Scientific Foundation SPIROSKI, Skopje, Republic of Macedonia
Open Access Macedonian Journal of Medical Sciences. 2023 Jan 02; 11(C):80-83.
https://doi.org/10.3889/oamjms.2023.11257
eISSN: 1857-9655
Category: *C - Case Reports*
Section: *Case Report in Surgery*

Vertebral Hydatid Cyst: A Case Report

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Edited by: Ksenija Bogoeva-Kostovska
Citation: Abdennadher A, Jemai N, Ayari R, Mallat Y, Raboudi M, Ben Romdhane N, *et al.* Vertebral Hydatid Cyst: A Case Report. Open Access Maced J Med Sci. 2023 Jan 02; 11(C):80-83. https://doi.org/10.3889/oamjms.2023.11257
Keywords: *Echinococcus*; Hydatid; Vertebral hydatid cyst
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Received: 13-Nov-2022
Revised: 26-Nov-2022
Accepted: 28-Nov-2022

Funding: No external funding was received for this study
Competing Interests: Authors have declared that no competing interests exist

Abstract

BACKGROUND: Hydatid disease is caused by the larval form of *Echinococcus granulosus*. The reported occurrence of bony hydatidosis is 0.5–3% of all the cases and 50% of them affect the spine. Nevertheless, lumbar vertebral involvement is extremely rare. The most common occurrence is the dorsal level.

CASE REPORT: We present the case of a 32-year-old-military dog handler, who presented in 2018 to the urology department with an abdominal retroperitoneal mass. The MRI showed multilocular cystic lesions. The patient underwent surgery, and all of the cysts were removed. The anatomopathological evaluation concluded to hydatic cysts. He was briefly relieved from his symptoms. He was addressed to our department in 2020 suffering from lumbar-radicular pain and functional impotence of both lower limbs. MRI was performed showing multiple big cysts with inhomogeneous contents. CT scan showed destruction of the l5 vertebra. The patient underwent surgery. Initially, we performed through a posterior approach, an L4 to S1 laminectomy, posterior stabilization, and then total L5 corpectomy, anterior l4-s1 fusion through a xipho-pubic laparotomy after 2 months. Antihelminthic therapy was administrated. The patient's symptoms completely disappeared. No signs of reoccurrence were noted at the 2-year follow-up.

CONCLUSION: The primary extrahepatic cystic echinococcosis of bone is an extremely rare disease. Diagnosis can be long and difficult. It can lead to serious complications and should be highly considered in case of a cystic vertebral lesion in an endemic region. Spinal involvement is extremely rare but potentially curable with surgery and anthelmintic drug therapy.

Introduction

Hydatid disease is caused by the larval form of *Echinococcus granulosus*. *E. granulosus* uses men, sheep, and cattle as an intermediate host. The dog is the common definitive host, transmitting it to humans by direct contact. The reported occurrence of bony hydatidosis is 0.5–3% of all the cases and 50% of them affect the spine [1].

Nevertheless, lumbar vertebral involvement is extremely rare. The most common occurrence is the dorsal level.

We present an extremely unusual case of primary spinal lumbar/retroperitoneal hydatidosis, treated with total vertebrectomy with satisfactory results.

Case Report

We present the case of a Tunisian 32-year-old military dog handler, with no medical history, who presented in 2018 to the urology department with an abdominal retroperitoneal mass. An MRI was performed and documented many multilocular cystic lesions in contact of iliopsoas muscle and at the L4–L5 space [Figure 1]. He underwent surgery through an extraperitoneal approach (Leriche) [Figure 2]. All of the cysts were removed. The anatomopathological evaluation concluded to hydatic cysts.

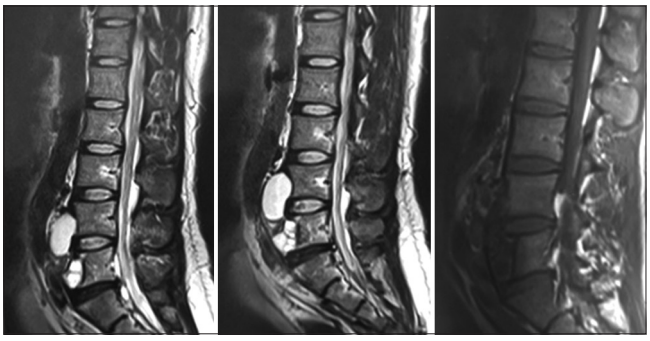

Figure 1: MRI findings of 2018

The patient was briefly relieved from his symptoms. He was addressed to our department in 2020.

