

Hydatid Cyst of the Spine: A Rare Case Report

Spinal Kist Hidatik: Nadir Bir Olgu Sunumu

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ABSTRACT

Hydatid cyst is a zoonotic disease that can affect multiple organs and is difficult to diagnose and treat. Spinal hydatid cyst (SHC) is a rare hydatid cyst involvement observed in 1% of all cases. It can induce various neurological symptoms depending on the region of the involvement. Paraplegia is one of the most prevalent neurological symptoms. In this case report, a 63-year-old male patient with bilateral lower extremity paraplegia was operated on by neurosurgery and diagnosed with SHC at the level of Th 11 vertebra in the pathological examination of surgically removed materials. Thus, we aimed to emphasize the significance of pathological and microbiological examination in the differential diagnosis of spinal disorders.

Keywords: Spinal hydatid cyst, hydatidosis, echinococcosis, paraplegia, albendazole

ÖZ

Hidatik kist, çeşitli organ tutulumlarına neden olabilen, teşhisi ve tedavisi zor olan zoonotik bir hastalıktır. Spinal hidatik kist (SHK), tüm olguların %1'inde görülen nadir bir hidatik kist tutulumudur. Tutulumun yerine bağlı olarak çeşitli nörolojik semptomlara neden olabilir. Parapleji, bu nörolojik semptomların en yaygın olanlarından biridir. Burada, bilateral alt ekstremitte parapleji nedeniyle beyin cerrahisi tarafından opere edilen ve cerrahi olarak çıkarılan materyallerin patolojik incelemesinde T11 seviyesinde SHK tanısı konan 63 yaşında bir erkek hasta olgusu sunulmaktadır. Biz bu olgu ile, omurga hastalıklarının ayırıcı tanısında patolojik ve mikrobiyolojik incelemenin önemini vurgulamayı amaçladık.

Anahtar Kelimeler: Spinal kist hidatik, hidatidoz, ekinokokkoz, parapleji, albendazol

INTRODUCTION

Hydatid cyst (CH), Cystic echinococcosis, hydatidosis or hydatid disease is a zoonosis caused by the larval stage of *Echinococcus granulosus* or *E. multilocularis* and occurs worldwide. Although disease is common and on the notifiable list of diseases in our country, its incidence is not fully known because it can remain asymptomatic for years and is on the notifiable list of diseases. Most of the epidemiological data is based on reported cases, hospital records and studies based on serological methods and does not reflect the truth. Also, the absence of disease in most seropositive cases limits the value of studies based on serological test results. Although group C disease reporting has

been in the notifiable infectious disease category in our country since 2005, there are approximately 500 notifications annually. The number of cases reported to the Turkish Ministry of Health between 2008-2012 is 3006; according to the Social Security Administration data Republic of Turkey in this period, the number of surgical procedures for these infections reported as 12556 (1,2). *Echinococcus granulosus* lives in the intestines of hosts such as dogs and wolves. It is swallowed by intermediate hosts and thrown in the fecal material; disease occurs in humans by eating animals such as sheep, cattle and goats or by accident. Contamination occurs through eating contaminated food without cooking, drinking water and breathing. It may involve all organs (kidney, spleen, brain,



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musculoskeletal and heart), especially the liver and then the lungs (3,4). While 0.5-2.0% of all reported cases occur in the musculoskeletal system, 50% of the cases are spinal hydatid cysts (SHC) (4).

In this case report, it was aimed to emphasize the importance of CH that although rarely, can cause spinal involvement, cause paraplegia, and should be considered in the differential diagnosis of undiagnosed mass lesions, especially in cystic structures.

