

Combined Anterior and Posterior Approach for Sacral/Retroperitoneal Hydatid Cyst Disease: Case Report

Sakral/Retroperitoneal Kist Hidatik Hastalığına Kombine Anterior ve Posterior Yaklaşım: Olgu Sunumu

ABSTRACT

Achieving complete resolution of spinal hydatid cyst disease is quite challenging when bone is involved. Many authors reported the poor outcome of posterior decompression and laminectomy for intraosseous spinal hydatid disease. In an attempt to avoid a similar poor outcome, hydatid cysts were reached via both anterior and posterior surgical approaches in our patient. A 73-year-old man presented with complaints of low back and right leg pain. Symptoms or signs of systemic hydatid cyst disease were absent. MRI demonstrated a cystic lesion in the presacral/retroperitoneal region, involving the body of the sacrum and sacral canal. Computed tomography images showed enlargement of the sacral foraminae. The multiseptated cysts and their contents were isodense with cerebrospinal fluid. The cysts were removed via an anterior extraperitoneal approach, using a paramedian vertical incision, and then were also approached posteriorly via bilateral S1 hemilaminectomy. No neurological deficits occurred following surgery. The patient's symptoms completely disappeared after this combination of aggressive surgery and antihelminthic therapy. The application of both anterior and posterior approaches to intraosseous sacral hydatid cysts may be preferred when faced with hydatid disease in this location.

KEYWORDS: Anterior extraperitoneal approach, Retroperitoneal region, Sacral hydatid cyst

ÖZ

Spinal kist hidatik hastalığı kemiği tuttuğunda kesin çözüme ulaşmak oldukça zordur. Birçok yazar, intraosseöz spinal kist hidatik hastalığı için posterior dekompresyon ve laminektominin kötü sonuç verdiğini bildirmiştir. Benzer kötü sonuçtan kaçınmak için, biz burada bir hastada hem anterior hem de posterior cerrahi yaklaşımı rapor ettik. 73 yaşında erkek hasta bel ve sağ bacak ağrısı şikayeti ile başvurdu. Sistemik kist hidatik hastalığına ait semptom ve bulgular yoktu. MRI sakrumun korpusu ve sakral kanalı tutan presakral retroperitoneal bölgedeki bir kistik lezyonu gösterdi. Bilgisayarlı tomografi görüntüleri sakral foramenlerin genişlediğini gösterdi. Multiseptalı kistler ve onların içeriği BOS ile izodens idi. Kistler paramedian vertikal insizyon kullanılarak anterior extraperitoneal yaklaşımla ve sonra bilateral S1 hemilaminektomi yoluyla posterior yaklaşımla çıkarıldı. Cerrahi sonrası nörolojik defisit oluşmadı. Hastanın semptomları bu agresif cerrahi ve antihelmintik tedavi kombinasyonu sonrası tamamen düzeldi. İntraosseöz sakral kist hidatikte hem anterior hem de posterior yaklaşım kullanmak bu bölgede hidatik kist ile karşılaşınca tercih edilebilir.

ANAHTAR SÖZCÜKLER: Anterior extraperitoneal yaklaşım, Retroperitoneal bölge, Sakral kist hidatik

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INTRODUCTION

Echinococcosis involves the spine in 0.5–1% of all cases, and these represent about 50% of all skeletal hydatid cysts (6,10,26). These can occur by direct extension from pulmonary or pelvic infestation, or less commonly begin in the vertebral body (8,14,22). Spinal hydatid cysts occur most commonly in the thoracic region and less frequently in the lumbar region. Infestation begins in the center of the vertebral body and extends outwards into the epidural space. Hydatid masses are therefore epidural in 90% of cases (6). Sacral localization of hydatid cysts and their extension into the pelvic cavity are extremely unusual. In this location, they are difficult to eradicate with surgery and antihelminthic agents (6).

The most common radiological characteristic of an osseous hydatid lesion is a combination of multilocular cysts and reactive sclerosis, as in a honeycomb, involving a large expanse of bone. Osteolysis is usually seen, sometimes associated with expansion of the bone and thinning of the cortex (5,18,20,28).

