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HYDATID CYST OF SACRUM AFFECTING THE SACROILIAC JOINT: A CASE REPORT

SAKROİLİAK EKLEMİ TUTAN SAKRUM KİST HİDATİĞİ: OLGU SUNUMU

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SUMMARY

Objective: To describe a rare instance of a hydatid cyst that caused severe and progressive lower back pain and was misdiagnosed as sacroiliitis.

Introduction: Hydatid disease or hydatidosis is a serious human cestode infection worldwide with a characteristic geographic distribution. The liver and lungs are the most frequently involved organs. Bone involvement is seen in less than 4% of cases, and vertebral hydatid disease is uncommon. The symptoms of sacroiliac joint and lower back pain cause difficulties for differential diagnosis, and this rare condition may be misdiagnosed.

Case Report: We report a case of a 21-year-old woman with hip pain caused by a secondary hydatid cyst of the sacral and pelvic areas. The patient was diagnosed with sacroiliitis and sciatica in another center. As treatment for these failed, the patient was referred to our clinic. Physical examination and radiological tests revealed iliac bone lesions starting from the left sacral area and extending to the sacroiliac joint.

Results: Following medical treatment, curettage and grafting were performed. Macroscopic imaging and histopathological evaluation of the material that was removed from the lesion field were consistent with hydatid disease. No recurrence was detected and the patient was free of symptoms after 11 months of follow-up.

Conclusions: Bone hydatid disease can be misdiagnosed in the early periods. When it progresses, it results in destruction of the bone. In patients with a history of hydatid disease and with musculoskeletal complaints, the possibility of hydatid cysts should be kept in mind. There are ongoing debates as to whether bone grafting or acrylic cement are safer for the management of the remaining cavity, and we consider bone grafting to be the safer treatment.

Key words: Cyst hydatid, surgical treatment, spinal hydatid infection

Level of evidence: Case report, Level IV

ÖZET

Amaç: Ciddi ve ilerleyici bel ağrısına neden olan ve yanlış tanı almış nadir görülen bir kist hidatik vakasını tanımlamak.

Giriş: Hidatik hastalık ya da hidatidozis dünyada yaygın bir insan sestod enfeksiyonudur ve karakteristik coğrafik bir dağılıma sahiptir. Karaciğer ve akciğerler en sık tutulur. Vakalarda kemik tutulumu %4'den daha azdır. Vertebral hidatik hastalık nadirdir. Sakroiliak eklem bulguları ve bel ağrısı ayırıcı tanıyı zorlaştırır ve bu nadir durum yanlış tanıya neden olabilir. Olgu Sunumu: Sakrum ve pelvisin sekonder hidatit kistlerinin kalça ağrısına neden olduğu 21 yaşında bayan hasta sunuldu. Başka bir merkezde sakroileit ve siyatik ağrısı tanısı almış ve tedavisi başarısız olan hasta kliniğimize sevk edildi. Fizik muayenesi ve radyolojik testleri sol sakrumdan başlayıp sakroiliak kemiğe uzanan iliak kemik lezyonlarını açığa çıkardı. Sonuç: Medikal tedaviyi takiben, küretaj ve greftleme uygulandı. Lezyondan çıkarılan materyalin makroskopik görünümü ve histopatolojik değerlendirilmesi hidatit hastalık ile uyumluydu. 11 aylık takibinde nüks gözlenmedi ve hasta semptomsuzdu.

Tartışma: Erken dönemlerinde kemik hidatik hastalığı yanlış tanı alabilir. Hastalık ilerler ve kemiği destrükte eder. Hidatitk hastalık öyküsü olan ve kas-iskelet şikâyetleri olan hastalarda hidatik kist hastalığı ihtimali akılda tutulmalıdır. Debride edilen vertebrada kalan kavitenin kemik greftiyle doldurulmasının daha güvenilir olduğuna inanıyoruz.

