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A case of cauda equina syndrome: Sacral hydatid cyst

Mohammad Asef Adelyar*; Ramin Saadaat; Samea Hesham; Ahmadullah Danish; FarukhSiar Farooqi; Tahmineh Aini; Besmellah Najrabi; Wahidullah Momand; Hamid Wardag; Hedayatullah Hayat

***Corresponding Author: Mohammad Asef Adelyar**
Department of Abdominal Surgery, Aliabad Teaching Hospital, University of Kabul Medical Science, Afghanistan.
Tel: 0093783210709; Email: dr.asef.adelyar@gmail.com

Abstract

Primary hydatid cyst of the bone is rare, constituting only 2-5% of all hydatidoses. When spinal hydatidoses occur, they are most commonly found on the thoracic vertebrae.

Here we present a case of a 30-year-old female, who had been suffering from lower back pain for 3 years and caudaequinasyndrome for 2 months. After initial evaluation, magnetic resonance imaging (MRI) revealed a lesion in the left aspect of the body of lumber 5 (L5) and sacral 1 (S1) vertebrae. A laparotomy through a lower midline incision was performed and a retroperitoneal sacral hydatid cyst was revealed. The mass was then excised incompletely requiring a posterior median vertical skin incision from L5 to S1, with laminectomy of L5 and S1 for complete excision of the hydatid cyst and neural decompression.

Hydatid cysts should be considered in the differential of sacral cystic masses as they could cause devastating sequelae (anaphylactic shock, spillage of contents in the peritoneal cavity and other organs, with destruction of the bone and neurological defects).

Keywords

Caudaequina syndrome; foot drop; hydatid cyst; low back pain; sacrum; urinary retention.

Abbreviations

CT scan: Computed Tomography Scan; ED: Emergency Department; Ig G: Immunoglobulin G; IHA Test: Indirect Haemagglutination Test; L5: Lumber 5; MRI: Magnetic resonance imaging; S1:Sacral1.

Introduction

Hydatid (GREEK for 'watery cyst') disease was first recognized by Hippocrates over 2000 years ago [1]. According to Islekel et al bone disease was first described by Bidloo in 1708, with the first description of spinal hydatid in 1807 by Churrier and the first surgical intervention was reported by Reydellet in 1819 [2].

A hydatid cyst with bony involvement is a rare occurrence (0.5-4%). Spinal involvement is seen in 30-50% and pelvic involvement in 20% of these cases [3].

Hydatid disease is often a parasitic infection caused by larvae of the tapeworm Echinococcus. Echinococcusgranulosus is the species that most commonly infects humans. The infestation is prevalent in sheep and cattle farming communities of the Mediterranean, Asia, North and East Africa, South America, Australia, and the Middle East. The parasite requires two hosts in its life cycle: a definitive host (usually a dog) and an intermediate host (e.g. humans). Humans are infected by direct contact with an infected definitive host or by ingestion of contaminated food. Echinococcus embryos then migrate through intestinal mucosa, enter intestinal venules and lymphatics, and reach the liver in 60% to 70% of cases. If the embryos bypass the liver, they enter the systemic circulation and are carried by the blood stream to any organ or tissue in the body [4]. In the spine, the spongy part of the vertebra become a preferred site of infestation [5].

With regard to the diagnosis, clinical history, laboratory evaluation such as serological detection of specific iimunoglobulin G (IgG) antibodies, radiologic investigations, and histopathologic analysis of the biopsy specimen are of paramount importance [6].

The surgical removal is often technically difficult. Simple cyst puncture is not a solution as finding which compartments in the vertebrae the cyst is accessible. Also, it is vital to excise the cyst together with its wall without rupturing it in treatment. Rupture of the cyst may result in dissemination and chronic recurrence [5]. The infestation can erode the spinal column, eventually leading to its destruction and neurological deterioration. The prognosis can be poor, comparable to that of malignancies [2].

Here, we present a very rare case of sacral hydatid cyst and the resulting surgical excision.

