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CASE REFOPT / *OLGU SUNUMU*

SPINAL HYDATIDOSIS. REPORT OF A CASE TREATED BY THREE LEVEL CORPECTOMY, ANTERIOR AND POSTERIOR INSTRUMENTATION

OMURGA HİDATİTOZU: ÜÇ SEVİYE KORPEKTOMİ, ANTERİOR VE POSTERİOR ENSTRÜMANTASYON UYGULANAN BİR OLGU SUNUMU

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SUMMARY:

Hydatid disease is caused by larval form of the flatworms Echinococcus granulosus and Echinococcus multilocularis. Bone involvement is extremely rare (2 %), with vertebral column being infested in half of these cases. Thoracic vertebra is the most affected site.

24 years old female presented with severe back pain exacerbated on walking. Radicular symptoms were absent, there was no incontinence or motor-sensory deficit. She had underwent 2 biopsies at another institution with inconclusive results. CT scans and MRI revealed extradural intraspinal paraspinal hydatid disease at T8-9-10 levels. Patient received 4 courses of Albendazol treatment before surgery. She underwent 2 staged anterior and posterior surgery. Decompression and excision was achieved by laminectomy and 3 level corpectomy. Stabilization was achieved via rod-screw construct and titanium cage.

No complications occurred throughout and after the surgery. Patient was mobilized on the first postoperative day. Her pain eased and she remained symptom free till now. IHA levels remained lower than those of before the surgery.

Spinal hydatid lesion needs to be approached as if a malign tumour. Wide resection with combined anterior and posterior approach is necessary for successful treatment and decompression of vertebral and paraspinal hydatid disease. This is the first case where a multilevel spinal hydatid disease is treated with 3 level corpectomy. Multilevel spinal involvement should not be a reason to avoid surgical treatment if the lesion can be excised with three level corpectomy. Andazol therapy prior to surgery is beneficial in decreasing the viability and size of the cyst.

Key Words: *Spinal Hydatidosis, Corpectomy, Echinococcus, surgical treatment*

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

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INTRODUCTION:

Hydatid disease is the name given to infestation by larval form of the flatworm species Echinococcus granulosus and Echinococcus multilocularis. Whilst E. granulosus causes the more prevelant hydatid cyst, E. alveolaris is responsible for the rare but more aggressive alveolar form. Hydatid disease is endemic in Meditterranian region, Middle East, Africa and India [6,9,23]. However, due to indolent course of the disease, patients infested in endemic regions may present with delayed onset of symptoms in non-endemic areas, causing a wider range of distribution and late diagnosis of the disease [7,12,20,21]. The definitive host of the parasite is dogs which carry the adult form of the cestode and shed the eggs in their faeces. Intermediate hosts such as sheep, other ruminants and humans are infested by ingesting the eggs expelled with the dog faeces. Eggs hatch in the intestine of the intermediate host and larvae enter the portal circulation through intestinal mucosa, reaching the liver and lungs (90%), causing cystic disease of these organs [2,3,21].

Larvae may enter systemic circulation by permeating these two filtering organs or by portal shunts, causing hydatid disease in any organ (10%). Bone involvement is extremely rare (2%), with vertebral column being infested in half of these cases [6,10,15,18-20] Bone involvement may result from direct invasion or hematogenous spread. Thoracic vertebra is the most affected site, followed by lumbar, sacral and cervical vertebra. Spinal hydatidosis may be classified as primary intramedullary, intradural extramedullary, extradural intraspinal, vertebral, and paravertebral according to Braithwaite and Lees [4].

We present a case of multilevel thoracic extradural, intraspinal, vertebral and paravertebral hydatid disease treated with two staged anterior and posterior decompression. To our knowledge, this is the first case in literature where a multilevel spinal hydatid disease was successfully treated by 3 level corpectomy.

CASE REPORT:

24 years old female patient presented with severe back pain exacerbated on walking. Symptoms were present for 2 years, but in the last 6 months severity of the symptoms had reached a level where patient could not walk. Radicular symptoms were absent, and there was no incontinence or motor-sensory deficit.

