

Case Report

Primary Sacral Hydatid Cyst Mimicking a Neurogenic Tumor in Chronic Low Back Pain: Case Report and Review of the Literature

Manuel Segura-Trepichio, Jose Manuel Montoza-Nuñez, David Candela-Zaplana, Josefa Herrero-Santacruz¹, Fernando Pla-Mingorance²

Departments of Orthopedic Surgery, ¹Anatomical Pathology and ²Radiology, Vinalopó University Hospital, Alicante, Spain

ABSTRACT

Hydatid disease is caused by infection of *Echinococcus granulosus*. Bone hydatid cyst presentation without hepatic affectation is infrequent and occurs in 0,5-2% of cases. This rare condition makes clinicians not always aware of the disease, and as a result, misdiagnosis of spinal echinococcosis is common. We present a case of a 48-year-old female patient with primary sacral hydatidosis. Chronic low back pain radiating to the left buttock was the only symptom. The magnetic resonance imaging (MRI) suggested a neurogenic tumor versus giant cell tumor. Biopsy and pathological study revealed a hydatid cyst. Anthelmintic and surgical treatment was performed. At 12 months after surgery, the patient is free of recurrence. In patients with chronic low back pain and a MR suggestive of neurogenic tumor, spinal hydatid cyst should be considered in the differential diagnosis. It is recommended the assistance of an anesthesiologist during biopsy to avoid an anaphylactic shock.

KEYWORDS: Echinococcosis, giant cell tumor of bone, hydatid disease, low back pain, spinal cord neoplasms

INTRODUCTION

Hydatid disease (HD), also known as cystic echinococcosis, is caused by infection with the larval stage of *Echinococcus granulosus*.^[1] The disease has the highest incidence in countries where sheep are raised with the help of dogs. In endemic regions, human incidence rates for HD can reach >50/100,000 person-years, and prevalence levels as high as 5%–10% may occur in parts of East Africa, Central Asia China, Argentina, and Peru.^[2] The adult tapeworm, which inhabits the small intestine of dogs, produces eggs which contain the larval tapeworm (oncosphere). Eggs excreted in the feces are dispersed widely in the environment. Intermediate hosts (sheeps and humans) become infected by accidental consumption of water or food that has been contaminated. Oncospheres penetrate the intestine and are carried through bloodstream where they are filtered out in various organs.^[3] The most common hydatid cyst sites in humans are the hepatic filter, 60%–70% of cases, followed by lung and brain. Bone hydatid cyst presentation without hepatic affectation is infrequent and occurs in 0,5-2% of cases, half of which infest the spine.^[4] Primary involvement of the sacral spine, as in the current case, is very rare.

The first case was described in 1951.^[5] Since then, few cases have been reported in literature [Table 1]. As a result, sacral hydatid cyst (SHC) is characterized by clinical latency, late diagnosis, and therapeutic difficulties. We document diagnosis and treatment of a patient with a

primary SHC and is compared with other cases published in literature.

CASE REPORT

A 48-year-old woman from South Mediterranean region was admitted at our center because of low back pain radiating to the left buttock. She had no improvement with analgesia and physiotherapy for a year. Neurological examination showed no motor, sensory, patellar, or Achilleian reflexes' deficits, and no bladder disorders. The patient had a lumbar radiography without abnormalities. The magnetic resonance (MR) revealed a cystic lesion with cerebrospinal fluid-like signal in contact with left sacral roots extending through sacral holes reaching iliac muscle [Figure 1]. The MR image suggested, as the first option, a neurogenic tumor (sacral neurofibroma or schwannoma), as the second, giant cell tumor (GCT) or chordoma. After consultation with radiologists and pathologists, a computed tomography-guided biopsy was performed with an 11-gauge bone access needle. During biopsy, a greenish-yellow aspirate was obtained. Pathological study revealed hydatid cysts presence, with an inner germinal layer, surrounded by an outer layer of chitin [Figure 2]. Specific

Address for correspondence: Dr. Manuel Segura-Trepichio, Department of Orthopedic Surgery, Vinalopó University Hospital, 03203 Elx/Elche, Alicante, Spain. E-mail: manusegura5@gmail.com

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enzyme-linked immunosorbent assay (ELISA) serological tests proved negative. Before surgery, treatment with oral albendazole (400 mg/12 h) and praziquantel (100 mg/8 h) for 2 months was given to the patient.

