

Hydatid cyst of the gluteal muscle: A case report and literature review

Kyste hydatique du muscle fessier: A propos d'un cas et revue de la littérature

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ABSTRACT

Introduction: Echinococcosis is a relatively widespread anthrozoosis in endemic regions, preferentially affecting the liver and lungs. Although rare, it can sometimes be localized in the muscles. The clinical symptoms are insidious and not very indicative, often leading to a delayed diagnosis. We reported a case of a hydatid cyst located in the gluteal muscle.

Observation: This was a 52-year-old female patient admitted for the appearance of a swelling in the left buttock region, progressively increasing in size. The radiological exam, revealed a large simple cyst originating from the gluteal muscle with purely liquid content. A surgical excision was performed, and the parasitological examination of the hydatid fluid confirmed the diagnosis.

Conclusion: Hydatid cysts in soft tissues are rare, slow-developing tumors with local extension. This diagnosis should be considered, especially in individuals from endemic countries. The treatment is primarily surgical, but the best way to combat hydatid disease, regardless of its location, remains prevention.

Key words: Cyst, Echinococcosis, Endemic, Gluteal muscle

RÉSUMÉ

Introduction: L'échinococcose, est une anthrozoose cosmopolite relativement répandue dans les régions endémiques, touchant préférentiellement le foie et les poumons. Bien que rare, l'échinococcose peut parfois se localiser dans les muscles. La symptomatologie clinique est insidieuse et peu évocatrice, ce qui entraîne souvent un retard diagnostique. Nous avons rapporté un cas de kyste hydatique localisé au niveau du muscle glutéal.

Observation: Il s'agissait d'une patiente âgée de 52 ans, admise pour apparition d'une tuméfaction de la région fessière gauche augmentant progressivement de volume, le bilan radiologique a montré un volumineux kyste simple prenant naissance au niveau du muscle glutéal, et dont le contenu est purement liquidien. Une exérèse chirurgicale a été réalisée et l'examen parasitologique du liquide hydatique a confirmé le diagnostic.

Conclusion: Le kyste hydatique des parties molles est une tumeur rare, à développement lent, et à extension locale. Il faut penser à ce diagnostic, surtout chez des sujets originaires d'un pays à endémie élevée. Le traitement est essentiellement chirurgical, mais le meilleur moyen pour lutter contre la maladie hydatique, quelle que soit sa localisation, reste la prévention.

Mots clés: Echinococcose, Endémie, Kyste, Muscle glutéal

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INTRODUCTION

Echinococcosis, also known as hydatid disease (HC), is a cosmopolitan anthroponosis of humans and certain mammals caused by the development of the larval or hydatid form of a dog tapeworm called echinococcus granulosus (1). It is relatively widespread in endemic regions, notably in the countries of the Mediterranean basin, North Africa and Latin America (1, 2). Although the liver (60%) and lungs (20%) are the most common sites, other organs can also be affected by this parasitosis (2). Muscle localization is rare, even in endemic regions where it does not exceed 3% (1).

Musculoskeletal HCs, particularly those located in the gluteal region, are extremely rare. This rarity is attributed to an unfavorable environment for the parasite in the muscular tissues (3).

Generally, these HCs are presented as a persistent and painful mass in the affected area. It is crucial to consider this diagnosis, especially in patients with a history of contact with dogs and those residing in sheep-raising regions or rural areas (3).

We reported a case of secondary localization of a gluteal muscle HC in a patient previously treated for a pulmonary HC.

OBSERVATION

A 53-year-old female living in the countryside, with a history of contact with dogs, underwent surgery in 2022 for a pulmonary HC. The patient was admitted for consultation in February 2024 due to a swelling in the left gluteal region, which had been progressively increasing in size over the past three years.

Clinical examination on admission revealed a conscious, hemodynamically and respiratory stable, afebrile patient. The patient has a mass in the left gluteal region, asymptomatic at rest, non-inflammatory in appearance, soft in consistency and mobile in relation to the superficial plane. There were no adenopathies, and the rest of the physical exam was unremarkable.

A standard X-ray of the pelvis showed a soft-tissue tumour with no bone lesions or calcifications. The ultrasound revealed a well-defined cystic lesion with anechoic characteristics.