Case Report

A 30-year-old female patient presented to the emergency department of Ali Abad Teaching hospital with manifestations of caudaequina syndrome with symptoms of left foot drop, constipation, urinary retention, loss of perineal sensation and pelvic pain for two months. With further investigation, she also complained of chronic lower back pain for three years. Physical examination of the abdomen showed a fixed mass in the left lower quadrant and rectal examination showed decreased rectal sphincter tone. Neurological examination revealed decreased hip extensor and abductor strength (3/5) along with the muscles of the left ankle (1/5). There was also decreased tone of the posterior muscles of the left leg but with normal sensation.

Ultrasonography of the abdomen and pelvis revealed a left sided pelvic complex mass with possible adnexal origin measuring 9.8 x 8.8 cm. MRI reported an ill-defined lytic lesion in the body of L5 and S1 vertebrae towards left side with associated large soft tissue component (72 x 85 x 145 mm), and a plaque like soft tissue in the spinal canal posterior to the body of L5 and S1 vertebrae (Figure 1). The possible radiological diagnosis suggested was a complex ovarian mass, teratoma, chordoma, or cystic neuroma.

Plans were then made for an exploratory laparotomy and excision of the mass. A laparotomy through a midline incision from the umbilicus to symphysis pubis was created and the cystic mass in the retroperitoneum was observed. The cyst originated from the L5/S1 area of the spine and occupied and eroded the left sacral wing with involvement of the left psoas muscle. After mobilizing and incising the cyst, the cyst with its multiple daughter cysts were excised as much as possible (Figure 5) and then copiously irrigated. Not any other abnormal pathology was noted and a surgical drain was placed and the abdomen closed. The patient was then placed prone and a median vertical incision was performed from L5 to S1. A posterior bilateral L5 and S1 laminectomy was also done (Figure 2, 3 and 4). Before removing the remaining hydatid cyst, copious irrigation of the area was performed with hypertonic saline solution. The cyst was then removed in its entirety. Another drain was placed posteriorly. No complications occurred during the procedure. The mass was sent for histopathological examination with confirmation of hydatid cyst. On post-operative day seven, the patient was discharged from hospital in stable condition with albendazole therapy for six months. At one month follow up, the patient showed recovery of perineal sensation and anal tone, and resolution of urinary retention. At two months she showed increased ankle and foot strength, tone and mobility.

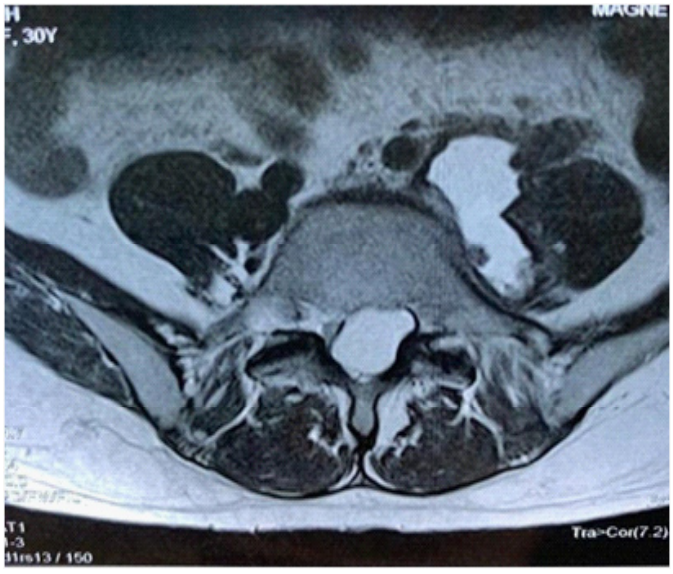

Figure 1: MRI of the pelvis revealed a multi lobulated cystic structure in the left side of the pelvis involving the left sacral wing and extending to the spinal canal.