Surgical treatment was performed using a dual approach to the lumbosacral spine.

First stage: Ilium anterior approach, dissecting external oblique, internal oblique, and transversus abdominis. The cyst extended to the psoas muscle and left sacral foramina [Figure 3]. During dissection of the cyst, the wall was broken and the operating field was irrigated with hypertonic saline (7.2% NaCl).

After the anterior approach, the patient was prepared for the second posterior spine approach to remove the sacral root origin of the sacral cyst [Figure 4]. Lumbopelvic instrumentation and bone grafting (femoral head) to fill the bone defect in the sacrum were added [Figure 5].

The patient continued treatment with albendazole (400 mg/two times a day) and praziquantel (40 mg/kg day) 3 months after surgery. At 12 months after surgery, the patient is free of symptoms and is complete autonomous for performing the basic activities of daily living.

DISCUSSION

Misdiagnosis of sacral echinococcosis in MR is common [Table 1].

In the present case, MR suggested a sacral neurogenic tumor. Neurofibromas, or schwannomas, can grow through the neural foramina outward from the sacral canal, and a giant mass may form anterior to the sacrum. Ogose *et al.* reported a presacral schwannoma with purely cystic form with similar MR characteristics to a SHC.^[24] On MR, benign schwannomas

have smooth tumoral margins and are isointense with muscle on T1-weighted images and hyperintense on T2-weighted images.^[25] In SHC, MR shows a cystic lesion with similar characteristics to cerebrospinal fluid with a thin peripheral enhancement after contrast administration. The sacral hydatid cyst (SHC) capsule is best seen on T2- and proton density-weighted images.^[26,27]

Hemama *et al.* reported a SHC as a meningocele on MR.^[8] Anterior sacral meningocele is a protrusion of the meninges through a defect in the anterior aspect of the sacrum into the retroperitoneal space. This lesion is seen with equal signal intensities of cerebrospinal fluid on all MR sequences and is usually unilocular.^[28]

Song *et al.* mistook a SHC with a GCT on MR.^[15] The most common GCTs site is distal femur and proximal tibia in 50%–65%, but can affect sacrum in 4%–9% of cases.^[29] The MR T2-weighted images show a heterogeneous high signal with areas of low-signal intensity (variable) due to hemosiderin or fibrosis. High signal in adjacent bone marrow thought to represent inflammatory edema.^[30]

Yegen *et al.* reported a SHC as a sacrococcygeal chordoma on MR.^[22] The sacrum is the most common location, accounting for approximately 30%–50% of all chordomas.^[31] Because chordomas lie in bone, they are usually extradural and induce bone destruction. In the MR, destructive lytic lesion with expansile soft-tissue mass can help distinguish chordoma from SHC.

We must be very careful with the SHC biopsy. Puncture of the cyst could cause the migration of parasites to the bloodstream causing an anaphylactic shock; hence, the assistance of an anesthesiologist during biopsy is recommended.^[32]

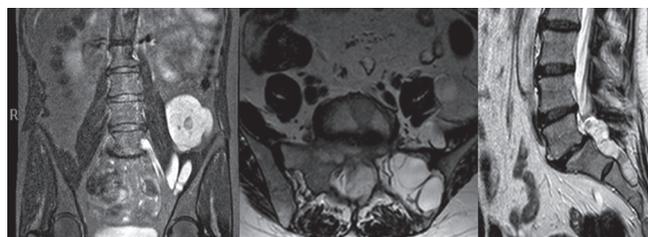


Figure 1: Magnetic resonance T2-short-tau inversion recovery. Coronal, axial, and sagittal planes. Cystic lesion with cerebrospinal fluid-like signal in contact with left sacral roots extending through sacral holes reaching iliac muscle