He presented with lumbar-radicular pain and functional impotence of both lower limbs.

Physical examination showed sensory deficits on the right lower leg and foot with no sphincter disorders.

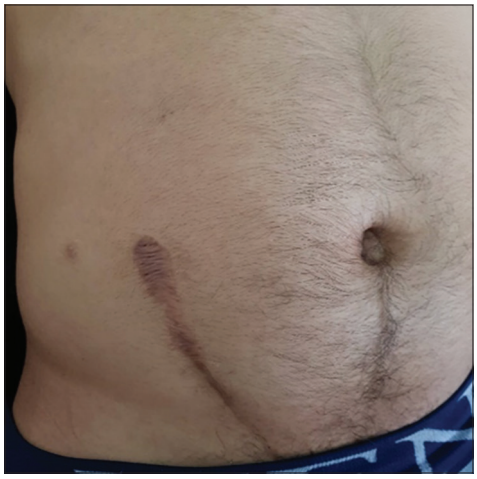

Figure 2: Leriche scar

The patient could stand up and walk with difficulty. Lasegue's sign and Mingazzini test were positive on both sides. There were no other abnormalities.

A lumbosacral MRI was performed showing multiple big cysts with inhomogeneous contents. The lesion has increased in volume. It is now extending from the right para-vertebral region to the L4–S2 midline.

The examination documented many cystic lesions with multilocular aspect [Figure 3] repressing the aorta and the inferior vena cava.

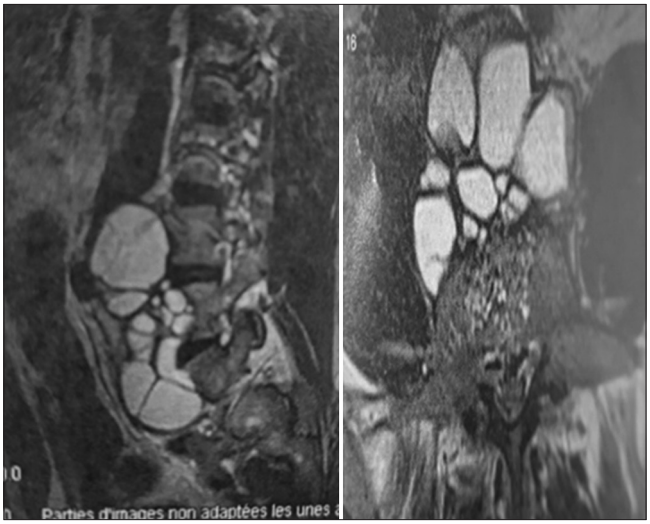

Figure 3: MRI findings of 2020

CT scan showed destruction of the L5 vertebra [Figure 4]. The anterior surfaces of L4 and L5 vertebrae were scalloped suggesting the cyst's long-standing nature.

The patient underwent surgery. Initially, we performed through a posterior approach, an L4 to S1 laminectomy, posterior stabilization with pedicular screws, and bone grafting [Figure 5]. Postoperatively, the patient received albendazole for 2 months.

He was relieved from his neurological signs.

After 2 months, the patient underwent a second surgery. This time we performed a total L5 corpectomy

