EPIDURAL HYDATID CYST OF THE LUMBAR SPINE: A Case Report

Murat HANCI*, Önder AYDINGÖZ**, Taner TANRIVERDİ*, Naci KOÇER***

ABSTRACT

Primary spinal extradural hydatid disease is a rare entity that may lead to severe, acute-onset neurological deficits. In this discussion, a case with an extradural hydatid cyst of the lumbar spine has presented. Neuroradiological studies of the lumbar area revealed an extradural cystic lesion. At surgery, echinococcosis was revealed and subsequent histopathological examination also confirmed the diagnosis. After the surgery, albendazole therapy was administered and the patient had complete recovery despite perforation of the cyst wall has occurred during the operation. We believe that administration of albendazole after surgery is effective to prevent recurrences and although a single case is not sufficiently promising, the prognosis is not as tragic as it was formerly thought to be.

Key words: Albendazole, Echinococcus, Extradural hydatid disease, Hydatid cyst

ÖZET

LOMBER OMURGANIN EPİDURAL HİDATİK KİSTİ: OLGU SUNUMU

Primer spinal ekstradural hidatid hastalığı, şiddetli, akut başlangıçlı nörolojik defisite yol açabilen nadir bir olaydır. Bu çalışmada, lomber omurgada ekstradural hidatik kistli bir olgu sunulmaktadır. Lomber bölgenin nöroradyolojik incelemesi, ekstradural kistik bir lezyon ortaya koymuştur. Cerrahi sırasında, ekinokokozis saptanmış ve histopatolojik değerlendirme, tanıyı doğrulamıştır. Cerrahi sonrasında, albendazole tedavisi uygulanmış ve kist duvarı operasyon sırasında perfore olmasına rağmen hasta tam iyileşme göstermiştir. Operasyon sonrası, albendazole verilmesinin rekürrensi önlemede etkili olduğuna inanıyoruz ve tek bir olgunun sonucu yeterince umut verici olmasa da, prognoz daha önceleri düşürüldüğü kadar trajik değildir.

Anahtar sözcükler : Albendazole, Ekinokokkus, Ekstradural hidatid hastalık, Hidatik kist

INTRODUCTION

Hydatid disease, or hydatidosis in general, is an uncommon helminthic infestation caused by a cestode, echinococcus. It is the larval stage of Echinococcus granulosus (E. granulosus) that causes the disease in man. It may affect various organ systems in the body and primary involvement of the bone is extremely rare (0.5 % to 4 %). The spine (50 %) and pelvis are the predilection sites of involvement (9).

Although the mode of spread to the spine has not been clearly defined, the disease usually affects the spine as a direct extension from pulmonary lesions or less often begins primarily in the vertebral body (1,9). Primary extradural hydatid cyst disease is said to be rare or an exceptional possibility (1,10).

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CASE

A 12-year-old male presented with a 4-month history of low back pain and sciatica on the right side, which did not respond to routine treatment. The back pain was dull and constant and aggravated by standing and walking.

The neurological examination revealed minimal weakness (4/5) of the hip flexion, hip adduction and knee extension of the right side with the abolition of the patellar reflex on both sides. Hypoesthesia was noted in the anterior lower half of the right thigh. The sphincters were intact.

Laboratory investigations disclosed slight increase in erythrocyte sedimentation rate (33 mm/h) and no eosinophilia. Special laboratory investigations including Casoni intradermal test and specific IgG of E. granulosus by ELISA were negative.

On neuroradiological examination, plain x-rays of the lumbar spine disclosed interpedicular widening and thinning of both pedicular contours medially at the level of L4 vertebra. A lumbar myelogram showed an extradural compression of the dural sac anteriorly and scanty amount of contrast material below the L4 level (Figure 1a-b).