Magnetic resonance imaging (MRI) revealed a voluminous, well-limited, lobulated cystic formation in the left gluteal soft tissues, originating in the gluteus maximus muscle, with purely fluid content, hypodense in T1 and frankly hyperdense in T2, without endocystic tissue septum or bud, measuring 72X80X96 mm in diameter, with peripheral enhancement, making it a stage I of Gharbi's classification (4) (Figure 1).

Faced with a muscular localization of the HC, complete excision of the cystic formation was performed electively, after protective measures had been taken using hydrogen peroxide-soaked drapes. The procedure involved isolation of the cyst, followed by puncture-aspiration which revealed a rock-clear fluid. Next, the proligeral membrane was extracted, followed by abundant irrigation with

hydrogen peroxide. Pericystectomy was performed to remove the entire HC shell with removal of some cyst muscle adhesions, followed by suction drainage of the residual cavity and subcutaneous tissue (Figure 2).



Figure 1. Coronal (a) and axial (b) T1 MRI sections of the left gluteal soft tissue show a cystic formation in the gluteus maximus muscle (arrow). MRI: Magnetic resonance imaging.

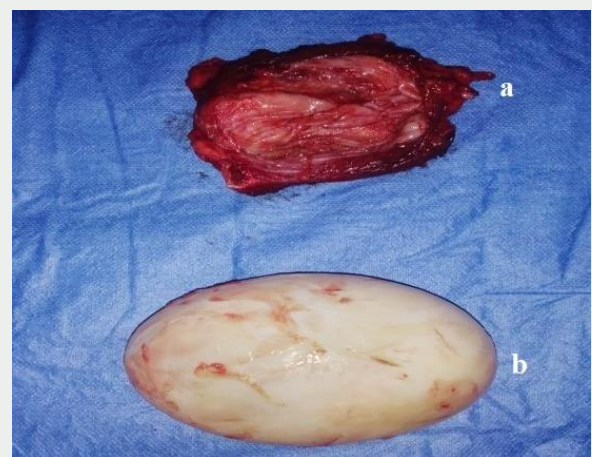


Figure 2. Aspect of a surgical specimen showing a thick peri-cyst (a) and a thin cystic membrane containing intact hydatid fluid (b).

The cyst puncture fluid was subjected to a parasitological study to confirm the diagnosis. To do this, the fluid was divided into three hemolysis tubes, then centrifuged. The resulting sediment was examined under a microscope at X10 and then X40 between slide and coverslip, revealing the presence of scolex and hooks (Figure 3).

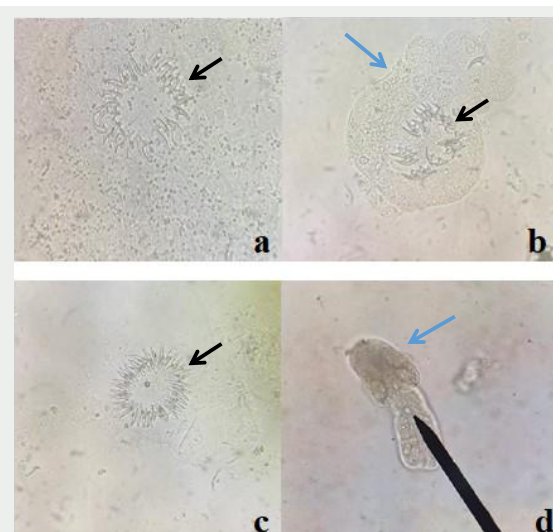


Figure 3. Parasitological examination of puncture fluid showing the presence of hooks (black arrow: a, b and c) and scolex (blue arrow: b and d).

Postoperatively, adjuvant albendazole-based medical treatment was prescribed to prevent recurrence. No evidence of recurrence or complications was seen during follow-up.

DISCUSSION

HC in the gluteal muscles present a diagnostic challenge due to their rarity and similarity to other muscular masses (3).

