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Percutaneous Aspiration Injection and Re-aspiration as A Minimally Invasive Treatment for Spinal Cystic Echinococcosis: A Case Report

Spinal Kistik Ekinokokkozisde Minimal İnvaziv Tedavi Olarak Perkütan Aspirasyon İnjeksiyonu ve Reaspirasyon: Olgu Sunumu

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ABSTRACT

Cystic echinococcosis is a parasitic disease with significant importance for public health in endemic regions. Spinal cystic echinococcosis, however, is a rare form that may lead to severe complications due to its localization. In this manuscript, we presented a 16-year-old male patient who admitted with abdominal and back edema for 2 months, evaluated with preliminary diagnoses of Pott's abscess and malignant mass, subsequently diagnosed with spinal cystic echinococcosis. It was concluded that cystic echinococcosis should be considered in differential diagnosis of large cystic masses and percutaneous aspiration, injection, reaspiration method might be a safe and effective treatment option particularly for cases of complicated spinal cystic echinococcosis.
Keywords: Cystic echinococcosis, spinal, child, percutaneous aspiration injection and re-aspiration

ÖZ

Kistik ekinokokkozis, endemik bölgelerde halk sağlığı açısından büyük öneme sahip bir paraziter hastalıktır. Spinal kistik ekinokokkozis ise çok nadir görülen ve yerleşim yeri sebebiyle ciddi komplikasyonlara yol açabilen bir formudur. Bu makalede 2 aydır karın ve sırtta yaygın şişlik şikayeti ile başvuran, Pott apsesi ve malign kitle ön tanılarıyla değerlendirilen spinal kistik ekinokokkozis tanısı alan 16 yaşında erkek hasta sunulmuştur. Sonuç olarak, büyük kistik lezyonların ayırıcı tanısında kistik ekinokokkozisin akla getirilmesi gerektiği ve perkütan aspirasyon, enjeksiyon, reaspirasyon yönteminin özellikle kompleks spinal kistik ekinokokkozis olgularında güvenli ve etkili bir tedavi seçeneği olabileceği kanısına varılmıştır.
Anahtar Kelimeler: Kistik ekinokokkozis, spinal, çocuk, perkütan aspirasyon enjeksiyon ve reaspirasyon

INTRODUCTION

Cystic echinococcosis (CE) is a serious disease caused by the *Echinococcus granulosus* tapeworm (1). Infections with *E. granulosus* complex parasites are frequently acquired during childhood, yet the cysts they generate might take several years to grow to a size detectable enough to cause symptoms. These distinct cystic lesions predominantly manifest in the liver (70%) and the lungs (20%), but other parts of the body can also be affected (2,3). Spinal CE, a rare and serious condition with a high risk of recurrence (1,4).

Herein we present a case of spinal CE who initially presented with a massive abdominal mass and was successfully treated with percutaneous aspiration, injection, and re-aspiration (PAIR).

CASE REPORT

A previously healthy 16-year-old male was referred to our clinic for evaluation of a painless, large mass covering the left abdomen and back. His family reported that the mass had been enlarging over the past two months. There was no history of animal

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contact, weight loss, or night sweats, and the patient had no known medical conditions or exposure to tuberculosis. The patient was residing in a town within Central Anatolia Region.

On admission, vital signs were normal. Physical examination revealed a 9x10 cm swelling in the left lower quadrant of the abdomen and a 12x10 cm mass on his back. Vertebral tenderness was noted upon deep palpation of the L2-L3 vertebrae.

Laboratory tests revealed a white blood cell count of 5,880/mm^3, hemoglobin level 13.7 g/dL, platelet count 291,000/mm^3. C-reactive protein of 3.7 mg/L (normal range <5 mg/L) and an erythrocyte sedimentation rate of 24 mm/h (normal range <20 mm/h). Serum transaminases, electrolytes, creatinine, immunoglobulins were normal and HIV serology was negative. Blood culture, brucella immunocapture test, tuberculin skin test and cyst hydatid indirect hemagglutination test were requested.

Abdominal ultrasonography revealed a large mass with dense cystic regions and thick walls. The mass originated from the lower vicinity of the left kidney, occupied the left lumbar lobe and lower quadrant, and extended towards the proximity of the bladder. Subsequent abdominal magnetic resonance imaging (MRI) showed cystic lesions that were multilobulated and septated, originating from the paravertebral region, causing displacement of the left kidney towards the anterosuperior direction. The MRI further revealed the mass extending into the iliopsoas muscle and the subcutaneous tissue of the lumbar region, measuring 12x12x23 cm (Figure 1a). There were T2 hyperintense lesions in the medullary cavity of lumbar 2nd and 3rd vertebra consistent with bone involvement. These findings were compatible with Pott's disease. Tuberculin skin test and brucella immunocapture tests were negative.

