

Case series

Primary hydatid cyst of the thigh: about four cases



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Abstract

The hydatid echinococcosis is preferentially localized in the liver. Muscle localization is very rare, representing less than 1% of the locations of human hydatid echinococcosis. The clinical symptomatology is insidious and not very suggestive, causing a frequent diagnostic delay. The diagnosis is confirmed by imaging: ultrasonography and/or magnetic resonance imaging. The curative treatment remains however surgical. We report a retrospective study over a period of 3 years including 4 cases of primary hydatid cysts of the thigh at the Service of Traumatology-Orthopedics B4 of Hassan II CHU all treated surgically.

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Introduction

The hydatid cyst or hydatidosis is an anthroponosis due to the larval form development in humans of tenia (*Echinococcus granulosus*). It is endemic and constitutes a real public health problem in Morocco. The preferred locations for human echinococcosis are the liver which account for 60% of cases. Other sites of development are the lung (20%), and less frequently the spleen, heart, kidney, central nervous system, bone, and soft tissue [1,2]. The muscular involvement of hydatidosis is an uncommon event, representing less than 1% of human echinococcosis localizations [3,4], because the presence of lactic acid is an unfavorable environment for its growth. The clinical symptomatology is insidious and not very suggestive, causing a frequent diagnosis delay. The hydatidosis muscular can have several imagery aspects that must be known to make the diagnosis preoperatively and prevent the appearance of many serious complications which can be at the origin of diagnostic problem in particular in the absence of pathognomonic radiological signs. The objective of this research article is to point out that, regardless of the site involved, this zoonotic infection should always be considered in the differential diagnosis of any cystic lesion especially in endemic areas.

Methods

This is a retrospective descriptive study spread over a period of 3 years (from June 2015 to June 2018) performed at the Department of Traumatology-Orthopedics B4 of CHU Hassan II of Fez and including 4 cases of primary hydatid cysts of the thigh diagnosed and treated all surgically in our service and we have excluded all cases whose the localization was secondary or in another muscular locations on the rest of the human body. We have identified all the epidemiological,

clinical and radiological data that have been analyzed to better detail the characteristics of these localization. All our patients were operated using the same operative technique consisting on a pericystectomy in one bloc and they were evaluated at 3 weeks in postoperative, then at 6 weeks after results of histopathological examination.

Results

The mean age of our patients was 43 years (range 37 to 55 years) with a sex ratio of 0.33 (1M/3F). All patients who lived in rural areas where raising sheep and contact with dogs was common. Patients consulted for a tumefaction of the soft tissue evolving over more than 3 years which increased in volume progressively (Figure 1). The muscle focus was usually unique and primary. The proximal muscles of the limbs were mainly involved. Ultrasonography was performed for all our four patients and suggested the diagnosis in all cases. The typical characteristic was a liquid echostructure in an endemic context. Magnetic resonance imaging was performed in three of our four patients whose location was deeper (Figure 2). All patients underwent abdominal ultrasound and chest x-ray for other locations and returned negatively. Surgical treatment was used in all cases consisting on a pericystectomy in one bloc (Figure 3) under general anesthesia. It allows us to carry the entire cyst without breaking the wall. In macroscopic aspect, the cystic lesions contents were yellowish, bound and multivesicular (Figure 4). All the parts were sent to the laboratories for histological confirmation. The immediate postoperative course was favorable and no complication was observed. Histopathological examination of the specimen revealed hydatid cyst and showed an inflammatory tissue reaction surrounding "parasite-like cyst". Scoleces were also detected within the surgical specimen. After surgery, all our patients were treated with an additional 1-month course of Albendazole. The resumption of walk was started the day after

the operation and there was no problem observed. At the last follow-up (18 months), clinical, biological and ultrasound surveillance did not indicate local or distant recurrence.