Hydatidosis is a worldwide disease, affecting both humans and many mammals (5). Dogs, wolves and some jackals serve as definitive hosts (5). These animals contract the infection by ingesting contaminated viscera, such as liver or lungs, from sheep, which act as intermediate hosts (5). Sheep, the main reservoirs of the *Echinococcus granulosus* tapeworm, become infected by grazing on grass soiled by dog faeces, which contain the parasite's eggs (5). Human is only an accidental intermediate host, who can contract the infection either directly from contact with dogs or indirectly through ingestion of contaminated food, thus creating an epidemiological impasse (6, 7).

Hydatidosis affects a wide range of organs in humans, the most common being the liver and lungs (2). However, this parasitosis can also affect other organs, notably the

muscles (2). Muscle hydatidosis is rare, and its frequency is poorly defined (between 1% and 4% of all localizations (2)). Despite its rarity, muscle hydatidosis ranks third after liver and lung ones (2).

Several factors explain the rarity of HC muscular localization (6). First, the hepatic and pulmonary barriers are effective in preventing the migration of hexacanth embryos into the systemic circulation. Second, the muscular environment is not conducive to the growth of hydatid larvae, due to the production of lactic acid, which is toxic to the parasite, and the muscle contractions that prevent its attachment (6, 7).

Muscular HC is generally located in the proximal muscles of the lower limbs, which may be due to the high vascularity at this level favouring the arrival of the tapeworm (8).

The rare cases reported in the literature are generally primary, and may affect various regions such as the upper limbs, vertebrae, diaphragm and psoas muscle (8). Muscle involvement is usually isolated, although in around 8% of cases it may be associated with other localizations (8).

Gluteal localization is exceptional, even in endemic countries, which makes the present observation special. In the literature, sporadic cases of HC in the gluteal muscle have already been described in the Maghreb region and worldwide (Table 1).

Table 1. Reported Cases of Hydatid Cysts in the Gluteal Muscle in North Africa and Worldwide

1 st author (Reference)	Country	Age and sex	Diagnosis	Symptoms	Treatment	Outcome
El Alaoui (3)	Morocco	29; male	MRI and serology	Painful mass	Surgical excision and albendazole	Recovery without recurrence
Moujahid (9)	Morocco	29 and 60 years ; females	CT scan	Swelling	Surgical excision	Recovery
Abdellaoui (10)	Morocco	45 years ; male	CT scan	Painless swelling	Surgical excision	-
Sghaier (11)	Tunisia	41 years; female	MRI and serology	Pain with swelling	Surgical excision	Recovery
Rabhi (12)	Tunisia	82 years; female	MRI / serology	Painful swelling	Surgical excision and medical treatment	Recovery without recurrence
Ben Ismail (13)	Tunisia	41 years; male	CT scan	Functional impairment with pain	Surgical excision and medical treatment	Recovery without recurrence
Niazi (14)	Syria	47 years; male	MRI / Serology	Numbness, sciatica, and foot drop	Surgical excision and medical treatment	Sacrocutaneous fistula
Fahl (15)	Lebanon	38 years; female	CT scan / serology	Painful swelling	Surgical excision and medical treatment	Recovery without recurrence
Iacob (16)	Romania	53 years; female	MRI / Serology	Pain with functional impairment	Surgical excision and medical treatment	Recovery without complications
Adiyeke (17)	Turkey	36 years; female	CT scan	Pain with mass	Surgical excision and medical treatment	Recovery
Flores (18)	Portugal	73 years; female	CT scan / serology	Left lower limb pain	Partial surgical excision and albendazole	Stability of lesions under albendazole
Seyedsadeghi (19)	Iran	29 years; male	CT scan	Progressive painful swelling	Surgical excision and albendazole	Recovery without recurrence
Ferjaoui (20)	India	67 years; female	MRI	Pain with swelling	Medical treatment only (patient refused surgery)	Monitoring every 3 months

CT: computed tomography. MRI: Magnetic resonance imaging.

The clinical manifestations of muscular HC localizations are not specific. They generally begin insidiously, with the appearance of a well-defined, painless swelling of variable size and soft consistency, while preserving the patient's general condition (8). This was the case for our patient. Muscular echinococcosis can also mimic other conditions such as a lipoma, calcified haematoma,

malignant or benign tumour, or even a hot abscess in the case of fissuring or superinfection (8).

