Primary hydatid disease of the thigh. A rare location

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Summary Primary muscle hydatidosis is very rare, accounting for less than 1% of hydatid cyst locations. Clinical symptoms are insidious and non-specific causing a frequent delay in diagnosis. Intramuscular hydatid disease can cause a variety of diagnostic problems, especially in the absence of typical radiologic findings. We report the observation of an 82-year-old man consulting for inguinal tumefaction with radiological exploration suggestive of hydatid cyst of the adductors muscles. Magnetic resonance imaging (MRI) is helpful in diagnosis, since it reveals a very suggestive aspect and demonstrates the relationship between cysts and adjacent structures. Treatment of muscle echinococcosis is based on surgery, which is curative and incurs a low risk of local relapse.

Introduction

Muscle infestation by Echinococcus is a rare entity, even in endemic countries, where its frequency is estimated at less than 3% [1—3]. It is often asymptomatic and progresses slowly, which can delay diagnosis. Soft tissue hydatidosis can have several imaging aspects, which must be known to make the diagnosis preoperatively and prevent the onset of sometimes serious complications [4,5].

We report the rare observation of a hydatid cyst of the adductors of the thigh and discuss the MRI arguments suggesting the hydatid origin of a soft tissue mass as well as the therapeutic options, particularly surgery, for muscular echinococcus disease.

Observation

An 82-year-old man living in a rural area, with no notable history, was hospitalized for exploration of a tumefaction of the internal left thigh that had appeared 5 months before, progressively increasing in volume. The patient was apyretic, in good general health, presenting a voluminous mass measuring 10 × 5 cm on the internal side of the root of the left thigh occupying the thigh's medial compartment (Fig. 1). This mass adhered to the deep structures and was slightly
sensitive to palpation, with normal-appearing skin and no signs of nerve or vascular compression.

The standard X-rays demonstrated soft tissue thickening with no calcifications or bone abnormalities. Ultrasound of the left thigh showed an intramuscular, hypoechogenic heterogenous formation of the internal side of the root of the thigh measuring 10 cm at its widest point, with regular contours and containing multiple vesicles, suggesting the diagnosis of a multivesicular muscular hydatid cyst (Fig. 2).

The thoracic radiograph and abdominal ultrasound were normal. The hydatid serology was negative.

MRI, used to more thoroughly explore how the mass was situated in relation to the neighboring vascular and nerve structures, demonstrated at the internal side of the root of the left thigh, a rounded cystic formation with a low signal on the T1-weighted images, containing multiple vesicles with a clear hypointense signal compared to the rest of the cystic fluid (Fig. 3). On the T2-weighted images, the lesion showed a clear hyperintense signal (Fig. 4). The cysts were surrounded by a thin peripheral wall showing a hypointense signal on all sequences. The T1-weighted fat-sat sequences after gadolinium injection showed relatively substantial peripheral enhancement (Fig. 5). This cystic formation had developed within the medial compartment of the thigh, pushing the superficial femoral blood vessels upward and inward without invading them and pushing the sciatic nerve backward.

The patient was treated with monoblock excision of the cyst (Fig. 6) with no complications in the immediate follow-up period. The histological examination confirmed the diagnosis of muscular hydatid cyst.

At the last follow-up at 20 months, clinical, biological, and ultrasound monitoring demonstrated that there was no local or visceral recurrence.
Discussion

The primary muscular hydatid cyst is rare even in endemic zones. Its frequency varies from 1 to 5% [2,3]. Hexacanth embryos entering via the digestive tract are most often stopped by the liver and lung acting as filters. A very small number of hexacanths arrive in the coronary circulation where they spread throughout the body. The infrequency of muscular locations is explained by the fact that continual muscular contractions and the production of lactic acid prevent scolex implantation [1,3—7].

A number of muscular locations have been described, with predominant involvement of the neck, trunk, and limb root muscles such, as we observed in our patient. This can be explained by the rich vascularization of these areas [2,4—6].

The clinical symptomology is insidious and nonspecific, frequently causing a delay in diagnosis. The nonspecific clinical picture can be summarized as a painless noninflammatory tumefaction progressively increasing in volume over several years while preserving the patient’s good general health [2—4]. However, a certain number of cysts are revealed by complications such as nerve compressions or infections simulating an acute abscess or a malignant tumor [3,8].

