

Recurrence of Primary Costal Echinococcosis with Spinal Cord Compression

Omurilik Basısı ile Nüks Eden
Primer Kostal Ekinokokkozis

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Abstract: Primary hydatid disease of the rib is very rare, even in endemic areas. We describe a patient who presented with recurrent costal hydatid disease that caused spinal cord compression and progressive paraparesis 7 years after the initial cyst removal surgery. In the first operation, cysts were removed but no bone was resected. To prevent recurrence of costal hydatid disease, we recommend wide bone resection in addition to extirpation of the cysts.

Key Words: Echinococcosis, hydatid disease, rib, spinal cord compression

Özet: Kostanın primer hidatik kist hastalığı endemik bölgelerde dahi nadir rastlanan patolojilerdendir. İlk ameliyatından yedi yıl sonra omurilik basısı ve ilerleyici paraparezisi ile rekürrens oluşan kostal hidatik kist hastası sunulmuştur. İlk ameliyatta kistlerin kemik eksizyonu yapılmadan çıkarılması rekürrens nedeni olabilir. Biz, kostanın primer hidatik hastalığında rekürrensi önlemek amacı ile, kistlerin çıkartılmasına ek olarak geniş kemik ve yumuşak doku rezeksiyonunu önermekteyiz.

Anahtar Kelimeler: Ekinokok, kist hidatik, kosta, spinal kanal kompresyonu

INTRODUCTION

Human hydatid disease is caused by *Echinococcus granulosus*. The dog is the definitive host for this parasite. The adult worm lives in the small intestine and discharges a large number of eggs in the feces. The domestic intermediate hosts are cattle, sheep, and goats, but man may also be infected. The intermediate host swallows the eggs and the larva develops, leading to hydatid disease. Bone involvement is unusual, but typically occurs at more

highly vascularized sites, such as the vertebrae, long-bone epiphyses, and ilium. The larval form reaches the bone, penetrates the spongy tissue, and grows in the direction of least resistance, infiltrating and damaging the tissue like a tumor (2).

Advanced imaging techniques, such as computed tomography (CT) and magnetic resonance imaging (MRI), are the best diagnostic tools for these cases. The prognosis is poor when bone is involved, even in patients who undergo extensive medical and

surgical treatment. In this paper, we present an unusual case of recurrent costal hydatid disease that extended into the thoracic cavity and spinal canal.

CASE REPORT

Seven years before he was admitted to our center, a 62-year-old male from a rural area presented to another institution with the complaint of back pain. He had no neurologic deficits at the time. A chest x-ray showed notching of the left 7th rib (Figure 1), and MRI demonstrated a multiloculated costal lesion. The lesion was hypointense on T1-weighted images and extended posteriorly into the paravertebral muscles (Figure 2). During surgery, the cysts were removed via a posterior approach and the area was flushed with saline. No bone was removed. The anterior cortex of the affected rib and the pleura were found to be intact. The pathological diagnosis was *E. granulosus*.

The patient came to our clinic 7 years post-surgery with the complaints of back pain, weakness in the lower extremities, and urinary incontinence. Neurological examination revealed 3/5 paraparesis, increased deep-tendon reflexes, and a positive

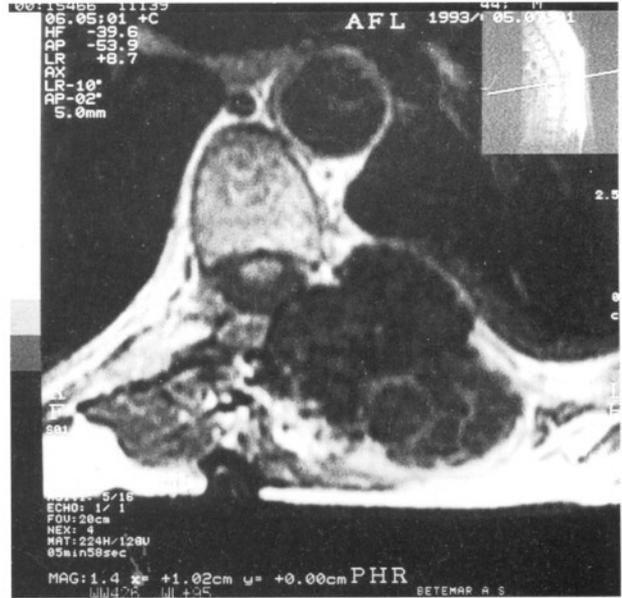


Figure 2: An axial T1-weighted MR image from the initial investigation shows multiple hydatid cysts in the left seventh rib, with the lesion extending posteriorly into the paravertebral muscles.