Anahtar Kelimeler: Kist hidatit, cerrahi tedavi, omurga kist hidatiti

Kanıt Düzeyi: Olgu sunumu, Düzey IV

INTRODUCTION

Echinococcosis is a zoonotic infection caused by *Echinococcus* species, and is one of the most important helminthic diseases worldwide. *Echinococcus granulosus* is a small tapeworm that requires humans as intermediate hosts. Human infection is caused by ingestion of the tapeworm eggs. Involvement of the liver and lungs is most common[7]. Bone hydatid disease is a rare pathology, and bone involvement represents only 1–2% of all hydatid cyst cases. The iliac bones and the spine are the most common sites[3]. This case report describes a rare instance of a hydatid cyst that caused severe and progressive lower back pain (LBP) with S1–2 neural foramen involvement, without neurological dysfunction.

CASE REPORT

A 21-year-old female presented with hip and LBP. She described the pain as radiating towards the left thigh with no change when resting. In her medical history, six years previously she had been operated on three times for lung hydatid cysts, and she has also been treated with antiparasitic therapy after surgery. She was followed up and treated for sacroiliitis and sciatica because of the left hip pain for six months at another center.

At presentation, her vital signs were normal. She had lower back pain and a normal range of motion of the lower extremities. The straight leg raising test was positive on the left side at 70–80°. The results of neurological examination of the lower limbs were normal. The laboratory findings were consistent with increased levels of lymphocytes, eosinophills, and C-reactive protein. An indirect hemaglutination test was positive for hydatid cysts.

A chest X-ray and thorax computerized tomography (CT) revealed increased density and consolidation on both lower lobes of the lungs. An abdomen CT revealed cysts that were compatible with type 2 and type 3 hydatid cysts on the hepatic parenchyma. A plain pelvic radiograph revealed an expansive destructive lesion on the left iliac bone and sacrum extending to the sacroiliac joint (SIJ) (Figure-1A).

A pelvic CT showed multilobular cysts located in the left hemi-sacrum, expanding to the S1–2 neural foramen and extending to the iliac bone and SIJ (Figure-1B).