Sero logical examinations are often negative. Usually, hydatid serology is only positive in cases of infection or fissuration of the cyst [1,4].

Ultrasound is most often the key examination for orienting the diagnosis of any tumefaction of the soft tissues. It specifies the fluid nature of the tumefaction and its seat [2,7,8]. In typical cases, the diagnosis of hydatidosis can be made by showing a more or less heterogenous liquid formation with visualization of daughter vesicles, as in our case [2,4]. However, there are atypical forms in which the lesion is either mixed or a solid pseudotumor with or without rounded transonic elements within [5].

The advantage of computed tomography (CT) resides in its ability to enumerate the cysts and to demonstrate their size and topography as well as their relation with the vascular and nerve structures [1,3]. However, CT presents the same diagnostic problems as ultrasound if the mass is not clearly liquid and the vesicle and/or membrane structure cannot be patently identified [2,5].

MRI is the first-choice diagnostic method in hydatid disease of the soft tissues. With its high contrast resolution, it provides a better study of the locoregional extension of the lesion and its relations with the nerve and vascular pedicles, while providing a meticulous analysis of the cyst walls [3,4,7,8]. In soft tissues, the hydatid cyst typically contains a number of daughter vesicles stemming from the proliferation of the endocyst (proligerous membrane), giving an aspect of a cyst within a cyst. These daughter vesicles show a clear hypointense signal compared to the rest of the cystic fluid on T1-weighted sequences and show a hypo- or hyperintense signal on T2-weighted images depending on whether scolex are present [5,9]. These signs are perfectly illustrated in our observation. However, these daughter vesicles are only present in 30% of cases [2,9].

The pericystic hypointense radiolucent line, clearer on the T2-weighted images, demonstrates the hydatid or endocyst membrane and a second collagen-rich membrane resulting from the reaction of the host to the parasite infection, which is the pericyst [5,9]. Detachment of the
proligerous membrane is demonstrated by a linear or ribbon-like hypointense signal inside the cyst, corresponding to the serpent sign. It shows the involution of the hydatid cyst and illustrates the detached and collapsed parasite membrane [2,5].

After injection of gadolinium, the cysts can present moderate peripheral enhancement because of the pericyst's vascularization. However, with cyst superinfection, greater enhancement is noted related to the hypervascularization of the pericyst as well as the acute inflammation of the surrounding soft tissues [1,2].

Ultrasound- or CT-guided puncture biopsy can be useful in diagnosing doubtful and atypical cases while running no major risks according to several authors [3,5,6]. However, the results of the microscopic analyses of fine-needle biopsy are not always conclusive [10].

Treatment of muscular echinococcosis is surgical. The first-choice technique is pericystectomy, removing the entire cyst without breaking through the wall. Intraoperative precautionary measures using fields soaked with hypertonic saline placed on the wound edges to prevent local dissemination of the scolex [6,10,11]. Yet, excision of hydatid cysts of the soft tissues can sometimes be problematic because of the absence of cleavage lines, particularly when the cyst is infected, and adherence to the blood vessels and nerves, which can be particularly tight, making complete excision difficult [4].

For some authors, percutaneous techniques using puncture, aspiration, alcohol or sclerosing product injection, and reaspiration or percutaneous drainage with injection but no reaspiration are a well-validated alternative to surgical excision for selected patients [10,11].

The value of medical treatment with benzimidazole derivatives (albendazole) in single locations of the locomotor system is still being debated in view of their poor dissemination in the cystic fluid [11]. This treatment is reserved for inoperable cases or as a complement to surgery when the cyst is complicated by rupture [1,6—10].

**Conclusion**

The hydatid cyst of the soft tissues is a rare, slowly developing tumor with local extension. This diagnosis must be considered, particularly in subjects from highly endemic countries. MRI is the most useful imaging tool in hydatid disease of the soft tissues whenever ultrasound or CT do not sufficiently characterize the hydatid cyst and to detail its involvement with the surrounding structures. Pericystectomy is the first-line treatment, but the best means to control hydatid disease, whatever its location, remains prevention.

**Conflicts of interest**

None.

**References**