Case report

An unusual presentation of hydatid cyst: A tight mass a case report with a literature review

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A B S T R A C T
Hydatidosis is a worldwide infectious disease caused by the larval form of a parasitic tape-worm of helminths affecting mainly the liver and lungs in cattle- and sheep-raising regions. Muscle localization remains extremely rare and its diagnosis is sometimes challenging. We report the case of a 45-year-old man presenting with a swelling of the left thigh gradually increasing in size. Clinical examination was non-specific, sonographic evaluation suggested the diagnosis, and the patient underwent successful surgical treatment. Intramuscular hydatid cyst is an uncommon disease even in endemic countries, usually confused with other lesions which is a source of delayed treatment and fatal complications. MRI is the gold standard allowing diagnosis and providing the characteristic aspects which are of major importance for optimal management.

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Introduction

Hydatidosis is a global parasitic disease caused by accidental contamination by a cestode of the Echinococcus granulosus type. Infestation occurs either directly by ingestion of the taeniae eggs contained in the feces of dogs, considered the definitive hosts or through the consumption of contaminated food such as sheep offal. It represents a public health issue in the countries where it is rife. However, the resurgence of immigration led to increasing new cases reported by developed countries. Muscle location is uncommon even in endemic countries [1]. The preoperative radiological diagnosis, provided by ultrasound and MRI is of utmost importance to take precautions and avoid certain procedures that could cause fatal complications.

Clinical Presentation

A 45-year-old man, presented with swelling of the left thigh that had been increasing in volume for 5 months and
Fig. 1 – Axial T2-weighted image of the thighs: multiples well defined cystic lesions of the left vastus lateralis muscle, surrounded by a hy-pointense rim, and containing hyper-intense daughter cysts (arrows).

Fig. 2 – Axial T2-weighted image of the thighs: multiloculated cystic lesions of the left vastus lateralis muscle, surrounded by a hypointense rim (arrows).
becoming progressively more painful. The patient had no past clinical history. The physical examination revealed no fever and no pulmonary, digestive or cutaneous symptoms. The standard laboratory tests showed no abnormalities.

Ultrasound monitoring of the left thigh revealed a large multilocular cyst, measuring 15 cm in diameter and 21 cm in length. MRI demonstrated multiple well-defined hyper signal T2 lesions (Figs. 1 and 2), within the left vastus lateralis muscle, containing hyper-intense daughter cysts (Figs. 3 and 4) and surrounded by a hypointense rim that enhanced intensely (Fig. 5). No abnormal bone marrow signal intensity (Figs. 1 and 2), evidence of fracture or bone edema was seen (Fig. 2). Femoral neuro-vascular bundle and sciatic nerve appeared intact.

This picture raised the suspicion of a hydatid cyst, although no high risk of echinococcosis contamination was evident. The diagnostic was confirmed by serological testing with a positive ELISA and Western blot test.

A pericystectomy considering hydatid cyst as a provisional diagnosis was chosen as treatment covered by 400 mg of Albendazole per day for 10 days before and 3 weeks after.

**Discussion**

Hydatidosis is a disease that remains endemic in many countries in Asia, Africa, Middle East, Mediterranean and South America [2]. It is an anthropo-zoonosis fed by illegal slaughter of animals, failure to treat large numbers of stray dogs, and poor hygienic conditions as well as the lack of awareness. The usual localization is often hepatic or pulmonary. Musculoskeletal involvement as a primary location, is rarely seen even in endemic countries with a frequency that does not exceed 5% [2]. This low rate is due to the liver detoxification function, to muscle contractions preventing larve fixation, and to the secretion of lactic acid with a toxic action. Among the reported muscle locations, proximal muscles seem to be the most affected probably due to a richer vascularization [3,4].

Clinical expression depends on the site, the effect on adjacent structures and complications. Initially asymptomatic, the mode of revelation is most often a painless swelling of the soft tissues. This slow evolution can suggest a myositis or a calcified hematoma. However, complications of superinfection, nerve compression, rupture and allergic reactions lead to an

Fig. 3 - Sagittal T2-weighted images of the left thigh: intramuscular extensive multivesicular cystic lesion (arrows) with multiloculated anterior hyperintense cystic lesions (asterix).
inflammatory expression which can simulate an abscess or a malignant tumor, a source of diagnostic delay [1].

During muscle invasion, biology reveals occasionally a non-specific eosinophilia and serology is inconstantly positive with many false positives up to 80% of cases [5].

