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Case Report

Vertebral hydatid cyst infection. A case report

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ABSTRACT

Hydatidosis is a common parasitic zoonosis in Middle Eastern, African, and Mediterranean populations whit primarily and well known involve of liver and lungs, but some complications are extremely rare and underrated. Particularly Hydatid cystic disease of the skeletal is one of the rarest clinical manifestations and when occurs involve in almost 50% of cases the spine. This manifestation is extremely debilitating, hard to correctly identify and manage. We want to underline this rare involve of spine to avoid misdiagnosis and complications.

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Introduction

Hydatidosis is an infection disease caused by the larval stages of a little endoparasitic flatworm belonging to the Class Cestoda, or better it is a 'true tapeworm' of Subclass Eucestoda, genus *Echinococcus* in the family Taeniidae.

Four forms of genus *Echinococcus* have medical relevance:

- *Echinococcus Granulosus*
- *E. Multilocularis*
- *E. Vogeli*
- *E. Oligathrus* (but is extremely rare)

The course of infections requires several intermediate hosts (usually herbivores), who are however different between

the various forms of echinococcosis and ends in the definitive host who is often a Canidae.

Humans are accidental hosts, because they are usually a dead end for the parasitic infection cycle.

The most common form is cystic echinococcosis (CE) also known as unilocular echinococcosis, caused by *Echinococcus granulosus*. The second most common form is alveolar echinococcosis, (also known as alveolar colloid of the liver, alveolar hydatid disease, alveolococcosis, multilocular echinococcosis, "small fox tapeworm"), caused by *Echinococcus multilocularis* and the third is polycystic echinococcosis (also known as human polycystic hydatid disease, neotropical echinococcosis), caused by *Echinococcus vogeli* and very rarely,

In the human, the characteristic cystic lesions are mainly found in the liver for 70% and in the lungs for 20%, in the 3% of

Abbreviations: CE, cystic echinococcosis; FDG, 18F-fluorodeoxyglucose; PET-CT, Positron Emission Tomography - Computed Tomography.

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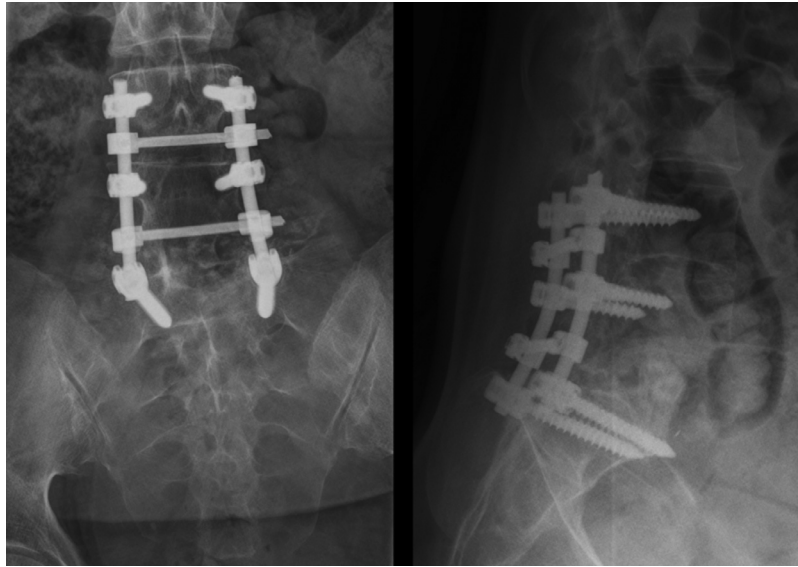


Fig. 1 – Details of lumbar spine X-Ray showing the surgical outcomes and no others findings.

all cases is involved the central nervous system, but virtually any part of the body may be affected. The vertebral column (which is involved in 50% of the 0.5%-4% of cases affecting the bone) [2–4] are particularly vulnerable given the sequelae of their involvement [5].

We report a hydatidosis case of the column in a woman of 39 years old, with severe lumbar-radicular pain and deficit of deambulation.

Case report

A 39-years-old female has developed increasing lumbar-radicular pain and functional impotence of lower limbs. The patient had a history of lumbar-radicular pain since 2005 caused by many hydatid cystic lesions treated with corpectomy on L5, disc decompression and stabilization with screws and plates. The parasitological examination of the lesions has documented the presence of *Echinococcus granulosus*.

Actually, the patient can't stand up, the Lasegue's sign is positive on the right and Mingazzini test is positive on either side, especially on the right, and she has right foot dorsal flexion deficit.

