Case report

Infected primary hydatid cyst of the right lumbar paraspinal muscles: A rare case in a ten-year-old child

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Introduction and importance: Most of the time, intramuscular hydatid cysts are secondary. The unusual topographies are a source of diagnostic delay and treatment difficulties. The aim of our case report is to put forward the features of this uncommon presentation and to propose a therapeutic management.

Case presentation: We report the case of a primary hydatid cyst located in the right lumbar paraspinal muscles in a ten-year-old girl, in good health and without a pathological medical history. The patient presented with a discreet right lumbar swelling of firm consistency, painless and without signs of inflammation. Ultrasound and magnetic resonance imaging suggested the diagnosis of an intramuscular paravertebral hydatid cyst. The hydatid serology was positive. The patient had en-bloc resection of the cyst. He was infected with pus. The histopathological examination confirmed the diagnosis of muscular hydatidosis. Medical treatment was started. Three years after the operation, the MRI confirmed that was no recurrence.

Clinical discussion: Echinococcus is a parasitic cestode that can infect dogs and other pets and farm animals, with humans as incidental hosts. Muscular hydatid localization, secondary to hematogenous dissemination, is rare, even in endemic countries. The combination of medical treatment with surgery ensures the best condition to avoid recurrence.

Conclusion: In regions where hydatidosis is endemic, a tumor in any part of the body should be considered a hydatid cyst until proven otherwise.

1. Introduction

Hydatid cyst is zoonotic infection caused by Echinococcus granulosus, transmitted accidentally to the humans. In order of frequency, the organs affected are the liver and lungs although it may involve the heart, kidneys, spleen and brain and rarely skeletal muscles [1]. This case report describes such a rare case for two reasons. The first is localization in the lumbar paravertebral muscles in children. The second is that the cyst contents are infected. We suggest the management of these hydatid cysts and discuss the best approach to avoid recurrence.

This work has been reported in line with the SCARE criteria [2].

2. Case report

A ten-year-old girl consulted in paediatric orthopaedics department for newly discovered right lumbar paravertebral swelling with local back pain. The swelling is firm, painful, fixed in relation to the subcutaneous plane, 11 cm long and without signs of inflammation. The temperature was 38.3 °C. The neurologic examination and the remainder of the physical examination are normal. The patient comes from a rural area and confronted domestic dogs. The x-ray of the lumbar spine is without abnormalities. Laboratory tests showed a normal total white-cell count (7800 mm³; reference range, 4000 to 10,000) with an elevated eosinophil count 54% (reference range, 0 to 5%), a C-reactive protein level of 20 mg per liter (reference value, <5) and an erythrocyte sedimentation rate of 23 mm in the first hour (reference value, <10).

Ultrasound showed a clean-walled cystic lesion with a detached inner wall with mixed anechoic and echogenic content. The ultrasound appearance is suggestive of a hydatid cyst type. MRI of the spine found a thickened cystic formation with clean walls at the height of the L4 L5 vertebrae (Fig. 1) and an inflammatory response in adjacent muscles (Fig. 2). The hydatid serology was positive. The clinical, biological and...
Radiological evidence was in favour of an infected muscular hydatid cyst. The extension workup, including abdominal and cardiac ultrasound and chest x-ray, was normal.

In collaboration between infectious disease doctor and orthopaedic surgeon, the patient was prescribed Albendazole. This medical treatment is continued for one month. Then the patient was operated, en-bloc resection of the lesion was performed. But there was a hole (Fig. 3) through which the pyogenic contents of the hydatid cyst contaminated the surgical site (Fig. 4). After its removal, operative cavity was thoroughly washed with 20% hypertonic saline and aspiration thereafter. The germ isolated is a Corynebacterium group G-2. Histopathological examination confirmed the pre-operative diagnosis (Figs. 5, 6). Albendazole treatment at a dose of 15 mg/kg/day divided into two doses was continued for six months after surgery. Antibiotic treatment Clavulanic Acid-Amoxicillin for the infection was started for two weeks. Monitoring of treatment tolerance was based on complete blood counts, liver enzymes, and renal function every three months. A control MRI performed after three years of surgery confirmed the absence of...

Fig. 1. Ten-year-old child who consulted for a right lumbar paravertebral swelling. MRI was in favour of an infected hydatid cyst. Axial T1 MRI injected slice showed intense contrast enhancement in the cystic capsule and adjacent soft tissue.

Fig. 2. Axial T2 MRI slice: hypersignal of the fluid contents of the cyst, collapse of the cystic wall (arrow) and outflow of the fluid (asterisk).
recurrence of the hydatid cyst (Fig. 7).

3. Discussion

*Echinococcus* is a parasitic cestode that can infect dogs and other pets and farm animals, with humans as incidental hosts. Infection can cause cystic lesions in the liver and lungs and also in the central nervous system and bones. Muscular hydatid localization, secondary to hematogenous dissemination, is rare, even in endemic countries [1,3,4].

We describe the case of an infected primary hydatid cyst of the lumbar paraspinal muscles and elaborate on their management. The theory for primary hydatid disease to arise in the paraspinal muscles suggested that the larvae may directly infiltrate the inferior vena cava system through the intestinal wall venous connections rather than entering the portal circulation [5].

