

# VERTEBRAL HYDATID DISEASE AND ITS TREATMENT BY ANTERIOR-POSTERIOR RADICAL EXCISION, FUSION AND CHEMOTHERAPY WITH ALBENDAZOLE. CASE REPORT - ELEVEN YEARS FOLLOW - UP RESULT.

OMURGA KİST HİDATİĞİ VE ALBENDEZOL, ANTEROR VE POSTERİOR RADİKAL EKŞİZYON VE FÜZYON İLE TEDAVİSİ: OLGU SUNUMU VE 11 YILLIK TAKİP SONUÇLARI

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## SUMMARY

Hydatid cyst is a zoonosis caused by the larval form of parasitic tapeworm *Echinococcus granulosus*. We present a case of vertebral hydatid cyst with paravertebral abscesses operated 11 years ago. A 32 year old woman presented multiple giant paravertebral abscesses at the level of T11-T12 and L1 vertebrae and pathological fracture of L1 vertebra because of vertebral hydatid cyst. Posterior instrumentation and fusion followed by anterior L1 corpectomy and fusion were done. Patient was pain-free at eleven-year follow-up. There was no radiological evidence of relapse. Hydatid disease of the spine is rare, misdiagnosis and therefore inadequate treatment and recurrence is frequent. Maintaining the stability of the spine and achieving a fusion mass is important in the decision of surgical technique in vertebral type of hydatidosis.

**Key words:** *Echinococcus granulosus*, hydatid cyst, corpectomy, anterior fusion.

**Level of evidence:** Case report, Level IV.

## ÖZET

Hidatit kist *Echinococcus granulosus*'un larva formu ile oluşan bir zoonozdur. Bu çalışmada 11 yıl önce paravertebral apse ile birlikte vertebral hidatit kist nedeni ile ameliyat edilen olgu sunulmuştur. 32 yaşındaki kadın hastada T11-T12 ve L1 seviyelerinde yaygın paravertebral apse, L1 seviyesinde vertebral hidatit kiste bağlı patolojik kırık saptandı. Posterior enstrümantasyon ve füzyon, anterior L1 korpektomi ve füzyon uygulandı. Bir yıl sonraki kontrolde hastanın semptomları tamamen düzelmişti. Radyolojik tetkiklerde apse nüksü görülmedi. Oldukça nadir rastlanan vertebra kist hidatit tanısı ve tedavisi zor bir enfestasyondur. Nüks ihtimali oldukça yüksektir. Omurganın stabilitesinin temin edilmesi ve füzyon kitlesinin elde edilmesi vertebral hidatidozun cerrahi tedavi kararında oldukça önemlidir.

**Anahtar Kelimeler:** *Ekinokokus granülozus*, hidatit kist, korpektomi, anterior füzyon.

**Kanıt Düzey:** Olgu sunumu, Düzey IV.

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## INTRODUCTION:

Hydatid cyst is a zoonosis caused by the larval form of parasitic tapeworm *Echinococcus granulosus* (5). *E. granulosus* mostly involves liver (50 % – 60 %) and lungs (20 % – 30 %), however, any organ of the body can also be involved (10 %) (7). Hydatid cyst of bone includes 1% of all cases, 50% of this involves vertebrae and most commonly is localized to the thoracic spine and, in decreasing order of frequency, to the lumbar, sacral, and cervical spine (6,9). Diagnosis and treatment of this rare disease is difficult. Patient's history, echinococcal agglutination test, MRI is important for diagnosis. Management of vertebral hydatid disease is much more difficult than soft tissues. It has a very high rate of recurrence due to difficulty of the radical excision. We present a case of vertebral hydatid cyst with paravertebral abscesses operated 11 years ago with anterior corpectomy, total laminectomy and posterior instrumentation.

