**(2):271–3.
12. Govender TS, Aslam M, Parbhoo A, Corr P. Hydatid disease of the spine: A long-term followup after surgical treatment. **Clin Orthop**. 2000;**(378)**:143–7. DOI: 10.1097/00003086-200009000-00023