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Widespread Hydatidosis of Sacroiliac Bones and Retroperitoneum A Case Report

Abstract: Hydatid disease is one of the oldest diseases known to humankind. In 85-95% of the cases, the liver and/or the lungs are involved and only 5-15% of other organs.

The case presented here is a 38-year-old man with hydatid disease of lower lumbar vertebrae, sacrum, lumbosacral spinal canal, and left iliac bone; with extension to retroperitoneum and piriformis, sacrospinalis, iliacus, iliopsoas, and quadratus lumborum muscles; without involvement of liver or lung, as diagnosed by MRI and CT scan and confirmed by surgery.

Keywords: hydatid cyst, echinococcosis, skeletal hydatidosis

Introduction

Hydatid disease is an infestation by the larvae of *Echinococcus granulosus*. Although the parasite may affect any organ, lung and liver are the most frequently involved sites.^{1,2} In the spinal column, hydatid disease usually begins in the vertebral body with predilection for the thoracic and lumbar spine.^{2, 3} Sacral involvement is rare in hydatid disease.² Intra- and retroperitoneal hydatid disease is usually the result of spontaneous, traumatic or surgical rupture of a hepatic cyst.⁴

Although there have been reports of sacral, spinal canal, retroperitoneal, and musculoskeletal hydatid disease, with extension to two or more adjacent anatomic locations^{1, 3}, to the best of our knowledge, such an extensive spread with involvement of all these locations together has not been reported before.

Case Presentation

A 38-year-old Afghan shepherd was referred to our hospital with a history of recurrent pain in his low back and lower extremities since 8 months ago. Bladder incontinence and bowels disturbance, with progressive radiculopathy of the lower limbs were added to his problems since 6 months earlier. He had a history of laminectomy at L4-L5 level in another country for the same problem, before his new admission. On physical examination, there was weakness in lower limbs mostly on the left side. Left flank, along the paraspinal muscles was also tender. A midline surgical scar due to a previous laminectomy was evident on the back.

Routine laboratory investigations and chest radiography were normal. Provisional clinical diagnosis was the recurrent lumbar disc herniation.

MRI of lumbosacral spine was performed to evaluate recurrent disc herniation which revealed a large, complex multiloculated, multi-septated, predominantly cystic lesion, hypointense on T1-weighted and hyperintense on T2-weighted images with a hypointense capsule involving the L5, left sacral ala, sacral neural foramina and the adjacent iliac bone. (Figures 1, A and B)

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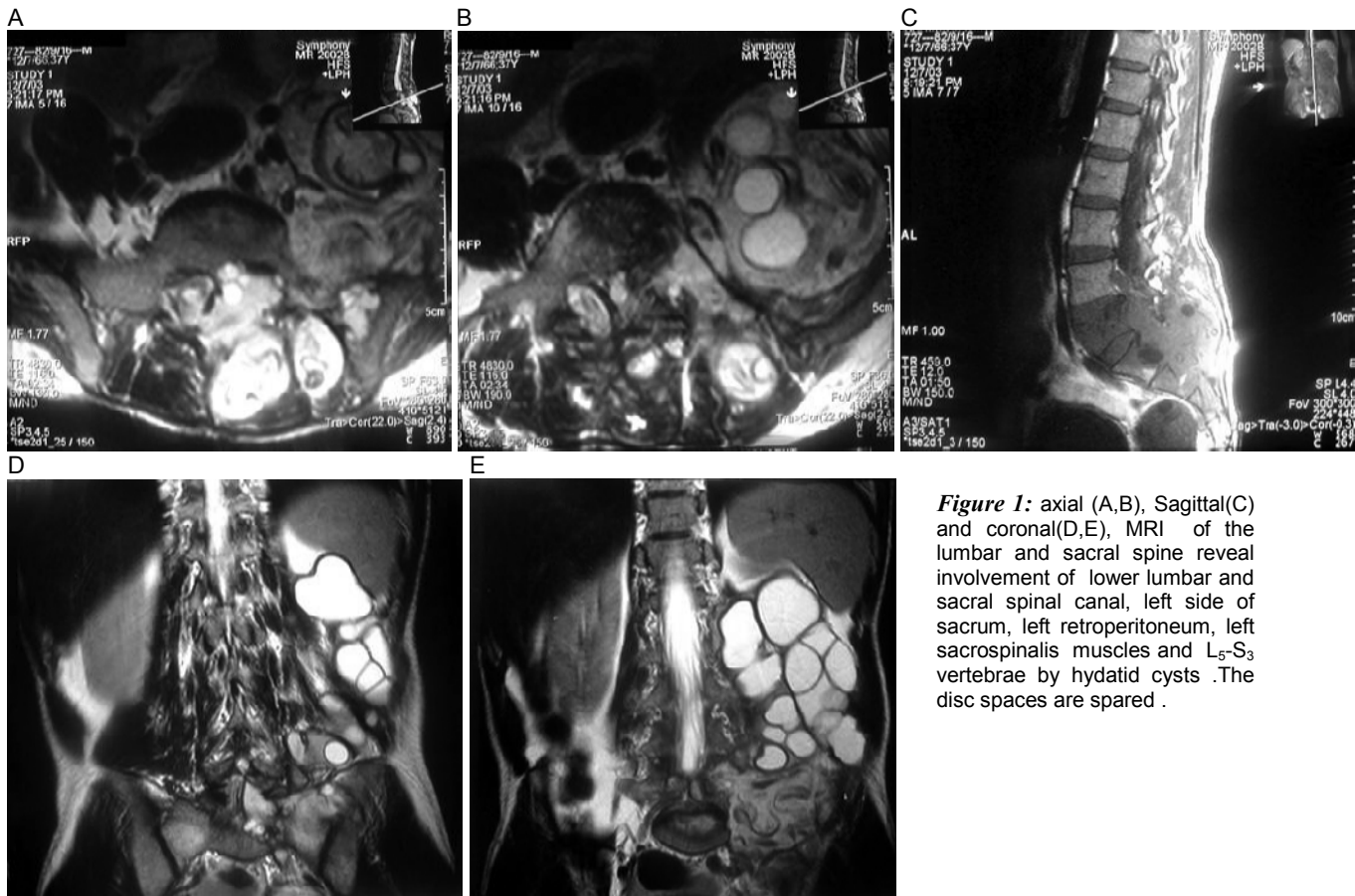