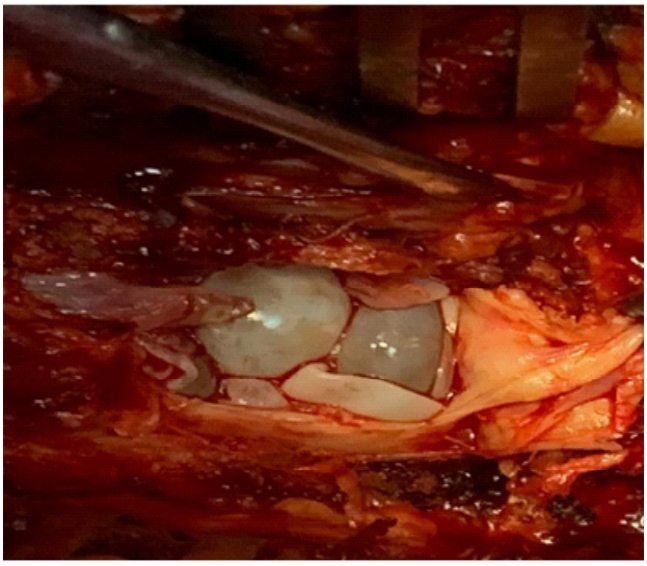

Figure 2: An intra-operative image of hydatid cyst with multiple daughter cysts (posterior approach).

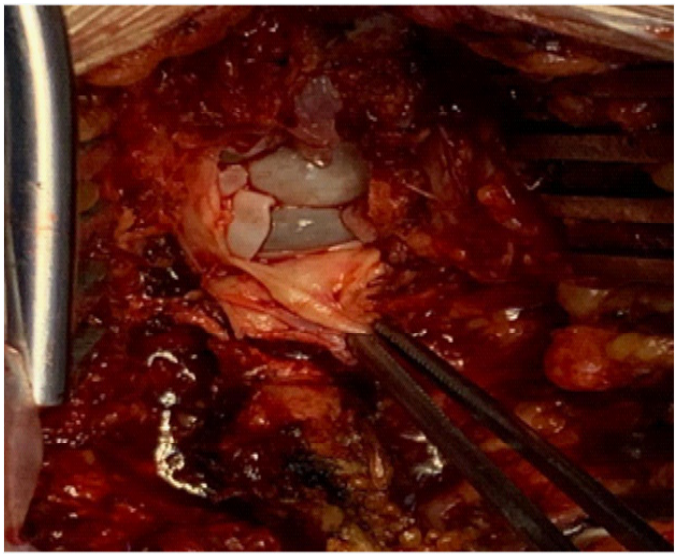

Figure 3: An intra-operative image of hydatid cyst (posterior approach).