We present an extremely unusual case of primary spinal sacral/retroperitoneal hydatidosis. The patient underwent an anterior extraperitoneal approach for the presacral retroperitoneal component and S1 bilateral hemilaminectomy for removal of cysts in the spinal canal.

CASE HISTORY

A 73-year-old man presented with a 2-month history of progressively worsening lower back pain radiating to his right foot. On examination, sensory deficits were present on the right lower leg and foot. He had no bladder or bowel symptoms. The general physical examination was normal.

Computed tomography (CT) images demonstrated a large, multiloculated, lytic lesion that expanded anteriorly through the S1-S3 vertebral bodies, causing extensive destruction of the sacrum, and extending into the sacral canal at the S1 and S2 levels (Figure 1). The sacroiliac joint was normal bilaterally.

Magnetic resonance imaging demonstrated a multilobular cystic lesion with thin and regular cyst walls. The cyst contents were hypointense on T1-weighted images, hyperintense on T2-weighted images, with an intensity similar to that of cerebrospinal fluid. The mass also encroached on the

spinal canal, compressing the dural sac at the sacrum but preserving the intervertebral disc. The large cyst extended into the pelvic retroperitoneal cavity (Figure 2). An intrapelvic multilobulated cyst measuring 80x40 mm was visualized with ultrasonography. Further imaging by ultrasound and/or CT did not reveal the involvement of any other organs, including the liver, lung, spleen, brain, and other parts of spinal canal.

Total blood cell count, erythrocyte sedimentation rate, complete biochemical serum and urine parameters, coagulation tests, and serum protein electrophoresis were normal.

Twice daily doses of albendazole 400 mg were used for one week and the patient underwent surgical intervention afterwards. The patient was placed in the supine position under general

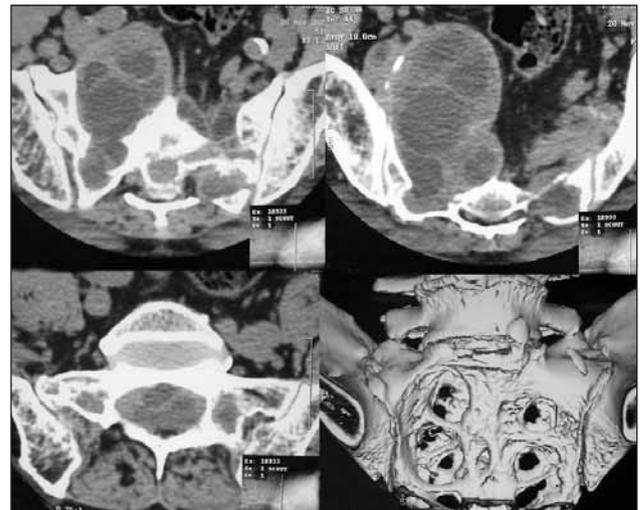


Figure 1: Computed tomography images demonstrated a large, multiloculated, lytic lesion that expanded anteriorly causing extensive destruction of the sacrum, and extended into the sacral canal at the S1 and S2 level. The sacroiliac joint was normal bilaterally. Massive destruction of the anterior sacral segments was seen on the reconstructed 3D CT image.



Figure 2: Magnetic resonance T1- and T2-weighted sequences show a large, multiloculated, cystic lesion involving the sacrum and S1 vertebra with extradural components.

endotracheal anaesthesia with an adjustable block underneath the lumbo-sacral junction. A right-sided paramedian skin incision was made from the umbilicus to the symphysis pubis. The superficial sheet of the rectus abdominis muscle was vertically cut and the muscle was retracted to the left. The abdominal muscles were separated from the retroperitoneal fat to the medial side of the psoas muscle (Figure 3). The peritoneum was dissected from the right abdominal wall. The right iliac artery and vein were retracted to the right (Figure 3). The ureter was identified and protected during the dissection. The cyst was then directly exposed in the right presacral/retroperitoneal region and the liquid content of the cysts was carefully aspirated followed by injection of 3% hypertonic saline into the cysts. After waiting for five minutes, the cysts were opened and the echinococcus vesicles removed (Figure 4). Copious irrigation of the surgical field was then performed with 3% hypertonic saline solution. Cysts were located in the retroperitoneal, intrasacral regions and the sacral canal and were therefore aspirated multiple times, with some areas accessed for irrigation and removal only with a 30° scope (1). This anterior extraperitoneal approach afforded adequate exposure deep in the presacral space. A surgical drain was left in the large emptied space just in case we desired to do additional irrigation with 3% hypertonic saline, if residual cysts were seen on post-operative CT images.