CASE REPORT

A 63-year-old male patient living in rural area and dealing with animal husbandry, was admitted to the neurosurgery outpatient clinic of our center due to loss of bilateral lower extremity strength for the last one month and complete motor function loss, paraplegia and immobilization that developed in the last one week. He had been describing swelling in the back area for 3-4 years. It was learned that the swelling was tried to be excised with local anesthesia as pre-diagnosis of lipoma, but it was not successful. He had known diagnoses of diabetes mellitus, hypertension, chronic obstructive pulmonary disease, previously cerebrovascular disease and carotid artery stenosis. Vital signs at first admission was normal, except oxygen saturation was 98% under nasal O₂ treatment at 3 Lt/min. Physical examination revealed a distinct, painless swelling in the thoracolumbar region, which could be detected by inspection and palpation. In his neurological examination, bilateral paraplegia was found in the lower extremities, but no motor deficit in the upper extremities. The patient had urinary and fecal incontinence. On his chest examination revealed bilateral expiratory wheezing in both lungs. Laboratory test results in the patient's first application were normal expect white blood cells count with 14.500/mm³.

In thoracic (Th) magnetic resonance imaging (MRI) before admission and in Th computed tomography (CT) taken at our center, extradural mass lesions at Th 10-11 levels, initially thought to be compatible with metastasis, involving the dorsal colon, right pedicle and lamina and soft tissue densities were observed (Figure 1). In abdominal CT there was a hypodense cystic mass lesion with necrotic, hyperdense calcific foci extending towards the spinal canal and invading the spinal cord at the level of the Th 11 vertebra, originating from the right posterolateral part, causing bone destruction in the vertebral corpus and posterior elements, extending from the inferior to the right lower quadrant in the thorax, invading the paravertebral muscle groups.

As a result of the imaging, the differential diagnosis was considered to include SHC, and the indirect hemagglutination (IHA) test for CH was requested and it was found positive at 1/1280 titer. After this stage, the patient was consulted in terms of treatment recommendations and preoperative evaluation. With the pre-diagnosis of CH, we recommended that albendazole 2x400 mg/day (oral) treatment be initiated and the operation was postponed for two weeks.

Rose Bengal, wright, wright containing anti-human globulin (coombs wright), 2-mercaptoethanol, VDRL-RPR, quantitative VDRL, Treponema pallidum hemagglutination, heterophile antibody, acid resistant bacteria staining by Erlich Ziehl Nielsen technique tests were requested in terms of some other infective diseases that should be considered in the differential diagnosis while preoperative preparations were made. These examinations were negative.

The patient was operated approximately 3 weeks after his first admission. White membrane cysts draining spontaneously at intra-operative Th 11 level were seen. It was observed that the cysts destructed the Th 11 vertebral right lamina, the right facet joint and the right rib and went deep enough to almost reach the pleura. Appropriate samples were taken from these cysts and frozen for examination, pathology and microbiology. The cysts were cleaned with appropriate washing and excision, after the hemovac drain was placed, patient was closed and the operation was terminated. Frozen examination was evaluated as compatible with CH. A diagnosis of CH was made in the pathological examination of the bone biopsy abortion material sent intraoperative afterwards (Figure 2, 3).

The patient was consulted to us for discharge on the postoperative 9th day. In the postoperative period, the patient's paraparesis continued and was still immobile. The wound looked clean. The patient, who had been receiving albendazole 2x400 mg/day (oral) treatment for 28 days due to CH, was discharged with the recommendation of outpatient follow-up. In the outpatient clinic visit 1.5 months later, CH IHA test titer decreased to 1/320. Continuation of his current treatment was recommended. He was called for control again 15 days later. In the next period, detailed



Figure 1. Extradural cystic lesions at Th10-11 levels (vertebral MRI)

Th: Thoracic, MRI: Magnetic resonance imaging

information was given to the patient about the necessity of regular serology and imaging follow-up for CH every 3-6 months.

DISCUSSION

Although CH disease often causes liver and lung involvement, it may rarely (1-2%) cause bone involvement. The incidence of the disease is very low due to the accumulation of *Echinococcus* larval forms filtering from the liver/lungs into bone tissue. Vertebral involvement has also been shown in 45-50% of patients with bone involvement (1-5). Among the *Echinococcus* species, *E. granulosus* is the most common form, while *E. multilocularis* causes rare alveolar form. Since they can invade the neighboring tissue where they are located, they often misdiagnosed as carcinoma or sarcoma. Also, sometimes parts of the parasite can metastasize to distant areas. Initial imaging studies are often highly suspicious for carcinoma or sarcoma (4). The first SHC case was reported in 1948, and reports of cases continue worldwide today. It was suggested that; if a patient had involvement in any area other than liver or lung, other organs should be scanned by imaging methods (5). Paraplegia has been reported in 66-70% of cases with vertebral involvement (4,6). In a pooled analysis by Suntutur and Çavuş (7); 86 cases of SCH from our country were examined. 48 (56%) of the cases were male, the mean age was 36.4±16.8 (8-73 years) years. The most common complaints were back pain (83%), walking difficulty (47%), urine and/or stool incontinence (27%) and numbness in the legs (22%). In the case presented, there was bilateral paraplegia and complete motor function loss were found in the lower extremities, similar to the literature. A suspected image of metastasis or SCH was detected in the MR and CT examinations of the patient.