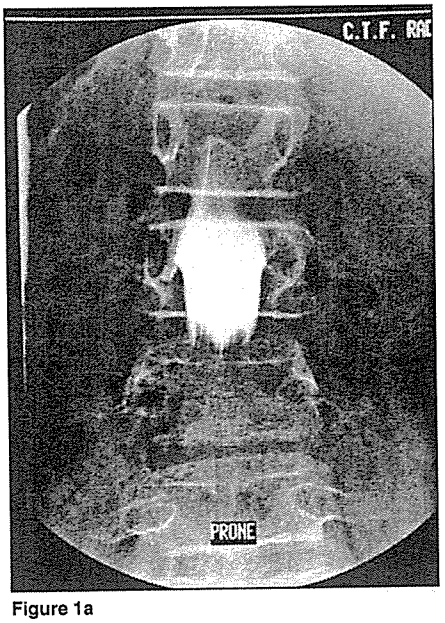

Figure 1a

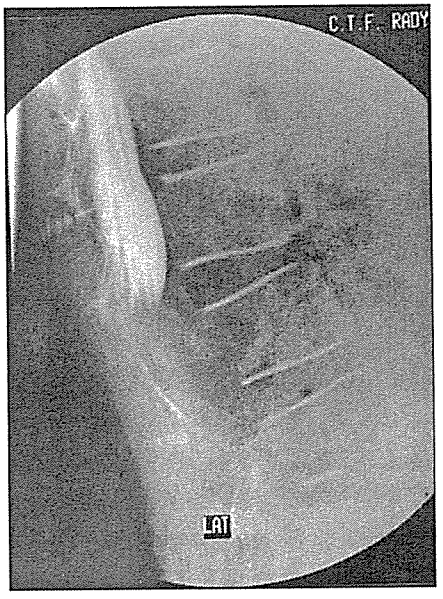

Figure 1b

Figure 1.Prone (a) and a lateral (b) sections of myelogram show complete extradural type block of contrast medium at L4 level.

Computerized tomography (CT) (Figure 2)

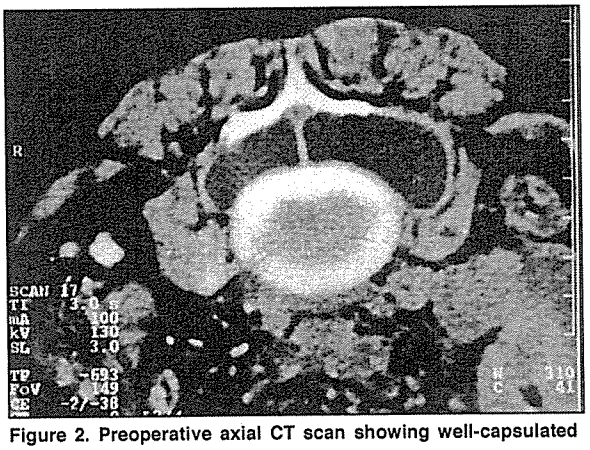

Figure 2. Preoperative axial CT scan showing well-capsulated intracanal huge cyst.

and magnetic resonance imaging (MRI) (Figure 3a-b) of the lumbar spine demonstrated a well-capsulated cyst located mainly in the epidural space, extending to the neural foramina bilaterally at the L4-5 level. Additionally, T1-weighted sagittal MR images clearly showed the intraosseous invagination of the epidural encapsulated homogeneous cyst (Figure 4).