An XR of the column was performed which excluded post-traumatic alterations and only showed the previous surgery outcomes Fig. 1. Due to the pain increasing, she was hospitalized. A lumbosacral MRI was performed (Intera 1.5 T; Philips Healthcare, Best, The Netherlands, with sagittal T1 TSE, T2 TSE, STIR sequences, axial T2 SPIR and T1 TSE and next to endovenous injection of Gadolinium was acquired axial and sagittal T1 TSE) to properly investigate the clinical status of the patient and to find the cause of her lumbar pain.

The exam documented many cystic lesions with multilocular aspect Fig. 2, without any post-contrastographic enhancement Fig. 3, with extension into spinal canal at L4-L5

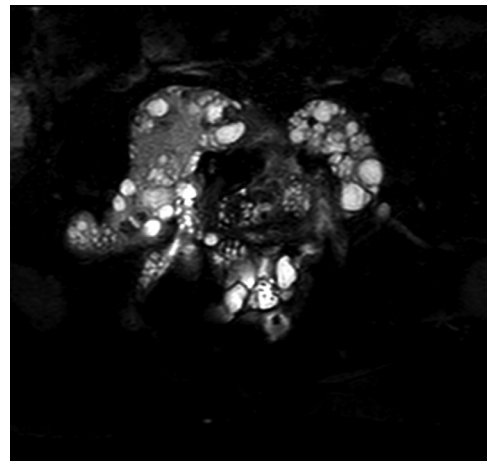


Fig. 2 – Axial sequence T2-SPIR (Spectral Presaturation with Inversion Recovery) of lumbar spine at L4 level showing multiple cysts.

level, paravertebrale on the both side and into the para-spinal soft tissues Fig. 4.

Probably, this is a recurrence of the same infection, not completely eradicated after the surgery that the patient underwent in 2005 and medical therapy, followed only for a few months due to lack of adherence to therapy.

In subsequent days a PET-CT scan with ^{18}F -fluorodeoxyglucose (FDG) was performed. The CT confirmed the cystic lesions on spine and demonstrated multiple ubiquitous pulmonary cistic-like lesions Fig. 5, without contrast enhancement. All these findings didn't show any form of uptake of FDG Fig. 6.

Albendazole therapy was started at a dose of 10-15 mg/kg/day for at least 6 months. The patient is currently in follow-up.



Fig. 3 – Sagittal T1 TSE (Turbo Spin Echo) post gadolinium injection showing no pathological enhancement.

The signs of previous surgery did not create problems for the diagnosis of the recurrence of the infection and did not generate artifacts related to the magnetic field of the reso-

nance. In relation to the anamnestic data and the MRI images it was easy to formulate a correct diagnosis.

Discussion

Hydatid disease is a worldwide public health problem. Medical literature demonstrates that bone Echinococcosis is underrated due to medical community lack of awareness, and a high incidence of misdiagnosis that lead to damage in term of therapy outcome, complications and economy impact. According to data reported in an Italian study, the estimated average national financial burden is 4 million euros per year [4]. Disease incidence is extremely low because the liver and lung trap most of the larvae and skeletal hydatidosis results from the deposition of the larval forms filtered out of liver/lungs into the bone tissue [1]. From the literature review [6] in a period between 2012 and 2017, few cases of bone echinococcosis have been documented, and only 3 cases of spinal disease were from Europe. Incidence of bone hydatid disease is reported to be 0, 4%-5% from various studies and the vertebral column is involved in $\geq 50\%$ of these cases, producing a particularly vulnerable and severe disease and sequelae. Based on published data, [7], the frequency of involved spine levels shows a predominant involvement of the thoracic (45%-50%) and the lumbar spine (20%-39%). The involvement of the individual vertebral levels is rather gradual with an ascending decline. Primary isolated bone hydatid disease is possible but is a very rare occurrence.

Hydatid disease of spine usually spreads by direct extension from primary pulmonary, abdominal and pelvic infestation. In these cases, primary lesions are well discernible on radiographic images.