Neumayr et al. (8) analyzed the SHC literature reported between 1965 and 2012, in a review study that included 189 publications in which 367 publications were reviewed. As a result of the analysis, it was reported that the mean age was 35 years (3-77), and the prevalence was higher (56.9%) especially in male patients. The presented case was also 63-year-old male patient.

The most common locations of spinal HC are the Th region (range 45-50%), followed by the lumbar (range 20-39%), sacral (20%) and cervical (10%) areas. The reason for the involvement

in the Th and lumbar regions is thought to be due to the intense local vascularization and the rich blood circulation of the spongy vertebral bones. The presented case also had Th 11 involvement.

Often specific in routine laboratory tests changes are not seen in CH cases. Often liver function tests detected as normal or rarely may increase the level of transaminases and cholestasis enzymes due to biliary tract involvement (9). No abnormality was found in our patient's laboratory examinations other than serology. Laboratory tests, immunological methods and radiological imaging methods are used for diagnosis. Secondary infection in the cyst can develop; may be fistulized to neighboring organ; allergic reactions may develop due to the allergen content of the cyst (9). The diagnosis of the presented patient was made both serologically, radiologically and pathological examination of the the tissues taken peroperatively.

In the differential diagnosis of SCH, pathologies such as pyogenic infections, tuberculosis, metastatic malignancies, fibrous dysplasia, multiple myeloma, giant cell tumor and enchondroma should be considered (4-6). No abnormality was found in the tests performed for the differential diagnosis of the presented case.

Braithwaite and Less (6), stated according to their radiological involvement; SCHs divided into 5 groups as intramedullary, intradural extramedullary, extradural, vertebral, and paravertebral involvement that spread to spinal structures. While the first 3 groups are extremely rare, the last two groups affect spinal neural structures and are common. In our case, the involvement was primarily in the spinal region and other examinations performed and no other organ involvement was detected.

SHC usually progresses with dorsal vertebral involvement and is mostly multiple involvement. In spinal involvement, the vertebral corpus is initially involved, usually through a port-venous shunt. *Echinococcus* embryos grow multilocular in vertebral bodies, causing tumor-like infiltration and damage. Subsequently, it invades the pedicle and lamina. Finally, it spreads across the cortex to surrounding soft tissues and bone structures (4-6). CT and MRI findings of presented case were typical for SCH.

SHC infection may cause various symptoms such as pain, sensory defects, motor loss, neurogenic bladder, and bowel dysfunction, Paraplegia depending on the region it is involved due to direct

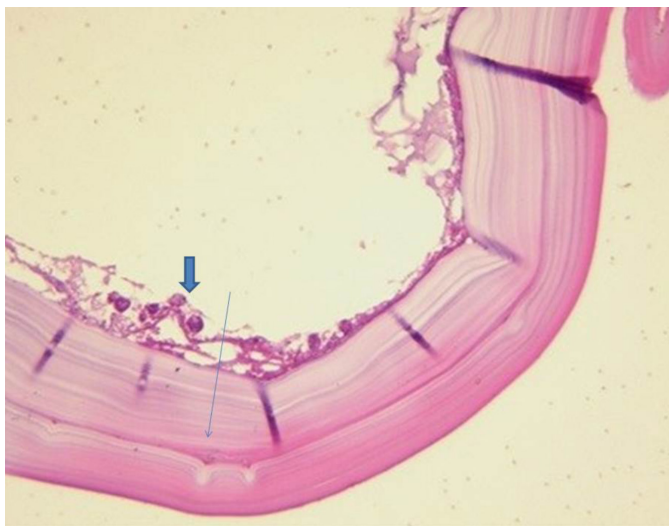


Figure 2. The germinative membrane and scolexes seen in the microscopic examination of the bone biopsy material