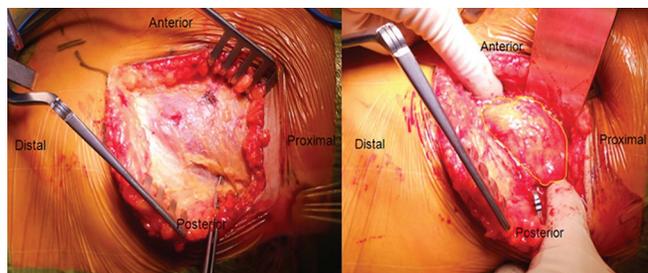


Figure 3: Ilium anterior approach, dissecting external oblique, internal oblique, and transversus abdominis after which the hydatid cyst was found

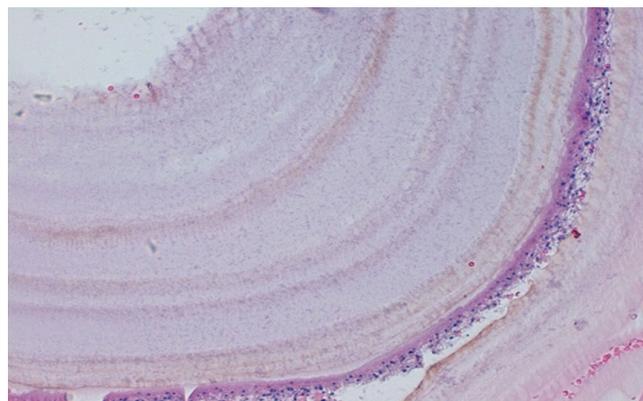


Figure 2: Hydatid cyst (cyst wall, inner germinal layer) (H and E, ×200)

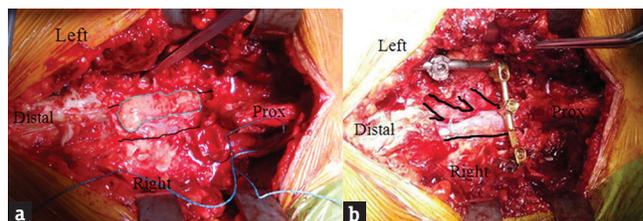


Figure 4: Posterior approach. (a) Hydatid cyst (highlighted in blue) on top of the dural sac. (b) First and second left sacral roots, after removing the hydatid cyst (highlighted in black)

Table 1: Primary sacral hydatid cyst articles by country. Sociodemographic data, presentation, and management