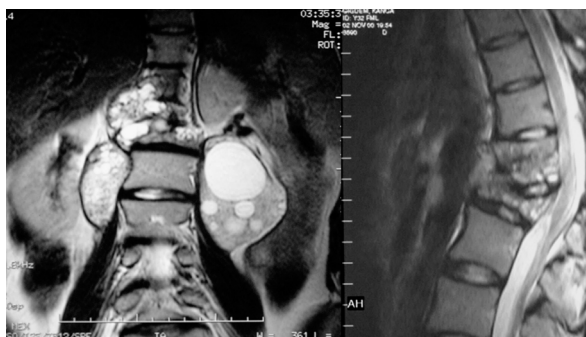
## CASE REPORT:

A 32 year old woman presented with back and low back pain sought on September 2000. She had a previous surgery 3 years ago on 1997 in another hospital and partial laminectomy was applied with the diagnosis of hydatid cyst. Neurologic examination, routine laboratory investigations and chest radiography findings were normal. Severe pain was the major complaint.

Recurrence of previous lesions was detected in laminectomized site and surrounding soft tissue posteriorly. Patient had multiple giant paravertebral abscesses at the level of T11-12 and L1 vertebrae and pathological fracture of L1 vertebra (Figure-1.a-b).

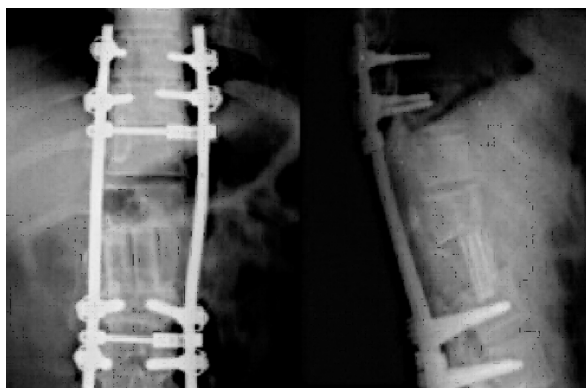


**Figure-1.a.** Preoperative radiographies showing pathological fracture of L1 vertebra,



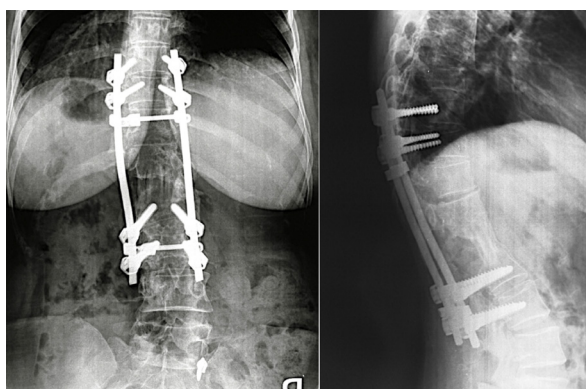
**Figure-1.b.** preoperative MRI showing multiple giant paravertebral abscesses at the level of T11-12 and L1 vertebrae.

Total laminectomy, posterior instrumentation and posterolateral fusion followed by anterior approach with left thoracotomy, paravertebral abscess drainage and decompressive L1 corpectomy were done. Fibular and costal bone grafts were used for anterior fusion. In another session right thoracotomy was done and other paravertebral abscess was excised. The patient had intermittent albendazole medication for 6 months (Figure-2).



**Figure-2.** Early postoperative AP and lateral radiographies.

Patient was pain-free at eleven-year follow-up. There was no sagittal-frontal plain imbalance (Figure-3). Bony fusion was achieved anteriorly-posteriorly. There was no radiological evidence of relapse.



**Figure-3.** Eleven years follow-up radiographies. Fusion and maintenance of both sagittal and frontal balance without recurrence can be seen.