The patient was placed in the prone position with an adjustable block underneath the thorax to begin the second (posterior) approach. A median vertical skin incision was made from the L5 to the S3 vertebra. A posterior bilateral S1 hemilaminectomy

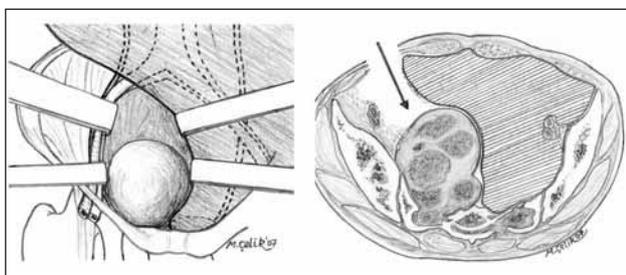


Figure 3: (Left) Schematic drawing of the anterior retroperitoneal approach of the pelvis. The avascular path is between the peritoneum and the psoas muscle. (Right) Cross-sectional schematic representation of the retraction of the retroperitoneal vascular and urinary elements for accessing the presacral cyst.



Figure 4: Some of the cysts retrieved during surgery, showing great variation in size.

was then performed. Copious irrigation of the surgical field using 3% hypertonic saline solution was performed before the cysts were removed. No intraoperative complications or need for transfusion occurred. An additional drain was placed posteriorly for the same reason as discussed above. No neurological, bladder, or bowel symptoms occurred in the immediate postoperative period. The patient received broad-spectrum antibiotics (1 gr cefazolin IV pre- and postoperatively) in addition to daily albendazole (800 mg daily in two divided doses for 3 months). Histopathological examination of the cyst wall confirmed the diagnosis of a hydatid cyst. Postoperative pelvic CT scans confirmed gross total excision of the cyst (Figure 5). The patient was ambulatory with neither residual lower back nor leg pain when discharged home on the postoperative seventh day. The patient continued to show an excellent recovery, without clinical or MRI evidence of hydatid disease recurrence on subsequent clinic visits.

DISCUSSION

Spinal hydatid disease manifests itself through symptoms and signs related to compression of the cysts on other structures; no specific pathognomonic symptoms or signs exist (4,22). As in our case,

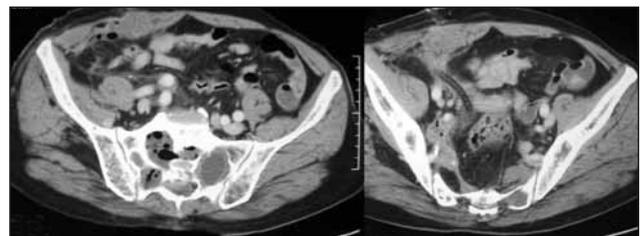


Figure 5: Postoperative pelvic CT scans confirmed the gross total excision of the cysts.

patients may present with radiculopathy, myelopathy and/or local pain owing to bony destructive lesions and pathological fractures (19). Spinal involvement is believed to occur through vertebral-portal venous anastomosis, first involving the center of the vertebra. The infection progresses as a multivesicular infiltration of cancellous bone that may involve the vertebral bodies, pedicles, and laminae. Intervertebral discs are usually spared because cyst growth is confined to the periosteum (7,12,13,21,29,30).

Successful treatment of spinal hydatid disease necessitates intense systemic antihelminthic therapy combined with aggressive surgery (20). Complete surgical removal of the cysts is the most common initial treatment for spinal hydatid disease. Simple decompression with laminectomy is performed most frequently, because of its low morbidity and mortality, but it is not free of accompanying risks. However, access to the vertebral body through this posterior approach is limited, resulting in poor outcomes, especially in recurrent disease. Despite treatment, the disease frequently relapses with progressive destruction of the vertebral column and neurological deterioration (15,21,22,23).

