

Primary Spinal Hydatid Disease

Primer Spinal Hidatik Hastalığı

ABSTRACT

Primary spinal hydatid disease is rare. Spinal hydatid disease should be considered in the differential diagnosis of spinal cord compression syndrome in endemic countries and evaluated with imaging and serology. Our case was a 34-year-old man. The patient presented with progressive back pain for 8 months and lower extremity weakness for 3 months. Neurological examination was suggestive of upper motor neuron type of paraparesis. Magnetic resonance images of the thoracic region showed an intradural multicystic lesion. The mass was explored with T 10-11 laminectomy. It had displaced the cord to the right side. The fluid was clear and did not contain pus. The lesion was easily dissected from the cord and was resected totally. The pathological diagnosis was hydatid disease.

KEYWORDS: Hydatid disease, Spinal mass, Paraparesis

ÖZ

Primer spinal hidatik hastalığı nadirdir. Spinal hidatik hastalığı, endemik bölgelerde spinal kord kompresyonu sendromunun ayırıcı teşhisinde düşünülmeli, görüntüleme ve seroloji yapılmalıdır. Bizim hastamız 34 yaşında erkektir. Hastada 8 aydır progresif sırt ağrısı ve 3 aydır alt ekstremitelerde güçsüzlük vardır. Nörolojik muayenede üst motor nöron tipinde paraparezi saptandı. Torakal alanın manyetik rezonans görüntülemesinde intradural multikistik lezyon görüldü. Kitle T10-11 laminektomi ile ortaya kondu. Kitle kordu sağ alana doğru deplase etmişti. Sıvı berraktı ve pü içermiyordu. Kitle korddan kolaylıkla diseke edildi ve total rezekte edildi. Patolojik teşhis hidatik hastalık idi.

ANAHTAR SÖZCÜKLER: Hidatik hastalığı, Spinal kitle, Paraparezi

Hikmet TURAN SÜSLÜ

Ayçiçek ÇEÇEN

Alp KARAASLAN

Ali BÖREKÇİ

Mustafa BOZBUĞA

Dr. Lütfi Kırdar Kartal Training and Research Hospital, Neurosurgery Clinic, Istanbul, Turkey

Received : 12.02.2009

Accepted : 09.03.2009

Correspondence address:

Hikmet TURAN SÜSLÜ

E-mail: hikmets1972@yahoo.com

INTRODUCTION

Hydatid disease (HD) is a worldwide zoonosis produced by the larval stage of the *Echinococcus* tapeworm. Hydatid disease (HD) can develop anywhere in the human body. The liver is the most frequently involved organ, followed by the lungs. Furthermore, secondary involvement due to hematogenous dissemination may be seen in almost any anatomic location (e.g., lung, kidney, spleen, bone, brain) (7). Bone involvement is a rare complication of this disease and the frequency of osseous involvement in HD is 0.5%– 4% (6,7,10). Primary spinal HD accounts for less than 1% of all HD (6). We report a patient with primary thoracic intradural extramedullary hydatid cyst. The clinical presentation, diagnosis and surgical treatment are discussed.

CLINICAL DETAILS

We report a 34-year-old man, who presented with progressive back pain for 8 months and lower extremity weakness for 3 months. Neurological examination was suggestive of upper motor neuron type of paraparesis grade 3/5. Magnetic resonance images (MRI) of the thoracic region showed an intradural extramedullary multicystic lesion with a regular contour. The lesion gave a low signal on T1-weighted and high signal on T2-weighted images. The lesion demonstrated no rim enhancement after contrast material injection (Figure 1 and 2). The spinal cord was compressed and displaced anterolaterally. Abdominal ultrasonography and MRI of the cranium, cervical and lumbar region were normal. The erythrocyte sedimentation rate (ESR) and C-reactive protein (CRP) values were 80 mm/h and



Figure 1: T2-weighted images of the thoracic region show an intradural extramedullary multicystic lesion with a lower-signal cyst wall. The spinal cord was compressed and displaced anterolaterally.

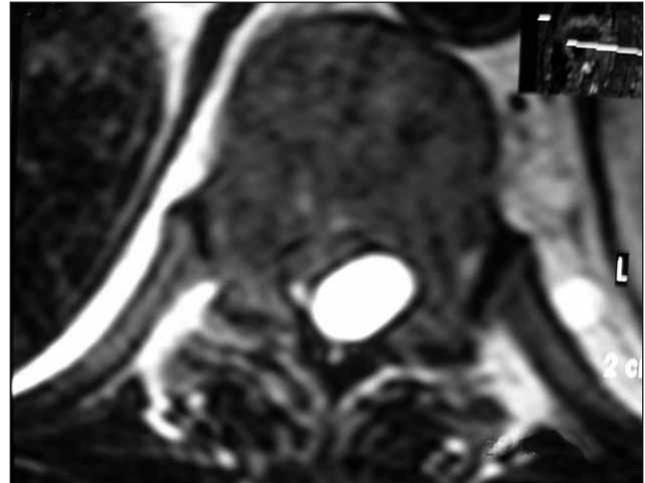


Figure 2: Axial MRI showing intraspinal cystic lesion (hyperintense on T2 weighted images).

was 48 mg/dl, respectively. Other biochemical values were normal. The case was explored with T10-11 laminectomy. Opening the dura, a yellowish white, solid, clearly defined, multi-lobulated (by tiny septa containing cystic fluid in its compartments) lesion appeared. It had displaced the cord to the right side. The fluid was clear and did not contain pus. The lesion was easily dissected from the cord and was resected totally. The operation area was soaked with 3% normal saline for 10 minutes. Pathological examination revealed that the cuticular membrane had a lamellar, acellular, homogenous eosinophilic appearance. In addition, soft tissue pieces showing fibroblastic proliferation, inflammatory cells, and infiltrated giant cells were observed. The pathological diagnosis was hydatid cyst. Albendazole was administered during the postoperative period (10 mg/kg/day orally). Partial weight-bearing was permitted at the end of second month. Full weightbearing was permitted at end of third month. At the one year postoperatively, the patient was pain free. All serological tests were normal. Radiological evaluation showed no evidence of disease recurrence.