Babinski's sign bilaterally. Plain x-rays showed bone destruction in the left sixth and seventh ribs, and widening of these ribs due to cystic lesions (Figure 3). MRI demonstrated hypointense cysts with iso- to mildly hyperintense cyst walls, and showed infiltration of the vertebrae and spinal cord compression. Thoracic CT revealed a multiloculated cystic lesion with destruction of the sixth and seventh ribs, and the laminae and transverse processes of the

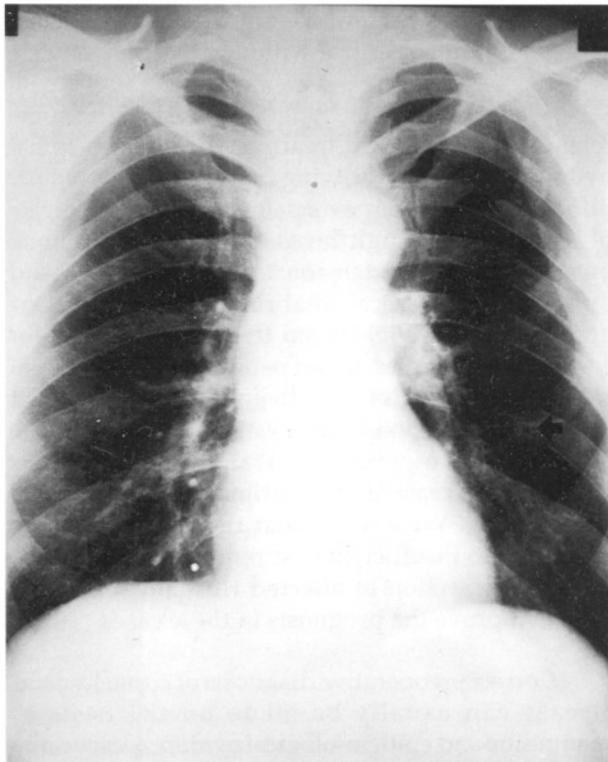


Figure 1: A plain x-ray taken during the initial case work-up shows notching of the left seventh rib.



Figure 3: A plain x-ray taken 7 years after the first operation shows involvement of the left sixth and seventh ribs.

T6 and T7 vertebrae. The lesion was compressing the spinal cord, and could be seen extending into the spinal canal and left thorax (Figure 4). The search for another systemic focus by abdominal and pelvic ultrasonography, lumbar and cervical MRI, and cranial CT was negative.

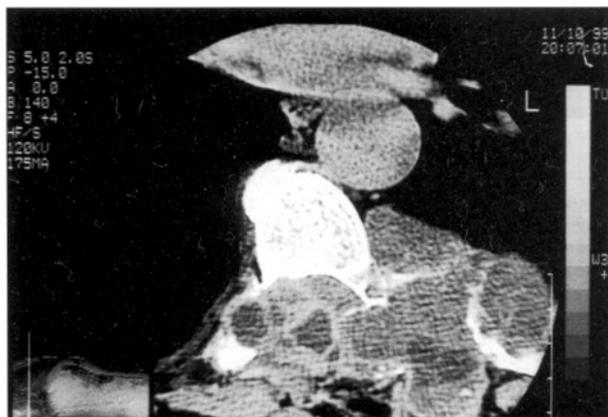


Figure 4: An axial CT image at the same level obtained 7 years after the first surgery shows bone destruction in the T7 vertebra and the seventh rib, and extension to the spinal canal and thorax.