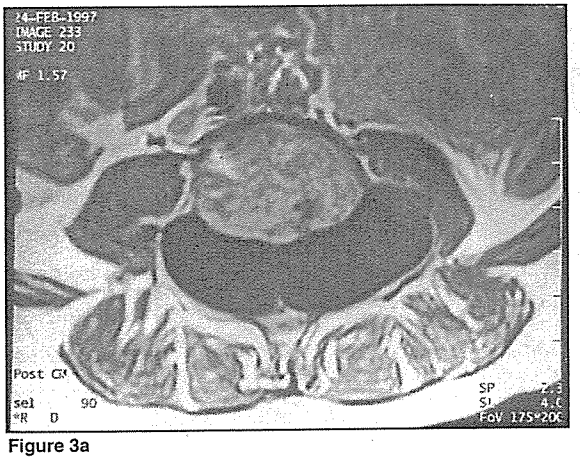

Figure 3a

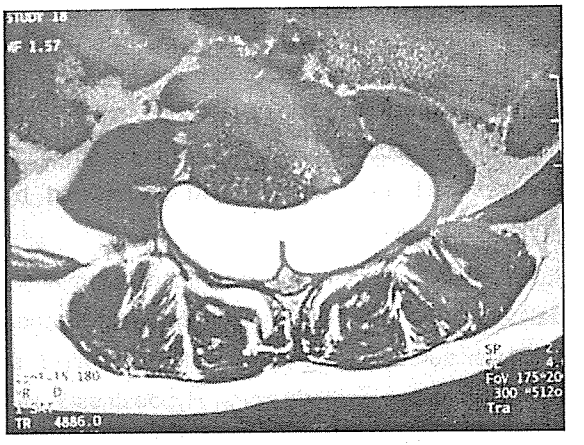

Figure 3b

Figure 3. Preoperative T1-weighted (a) and T2-weighted (b) MR images indicating a well-capsulated cyst located dominantly anterior epidural space, extending to the neural foramina bilaterally.

The patient underwent operation fifteen days after the admittance, and following complete L3 laminectomy, the ventrally located huge unilocular cyst was exposed. During the removal of the cyst, the thick, fibrous capsule was perforated, thus the surgical field was contaminated. Remaining content of the cyst was aspirated and shrinked cyst wall was separated from the adjacent dura and removed completely. Finally, the surgical field was irrigated with hypertonic saline solution. The patient was prescribed oral albendazole 10 mg/kg/day during postoperative 6 month period.

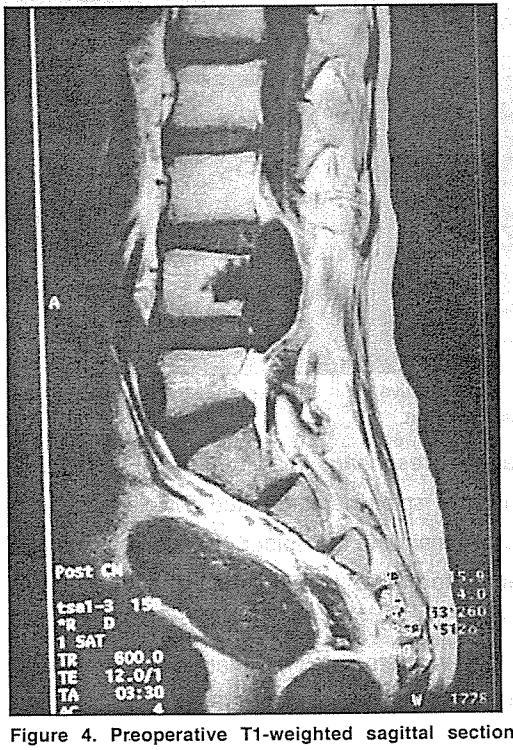

Figure 4. Preoperative T1-weighted sagittal section clearly demonstrates scalloping of the L4 vertebral body by the encapsulated homogeneous cyst.

Histopathological examination showed cuticular and germinative membranes of the cyst (fertile cyst of echinococcus containing daughter capsules) confirming the diagnosis of hydatid disease.

At 7 months postoperatively, MR imaging (Figure 5a-b) showed that, dural sac and caudal equine were normal. But, there was a pseudomeningocele, extending to the neural foramina bilaterally. It was probably secondary to the cerebrospinal fluid (CSF) leakage from the anterior dural defect that has occurred during the operation.

Four years after the operation, the patient was free from symptoms and there was no evidence of the parasite elsewhere in the body.