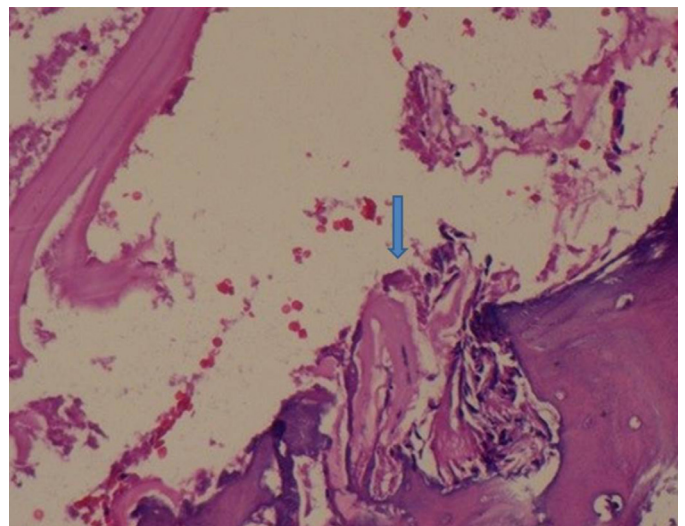


Figure 3. Mature bone trabeculae and germinative membrane between them

compression of the cyst, ischemic changes, or invasion of the cyst which is one of the most serious complication of the disease. In studies conducted, mortality was reported as 14-58% in cases with paraplegia, and the mean survival time after diagnosis was found to be 5 years. In our case; at the Th level, extradural, multiple cystic lesions destructing the dorsal column, right pedicle and lamina of the vertebra and invading the paravertebral muscle groups were detected. In his clinic, he had been describing loss of lower extremity muscle weakness and also incontinence for the last month and complete motor loss for the last week.

There are case reports of recurrence even with medical or surgical treatment. Albendazole and 6 month treatment duration was the most preferred treatment protocol in our country, but even despite the medical treatment, 51% of the patients have at least one relapse was observed (5,7,10). For example, Diktaş et al. (5) reported a 70-year-old male case who was operated 11 times for a CH with pelvic and spinal involvement. El-On (10) reported; a 53-year-old male patient with a severe destructive lesion of the L4 vertebral body caused by *E. granulosus*, who underwent two operations and was treated continuously with albendazole followed by praziquantel, but no eradication could not be achieved despite long-term chemotherapeutic therapy combined with surgery.

Spinal CH treatment is particularly difficult in patients with vertebral and paraspinal involvement, spinal instability and recurrence. Control examination, radiology and serology follow-up are required at regular intervals. Cysts should be removed without rupture during surgery and the surgical site should be irrigated with hypertonic saline solution to reduce the risk of recurrence (11). The recommended duration of anthelmintic therapy is 4-6 months (3,11,12). The presented case treated with albendazole and operated approximately two weeks later of treatment. In the postoperative period, inpatient follow-up and treatment continued for about 10 days. The patient came for control once on the 15th day after discharge. Serological regression was detected, but the patient did not have a thorax CT, so radiological evaluation could not be evaluated. Detailed information on the necessity of regular serology and imaging follow-up for CHs was given to the patient at discharge and first check-ups in every 3-6 months. However, the patient did not come back for control after the first control.

Although this disease is rare worldwide, cases/case series reported from our country are available in the literature (4,5,9,11-16). This may be attributed to the fact that our country is one of the endemic regions for this disease.

In conclusion, SCH is one of the preliminary diagnoses that should be considered in the differential diagnosis of mass lesions with vertebral involvement. The importance of early diagnosis and treatment should be kept in mind for this disease which usually causes symptoms secondary to compression which can cause paraplegia and adversely affect the quality of life.

*Ethics

Informed Consent: Informed consent was obtained.

Peer-review: Internally peer-reviewed

*Authorship Contributions

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