A left thoracotomy was performed, and multiple cysts were removed via an anterior approach. The patient's left sixth and seventh ribs, and the left pedicle of T7 were removed. Small cysts were observed in the destroyed bone of T7 vertebral body, and we excised all bone tissue that appeared infected in order to prevent recurrence. The surgery site was covered with saline-soaked sponges during this operation. Left site of the dura in the spinal canal was visualized completely. However, postoperative CT showed residual cysts on the right side of the dura, so another operation was performed 2 weeks later. The affected lamina, spinous process, and right pedicle of the T7 vertebra were removed by posterior approach.

The pathological diagnosis was hydatid disease of the rib. After the two operations in our center, the patient reported less pain and showed improved motor strength. He was prescribed a 3-month course of albendazole therapy. Two months after discharge, his neurological examination was normal, and a recheck at 18 months showed no recurrence.

DISCUSSION

The reported incidence of primary hydatid disease of the bone ranges from 0.5-2%, and the spine

is affected in approximately half of all cases. Rib involvement in osseous hydatid disease is extremely rare (1,2,5). When it does occur, the cysts tend to be located in the posterior portion of the rib, and this is what we observed in our case. In later stages, the lesion breaks through the cortical bone to produce an extrapleural soft-tissue mass. In very advanced cases, the lesion involves not only the vertebra, but also extends into the spinal canal causing spinal cord compression.

Most cases of costal hydatid disease are reported as spinal lesions because patients usually do not seek medical attention until they develop neurologic symptoms (5). In the initial stage of disease, our patient's complaint was back pain unaccompanied by neurologic deficit. At that point, the infection was limited to the rib and paravertebral muscles. After cyst removal with no bone resection, the patient was symptom-free for 7 years. Hydatid disease may be confined to the bone for many years. Intervertebral discs, capsules, and ligaments are resistant to hydatidosis (6). In our case, the disease spread from the rib to the adjacent soft tissues and bony structures, and this may have resulted from direct contact over years. Based on this case, we propose that hydatidosis of the ribs and vertebrae may spread to other sites via the trans-cartilaginous route or by direct contact with nearby soft tissues.

The prognosis for hydatid disease of the spine remains poor. Surgical treatment of osseous spinal hydatidosis is not gratifying because it is extremely difficult to completely extirpate the cysts (7). The rate of recurrence is high because of extensive bone invasion, disease extension to the soft tissue, and difficulty with radical total removal of cysts. Our literature search yielded no treatment strategy for costal echinococcosis. In our patient's case, long-term remission was achieved with an initial surgery that involved extirpation of the cysts alone and no bone resection. However, costal echinococcosis reappeared 7 years later, this time with spinal canal involvement. We believe that the recurrence was likely due to insufficient cyst removal, and suggest that wide resection of affected ribs and soft tissue would improve the prognosis in these cases.

Correct preoperative diagnosis of costal hydatid disease can usually be made on the basis of anamnestic and epidemiological evidence, especially in countries where hydatidosis is endemic. Currently, MRI is the diagnostic tool of choice. On T1-weighted images, viable cysts appear as round low-intensity

lesions with iso- to mildly hyperintense walls. T2-weighted images show hyperintense cystic cavities with well-demarcated hypointense cyst walls. Our MRI findings are similar to those reported by Tekkok et al. (9). In contrast to the findings of Sinner et al., the T2-weighted images in our case showed a hypointense cyst wall, and we observed no gas bubbles on CT or MRI (8). CT of our case demonstrated low- to isodense cysts with hyperdense cyst walls. In addition to what is shown on MRI, CT scanning also reveals the precise anatomical location of these lesions; identifies paraspinous, intraspinal, or intrathoracic extension; and shows that the lesions do not enhance with intravenous injection of contrast material (4). As mentioned, MRI is the gold standard for diagnosing hydatid disease of the spine and rib, but we recommend that CT also be done preoperatively in order to identify any extension of the bony lesion and plan the appropriate surgery. In contrast to the opinions of Bhojraj et al., we believe that MRI may be more suitable than CT for postoperative detection of recurrence during follow-up (3).

In summary, patients with costal hydatid disease should be examined by both CT and MRI to demonstrate spinal involvement. Surgery for hydatid disease of the rib should include wide bone resection as well as extirpation of the cysts.

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