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CASE REPORT

# Sacroiliac Joint Hydatidosis Mimicking Ankylosing Spondylitis: A Case Report

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Hydatid cyst disease caused by *Echinococcus granulosus* is common in Mediterranean sheep-raising countries. Spinal involvement in hydatid cyst cases is very rare. The disease mainly affects the lungs and liver and, to a lesser extent, the brain and bones. Depending on location, the disease may mimic soft tissue and bone tumors. Osteolytic and inflammatory changes may imitate osteomyelitis and malignancies or rheumatologic diseases of the affected area. In this article, we report an unusual case of hydatid disease of the sacroiliac joint who was misdiagnosed with ankylosing spondilitis and under follow-up for two years. We also discuss the differential diagnosis of mass lesions of the sacroiliac joint. Hydatid cysts should be considered in the differential diagnosis in patients living in endemic countries.

Keywords: Echinococcosis; sacroiliac joint; spondylitis, ankylosing.

Echinococcosis is a parasitic disease caused in humans by *Echinococcus granulosus* and *echinococcus multilocularis*.[1] The disease mainly affects lungs and liver, however, bone, muscle, peritoneum, heart, kidney, and brain involvement may be also observed.[2] The incidence of musculoskeletal involvement is 0.5 to 2%.[3]

Skeletal involvement usually occurs by the dissemination of primary disease from the liver and lungs. Cases of vertebra, femur, tibia, and pelvic involvement have been previously reported.[4] Skeletal involvement often presents with sciatica, hip and groin pain. Differential diagnosis is mostly confirmed with imaging modalities. We report a rare case of hydatid disease with sacroiliac joint involvement mimicking ankylosing spondylitis (AS).

## CASE REPORT

A 20-year-old woman presented with complaints of left hip pain, which was persistant for two years. The pain was continuous and increasing at night. She did not suffer from morning stiffness or low back pain. Her past medical history revealed that she was diagnosed with AS two years ago. She was initially treated with sulfasalazine 2000 mg daily and naproxen sodium 750 mg daily however, symptomatic relief could not be achieved. She had a history of cystectomy five years ago, for lung and liver hydatid disease. She has been on albendazole treatment since cystectomy. On physical examination, the left hip range of motion was restricted by pain. The bilateral sacroiliac compression test was positive, and the neurologic examination of lower extremities were normal.

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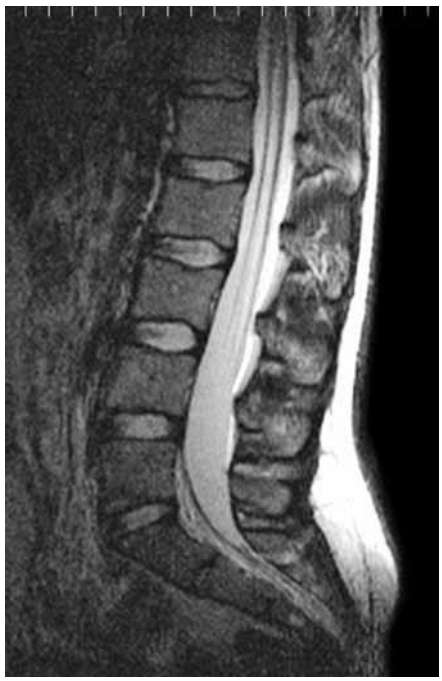

**Figure 1.** Lumbar magnetic resonance imaging showing normal findings.

Laboratory investigations including complete blood count, liver enzymes, and creatinine levels were within normal limits. Erythrocyte sedimentation rate (74 mm/h) and C-reactive protein levels (35.4 mg/L) were high.

There were no abnormal findings of the lumbar spine on magnetic resonance imaging (MRI) (Figure 1). Contrast enhanced sacroiliac MRI revealed lobulated, multivesicular cystic lesions, which originated from the second sacral vertebra, extending to the sacral ala and iliac wing at the third sacral level, which also filled the sacroiliac joint. Vesicular lesions in the sacroiliac joint and piriform muscles showed increased contrast uptake (Figure 2). Given the patient's medical history, bone hydatid disease and other cystic bone lesions were included in differential diagnosis.

Abdominal computed tomography (CT) and thoracic CT revealed cystic lesions of hydatid disease in both liver and lungs. The patient was evaluated by a thoracic surgeon and a general surgeon, who recommended albendazole treatment for the lung and liver lesions. The patient then underwent cystectomy for sacroiliac lesions. The cyst cavities were irrigated with hypertonic saline solution and polyvinylpyrrolidone, and then filled with bone allograft. No bacteria were cultured in the drainage fluid. High-dose albendazole (800 mg/day) was initiated for 28 days. The treatment was interrupted for two weeks. This cycle was repeated three times. Naproxen sodium and sulphasalazine were discontinued. Eight months after surgery, repeated sacroiliac CT showed postoperative bone allografts and sclerosis at the upper site of the joint. There were no cysts or findings of recurrence (Figure 3). The patient's pain was completely relieved. Liver and lung lesions also had a constant course during the follow-ups.