Figure 1: axial (A,B), Sagittal(C) and coronal(D,E), MRI of the lumbar and sacral spine reveal involvement of lower lumbar and sacral spinal canal, left side of sacrum, left retroperitoneum, left sacrospinalis muscles and L₅-S₃ vertebrae by hydatid cysts. The disc spaces are spared.

The lesion extended into spinal canal from L4 to S3 levels, compressing the thecal sac tightly. The left sacrospinalis, quadratus lumborum, iliopsoas, and piriformis muscles were involved by the cysts. The disc spaces were intact and no definite evidence of recurrent herniation was detected. (Figures 1, A-C)

Superiorly, the lesion extended to the left perirenal fat, displacing the left kidney superoanteriorly. Coronal T2W image, revealed this extension very well. (Figures 1, D and E)

Abdominopelvic ultrasonography showed a large cystic mass with multiple septae on the left side of retroperitoneal space. This cystic lesion extended all the way down to the pelvic floor. Liver, spleen and kidneys were normal.

Spiral CT scan with oral and IV contrast revealed a large, multiloculated cystic lesion with enhancing septae involving the same areas shown on MRI (Figures 2, A-C).

The imaging findings were confirmed by surgery and the pathology report indicated a hydatid cyst containing multiple daughter cysts.

The patient left the hospital in good conditions to take albendazole tablets for 10 weeks.

Discussion

Musculoskeletal hydatidosis occurs most commonly as an isolated finding without concomitant hepatic or pulmonary involvement in patients from endemic regions.⁵ In bones, the disease occurs mostly in highly vascularized areas of the skeleton such as the vertebrae, long bone epiphysis, ilium, skull and ribs.^{2,4,6} The lower lumbar vertebrae and left ilium were involved in our case.

Spinal hydatid cyst accounts for 1% of all cases of hydatid disease^{7,8}, occurring in approximately one half of the patients with osseous involvement because of portovertebral shunting.⁴ Extension into spinal canal results in cord compression.⁴ Intervertebral disc, ligaments and capsules are said to be resistant to hydatidosis and in most cases the disc is preserved.² We found the same findings in our case.

Primary retroperitoneal hydatidosis is extremely rare⁹, and only occasional case reports have appeared in literature. Retroperitoneal involvement was always thought to be secondary to rupture or spillage of liver hydatid cyst. In our case, the sacral and lower lumbar vertebrae were involved while the intervertebral discs were intact. Extensive retroperitoneal and muscular involvement was an unusual finding seen in our case.



Figure 2: Spiral enhanced CT scan of lower abdomen and pelvis with oral and IV contrast reveals forward displacement of left kidney by retroperitoneal hydatid cyst. The perirenal fat is involved. There is also involvement of L₃, left side of sacrum, sacral spinal canal, and iliopsoas, quadratus lumborum, sacrospinalis, piriformis muscles by the hydatid disease on the left side.

Regarding the patient's history of laminectomy, the unusual spread of the hydatid might have been due to the previous surgical manipulation. Unfortunately, the operation was done in another country and we had no access to its report.

The diagnosis of hydatid cyst relies on serologic tests and imaging features.⁴ Nowadays, MRI is usually the first imaging modality in patients with suspected myeloradiculopathy.¹⁰ It was the reason for performing MRI as a first imaging investigation in the present case. The presence of multiple cysts (daughter cysts) within a cyst which are of varying signal intensities is the characteristic imaging feature of this disease, as was demonstrated in our case. (Figure 1, A and B)

On T1-weighted images, the daughter cysts are more hypointense than the parent cyst and on T2-weighted images, the daughter cysts are of slightly higher signal intensity than the parent cyst.^{11, 12}

Sonography and CT scan are useful for delineating the location of the cyst but findings are nonspecific.¹³ CT scan will demonstrate well-defined cysts which may be single or multiple, uni- or multilocular, and thin or thick walled. More specific signs include visualization of calcification in the cyst wall, presence of daughter cysts, and membrane detachment.¹

The present case illustrates the imaging features of extensive lumbosacral, spinal canal, retroperitoneal and musculoskeletal hydatid disease. The hydatid cyst should be considered in the differential diagnosis of every cystic mass in every location, especially in the areas where the disease is endemic.

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