Primary Intraspinal Extradural Hydatid Disease Causing Radicular Compression

Omurga Kanalında Kök Basısına Neden Olan Birincil Ekstradural Hidatik Kist

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Abstract: Spinal hydatid disease is a rare entity and occurs most commonly as a result of direct extension from pulmonary lesions. Isolated intraspinal extradural location of the hydatid cyst is even rarer. We report a case of an isolated spinal extradural hydatid cyst presenting with radicular compression.

Key Words: Spinal hydatid cyst, magnetic resonance imaging

Özet: Omurilik ve omurga kanalında hidatik hastalığı oldukça seyrek görülen bir durumdur ve çoğunlukla akciğer lezyonlarının doğrudan kanala sirayet etmesi sonucu oluşur. Yalnızca omurilik ve omurga kanalında yerleşmiş ekstradural hidatik kist daha da seyrektir. Bu yazıda kök basısı bulguları gösteren yalnızca omurga kanalında yerleşmiş ekstradural hidatik kistli bir olgu sunulmuştur.

Anahtar Sözcükler: Omurga kanalında hidatik kist, manyetik rezonans görüntüleme

INTRODUCTION

Hydatid disease is the cystic stage of Echinococcus granulosus which affects human being. The definitive host of this tapeworm is usually the dog, although the fox, wolf, or the cat may occasionally be the host. The animal becomes infected by ingesting the viscera of the intermediate host, usually the sheep which are contaminated by the larvae of the worm. The definitive host harbors the worm in the lumen of its small intestine. Humans and sheep are infected by ingesting these eggs. The chitinous coat of the egg is then dissolved in the intestine of the intermediate host. Embryos are liberated and these pass through the intestinal wall into the portal venous system. Within 3 weeks these

embryos develop into larvae and then start producing cysts. About 75% of the larvae are trapped by the liver and 15% by the lung, with the remaining 10% passing to the rest of the body (3).

Hydatid cyst of the spine represents 1% of all cases with hydatidosis and is most commonly located in the dorsal spine (1,3,4,5,8,9). The disease either occurs by direct extension from a pulmonary infestation or less often begins primarily in the vertebral body (3,5,6,8). Primary isolated spinal extradural hydatid disease is an exceptional possibility (9,12). We herein report a patient with an isolated spinal extradural hydatid cyst that manifested itself as S1 radix compression.

CASE REPORT

A 30-year old woman was admitted to our clinic with the complaints of pain and hypoesthesia at her left lower extremity. Her complaints had started 6 months ago and increased gradually, and had no benefit from medical treatment. On physical examination plantar flexion of left foot was 4/5, left S1 dermatome hypoesthesic, and left Achille reflex was absent. Electromyography (EMG) revealed partial denervation of left S1 root. Sagittal and axial spinal magnetic resonance imaging (MRI) scans detected multiple hypointense cystic structures surrounded by a wide isointense mass, all located epidurally at the level of L5 and S1 which obliterated the epidural space and eroded laminae, pedicles, and the vertebral body on the left side (Figure 1).

The patient was operated with the diagnosis of cystic spinal mass. Left laminae of L5 and S1 were seen to be eroded by the mass lesion. This epidural multicystic mass was easily dissected from the bones and removed en bloc. Explored area was irrigated by hypertonic saline solution. Histopathologic diagnosis was hydatid cyst. Postoperative course was uneventful and mebendazole treatment was initiated. Patient's symptoms were relieved dramatically and she was symptom free 3 months after surgery.

DISCUSSION

Hydatid cyst of the spine makes 1% of all cases with hydatidosis and is most commonly located in the dorsal spine (3,4,5,8,9,10). The spinal hydatid disease might be caused by encroaching pulmonary lesions or may develop primarily in the vertebral body (4,6,8,9,10). Rarely, the disease begins from the extradural area; the embryo is possibly being carried through the porto-vertebral venous shunts (4).

According to Braithwaite and Lees (2), spinal hydatid cysts are classified into five types:1. Primary cysts of the spinal cord (intramedullary hydatid cysts), 2. primary intradural cysts (intradural extramedullary hydatid cysts), 3. primary extradural cyst, 4. hydatid cyst of the vertebrae, and 5. paravertebral lesions extending to the spinal column. The first three groups of hydatid are rare, and only sporadic cases have been reported. We can classify our patient as Type 3. Our case showed no evidence of pulmonary or chest wall hydatidosis on computed tomography, and there was no pathological finding on abdominal ultrasonography.

There are no pathognomonic signs or symptoms of spinal hydatid disease. The diagnosis of hydatid disease affecting the spine at an early stage is by no means simple. The progressive nature of spinal hydatidosis with the clinical presentation of cord or radix compression is not pathognomonic to the



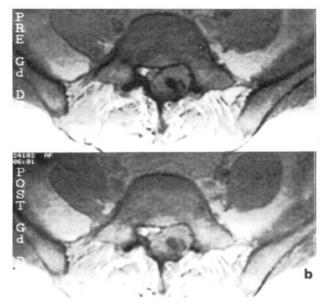


Figure 1, a and b. Pre-contrast and post-contrast sagittal (a) and axial (b) T1-weighted spinal MRI scans showing L5 and S1 mass lesion obliterating the epidural space and causing erosion of laminae and pedicles on the left side. The mass is composed of multiple hypointense areas and a wide isointense mass surrounding it.

disease. However, the clinician must be alert and bear in mind the disease when he has to deal with patients who have spent a long period in an endemic area or who have a positive medical history of hydatidosis (11).

The radiological diagnostic modalities include plain radiography, polytomography, myelography, computerized tomography, and more recently MRI. The findings on plain radiography and polytomography are nonspecific. Compression of the spinal canal by the invading mass can be demonstrated by myelography. Computerized tomography is less invasive than myelography, and can show bone destruction as well as paraspinal and intraspinal extensions of the lesion (5). MRI has recently been added to the arsenal of the radiological procedures, and has shown to have an interesting capability as a non-invasive modality for the study of the spinal canal and spinal cord in different planes (7,8,10,11). MRI is not only helpful in the diagnosis but also determines the surgical strategy by revealing extensions of the cysts. Although it has been reported that spinal hydatid cysts have typical MRI findings (8,10,11), MRI of our patient revealed hypointense areas and a large isointense mass surrounding them. Therefore, the lesion was not diagnosed as hydatid cyst preoperatively.

Decompressive laminectomy and removal of the cysts are considered to be the best surgical approach in the treatment of hydatidosis of the spine though long-term results are generally poor (3,5,8,9). It has been suggested that irrigation with hypertonic saline solution during the operation might be beneficial to prevent recurrence. Integrated surgical and medical treatment with mebendazole as a drug of choice is the preferred management of these patients (1,9,11).

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