For diagnostic and therapeutic purposes, the interventional radiology department performed several procedures, including a planned biopsy for differential diagnosis. During the biopsy, it was noted that the mass lacked solid characteristics. Instead, purulent to clear and colorless to pale yellow fluid was observed. A catheterization drain was then inserted, and daily irrigation was carried out via 1000 cc of normal saline. By the 6th day of treatment, the drained fluid transformed into a rock-colored water with white membranes floating in it (Figure 2). Hydatid cyst was diagnosed with histopathological examination. An indirect hemagglutination test revealed the presence of anti-hydatid cyst antibodies at a titer of 1/640. Other organs were screened and no cysts were detected.

Albendazole therapy was initiated. During the last visit, the patient was asymptomatic and he was discharged with a plan for albendazole therapy lasting a minimum of six months, along with regular follow-up visits. The control MRI at the six-month mark indicated the absence of any residual cystic lesions, with the observations interpreted as residual inactive areas (Figure 1b).

DISCUSSION

Spinal CE is a rare manifestation of hydatid disease, accounting for less than 1% of all cases. It can be difficult to diagnose without typical clinical, laboratory, and imaging findings because it can mimic other conditions, such as tuberculosis, bone tumors and metastatic

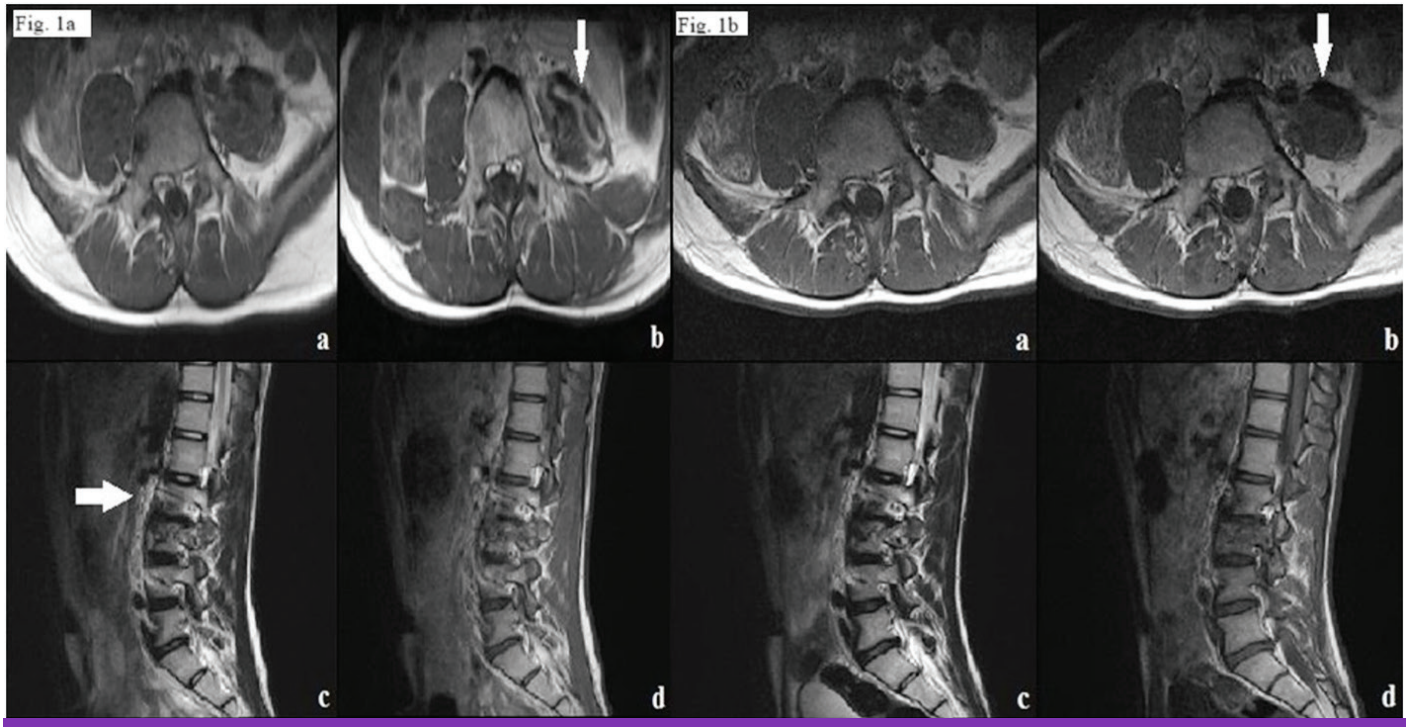

Figure 1a. Abdominal MRI reveals a multiloculated, septated cystic lesion (arrow) originating from the paravertebral region, extending into the medullary cavity of lumbar vertebrae and displacing the left kidney anterosuperiorly. (a) T1-weighted axial MRI (b) T1-weighted axial MRI with contrast (c) T2-weighted sagittal MRI (d) T1-weighted sagittal MRI with contrast; **Figure 1b**. There are several residual, inactive foci (arrow) in the paravertebral area, at the site of the previous hydatid cyst as revealed by the control MRI at month 6 after treatment. The bone involvement was stabil (a) T1-weighted axial MRI image (b) T1-weighted axial MRI with contrast (c) T2-weighted sagittal MRI (d) T1-weighted sagittal MRI with contrast

MRI: Magnetic resonance imaging

Figure 2. An image showing a container of drainage fluid, with white membranes floating in a rock-colored water

diseases. Upon reviewing radiological findings indicating a paravertebral cystic lesion, Pott's disease (PD) emerged as a potential differential diagnosis for our patient. PD, also known as tuberculous spondylodiscitis, is a type of osseous spinal tuberculosis (4,5). Back pain is the most common presenting symptom, while constitutional symptoms such as fever or weight loss may not be present (6). Computed tomography scans can show bone hydatid disease, which can sometimes be mistaken for tuberculosis, metastases, giant cell tumors, or bone cysts. However, MRI findings are more distinctive, especially in distinguishing spinal CE from other conditions (3,4,7). MRI shows well-circumscribed, cystic lesions, with CSF-like signal intensities, hypointense on T1-weighted imaging, and hyperintense on T2-weighted imaging. T2-weighted images show a low-intensity rim surrounding the homogeneous hyperintense cyst contents. The cyst wall may be thin and regular, isointense, or demonstrate a slightly lower signal than its contents. A markedly hypointense cyst wall on T1- and T2-weighted MRIs is characteristic of hydatid disease (8).