Discussion

Echinococcosis or hydatid disease is a cosmopolite antrozoosis common to humans and several mammal species. The disease results from the development of the larval or hydatid form of canine tenia (*Echinococcus granulosus*) in the organism. Hydatid cysts are rarely found in muscles even in endemic areas. Its frequency varies from 1 to 5% [3,5]. Hexacanth embryos that enter the digestive tract are most often stopped by the filters that make up the liver and lungs. A very small quantity arrives even in the great circulation where they disseminate throughout the body. The rarity of muscle sites is explained by the fact that continual muscular contractions and lactic acid production impede the implantation of scoleces [5-7]. Several factors could explain the exceptional nature of the muscular localizations of hydatid cysts: efficacy of the hepatic and pulmonary barriers, muscular environment unfavorable to the growth of hydatid larvae [8]. A lot of muscles localizations have been described, the involvement predominates in the muscles of the neck, trunk and root of the limbs [3,9,10]. The predominant localization in the proximal muscles of the lower limbs could be explained by the volume of the muscle mass and the richness of its blood supply [8]. The incubation period can last from five to 20 years and symptoms occur several years after exposure [11]. The clinical symptomatology is insidious and not very suggestive, causing a frequent diagnostic delay. The clinical presentation, which is not very specific, can be summed up as a non-inflammatory, painless tumefaction gradually increasing in volume over the years, with preservation of the general state [3,10].

However, a number of cysts are revealed by complications such as nerve compression or infections simulating a hot abscess or a malignant tumor [5,12]. Imaging tools such as ultrasound and computed tomography (CT) reveal a calcified cystic wall and microcalcifications within daughter cysts with a fluid density between the cyst and surrounding organs [13]. Magnetic resonance imaging (MRI) interpretation of hepatic KH is well known, but diagnosis is more difficult in soft tissue because imaging interpretations are not well described [4,13,14]. It allows, thanks to its high resolution in contrast, to better study the locoregional extension of the lesion and its relationship with the vasculonervous pedicles, while offering a careful analysis of the cystic walls [5,6,12]. Various serological tests can help in the diagnosis of echinococcosis. However, serology alone is not sufficient for diagnosis. The sensitivity and specificity of serological tests are not known in patients with hydatid diseases. This serology may be useful for monitoring treatment when it is positive [15]. It is important to establish the diagnosis preoperatively to reduce the risk of anaphylactic shock or dissemination in case of puncture or accidental opening of the cyst during resection. Surgery is required for treatment, ideally by total pericystectomy on one bloc. It allows to carry the entire cyst without breaking the wall. Intraoperative precautionary measures using fields soaked with hypertonic saline on the margins of the operative wound help to prevent local dissemination of scoleces [16,17]. However, exeresis of hydatid cysts from the soft tissues is sometimes problematic: the usual absence of cleavage planes, especially when the cyst is infected and adhesions to the vasculonervous elements that can be particularly tight, makes it difficult to perform a complete resection [5]. Recent years have been marked by the development of percutaneous interventional radiology such as puncture-aspiration-injection and re-aspiration (PAIR), and percutaneous drainage without reaspiration, which has improved the hydatid cyst mortality and morbidity. Medical treatment with imidazoles has little efficacy for the treatment of muscular hydatid disease. However it is indicated in

inoperable cases or in addition to surgery when the cyst has complicated rupture [16].

Conclusion

The hydatid cyst of the soft tissues remains a rare entity, with progressive development. This diagnosis should be considered, especially in subjects from a high-endemic zones. MRI is the most useful imaging tool for hydatid pathology of the soft tissues whenever ultrasound and CT are insufficient to characterize the hydatid cyst and to determine its relationship to vascular and nerve neighborhood structures. Radical perikystectomy without rupture of the cyst is the treatment of choice but the best way to fight against hydatid disease, whatever its localization, is the prevention. The limits of our study were the reduced number of our population sample and the other muscular locations on the rest of the human body.

What is known about the topic

- The hydatidosis is endemic and constitutes a real public health problem;
- The preferred locations for human echinococcosis are the liver.

What this study adds

- To think of the hydatid cyst in the differential diagnosis of any cystic lesion occurring in a subject living in an endemic area;
- The importance of radical treatment by perikystectomy in a one bloc for better results and especially to avoid recurrences.