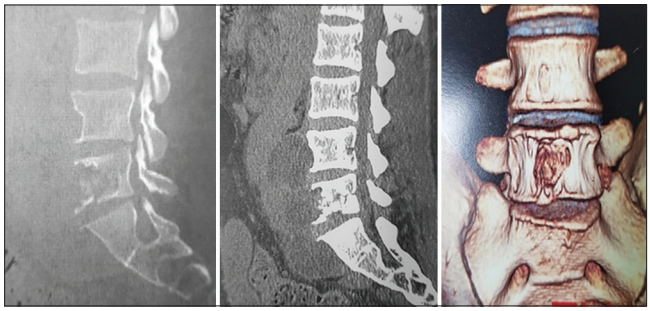

Figure 4: CT scan images

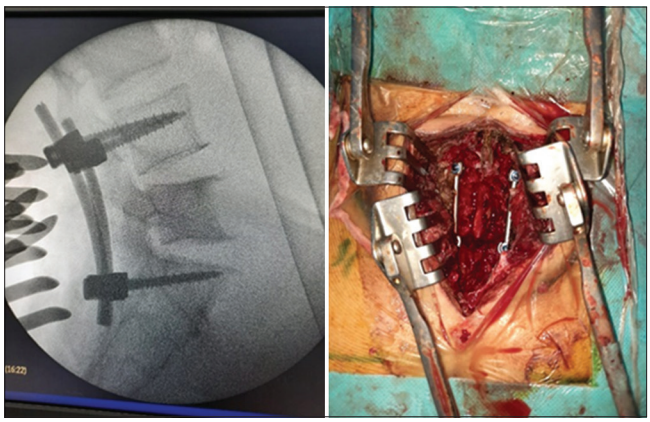

Figure 5: Posterior stabilization

thorugh a xipho-pubic laparotomy. 20% hypertonic saline solution was used for washing. The right iliac artery and vein were retracted to the left [Figure 6]. The ureter was identified and protected. The cyst was then directly exposed in the right retroperitoneal region. It was adherent to the right common iliac vein. The liquid content of the cysts was carefully aspirated, followed by an injection of 3% hypertonic saline solution into the cysts [Figure 6]. The corpectomy of L5 was performed. Then, L4–S1 fusion was performed using a pyramesh cage. No neurological deficits occurred following surgery.

On the past follow-up, 2 years postoperatively, the patient was satisfied with no lumbar pain nor neurological signs. No signs of reoccurrence were noted. A close follow-up is regularly performed

Discussion

Hydatid disease is a worldwide public health problem. It is mostly formed in the liver (60–70%) and the lungs (10–15%) [1]. The incidence of bone hydatid disease reported is 0.4–5%, involving the vertebral column in ≥50% of these cases [2].

There are two types of hydatidosis: primary, caused by hexacanth spreading by direct extension from primary pulmonary, abdominal, or pelvic infestation. Secondary contamination can be hematogenous, "per

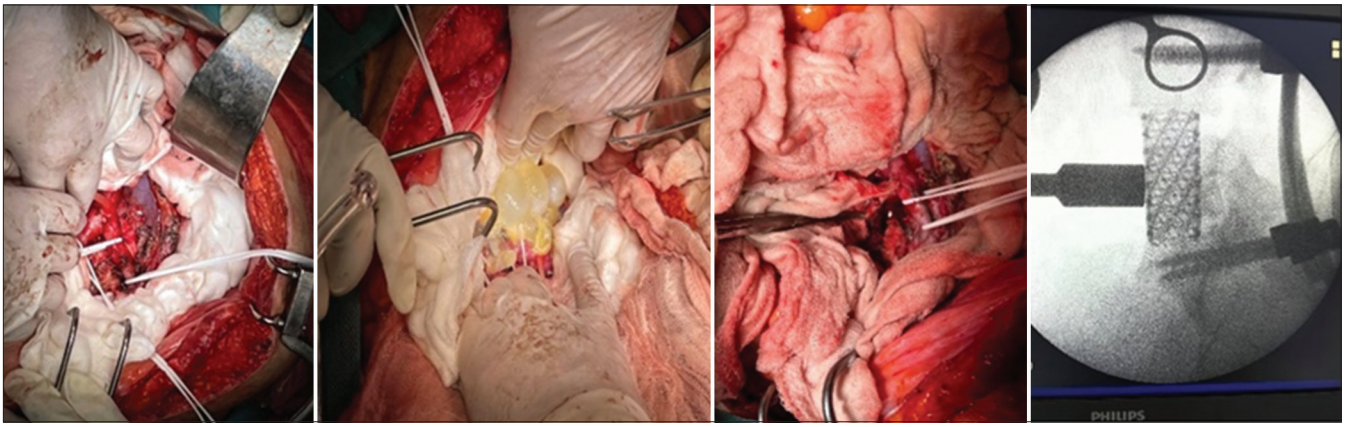

Figure 6: L5 corpectomy

contiguitatem" with direct invasion or "per continuitatem" by direct seeding of a hydatid cyst [3].