Six months before being referred to our hospital, she had been admitted to another institution and examined for thoracic tumour. Thoracic computed tomography (CT) scan had revealed a mass eroding the T10-11 vertebral bodies, right side posterior elements and right costovertebral junctions, extending along T9-10 T10-11 neural foramina into the spinal canal. Lesion was noted to be extradural, pressing the dural sac to the left (Figure-1). CT images had been interpreted as thoracic tumour and the patient had undergone ultrasound assisted percutaneous biopsy for two times, with inconclusive results. Thoracic magnetic resonance imaging (MRI) scan had been performed and it had revealed 55x60x68mm multilobulated, multicystic heterogenous mass, destructing the T9 vertebral body, right half of the T10 vertebral body, right pedicles, right facet joints, right laminas, and right costovertebral junctions of T9 and T10 vertebra. The lesion was noted to spread along T9-10 and T10-11 right neural foramina into the spinal canal, compressing the dural sac to the left. There was enhancement of the lesion following IV contrast administration. Thoracic kyphosis was increased (Figure-2).

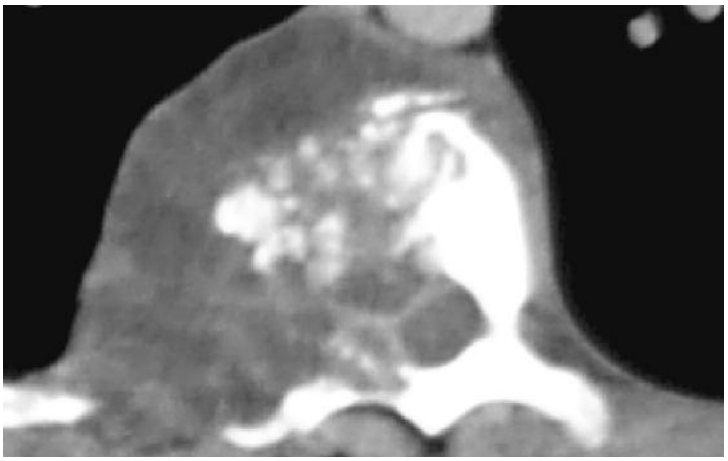

Figure-1. Axial image of preoperative CT scan suggesting a mass eroding the T10 vertebral bodies.

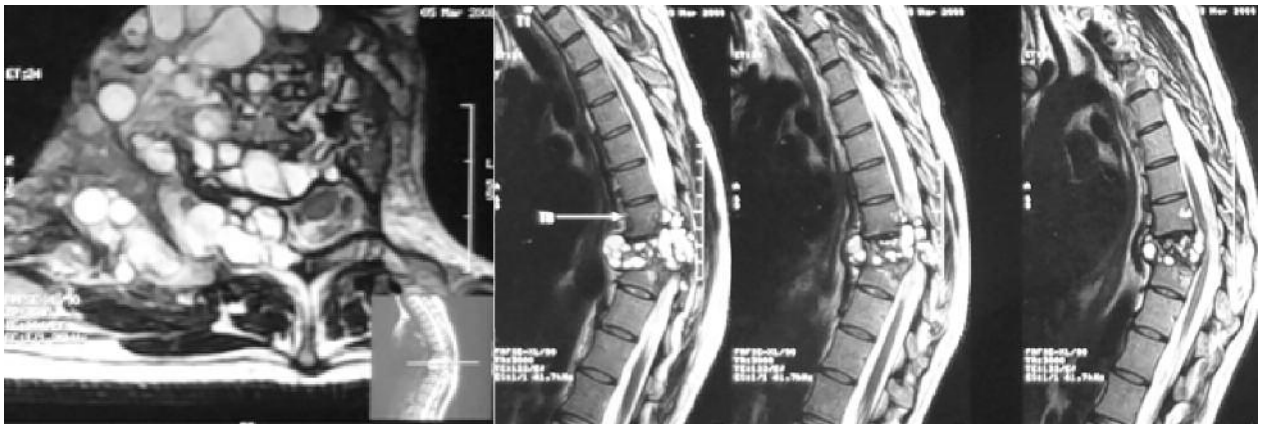

Figure-2. Axial and sagittal views of thoracic MRI scan. Grape-like shape of the Echinococcus spread and compression of the cord is clearly seen.