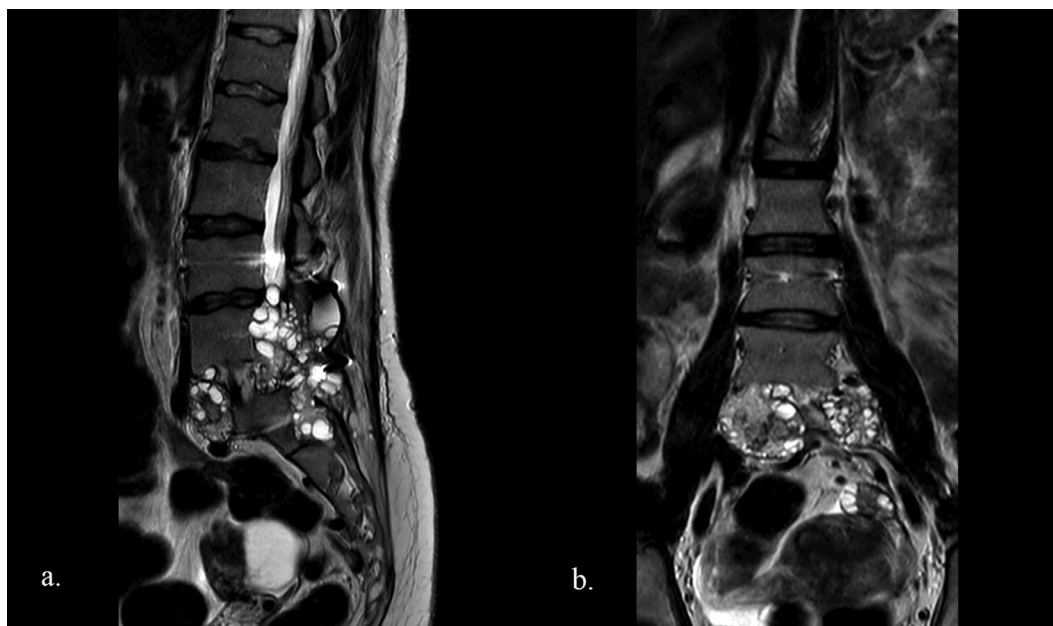


Fig. 4 – A. Sagittal T2 TSE (Turbo Spin Echo) and B. Coronal T2 TSE sequence showing the involvement of spinal canal and para-vertebral tissues.

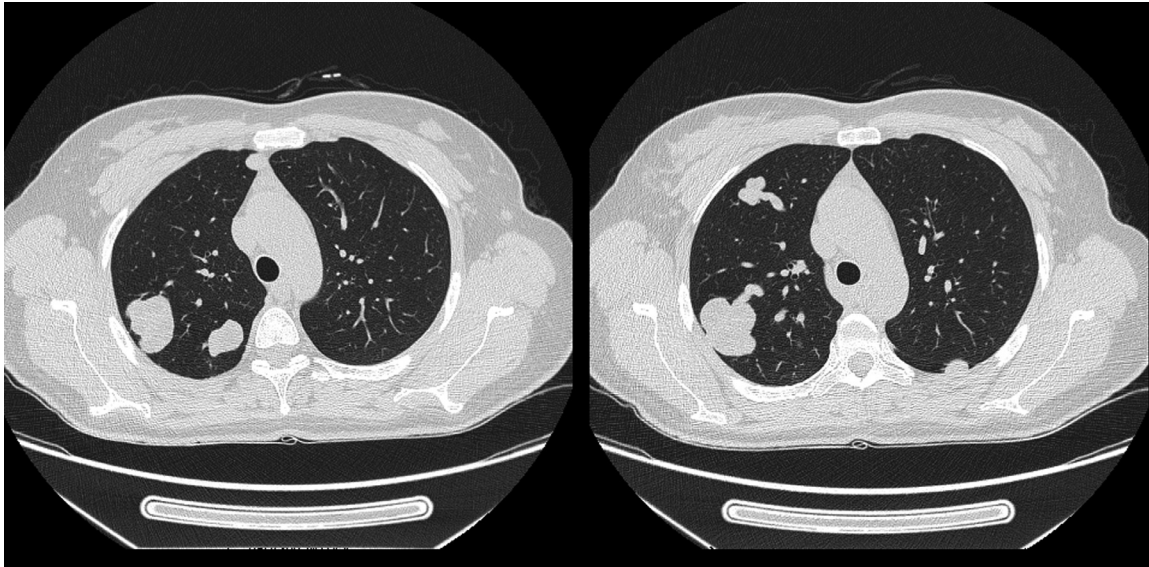


Fig. 5 – Axial tomography images of chest, performed during a PET-CT, showing multiple bilaterally cystic lesions.

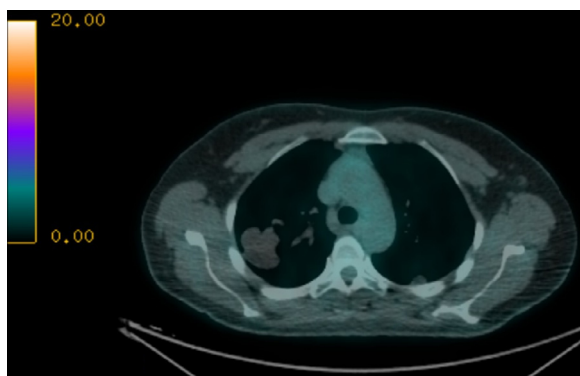


Fig. 6 – A fused PET-CT image that excludes pathological uptake of FDG (18F-fluorodeoxyglucose) of cystic lesions.