Imaging techniques are essential to raise the possibility of hydatid disease and to prevent inappropriate biopsy or aspiration. Imaging findings allow one to take intraoperative precautions in order to prevent any cracking or rupture causing dissemination which may result in severe anaphylactic reactions [6]. Ultrasound is a fundamental examination that determines the site and the echostructure of the swelling and allows one to classify it according to Gharbi’s staging. The anechoic cystic appearance with posterior acoustic enhancement, membranes detachment and daughter vesicles, is characteristic. Cystic sludge may have a pseudo-tissue aspect hypo or finely-echogenic. Atypical mixed or pseudo-tumor forms with or without rounded images remain difficult to characterize on ultrasound. The same difficulties can be encountered on CT imaging. Indeed, CT demonstrates associated bone lesions and calcifications with daughter vesicles that are rarer in hydatid muscle cyst compared to the hepatic [6]. Thus, as CT is facing the same technical problems as ultrasound, and does not provide additional information in atypical cases, MRI is the imaging of choice for the diagnosis of muscular hydatid cyst. The high resolution and multiplanar characterization allows evaluation of locoregional extension, vasculo-nervous relationships, analyzes the wall, and assesses lesions vitality. The typical appearance is that of multiple daughter vesicles, resulting from the proliferation of the proligorous membrane or endocyst producing an aspect of “intracystic cysts”. These daughter vesicles appear hypointense in T1 and T2 sequences when they are not viable, and in hypersignal T2 when they contain scolex. The cyst has hypointense appearance on T1 sequences and an intense hypersignal on T2 sequences. Its periphery describes a characteristic aspect called "Rim-sign" formed by the contrast of the juxtaposition of the fibrous endocyst which is hypointense T2, without enhancement and the pericyst, rich in collagen, showing a high T2 signal, and enhancing following injection of Gadolinium. The enhancement is moderate as it represents the vascularization of the pericyst which is a reaction of the host to the parasitic infection. The contrast enhancement becomes more important in case of superinfection secondary to inflammation and edema of adjacent soft tissues. This aspect is considered unusual because it is absent in other hydatid localizations, inconspicuous at CT-scan, and considered as a diagnostic argument [7].

Thus, the multi-planar and multi-sequence approach of MRI provides vital information for the treatment of muscle hydatid cyst that is primarily surgical. The planning of the
surgical act must be rigorous, with protection of the operating field by a saline-hypertonic-scoliodical solution or hydrogen peroxide to prevent recurrence. The monobloc excision of the pericyst is the ideal treatment that should be started whenever possible. Medical management is based on imidazole derivatives, prescribed for inoperable cases or in association to surgery in some situations [8].

**Conclusion**

MRI is the examination of choice for the diagnosis and the characterization of muscular hydatid cyst. It is paramount to avoid complications of heavy morbidity that are quite common if the diagnosis is not reached on imaging studies. That diagnosis should be suggested in any patient with otherwise unexplained swollen soft tissue in endemic areas. Hygiene measures, regulated slaughter, treatment of stray dogs, and awareness remain the best prevention tools.

**REFERENCES**

[1] Safoleas M, Nikiteas N, Stamatakos M, Safoleas C, Manti CH, Revenas C, et al. Echinococcal cyst of the subcutaneous tissue: a rare case report. Parasitol Int 2008;57(2):236–8.

[2] Jan Z, Zeb S, Shoaib A, Ullah K, Muslim M, Anjum H, et al. Hydatid cyst involving right pectoralis major muscle: a case report. Int J Surg Case Rep 2019;58:54–6.

[3] Tuna S, Duymus TM, Yanik HS, Durakbasa MO, Mutlu S, Erdem S. Hydatid cyst of biceps brachii associated with peripheral neuropathy. Int J Surg Case Rep 2015;8:150–3.

[4] Tekin R, Avci A, Tekin RC, Gem M, Cevik R. Hydatid cysts in muscles: clinical manifestations, diagnosis, and management of this atypical presentation. Rev Soc Bras Med Trop 2015;48(5):594–8.
[5] Lamine A, Fikry T, Zryouil B. L’hydatidose primitive des muscles périphériques. A propos de 7 cas. Acta Orthop Belg 1993;59:184–7.

[6] Thursky K, Torresi J. Primary muscle hydatidosis of the thigh: management of a complicated case with combination adjunctive albendazole and praziquantel chemotherapy. Clinical Infectious Diseases 2001;32(3):e65–e68.

[7] Alouini Mekki R, Mhiri Souei M, Allani M, Bahri M, Arifa N, Jemni Gharbi H, et al. Hydatid cyst of soft tissues: contribution of MRI (About three observations). J Radiol 2005;86(4):421–5.

[8] Mseddi M, Mtaoumi M, Dahmene J, Ben Hamida R, Siala A, Moula T, et al. Muscle hydatid cyst. J Ortho Restor Motor Appl Surg mai 2005;91(3):267–71.