Despite the inconclusive biopsy reports, she was diagnosed thoracic spinal tumour and referred to Neurosurgery Department at our hospital. She underwent CT guided biopsy to reveal the nature of the tumour mass. Scolex were seen on histological examination and diagnosis of hydatid diseasae was made. The patient was referred to Orthopaedics and Traumatology and Infectious Diseases departments for further treatment.

2 staged combined anterior and posterior surgery was planned due to multilevel involvement of the spine. The patient received 4 courses of albendazol treatment (200 mg 3 X 1). A final MRI scan before the initial surgery showed a decrease in the lesion size. The patient was operated via posterior approach where posterior instrumentation with pedicle screws at T5-6 and T12-L1 levels was performed. Decompression was achieved by laminectomies at the T9-10 level. Control MRI 3 months after the first surgery showed the persisting lesion with additional involvement of the T8 vetebral body, but no cord compression. Echinococcus IHA was positive at 1/4096 dilution.

Patient was hospitalized for the second operation as planned. The lesion was reached by anterior approach. It was noted that T8-9-10 vertebral bodies and paraspinal tissues were infested. Surrounding surgical field was packed with saline soaked gauze and cyst fluid was aspirated carefully without any spillage. Cyst space was irrigated with hypertonic saline followed by removal of the cyst wall from the surrounding tissues. T8-9-10 vertebral bodies were resected. Titanium cage of appropriate size packed with autogenous bone graft was placed in the bone defect formed. Anterior instrumentation between T7-T11 levels was performed with rods and screws (Figure 3 and 4).

Control MRI on May 2009 showed a 2 X 5 cm. cystic lesion in the corpectomy site. Recurrence was suspected and Echinococcus IHA was positive at 1/512 dilution. CT guided aspiration was performed and the results were negative. IHA levels fluctuated between 1/512 and 1/1024, but never reached the high levels those of before the surgery. The patient remained symptom free through the postoperative period.

DISCUSION:

Hematogenous spread of the disease begins in the blood rich cancellous center of the vertebral corpus, sparing the intervertebral disc, spreading to adjacent vertebra, spinal and paraspinal space. As a result of deficient host response, pericyst layer which is found in visceral hydatid cysts does not form in spinal hydatidosis, and a polycystic appearance developes [6,24]. This form may manifest on X-ray as a lytic lesion without periostela reaction. Although cortical bone is relatively resistant to echinoccocus infection, loss of mechanical support, diminished blood supply, increased tissue reaction and accumulation of mediators cause cortical deterioration. Subsidence of the spinal column results in deformities, with compression of the cord in advanced cases [5].

The most common symptom of the indolent bone hydatidosis is pain caused by the cyst's direct compression on the surrounding tissues. More than half of the spinal hydatisosis cases present with symptoms of cord compression. More than half of these cases have paraplegia at the time of

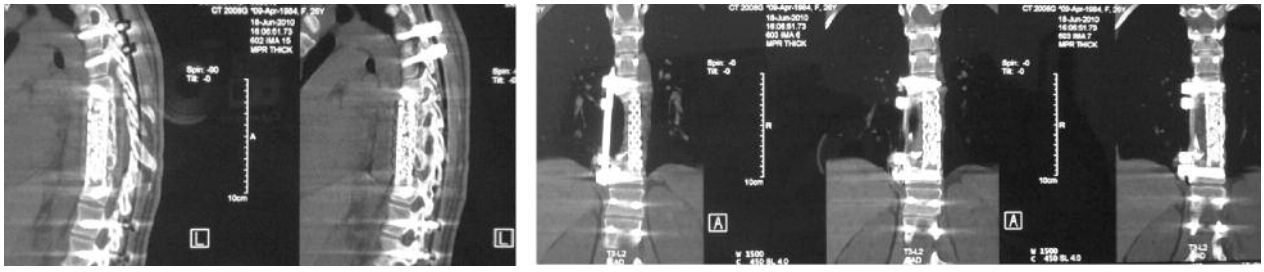

Figure-3. Post-operative 2 years. **(a)** Sagittal and **(b)** Coronal CT views. Three level corpectomy, cage implantation and anterior instrumentation performed.