## DISCUSSION:

Cystic hydatid disease is generally caused by a parasitic tapeworm, *Echinococcus granulosus*. Humans are infected by ingesting the eggs excreted by dogs, which are the definitive hosts. The liver and the lungs are involved in approximately 90% of cases, whereas bone involvement has been reported to be only 0.5 to 4%. Spinal involvement in hydatid disease is the result of portovertebral shunts. It usually starts in the vertebral body and grows slowly because of the resistant nature of bone. There are no pathognomic signs or symptoms of spinal hydatid disease other than the symptoms related to compression (1,6,8-9). Generally, the first symptoms are backache and radicular pain (6). Weaknesses of the limbs occur in the later phase of the disease. Paraplegia, the most serious complication of the disease, is caused by compression of the spinal cord by the cysts. In our case the first symptoms were severe back and low back pain without any neurological deficit.

Spinal involvement is found in 50% of the cases. The thoracic and lumbar spine are involved in 75% of cases, and neurologic deficit is reported to occur in 25 to 84% of cases (3,6). Spinal involvement has been classified by Braithwaite and Lees (3) into five types: 1) primary intramedullary hydatid cyst; 2) intradural extramedullary hydatid cyst; 3) extradural intraspinal hydatid cyst; 4) hydatid disease of the vertebrae; and 5) paravertebral hydatid disease. With regard to prognosis, the condition has a reported mortality rate of 50 % (2). Multiple vertebral involvement is unusual.

Diagnosis is usually difficult, and often is not made until clinical signs and symptoms of spinal cord or nerve compression appear. Even then, spinal hydatidosis is often misdiagnosed. Accurate diagnosis can be confirmed by eosinophilia and by positive results of a complement fixation test, a Casoni skin test, and an enzyme-linked immunoassay. MRI characteristically shows an image resembling a bunch of grapes. The cyst walls are thin and regular, with no septations. The presence of a markedly hypointense cyst wall on T1-weighted and T2-weighted images, and the absence of wall enhancement with gadolinium, are characteristic of hydatid disease. In the present case the MRI images showed severe bone destruction, kyphotic deformity and multiple giant paravertebral abscesses with characteristic signals at those affected three levels.

Differential diagnosis are tuberculosis, mycoses, pyogenic abscesses, and benign or malignant neo-

plasm of the spine. Missed diagnosis is frequent and could be devastating because of high mortality and morbidity (4). The treatment of choice is total surgical removal of the cysts without perforation of the cyst wall, and antihelminthic therapy with albendazole or mebendazole. Radical excision is almost impossible in hydatid disease of the spine because of the absence of distinct anatomic planes and the existence of neural structures so the local recurrence rate is high. Our case had a surgical procedure 3 years before our evaluation in another centre and only partial laminectomy was applied. The choice of surgical technique is important especially in type 4 vertebral hydatidosis to total removal of the cysts, correcting the deformity and maintenance of the correction.

The surgical excision of the mass depends on the location and extent of the lesion. The site and extent of the cyst, relation with neurological structures, presence of bone involvement and destruction, spinal instability and deformity is important in decision of surgical procedure type. Laminectomy with simple decompression is used most frequently (9). In subjects with spinal hydatidosis with involvements at one or more vertebrae accompanied by medium and severe kyphosis, decompression, fusion and instrumentation by simultaneous successive anterior posterior surgery is an effective and safe management method for removing the lesion, decompressing the spinal cord and nerve roots and effective kyphosis correction with solid fusion mass. The combination of this surgical procedure with chemotherapy (albendazole) for at least 6 months seems to be the most effective treatment method which also decreases recurrence.

Hydatid cyst should be kept as a differential diagnosis when encountered with a cystic lesion of spine. Hydatid disease of the spine is rare, misdiagnosis and therefore inadequate treatment and recurrence is frequent. Maintaining the stability of the spine and achieving a fusion mass is important in the decision of surgical technique in vertebral type of hydatidosis. Combined anterior-posterior radical excision, correction of deformities in both sagittal and frontal plains, stabilization with strut graft and titanium instrumentation, and chemotherapy with albendazole for at least 6 months for vertebral hydatid disease seems to be the most effective treatment method which also decreases recurrence.

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