DISCUSSION

Spinal lesions of cystic echinococcosis usually occur as isolated findings and without concomitant hepatic or pulmonary involvement. Primary vertebral HD can occur with direct portovertebral venous shunts (1). Spinal HD is classified into five groups: intramedullary, intradural extramedullary, extradural intraspinal, vertebral, and paravertebral (5,6,8,9). Infestation is thought to begin primarily in the center of the vertebral

body and subsequently to extend extradurally or paravertebrally. The thoracic spine is most frequently involved (50% of cases), followed by the lumbar (20%), sacral (20%), and cervical (10%) spine (11). Spinal hydatid cysts are usually situated in the dorsal region and generate medullary or radicular symptoms according to their location (1).

Spinal HD is usually difficult to distinguish from tuberculous spondylitis or chronic osteomyelitis. Some imaging characteristics have been described as typical of spinal HD. These include lack of osteoporosis and sclerosis in the host bone; absence of damage to the disk spaces and vertebral bodies and spread of the disease via a subperiosteal and subligamentous path; paraspinal extension; and, involvement of a contiguous rib (2,4,5,10). Computed tomography (CT) and MRI demonstrate a lesion with imaging characteristics similar to those of cerebrospinal fluid. In contrast to hydatid cysts located within the brain, spinal hydatid cysts demonstrate no rim enhancement after contrast material injection. The presence of a markedly hypointense cyst wall on T1- and T2-weighted images is characteristic of HD (1). The low-signal rim on T2-weighted images results from reactive fibrosis and degeneration around the parasitic membrane and correlates with the histopathological examination (3).

Treatment is difficult because of the progressive course of the bone involvement and the generally accepted algorithm for osseous HD. The general principle for surgical treatment is the removal of cyst contents without contaminating the patient, followed by appropriate management of any remaining cavity. The surgical area might be irrigated with chemical agents (3% normal saline) in an attempt to kill scoleces (1,8). Indications for chemotherapy include inoperable lesions, unwillingness of the patient to undergo surgery, and use as an adjunct to surgery. The combination of chemotherapy and surgical treatment has been found to be more efficient than surgical treatment alone (3). Mebendazole, albendazole, and antihelminthic drugs are used for chemotherapy. Albendazole has been found to be better absorbed than mebendazole and

exhibits superior efficacy against helminths (1). Despite treatment, the disease frequently relapses with progressive destruction of the vertebral column and neurological deterioration. Patients without any symptoms should be followed for the long-term and serological tests and radiographs should be used periodically to ensure that the disease has not recurred.

In conclusion, patients presenting with low back pain and/or radicular pain or monoparesis/paraparesis should be evaluated carefully pre-operatively regarding spinal surgery. Besides tumoral lesions, infectious lesions should also be kept in mind. The compartment of the mass, its localization (intradural, extradural, intramedullary, extramedullar) and clearly defined cystic lesions should alarm the physician regarding infection.

REFERENCES

1. Awasthy N, Chand K: Primary hydatid disease of the spine: An unusual case. *Br J Neurosurg* 19:425-427,2005
2. Beggs I: The radiology of hydatid disease. *AJR Am J Roentgenol* 145:639-648, 1985
3. Berk Ç, Çiftçi E, Erdoğan A: MRI in primary intraspinal extradural hydatid disease: Case report. *Neuroradiology* 40:390-392,1998
4. Braithwaite PA, Lees RF: Vertebral hydatid disease: Radiological assessment. *Radiology* 140:763-766, 1981
5. Ogüt AG, Kanberoglu K, Altug A, Cokyüksel O: CT and MRI in hydatid disease of cervical vertebrae. *Neuroradiology* 34:430-432, 1992
6. Pamir MN, Akalan N, Ozgen T, Erbenli A: Spinal hydatid cysts. *Surg Neurol* 21:53-57, 1984
7. Polat P, Kantarci M, Alper F, Suma S, Koruyucu MB, Okur A: Hydatid disease from head to toe. *Radiographics* 23:475-494, 2003
8. Pushparaj K, Sundararajan M, Madeswaran M, Ambalavalan S: Primary spinal intradural hydatid cyst. *Neurology India* 49:203-204, 2001
9. Sharma NK, Chitkara N, Bakshi N, Gupta P: Primary spinal extradural hydatid cyst. *Neurology India* 51:89-90, 2003
10. Torricelli P, Martinelli C, Biagini R, et al: Radiographic and computed tomographic findings in hydatid disease of bone. *Skeletal Radiol* 19:435-439, 1990
11. Tuzun M, Hekimoglu B: HD of the CNS: Imaging features. *AJR Am J Roentgenol* 171:1497-1500, 1998