The appearance of lumbar paravertebral swelling is the most common finding inconsistently associated with lower back pain. Some cases of intraspinal extension may be associated with myelopathy or cauda equina syndrome.

The biological tests reveal hypereosinophilia, but it is inconsistent since it is present in only 25% of cases and it is not specific. Serologic tests are valuable when they are positive. But in half of the primary intramuscular hydatidosis case serology is false negative because the capsule isolates the parasite from the host’s immune system [6]. A biological inflammatory syndrome associating an elevated C-reactive protein and an accelerated erythrocyte sedimentation rate is in favour of the infection of the cyst. Infection of the cyst with bacterial flora from bile or bronchial tree is usually caused by the communicating rupture. Infection develops only after rupture of both the pericyst and endocyst [7]. But the paravertebral muscle localization is isolated from any reservoir of contamination. Infection in this case can only be through the bloodstream. In recent experimental studies, it has been demonstrated the scolicidal effect of bacteria on fertile protoscoleces and this allows progress in the non-surgical treatment of certain hydatid cysts [8,9]. The consequence of infection is degeneration of the cyst, leading to partial or total calcification of the lesion.

Ultrasoundography should be the first preferred imaging modality for the detection of muscular hydatid cyst as being a non-invasive method. The MRI is currently considered the technique of choice; it is non-invasive and provides a better contrast resolution and multiplanar slices. It can reveal pathognomonic images of muscle hydatid cyst, such as a well-defined cystic mass with a thin cystic wall and membrane detachment [3]. The MRI can also show an oval mass, most often circumscribed without septa, of variable size, with low signal intensity on T1 weighted images and high signal intensity on T2 weighted images. This lesion is non-enhanced after injection of gadolinium and does not show perilesional edema. MRI also shows soft tissue involvement, best analyzed on coronal and sagittal slices.

The surgical excision of the muscular hydatid cyst gives good results. The tumor mass must be removed en-bloc without opening it so as not to sow cystic vesicles, sources of local recurrence or distant dissemination. In cases of accidental rupture of the cyst, the residual cavity should have local irrigation with aqueous iodine (10%), silver nitrate and hypertonic saline (20%) [10,11]. Percutaneous aspiration, infusion of scolicidal agents and re-aspiration (PAIR) performed under image guidance, is a great alternative for inoperable cases and cases with muscle localization [12]. Microscopic examination reveals the typical scolex and hooks. Molecular identification based on nucleotide sequencing of cytochrome C oxidase subunit 1 identifies *Echinococcus granulosus* [13].

The combination of medical treatment with surgery ensures the best
condition to avoid recurrence [14]. Albendazole is the mainstay drug for treatment of echinococcosis. According to a review of the literature, treatment of hydatid cyst with Albendazole may be associated with the prevention of recurrence and reduction of the size and death of the hydatid cysts [15]. It is also essential to prevent hypersensitivity reactions. In a subsequent randomized control trial looking at management of hepatic hydatidosis, it was found that the group receiving both pre and post-operative medication therapy with Albendazole showed better outcomes compared to groups who received either as a stand-alone [16]. For therapy in patients with muscular hydatidosis, variegated but little evidence is available regarding the timing and duration of Albendazole therapy [6]. The duration of the pre and postoperative treatment varies greatly according to the authors, between two days and two months preoperatively and between four weeks and thirty-six months postoperatively [6]. For sepsis, putting on antibiotic treatment combined with drainage helps to heal. The postoperative follow-up is done by clinical examination but especially by MRI every six months then every year.

4. Conclusion

Primary hydatid cyst of the lumbar paraspinal muscles can present as an isolated disease. Infection of the cyst can be seen in the muscle location. Through this case, it has been shown that the bloodstream is one of the modes of infection of hydatid cyst. Complete excision with Albendazole therapy and antibiotic represents the optimal treatment. Prolonged follow-up for recurrence is warranted.

Data availability

All data is available to readers.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Consent

Written informed consent was obtained from the patient’s parent for publication of this case report and accompanying images.

Ethical approval

N/a.

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Fig. 5. Histological image at ×20 magnification: Superinfected hydatid cyst.
Cystic wall: membrane hyaline eosinophilic acellular anhist and flaky (red arrow). Inflammatory infiltrate filling the cystic cavity (blue arrow).
Fig. 6. Histological image at ×40 magnification: Polymorphic inflammatory infiltrate made up of lympho-plasma mononuclear cells and eosinophilic and neutrophilic polynuclear cells.
Fig. 7. After 3 years, the T2 MRI in axial section did not show any hydatid recurrence but objectified fatty infiltration of the right lumbar paravertebral muscles (arrow).

Guarantor

Mohamed Zairi.

Research registration number

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CRediT authorship contribution statement

* Mohamed Zairi: Writing drafting the article, conception and design
* Rim Boussetta: Revising it critically for important intellectual content
* Ahmed Msakni: Revising it critically for important intellectual content
* Ghada Sahraoui: Reading and interpretation of histological images
* Ahmed Amin Mohseni: Supervision
* Mohamed Nabil Nessib: Final approval of the version to be published.

Declaration of competing interest

None.

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