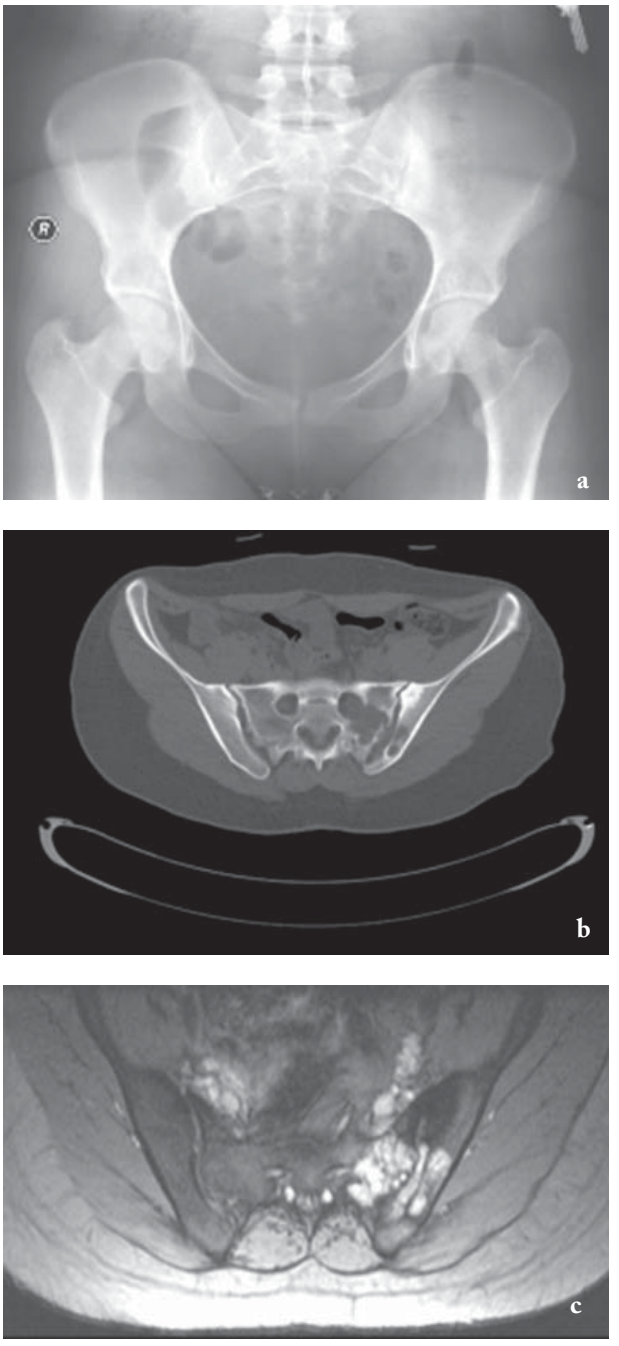

Figure-1. a. A plain pelvic radiograph showing an expansive destructive lesion on the left lateral sacrum extending through the right SIJ and iliac bone. **b.** Computerized tomography (CT) scans showing multiloculated cysts located in the left hemisacrum expanding the S1–2 neural foramen and extending to the iliac bone and SIJ. **c.** T1-weighted MRI revealing a hypointense cystic lesion in the S1 and S2 sacral ala, extending to the right SIJ and invading through the sacral canal with displacement of the S1 and S2 nerve roots.

A magnetic resonance T1-weighted image revealed a hypointense cystic lesion in the S1 and S2 sacral ala, extending to the left SIJ and invading through the sacral canal with displacement of the S1 and S2 nerve roots (Figure-1C).

Mebendazole treatment (100 mg twice daily) was administered for two weeks before surgery.In surgery, the bony window was removed from the posterior ileum over the cystic lesion. Multiple pearly-white capsulated cysts were enucleated. The affected sacroiliac joint and S1–2 sacral ala were curetted, and the area was irrigated with hypertonic saline and 10% polyvinyl iodine (Figure-2A, 2B).