Treatment for vertebral CE combines surgery and antiparasitic therapy. Removing intact cysts is essential, as cyst perforation can cause systemic spread and chronic recurrence (4). Decompression surgery is vital in managing these patients due to the increased risk of spinal cord compression (2). There is a case report in the literature detailing successful treatment of intra and para-sacral cysts using rigid endoscopy (9). Although a limited number of patients have been treated with PAIR combined with albendazole, one patient with a cervical vertebral hydatid cyst and severe clinical symptoms achieved complete resolution, while another patient with recurrent disease following complex surgery was successfully treated, resulting in the complete resolution of two vertebral cysts. The authors recommended the PAIR for extensive disease where radical removal is not feasible and where previous surgical interventions may exacerbate re-operation (10,11). We successfully treated our patient with PAIR procedure combined with albendazole, with no complications. Further research is needed to evaluate the long-term efficacy and safety of the PAIR for spinal CE. In an another study in which a total of 50 pediatric CE patients were evaluated, spinal CE was detected in two of them and they underwent surgical excision. They were treated with albendazole. No recurrence was observed (12). An 11-year-old male patient diagnosed with multiple CE in the liver and lungs presented with complaints of difficulty in walking and leg pain one year after discontinuing albendazole treatment and then he diagnosed with spinal CE. The patient underwent surgery and was then treated with albendazole for another 4 years. No recurrence was observed in the subsequent one-year follow-up. It was emphasized that recurrences and systemic spread should always be kept in mind (13). Albendazole or mebendazole treatment is essential to prevent recurrence. The optimal duration of treatment remains uncertain, and treatment should be individualized (14).

While serological tests serve as valuable tools in differential diagnosis, they can yield a high rate of false-negative results. The immune response tends to be higher in ruptured hydatid cysts. Serological tests often produce negative results when a cyst is aging, calcified, or no longer viable (3). Our patient exhibited a titer of 1/640 on the indirect hemagglutination test.

CONCLUSION

Hydatid cysts can manifest in diverse clinical presentations, and spinal hydatid cyst, although rare, represents a serious condition with significant morbidity and a complex treatment course. Patients with an abdominal mass of spinal origin should prompt consideration for hydatid cysts in the differential diagnosis, and the PAIR procedure appears to be a promising treatment option.

*Ethics

Informed Consent: Written informed consent was obtained from the parents. Approval was obtained from our patient's parents for the data to be published in the journal.

*Authorship Contributions

Surgical and Medical Practices: Ö.M.A., S.B., Concept: K.Y., Design: M.G., Analysis or Interpretation: Ü.K., Literature Search: M.G., Ü.K., Writing: Ö.M.A., K.Y., S.B.

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