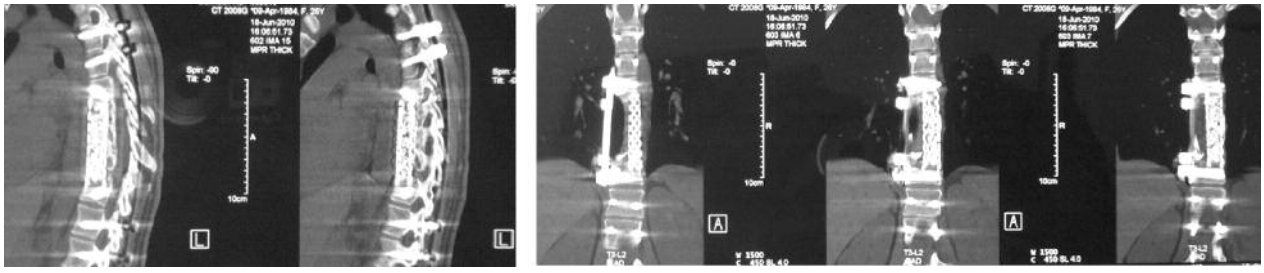

Figure-4. Postoperative 2 years. Lateral and AP XRs. .

presentation [8,11,13-15]. The mean interval from onset of symptoms till diagnosis is approximately 6 months.

If spinal hydatid cyst is suspected, a thorough history must be taken for any previous visceral hydatid disease, and the patient must be examined for liver and lung involvement. CT and MRI scans are mandatory for the diagnosis of spinal echinococcus. CT is especially useful to identify the damaged vertebral structures. MRI is useful for visualization of intraspinal and soft tissue spread. The cyst has the form of bunch of grapes without septation or clear delimitation. Intensity of the cyst fluid is same as the cerebrospinal fluid (CSF) on T1 and T2 weighted images. There is no Gadalinium enhancement of the lesion or the margins [1,16].

The conclusive diagnosis can be made only during the surgery. Presence of visceral cyst must warn the physician about the possibility of spinal hydatid disease. Biopsy or cyst aspiration of the cyst carries risk of anaphylaxis and dissemination, and should never be performed [25,26].

Medical treatment of osseous hydatid disease is less favorable than visceral forms due to poor bone penetration of antihelmintic chemotheuropatics. Treatment of choice is surgical resection where the cyst needs to be approached as if a malign tumour. Lesion must be completely removed, en bloc if possible, and necessary fixation should be performed. Cystic space and the surrounding surgical field must be irrigated with hypertonic saline. Whereas removal of the intradural and epidural cysts is possible following posterior decompression alone, wide resection with combined anterior and posterior interventions is usually mandatory for cases with vertebral and paraspinal tissue involvement [10,15,17,22].

This case is the first one in literature where a multilevel spinal hydatid disease is treated with 3 level corpectomy. Articles till now report maximum 2 levels of vertebra resection. We have achieved satisfactory fusion and stability by two staged anterior and posterior instrumentation and titanium cage with bone graft application. Multilevel spinal involvement should not be a reason to avoid surgical treatment if the lesion can be excised with three level corpectomy.

We have paid great attention to isolate the mass from surrounding tissue. No spillage of the cyst contents was noted during surgery, and so far there is no clinical or radiological sign of recurrence. However, previous aspiration and biopsy attempts might have diminished the efficacy of surgical intervention. Seeding may have occurred during biopsy, and there is no way to distinguish it from future recurrence.

Hydatid cyst of the spine is a serious disease with high morbidity. Advanced cases usually succumb to the complications of plegia and repeating surgeries. If the slightest doubt of spinal hydatid disease exists, patient must be referred to an experienced institution with adequate technical support before attempting any diagnostic or surgical intervention.

Preoperative antihelminthic drug therapy has succeeded to decrease the size of the lesion in this case. Although treatment of choice is surgical resection, we believe that albendazol therapy prior to surgery is beneficial in decreasing the viability and size of the cyst, thereby making the operation easier.

In this case, Ecchinococcus IHA showed to be a reliable indicator of the disease progress. It showed a rapid decline after the surgery and never returned to previous levels.

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