Bone involvement is often caused by the primary form. The embryos can reach the vertebrae through arterial circulation, lodging in any part of it. The parasite usually affects the more vascularized regions, like the vertebral body, and can extend to the dural sac, the spinal cord, and the paravertebral soft tissues. Infestation of the spine usually begins as a multivesicular infiltration of the spongy bone of the vertebral body that extends to the pedicles and laminae. Cyst growth usually respects the intervertebral disk since the cysts do not compromise the periosteum, at least in the early stages.

Vertebral hydatidosis was classified into five types by Braitwaite and Lees [4]: (1) primary intramedullary, (2) intradural extramedullary, (3) intraspinal extradural, (4) vertebral hydatidosis, and (5) paravertebral hydatidosis.

Primary retroperitoneal hydatid cyst without other organ involvement was first reported by Lockhart and Sapinza in 1958 [4]. In 1973, Mukherjee *et al* [5] reported nine cases, two had died due to anaphylactic reaction resulting from spillage during excision or biopsy done with the misdiagnosis of a retroperitoneal tumor.

In our case, the origin of the cyst is the retroperitoneal space.

Due to the lack of typical clinical characteristics and imaging criteria, the diagnosis is difficult and challenging. Early diagnosis is fundamentally facilitated by the epidemiological risk factors. Acute onset to protracted clinical courses are both possible in the progression of symptomatic illness. Pain is the most common clinical manifestation, followed by pathological fractures and medullar syndrome signs that indicate spinal cord compression.

There are no specific radiographic signs of bone hydatid disease. The primary function of CT is to screen the damaged vertebral structures. Hydatid cyst imaging is quite variable and may resemble giant cell tumors, tuberculosis metastases, or the normal pattern of a cystic lesion. The best imaging method is MRI. In MRI imaging, a hydatid cyst mainly has a single thin wall, and the contents have the same signal strength as the cerebrospinal fluid. The lesion shows no gadolinium uptake. More details about the lesion can be obtained by diffusion-weighted imaging, distinguishing between complex infected hydatidosis and abscesses.

Our CT and MRI findings lined up with those reported in the literature.

Visceral cystic echinococcosis was successfully treated with antihelminthic therapy using benzimidazole derivates (albendazole and mebendazole), despite recent research showing that response to treatment is strictly dependent on the stage of the disease and the size of the cysts. As a result, the efficacy of medical treatment may have been overstated. The use of benzomidazoles to treat or even prevent recurrences of vertebral cystic echinococcosis is being debated as a result of the lack of efficacy data on their usage in the treatment of bone cysts.

However, some authors have reported positive results of medical therapy with albendazole in patients with inoperable spinal cystic echinococcosis or disseminated disease [6].

In our case, the cysts were large with soft-tissue involvement and bone lesions, requiring a surgical management by posterior then anterior approaches with a total resection of L5 vertebra as well as the cysts.

The long-term outcome depends on the complete resection of all parasitic lesions, which is often hampered by the infiltrative nature of the disease.

Conclusion

Primary extrahepatic cystic echinococcosis of bone is an extremely rare disease. Diagnosis can be long and difficult due to the various and unspecific clinical features. It can lead to serious complications and should be highly considered in case of a cystic vertebral lesion in an endemic region. Spinal involvement is

extremely rare but potentially curable with surgery and anthelmintic drug therapy.

Ethics Approval and consent to participate

As per university standard guideline, participant consent and ethical approval have been collected and preserved by the authors.

Consent for Publication

Participant consent and ethical approval have been collected and preserved by the authors.

Availability of Supporting Data

The data supporting our findings are available and can be found in the orthopedics department of the Military Hospital of Tunis.

Authors' Contributions

All authors contributed to the elaboration of this study. Khalil AMRI and Rabie AYARI designed the study and wrote the first draft of the manuscript. Achraf ABDENNADHER managed the analyses of the study. Youssef MALLAT and Nacef JEMAI managed the literature searches. All authors read and approved the final manuscript.

Acknowledgments

To our mentor, idol and friend Khalil AMRI for his never-ending helping and compassion.

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