Laminectomy was performed routinely for spinal hydatid disease until recently, but it did not significantly affect the outcome (15). Many case reports and series in the literature document the poor outcome of posterior decompression and laminectomy for intraosseous spinal hydatid disease (17). We therefore preferred combined anterior and posterior surgical approaches, which we believed would offer a higher chance of successful eradication of the disease in the first intervention.

Posterior decompression alone is indicated only in cases with isolated involvement of the neural arch and in those with extraosseous extradural disease (11). Turtas et al reported a 50% recurrence rate after posterior decompression alone (30). Pure intradural or pure epidural lesions at all spinal levels are approached posteriorly. Surgical excision without intraoperative rupture results in cure in these cases (13,22).

Bone involvement is a major challenge for the surgeon, with spillage of scolices being unavoidable (3,13,21,22). Laminectomy alone may result in rupture of cysts and scolex spillage. Both physician and patient must keep in mind that hydatid disease

is highly prone to recurrence, and medical and surgical management plans must therefore be made accordingly (30). By accessing the sacral area through an anterior extraperitoneal approach, the intracavitary pressure of the cysts was decreased when they were opened. Thus the chance for scolices to disperse when the cysts were approached from the posterior was much decreased (the cysts had already been 'popped' from their anterior aspects). Control of the dispersion of the scolices is much easier from the anterior than from the posterior aspect, with compresses soaked with 3% hypertonic saline in both locations. The drain left in the space previously occupied by the cysts was useful for routine surgical drainage, but was not needed for recurrent hypertonic saline irrigation of the cyst area because the post-operative CT was free of residual cysts (27).

Anterior procedures as well as spinal stabilization have been reported among treatment options for spinal hydatid disease, but to date, little evidence exists to show that clinical outcomes have improved with stabilization devices or by using combined anterior and posterior surgery (16,17,23).

Regarding the indications for stabilization and fusion, fusion or stabilization is not recommended if only partial or complete laminectomy without excessive facet joint excision is performed (17). Preoperative instability was absent in our case and thus no fusion or stabilization was performed. The decision to add instrumented fusion depends on whether instability is already present or is created by the necessary surgical decompression (17). If stabilization with pedicle screws is performed, the surgeon should be aware that the disease might be iatrogenically seeded into the vertebral bodies if viable scolices are present at the time when the pedicles are probed before inserting the screws (17).

Surgery remains the definitive treatment for hydatid disease, although complete surgical removal is difficult in the spine (10). Total removal of the cysts without rupture appears to be effective in the prevention of late recurrences (10). Cysts were found in the sacral canal as well as retroperitoneal and intrasacral spaces in our case, and total excision of the cysts without operative rupture was therefore impossible. Radical excision is almost impossible in hydatid disease of the spine because of the absence of distinct anatomic planes, containment within small spaces having bony boundaries, and the existence of neural structures and the local

recurrence rate is therefore high (20). Rupture occurs most frequently in spinal hydatidosis, with an incidence as high as 44%, particularly if the cyst is in an extradural location (2,25).

Although their effectiveness is not proven, many surgeons prefer to use scoleocidal agents such as 3% hypertonic saline after removal of the cyst, hoping to prevent recurrence (24). We chose this option in our patient as well, and experienced no complications. Albendazole is the preferred antihelminthic agent in the treatment of hydatid disease, but the duration of treatment is controversial (10).

Despite optimal and medical therapy, recurrence and thus reoperations are generally needed to achieve clinical control of the disease and minimize complications (9). Strict follow-up is critical in the management of these patients. MRI scans should be done regularly during the postoperative period in order to ensure that any recurrence is detected early (19).

CONCLUSION

The outcome of conventional therapy consisting of antihelminthics with posterior decompression and laminectomy alone is poor for intraosseous spinal hydatid disease. Combined anterior and posterior surgery was successful in our patient and may be strongly considered to maximize the chance of success for this difficult to treat infection.

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