Spinal CE can be classified according to the route of spinal infection [7]:

1. Primary hematogenous spinal CE: hematogenous infection of spinal structures at primary infection.
2. Secondary hematogenous spinal CE: hematogenous infection of spinal structures following spontaneous or iatrogenic seeding from extraspinal CE.
3. Secondary ‘per contiguitatem’ spinal CE: direct invasion of spinal structures from extraspinal CE (mediastinal and paravertebral soft tissue, pleura, lung, ribs, pelvis, posterior paravertebral muscles).
4. Secondary ‘per continuitatem’ spinal CE: cerebral CE with spontaneous or iatrogenic seeding into the cerebrospinal fluid, leading to intradural spinal seeding.

Bone hydatid disease lacks specific radiological findings. CT has the primary role of screening the damaged vertebral structures. Imaging of hydatid cyst is quite variable and may have the typical pattern of a cystic lesion, exhibiting

no contrast enhancement or may be similar to tuberculosis metastases, or giant cell tumour. MRI is considered the best imaging tool. A hydatid cyst mostly contains a single thin wall and the contents have the same signal intensity as the cerebrospinal fluid in MRI imaging. The lesion shows no gadolinium uptake. Diffusion-weighted imaging can give more information about the lesion, as helping to differentiate between complicated infected hydatidosis from abscesses. Our CT and MRI results conformed to those in the literature.

The EC diagnosis is difficult and challenging due to the lack of typical clinical appearance and image characteristics. The epidemiological risk factors have a fundamental role in facilitating early diagnosis. The course of symptomatic disease might range from acute onset to prolonged clinical courses. The most frequent clinical presentation is pain followed by pathological fracture and symptoms associated with spinal cord compression, as medullar syndrome. This was reflected in our case where the patient presented only no specific symptoms and there was no alteration in laboratory excepted PCR increases.

Anthelmintic therapy with benzimidazole derivates (albendazole, mebendazole) showed its effectiveness in visceral CE although latest evidences concluded that response to treatment is strictly dependent from disease stage and cysts size; therefore, the effectiveness of medical treatment may have been overestimated. Efficacy data on Benzomidazoles in the treatment of bone CE are lacking, accordingly their use to cure or even prevent recurrences in vertebral CE is under discussion.

However, some authors have reported positive results of medical therapy with albendazole in patients with inoperable spinal CE or disseminated disease: in a study of 40 patients with inoperable vertebral hydatidosis treated with albendazole, El-Mufti and colleagues [8] reported a healing rate of 53% during a follow-up over 2 years. Islakel and colleagues [9] concluded that in patients with spinal CE submitted to surgery,

an additional medical therapy seems to delay the recurrence rate on long-term follow-up.

Some cases of spinal CE in which the lesions have been treated using a percutaneous approach are reported in literature. Foad and colleagues [6] described 2 cases of patients undergoing percutaneous CT-guided treatment according to PAIR approach (Puncture of the cyst, Aspiration of the cyst fluid, Injection of a scolical agent, and Re-aspiration of the cyst content).

Spektor and colleagues [9] reported a successful percutaneous treatment in a patient with vertebral hydatid cyst (C2-C3) extending into the vertebral canal and causing severe spinal cord compression. The procedure has led to rapid spinal decompression and clinical improvement. No adverse events were observed and follow-up over 1-year showed a complete resolution of clinical symptoms and no significant radiological findings were found. The authors concluded that percutaneous treatment could be particularly useful in cases of extensive disease where a radical surgery is impossible.

Conclusion

In summary, primary extrahepatic CE of bone is an extremely rare disease. The clinical course leading to diagnosis can be long and difficult because of the various and unspecific clinical features. Despite a significant progress has been made in the diagnosis and treatment of cystic echinococcosis, the predilection of the parasite for the spine in bone CE remain poorly understood. Spinal CE remains associated with a high risk of morbidity, disability and mortality, that is why imaging, serological tests and PCR should be done immediately to lead to an early diagnosis and a timely surgery to decompress the spinal cord and stabilize the spine. In addition, the intraoperative use of scolicides and pre-, peri- and postoperative use of albendazole are currently considered to be the treatments of choice for spinal CE. The long-term outcome depends on the complete resection of all parasitic lesions, which is often hampered by the infiltrative nature of the disease. We can conclude that the spinal hydatid cyst should be considered in the differential diagnosis when there are cystic lesions that cause osteolysis and spinal compression to provide the patient with optimal treatment and rapid recovery, and then rehabilitate him for his daily and working activities.

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