## DISCUSSION

Liver and lung involvement is common in hydatid disease, whereas other organ involvement is rare.[3]

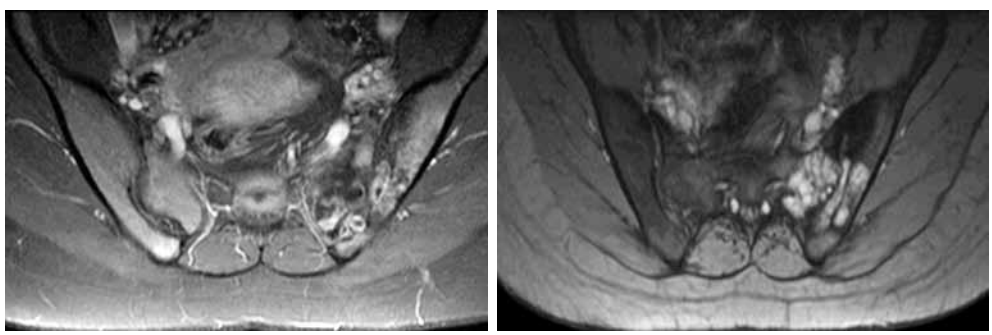

**Figure 2.** $T_1$ and $T_2$-weighted sequence of contrast enhanced sacroiliac magnetic resonance imaging shows mass lesion filling the left sacroiliac joint. Lesion is multicystic and uptakes contrast. However, the rim of the cyst is not contrast-enhanced. The cyst fluid has a high signal intensity.

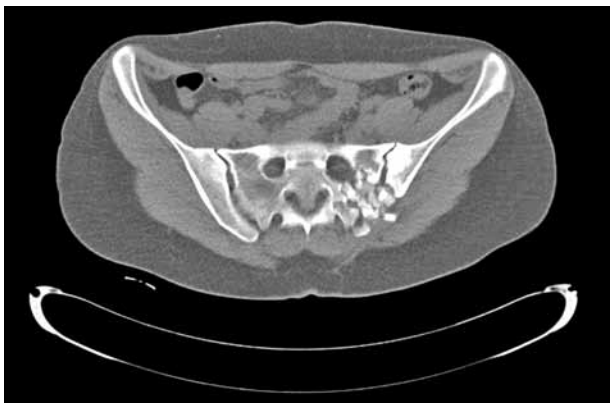

**Figure 3.** Eight-month post-excision sacroiliac computed tomography showing postoperative changes in the left sacroiliac joint due to bone allograft and sclerosis at the upper site of the joint.

The incidence of musculoskeletal involvement is about 0.5 to 2% with almost half of cases involving the spine. Our patient had a history of lung and liver hydatid disease five years ago. She later developed sacral bone, sacroiliac joint and piriformis muscle involvement. Bone involvement may be asymptomatic for a long time. Patients may present with pathologic fractures, severe pain, secondary infection or cord compression findings.[4] The mechanisms of bone fracture include compression or destruction of bone by induced osteoclast proliferation due to ischemia.[5] Nonunion of these fractures is very common. Sacral hydatid cysts become symptomatic with low back pain. Compression of the lumbosacral plexus causes pain in radicular pattern.[6] The disease is diagnosed at advanced stages when attention is focused on low back pain, since imaging of the sacrum is often delayed. Joint involvement occurs by the dissemination of the cyst from the adjacent bone. In our patient, spreading of cysts from the sacral bone to the sacroiliac joint was the most probable way. However, there was no pathologic fracture. Hip pain was her chief complaint.

Inflammatory back pain (IBP) is the main clinical symptom of AS, one of other spondyloarthritides. Inflammatory back pain has certain typical features like, morning stiffness, and aggravation of pain by immobility. It gives a chance to screen and diagnose the disease at an early stage. Radiological evaluation may be negative during the early period of the disease.[7] The diagnosis of AS is often delayed. This may be the consequence of the inability to separate IBP and mechanical back pain.[8] Calin's criteria[9] have recently been set up to define IBP. Four of the five criteria are enough for the definition of IBP: Starting before the age of 40 years, the pain continues more than three months, the pain begins slowly, stiffness in the morning and improving with exercise. More recently, Rudwaleit et al.[7] proposed a new set of criteria for IBP: morning stiffness lasting for at least 30 minutes, improving with exercise worsining with rest, waking from sleep (often in the second half of the night) and changing side. Our patient presented to us with unilaterally hip pain, which was awakening at night and continuous the whole day. She did not have back pain, morning stiffness or alternating buttock pain, which are all suggestive for IBP. The attention given to her medical history lead us to other diagnosis than AS.