Occasionally, the HC may manifest as signs of nerve compression or allergic reactions ranging from a simple skin rash to anaphylactic shock in the event of cyst rupture or puncture (8).

Biologically, high eosinophilia is not systematically present and has no significant diagnostic value, but can help orient the diagnosis (6). Hydatid serology, performed by immunofluorescence and Enzyme-Linked Immuno Assay (ELISA), is positive in around 50% of cases, which can also help confirm the diagnosis (6). It is also useful for monitoring treatment efficacy (6).

Ultrasound is an essential tool in the diagnosis of HCs, with an estimated sensitivity of 96% (6). Ultrasound images reflect the different stages of the disease according to Gharbi's classification (16) and reflect the progressive stage of the disease. Our patient presented a cystic ultrasound image with a purely liquid content, corresponding to type I of Gharbi's classification.

Computed tomography can be useful for assessing bone extension, particularly in pseudotumoral or unusually localized forms (6). However, MRI (16) is considered the examination of choice because of its ability to pinpoint the location and number of cysts, as well as providing a detailed assessment of lesions and their relationship with surrounding vasculonervous structures (16). In addition, MRI is of particular diagnostic interest in showing wall enhancement after gadolinium injection, which helps to exclude other diagnoses such as abscesses and soft-tissue tumours (16). However, the use of MRI should be reserved for cases where ultrasound cannot provide a definite morphological diagnosis (16).

Parasitological examination is of vital importance in confirming the diagnosis of HC, by demonstrating the presence of scolex and/or hooks in the fluid or material extracted from the cyst (4). Moreover, the results of this examination can influence the decision on the drug treatment to be used in addition to surgery (4).

The main treatment for hydatidosis is surgical, as this is the only radical way to confirm the diagnosis and guarantee a complete cure (5). During surgery, it is crucial to protect the surgical margins by using surgical drapes soaked in hypertonic saline or hydrogen peroxide to prevent local dissemination of the scolex (5).

Ideally, the surgical procedure consists of a monobloc excision of the cyst with a total pericystectomy (5, 21). However, when cleavage planes are absent, especially in cases of infection or deep adhesion of the cyst to surrounding vascular-nerve structures, cyst aspiration followed by pericyst resection may be necessary (5). This approach prevents the risk of accidental rupture of the cyst, which could release large numbers of viable scolex or, at worst, cause anaphylactic shock (21).

In the present case, we performed a monobloc surgical excision of the cyst with a total perikystectomy, taking care to observe the principles of protection to avoid rupture of the hydatid membrane at the surgical site.

Medical treatment with albendazole is recommended in conjunction with surgery, both pre- and postoperatively, to prevent recurrence (2). In cases where surgery is not possible, albendazole can be administered alone at high doses to minimize the risk of dissemination (2).

Combination therapies such as albendazole and praziquantel have been shown to be effective, but can have adverse side-effects, particularly on the lymphatic system (7). What's more, these treatments require a

prolonged course of treatment (7).

In a country like Morocco, where hydatidosis is endemic and constitutes a major public health problem (2), the ideal objective is to reinforce preventive measures. This mainly involves combating infestation among definitive and intermediate hosts, as well as raising public awareness and promoting good hygiene practices to reduce the risk of human contamination (22). This includes campaigns for deworming dogs, managing animal populations through veterinary surveillance, vaccination, and education of breeders, as well as the adoption of good hygiene practices such as handwashing and proper meat cooking (22).

CONCLUSION

The muscular location of HCs remains rare even in endemic regions, and is usually isolated. In the present case, the HC was located in the gluteal muscle and was secondary to pulmonary hydatidosis, which is also atypical. The clinical picture of hydatidosis is often nonspecific and may mimic a soft-tissue tumour, underlining the importance of considering this diagnosis according to the epidemiological and clinical context, especially in the presence of a soft-tissue mass. Ultrasound, possibly followed by MRI, are the main exploratory tools for confirming the diagnosis prior to surgery and for avoiding puncture. The main treatment is surgical intervention, ideally aiming for total peri-cystic resection without rupture, or alternatively resection of the protruding dome in case of difficulty.

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