DISCUSSION

Hydatidosis is generally encountered in underdeveloped countries where the rearing of sheep is a major industry and pork consumption is high (4). Compression of the spinal cord due to hydatidosis has reported to be as high as 14 % in Tunisia (3), 4.5 % in Morocco (3) and 3.8 % in Turkey (10). Men are more commonly affected than women. Primary involvement

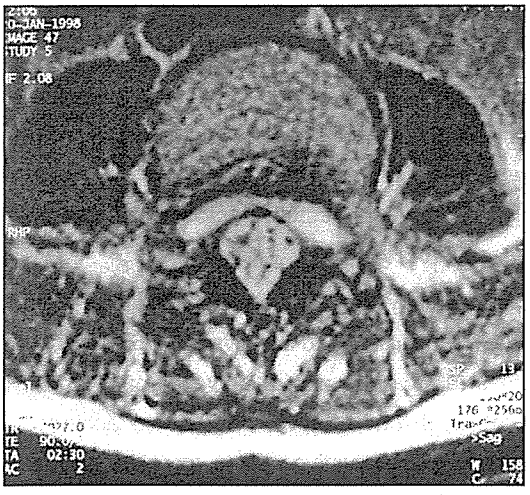

Figure 5a

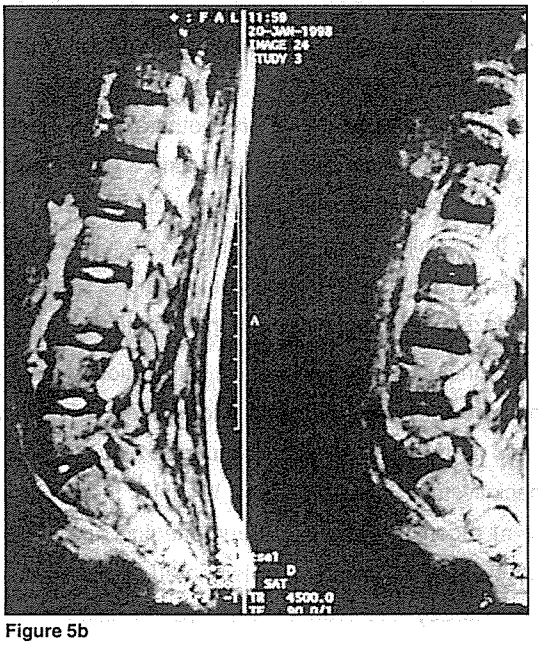

Figure 5b

Figure 5. Postoperative axial (a) and sagittal (b) MR sections indicating normal dural sac and cauda equina. Note also a pseudomeningocele extending to the neural foramina bilaterally due to the CSF leakage from the anterial dural defect during operation.

of the bone is extremely rare and occurs in 0.5 % to 4 % of the patients (9) but vertebral involvement accounts for nearly in 44-50 % (1).

A survey of that literature discloses that thoracal spine is the most commonly affected part of the spinal cord (50%) (1,4,10). Primary solitary extradural hydatid cyst without any other systemic involvement can occur in the presence of direct porto-vertebral venous shunts (6). In 1981, spinal echinococcosis was classified into five groups by Braithwaite and Less (5):

1) primary intramedullary cyst

2) intradural extramedullary cyst

3) extradural intraspinal cyst

4) hydatid disease of the vertebra

5) paravertebral hydatid disease

We think that, in the present case, the disease began from the extradural area and the embryo was possibly carried through the porto-vertebral venous shunts. Also, there was no systemic involvement and the adjacent ribs and paraspinal muscles were intact; there was scalloping of the L4 vertebral body. In these considerations, we classify our patient as type 3 .

The clinical picture of the disease is not pathognomonic and majority of the patients present with unremitting back pain. Sphincter disturbances, paresthesia, paraparesias, and eventually paraplegia may also be seen during the late phase of the disease (1,4).

Special laboratory tests including Casoni intradermal test or complement fixation tests are not usually diagnostic and have no value if only bone involvement is the case (6). This was also true for our case.