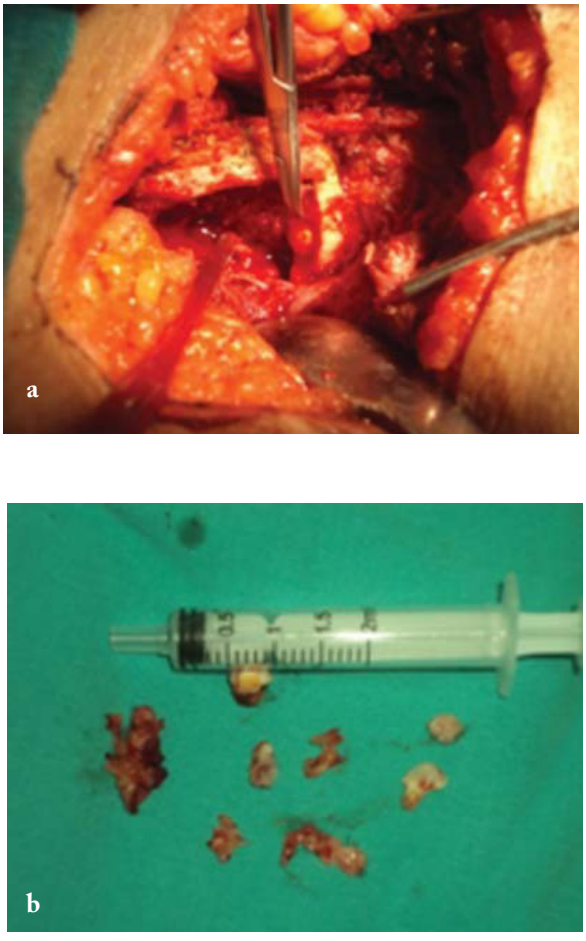

Figure-2. a. Cavity and hydatid cyst **b.** Enucleated hydatid cysts

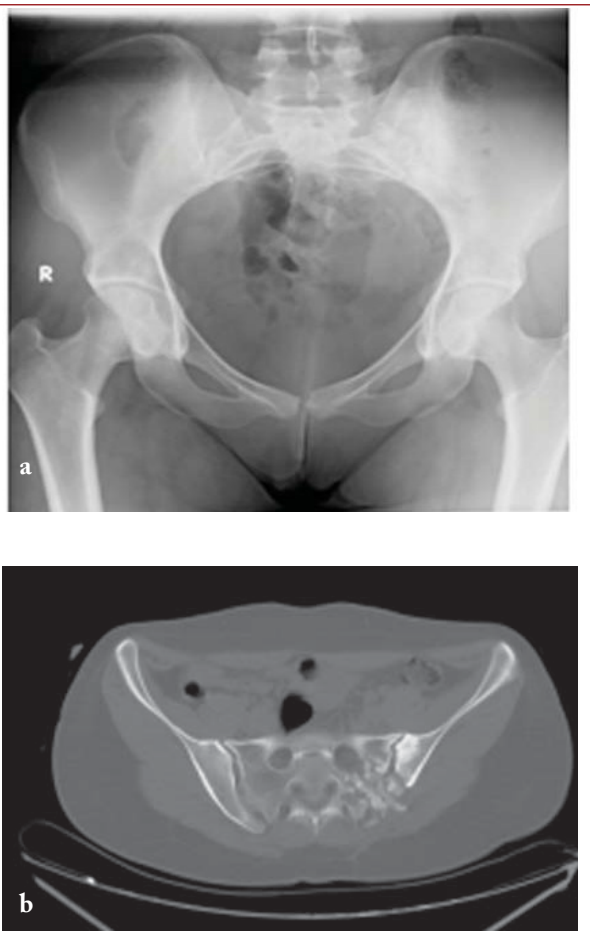

Figure-3. a. Plain pelvic radiograph 11 months after surgery. **b.** Computerized tomography (CT) scan showing the bone grafts are not yet completely reorganized in the left sacroiliac joint and sacrum. There are erosive changes and sclerosis in the left sacroiliac joint.

The remaining cavity was grafted with 60 cc allograft. A diagnosis of hydatid disease was confirmed by histopathological examination. The patient was mobilized with crutches in the postoperative period, allowing toe-touch weight-bearing. Mebendazole treatment was continued for four months. Eleven months after surgery, clinical and radiological examinations yielded no evidence of recurrence (Figure-3A, 3B). The patient had only a little discomfort of the left SIJ at full flexion.

DISCUSSION

Hydatid disease of the spine occurs in 1% of all cases of human echinococcosis, and is most commonly located in the dorsal spine (50%) [1]. The sacral and cervical spinal segments are the least commonly involved areas[2].

Due to the non-specific symptoms, such as lower back and radicular pain, misdiagnosis is common. In this case, the sacroiliac joint had been involved with the lower back pain, and so the patient had been followed by a rheumatology clinic for sacroiliitis. A positive history for previous echinococcosis is helpful for diagnosis, as seen in our case.

Surgical treatment of spinal echinococcosis involves selecting an approach that allows adequate removal of the cyst and cyst contents without spillage, management of the remaining cavity, and avoidance of hypersensitivity reactions to the hydatid antigen. Removal of the main cyst mass may not be effective, because a small daughter cyst can be left behind. Local application of disinfectants is useful to devitalize the cystic contents and prevent recurrence[4]. Hypertonic saline is the most frequently reported scolicidal agent used in spinal disease. We used hypertonic saline, and additionally polyvinyl iodine. The cavity was filled with polyvinyl iodine for ten minutes. We protected the surrounding tissues with hypertonic saline-impregnated sponges to prevent transmission.

Sapkas et al. suggested that osteosynthesis can be improved by the use of acrylic cement, considering that osseous grafts can be invaded by hydatidosis extension or recurrence[6]. Conversely, Pintilie et al. recommended bone grafting for management of the remaining cavity[5]. In this case, a large cavity occurred in the sacroiliac joint, sacral area and iliac wing, so we used a 60 cc allograft. There was no recurrence seen by computerized tomography eleven months postoperatively.

In conclusion, many cases of rare localizations of echinococcosis remain undiagnosed or misdiagnosed. Hydatid cysts should be kept in mind in the differential diagnosis of lower back pain, especially when treating patients who have a positive history or live in endemic areas.

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