Author	Country	n	Age	Gender	Presentation	Diagnosis	Treatment
Abdelhakim <i>et al.</i> , 2014 ^[6]	Tunisia	1	19	Male	Cauda equina	MRI	Albendazol + surgery
Najib <i>et al.</i> , 2012 ^[7]	Morocco	3	42	Male	Low back and leg pain	CT	Albendazol + surgery
			37	Male		MRI	
			25	Female			
Hemama <i>et al.</i> 2011 ^[8]	Morocco	1	9	Male	Paraparesis	MR (meningocele informed)	Albendazol + surgery
Patel and Shukla, 2010 ^[9]	India	1	19	Male	Low back pain	MRI (tarlov cyst informed)	Albendazol + surgery
Senoglu <i>et al.</i> , 2009 ^[10]	Turkey	1	73	Male	Low back and leg pain	CT MRI	Albendazol + surgery
Sasani and Ozer, 2009 ^[11]	Turkey	1	35	Male	Low back pain	MRI	Albendazole + surgery
Adilay <i>et al.</i> , 2007 ^[12]	Turkey	1	31	Male	Cauda equine	MRI	Albendazole + surgery
Bron <i>et al.</i> , 2007 ^[13]	Netherlands	1	27	Male	Low back and leg pain	MRI (diagnosis was missed initially)	Anti-helminthic + surgery
Joshi <i>et al.</i> , 2007 ^[14]	Spain	1	59	Female	Low back and leg pain	CT MRI	Albendazol + surgery
Song <i>et al.</i> , 2007 ^[15]	China	6	28, 3 (15–56)	Nonreported	Low back and leg pain	MRI (tuberculosis, metastasis, giant cell tumors informed)	Albendazol + surgery
Herrera <i>et al.</i> , 2005 ^[16]	Spain	5	70	Female	Cauda equina Low back and leg pain	CT	Mebendazole or albendazole + surgery
			57	Male		MRI	
			63	Male		Surgery	
			18	Male			
			20	Male			
Layadi <i>et al.</i> , 2005 ^[17]	Morocco	1	35	Man	Cauda equina	MRI	Surgery
Yilmaz <i>et al.</i> , 2004 ^[18]	Turkey	1	38	Female	Low back and leg pain	CT	Mebendazole + surgery
						MRI	
Chiboub <i>et al.</i> , 2001 ^[19]	Morocco	3	30-55	Female	Low back and leg pain	CT	Albendazole + surgery
				Female			
				Female			
Akhaddar <i>et al.</i> , 1999 ^[20]	Morocco	1	16	Male	Low back pain	CT + biopsy	Mebendazole + surgery
Pandey and Chaudhari, 1997 ^[21]	India	1	15	Male	Cauda equina	MRI	Surgery without mebendazole or albendazole
Yegen <i>et al.</i> , 1993 ^[22]	Turkey	1	46	Male	Low back pain + cauda equina	MRI (sacroccygeal chordoma informed)	Surgery without mebendazole or Albendazole
Cardona <i>et al.</i> , 1983 ^[23]	Spain	2	40	Female	Low back pain	CT computed tomography (tuberculosis informed)	Surgery + mebendazole
			26	Male			

MRI: Magnetic resonance imaging, CT: Computed tomography, MR: Magnetic resonance, CT: Computed tomography

The indirect hemagglutination test and the ELISA serology is 80%–100% sensitive and 88%–96% specific for liver cyst infection, but less sensitive for lung involvement (50%–56%) or other organs (25%–56%).^[33] In the present

case, the serological tests were negative. Other tests, such as immunodiffusion and immunoelectrophoresis, provide specific confirmation after a positive ELISA serology, and they were not used in this patient.^[34]

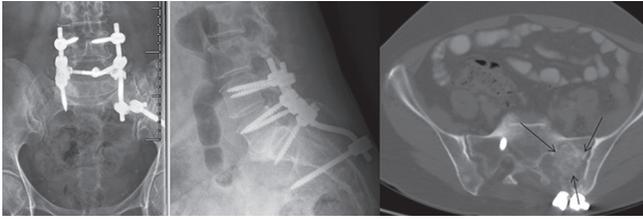


Figure 5: Lumbopelvic instrumentation and bone grafting (femoral head) to fill the bone defect in the sacrum (highlighted with black arrows)

Although medical treatment can decrease the size of cysts, the definitive treatment is complete surgical removal of the cyst.^[3] There are several antiparasitic treatment alternatives; however, the most effective therapy appears to be the combination of albendazole + praziquantel rather than albendazole alone.^[35,36]

CONCLUSION

In patients originating from endemic areas of HD, with chronic low back pain and an sacral MR suggestive of neurogenic tumor, meningocele, chordoma, or GCT, sacral spinal HD should be considered in the differential diagnosis. Treatment with surgery and albendazole + praziquantel is effective for removal of the cysts.

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Conflicts of interest

There are no conflicts of interest.

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