For radiological diagnosis, there is no pathognomonic feature of the hydatid disease of the spine. In the preoperative period, the disease might often be misdiagnosed as tuberculosis, pyogenic infection, and malignant disease of the spine (1). Plain x-rays may play an important role in the diagnosis of the vertebral hydatid cyst (10), but not in the presence of primary epidural involvement as in our case; but myelography can show the mass effect on the dural sac (1,24). Although CT provides assessment of bony and soft tissue involvement (1,2), there were no specific features suggestive of epidural hydatid cyst in

our patient. Thus, we feel that CT may not provide an adequate suggestion of the extent of the hydatid disease in the spine. Recent clinical studies have suggested that, MR imaging is the examination of choice when there is suspicion of hydatid disease (7,12). MR imaging can differentiate the disease from such other entities as tuberculosis, malignant and disc diseases. Paraspinal lesions are more clearly seen with the multiplanar (i. e., coronal and sagittal as well as transverse) imaging capability of MR imaging. The presence of a markedly hypointense cyst wall on T1- and T2-weighted images and absence of wall enhancement with intravenously administered gadolinium are characteristic of hydatid disease (bunch of grapes) (7). MR imaging in our case was not characteristic and the lesion resembled an arachnoid cyst on T1-weighted images on which the cyst content demonstrated CSF intensity. T2-weighted MR images, however, showed the homogeneous cyst content to be slightly more hyperintense than the CSF (11). Although MR imaging is useful in the diagnosis of primary extradural spinal hydatid cysts (8), it should be kept in mind that only the surgery and histopathological examination can provide the definitive diagnosis.

In the management of the spinal hydatidosis, surgical removal of the cyst without perforating the wall is considered to be the cornerstone of the treatment and remains palliative rather than curative. It is generally performed by means of a decompressive laminectomy (10). Anterior spinal decompression is advocated if the vertebral body is involved, as it is in most patients, and laminectomy does not allow for complete excision of the bony disease and thus recurrence is high (15). It is recommended that posterior decompression alone is indicated only when there is isolated involvement of the neural arch and in extraosseous extradural cases (7) like the one presented here. Intraoperative use of diverse scolicidal agents such as formalin, silver nitrate, or hypertonic saline is controversial (14). On the other hand, it has been known that irrigation with hypertonic solutions osmotically disrupt the cysts and kill the parasites (15). We think that further investigations are necessary to prove the help of such solutions; otherwise, they may cause necrosis of tissues and increase morbidity.

It is well-known that vertebral hydatidosis generally tends to be multifocal and invasive (1,5,15). Thus, there is almost always the risk of nerve damage and cyst perforation. If total removal can not be achieved, a benzimidazole derivative, such as albendazole or mebendazole, should be administered. Albendazole is considered to be more effective (13). It is a broad-spectrum antihelminthic drug with good oral absorption but may be hepatotoxic (13). There is no agreement concerning the duration of chemotherapy and most physicians recommend a course lasting 3 or 4 months (6,13). Our patient received oral albendazole 10 mg/kg three times daily for 6 months with a close follow-up for hepatic functions (13) and no complications related to albendazole treatment was seen in our patient.

In conclusion, echinococccal infestation must be considered in the differential diagnosis of cystic lesions of the spine with or without neurologic deficits, especially in the endemic areas. We think that with the help of recent advances in neuroimaging and with a good surgical technique supplemented with adequate chemotherapy, the prognosis of hydatid disease has improved. In this regard, MR imaging should be the investigation of choice because of a good topographic delineation which allows the establishment of the exact size of lesion. The treatment should be the combination of surgery and chemotherapy, the latter should be lasted 4 to 6 months. Although it is stressed in the literature that surgical removal of the cyst without perforating the wall should be the goal for not causing the dissemination, our patient with intraoperative perforation of the cyst wall could be

cured totally with the combination of surgery and oral albendazole lasting 6 months.

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