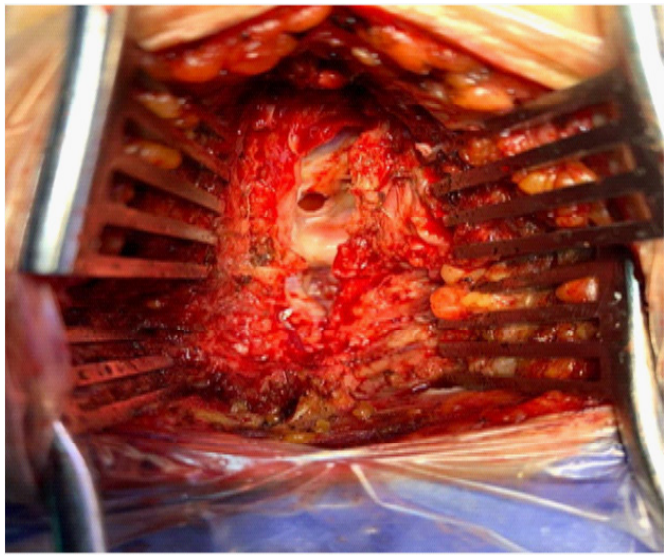

Figure 4: An intra-operative image of excision of hydatid cyst by laminectomy

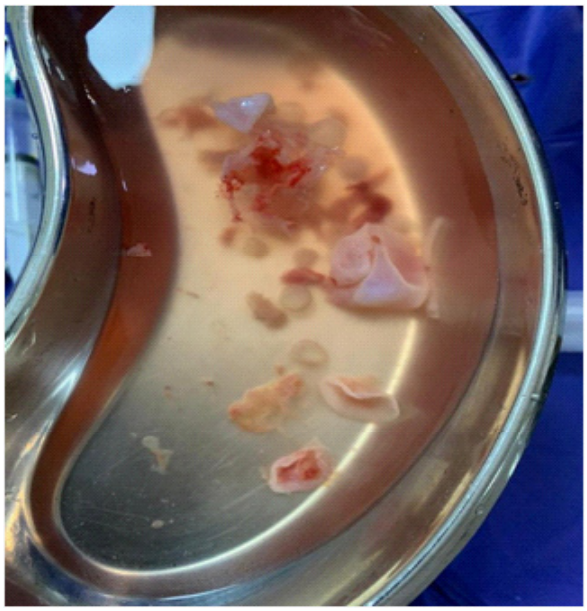

Figure 5: Multiple daughter cysts after excision.

Discussion

Bone involvement in hydatid disease is an incredibly rare occurrence [3]. In a series of 25 cases of vertebral hydatidosis the cyst was located in the cervical vertebrae in three, the thoracic vertebrae in 11, and the lumbar in five, and sacrum in six cases. In another series of cases of vertebral hydatidosis, the sacrum was involved in only one case. Approximately 90% of spinal hydatidosis cases are located extradurally. Intradural location is very rare and can appear like an arachnoid cyst. As there is no host reaction the cyst can grow to enormous size and remain asymptomatic. Recurrences and dissemination are common with the overall recurrence rate of 30-40%. When recurrence occurs, repeat excision should be attempted if feasible [7].

Spinal hydatid disease manifests itself through symptoms related to compression of the cysts on

other structures; no specific pathognomonic symptoms and signs exist [8]. The initial symptom is often back pain when the cyst invades the spinal canal after erosion of the bony cortex. Patients may present with radiculopathy, myelopathy and/or local pain owing to bony destructive lesions and pathological features. Spinal involvement is believed to occur through vertebral-portal venous anastomosis, first involving the center of the vertebra. The infection progresses as 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 [8].

It is of utmost importance that a correct preoperative diagnosis is made since all precautions must be taken to prevent dissemination and seeding of the surgical field. Deaths have been reported due to anaphylactic shock resulting from spillage during excision or biopsy after a mistaken diagnosis of a retroperitoneal tumour. In endemic regions, because of the diversity of its presentation the possibility of hydatid disease should always be considered for any growing mass in the body. Diagnostic techniques such as radiography, ultrasonography, computed tomography, MRI, and immunological tests are of value.

Conclusion

Hydatid cyst of the bone, even in the endemic areas, is a rare occurrence. The sacral hydatid cyst progression is insidious, often occurring with late diagnosis and bone destruction and neurological defect. A missed diagnosis of sacral hydatid cyst could be devastating and if laparotomy is done with inaccurate preoperative diagnosis, dangerous and life threatening complications like spillage of hydatid cyst materials in the peritoneal cavity with resulting anaphylactic shock could occur. Therefore, hydatid cyst must be kept in mind for all differential diagnosis of cystic masses in this area. Following excision, long-term follow-up is necessary due to high chance of recurrence. Despite it's a benign pathology the treatment must be aggressive and include complete removal of the cyst and avoid rupturing of the cyst contents, decompression of spinal cord, copious irrigation of the dissection site, and maintaining spinal stabilization.

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Authors Information: Mohammad Asef Adelyar[1]*; Ramin Saadaat[5]; Samea Hesham[1]; Ahmadullah Danish[1]; FarukhSiar Farooqi[1]; Tahmineh Aini[1]; Besmellah Najrabi[4]; Wahidullah Momand[1]; Hamid Wardag[2]; Hedayatullah Hayat[3]

[1]Department of Abdominal Surgery, Aliabad Teaching Hospital, University of Kabul Medical Science, Afghanistan.

[2]Department of Cardiology, Acomet Teaching Hospital, University of Kabul Medical science, Afghanistan.

[3]Department of Internal Medicine, Aliabad Teaching Hospital, University of Kabul Medical Science, Afghanistan.

[4]Department of Orthopedic Surgery, Aliabad Teaching Hospital, University of Kabul Medical Science, Afghanistan.

[5]Department of Pathology, French Medical Institute for Mother and Children, Kabul, Afghanistan.

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