Competing interests

The authors declare no competing interests.

Authors' contributions

All the authors have read and agreed to the final manuscript.

Figures

Figure 1: hydatid cyst of the adductor compartment: pre-operative aspects

Figure 2: MR Image of adductor compartment's hydatid cyst

Figure 3: perikystectomy in one bloc

Figure 4: perikystectomy in one bloc

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Figure 1: hydatid cyst of the adductor compartment: pre-operative aspects

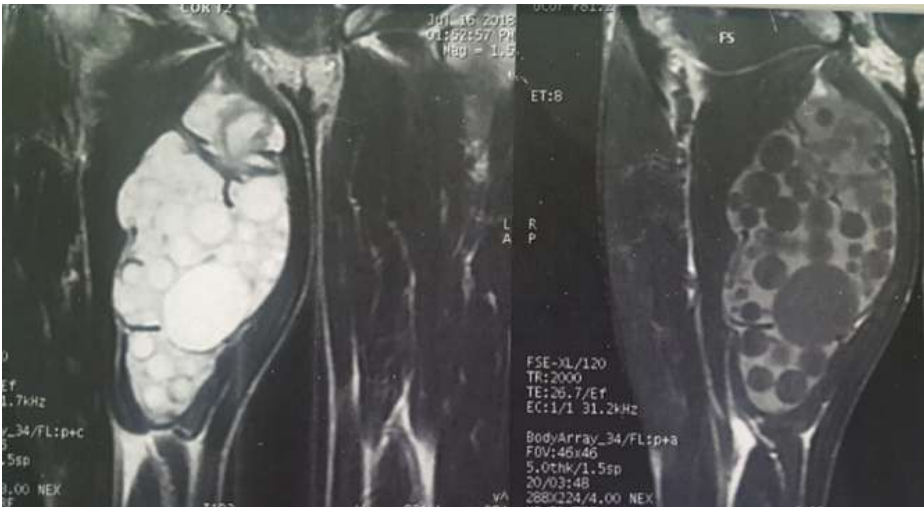


Figure 2: MR Image of adductor compartment's hydatid cyst



Figure 3: pericystectomy in one bloc

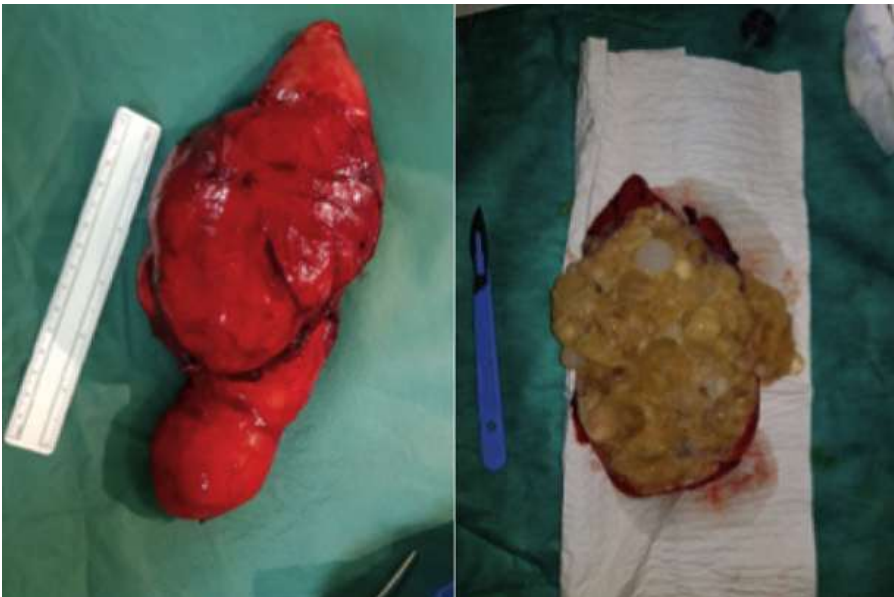


Figure 4: macroscopic aspect of the excised mass