Furthermore, MRI usually helps to define features of the cyst except calcifications.[4] Hydatid cyst appears as well formed, thin-walled and spherical lesions on MRI.[10] The fluid of the cyst is isointense with cerebrospinal fluid both on $T_1$ and $T_2$-weighted images and the wall of the cyst appears as a low signal intensity rim. In defining the wall of the cyst, $T_2$-weighted images are superior to $T_1$-weighted images. Fluid has a high signal intensity and, in contrast, the wall of the cyst has a low signal intensity on $T_2$-weighted images.[1] After intravenous contrast agent administration, the rim of the cyst shows no uptake. Calcification of the cysts wall is a very rare entity.[11] Computed tomography images resemble $T_1$-weighted MRI images. Magnetic resonance imaging of our patient revealed characteristics of hydatid cyst. Regular, thin-walled cyst and pericystic contrast uptake was determined. Chronic osteomyelitis, fibrous dysplasia of bone osteosarcoma, benign cystic lesions of the bone, brown tumor (hyperparathyroidism), and neoplastic lesions all should be included in the differential diagnosis.[12,13] Localization of lesions is also important. Although our patient did not present with inflammatory back pain, she had a presumptive diagnosis of AS and was medically treated for two years. This caused a delay in the diagnosis and unnecessary use of medication. Hence, atypical presentation of hip or low back pain and its irresponsiveness to anti-inflammatory medication warrants further investigation.

Yılmaz et al.[14] previously reported a similar case of hydatid disease of the sacrum affecting the sacroiliac joint. They reported a 38-year-old woman presenting with a three months of history of low back pain and sciatica on the right side. Numbness and a gradually developing weakness also accompanied these complaints. Unlike in our case, their patient did not have liver hydatid disease. Another difference was that our patient did not have neurological symptoms.

The diagnosis of the hydatidosis is mainly based on radiological and serological tests. Sensitivity and specificity of the serological tests vary according to the nature of antigen and the selected method. The serological tests used to detect antibodies against echinococcal antigens include, indirect hemagglutination, indirect immunofluorescence antibody test, latex agglutination test, solid face radioimmunoassay, immunoelectrophoresis, counter immunoelectrophoresis and enzyme-linked immunosorbent assay (ELISA). The serological tests used to define echinococcal antigens are, coagglutination test, counter current immunoelectrophoresis and ELISA. Antibodies persist for a long time after surgery. For this reason, antigen tests are more useful for the detection of relapses and new infections.[15]

Echinococcal disease causes very poor immunological response in the host. The sensitivity and the specificity of the serological tests for diagnosis differ in a wide spectrum; hence, the clinician should request at least two serological tests to increase the probability for a correct diagnosis. One of the tests should have a high sensitivity, while the other should have a high specificity.[15] We did not use any serological test for the diagnosis in our case. The diagnosis was almost certain after imaging, and it was confirmed by pathological examination of the surgical specimen.

Both medical and surgical treatment are applied for cases of spinal involvement. Excision should progress until healthy bone comes to surface. Scolicidal agents like hypertonic saline solution and cetrimide are often used to wash out the cavity to prevent recurrence, since contents of the cyst can cause anaphylaxis while performing surgical excision.[4] The nature of bone involvement is often ambiguous. In advanced stages, it becomes diffuse and invasive.[5] Recurrence is very high after surgery. In the first two years, the incidence of recurrence reaches eight to 22%.[16] Large lesions of pelvic bones have a poor prognosis due to the high risk of sepsis.[17] In our patient, lesions in the sacral bone and sacroiliac joint were completely excised and the remaining cavity was washed out with scolicidal agents. The cavity was filled with bone allograft to support the stability of the adjacent structures. No recurrence was detected for eight months following surgery. Albendazole therapy should be initiated prior to other medical treatment choices. Albendazole markedly improves almost 40% of cases.[18] In addition, there is evidence to suggest that benzimidazole after surgery for three months is beneficial.[19] Our patient underwent surgery twice for lung and liver disease. Although albendazole was given to the patient, recurrence was seen in the sacrum within three years after surgery. We repeated albendazole therapy after excision. Unfortunately, curative treatment is not always possible and recurrence is high.

In conclusion, medical history of a patient may give invaluable clues for challenging and unusual diagnosis as in this case. Echinococcal disease should be considered in the differential diagnosis of any cystic mass in endemic areas.

**Declaration of conflicting interests**

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