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
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 disease. Hydatid cysts are usually located at the thoracic level (52%), followed by lumbar (37%), and then cervical and sacral levels. Braithwaite and Lees have classified this disease into 5 types: 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 1807. The disease has a characteristic geographic distribution and is prevalent in most of the sheep-raising regions of the world. Echinococcosis in humans occurs as a result of infection by the larval stage of taenia cestodes of the genus Echinoccus. 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). 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, unicellular cyst that consists of an inner germinal layer of cells supported by a characteristic acidophilic-staining, acellular, laminated membrane of variable thickness. 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.
of the infected intermediate host. After ingestion, the protoscolices evaginate, attach to the intestinal mucosa and develop into adult stages in 32 to 80 days\textsuperscript{9}.

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

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

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

Figure 4: Histopathology of hydatid cyst showing inner germinal layer with daughter cysts and laminated chitinious layer
hydatid disease of spine can be explained through direct porto-vertebral venous shunt.

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 MRI 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 choice. 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 images. On T2W images they appear hyperintense with sharply defined, hypointense cyst wall.

The differential diagnosis of cystic lesion of sacrum includes developmental cysts epidermoid, dermoid, teratoma, neuenteric and retrorectal cystic hamartoma (tail gut cyst), anterior sacral meningocoele, 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 surgery. The treatment is essentially surgical and decompressive laminectomy with 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 of intra-operative hypertonic saline or 0.5% silver nitrate. If it ruptures during the procedure, the use of antihelminthic drugs such as praziquantel or albendazole is useful but may not prevent further spread or anaphylactic reaction. Surgery 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.

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