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MUSCULAR HYDATIDOSIS: REPORT OF A RARE CASE IN THE QUADRICEPS IN ALGERIA

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Abstract

Hydatid cyst (HC), caused by *Echinococcus granulosus*, is a significant public health issue in livestock farming areas. While common in the liver and lungs, primary skeletal muscle involvement is rare (1-5%). This report details an exceptional case of primary muscular HC in the quadriceps.

We present the case of a 14-year-old girl from a rural area with a several-week history of pain and swelling in her left thigh. Clinical examination revealed a firm, tender mass. Ultrasound showed a 60 x 38 mm intramuscular anechoic cystic lesion in the quadriceps (Gharbi Type I), suggestive of HC. Staging workup revealed no other hydatid localizations. Total pericystectomy was performed without rupture, complemented by perioperative Albendazole (10-15 mg/kg/day for 1 month pre- and 3 months post-surgery). Histopathological examination confirmed a muscular hydatid cyst, showing a thick, and laminated, acellular membrane characteristic of the hydatid cuticle. The patient's 18-month postoperative follow-up was favorable, with no recurrence.

Primary muscular HC of the quadriceps is an exceedingly rare diagnosis that should be considered for unexplained soft tissue masses, especially in patients from endemic regions. Imaging, particularly ultrasound and MRI, is crucial for suspicion, with histopathology providing definitive confirmation. Complete surgical resection (pericystectomy) combined with perioperative Albendazole is the standard treatment for favorable outcomes. Prevention key remains in controlling hydatidosis, treatment of dogs mainly pets.

Keywords: Algeria, Echinococcosis, Hydatidosis, Cestod parasite, Soft tissue mass, Treatment.

Introduction

Hydatid cyst (HC), or human hydatidosis, is a cosmopolitan parasitic infection caused by the cestode *Echinococcus granulosus* larval development, as a global public health problem, mainly in countries raising sheep farms (Agoumi, 2003). Countries includes most of the Middle East and North Africa, is one of the most ancient foci of the domestic cycle of HC and is recognized as one of the major hotspots of CE. There are 22 countries in the EMRO, where about 688 million people are living at risk of HC (Borhani *et al*, 2020). Humans are accidental intermediate hosts in the HC with the dog as the definitive host and the sheep (mainly) as the usual intermediate host. Human infection occurs through ingestion of parasite eggs present in the environment contaminated by canine feces, or by direct contact with infected dogs. After ingestion, the hexacanth embryo released in the intestine crosses the mucosal barrier and enters the portal circulation. The liver acts as a first filter, explaining its predominant localization of approximately 60% cases. Lung constitutes the second filter (approximately 30%). More rarely (about 10% cases), the embryo can bypass these two filters and reach the systemic circulation localizing in any organ or tissue (Amar *et al*, 2010). Other locations occasionally were in the spleen, kidney, pancreas, intraperitoneal space, heart, ovaries, prostate, incision scar, retroperitoneal space, thyroid, vesica urinaria, orbita, head and neck, chest wall, brain, musculoskeletal and soft tissue, breast, and axillary space (Yagmur and Akbulut, 2012).

Skeletal muscle involvement is a rare, but even exceptional, localization, with its frequency estimated between 1 and 5% of all human hydatidosis (Jerbi Omezzine *et al*, 2010). Mazyad *et al.* (1998) reported a spinal cord HC. This rarity is attributed to several factors: muscle contractility and local lac-

tic acid production are thought to create an environment unfavorable for the implantation and growth of the hydatid larva (Soufi *et al,* 2010). Muscular HC is most often primary and isolated. Clinically, it manifests insidiously as a painless or slightly painful swelling, slow-growing, which can mimic a benign or malignant soft tissue tumor (Abdellaoui and Bouabdallah, 2014). Symptoms appeared when the cyst was a significant size and pressed on the infected organ (Symeonidis *et al,* 2013). Imaging, particularly ultrasound and magnetic resonance imaging (MRI), plays a key role in preoperative diagnostic orientation (Garcia-Diez *et al,* 2000), while histopathological examination of the surgical resection specimen provides definitive confirmation.

The objective of this work is to report a rare case of primary muscular hydatid cyst located in the quadriceps muscle of an adolescent girl, and to discuss, through a literature review, the diagnostic and therapeutic particularities of this unusual localization.

Case presentation: A 14-year-old female patient without significant medical history, originating from a rural area was presented with pain associated with swelling of the left thigh evolving over several weeks. Clinical examination showed a firm, slightly mobile mass, tender on palpation of the left thigh anterosuperior aspect.

A soft tissue ultrasound showed an anechoic fluid collection (60x38mm), intramuscularly within the quadriceps muscle. This lesion was avascular on color Doppler mode and exerted a slight mass effect on adjacent structures. The ultrasound appearance was suggestive of a hydatid cyst (Type I of Gharbi classification). A staging workup including a chest X-ray and an abdominopelvic ultrasound didn't show any other hydatid localization.

Given the suspicion of an isolated muscular hydatid cyst, surgical intervention was indicated. A total pericystectomy, removing the cyst without rupture was performed under general anesthesia. Perioperative treatment with Albendazole® (10-15mg/kg/day) was given for a month before and 3 months after surgery or Mebendazole® was 40 to 50mg/kg of body weight per day for at least 3 to 6 months to prevent any recurrence (Kern *et al,* 2001). Bone hydatid cysts were less susceptible to chemotherapy and surgical treatment was indicated (Mazyad *et al,* 1999). The gross examination of surgical specimens showed a whitish cystic structure measured 5.5x3.5cm adjacent to fragments of muscle tissue. Specimen was fixed in 10% buffered formalin. After paraffin embedded, for histological sections and stained with H & E (Troyer and Babich, 1981). Microscopic examination revealed a cystic structure whose wall consisted of a thick, laminated, eosinophilic, acellular membrane, characteristic of the hydatid cuticle (laminated layer).

Informed consent was obtained from the legal guardians for this girl and associated images, ensuring patient anonymity. The surgical specimen was sent to Pathology Laboratory for kindly diagnostic confirmation.

Results

This structure was bordered by striated skeletal muscle tissue without signs of malignancy or major specific inflammatory reaction. The germinal layer (proligerous membrane) and scolices were not clearly identified on the examined sections. These findings confirmed the diagnosis of muscular hydatid cyst.

Postoperative follow-up at 18 months was favorable, with no clinical or ultrasound signs of local or distant recurrence. Details were given in figures (1 & 2).

Discussion

Muscular hydatidosis is a rare entity, accounting for only 1 to 5% of all localizations of hydatid disease (3), even in endemic areas (6, 7). The case showed localization within the quadriceps muscle, which is considered even more exceptional compared to other muscles such as the psoas, diaphragm, or limb muscles (Durakbasa *et al,* 2007). The rarity of this localization is explained by local factors unfavorable to the development

of the hexacanth embryo: the high vascularization of the muscle could paradoxically facilitate its rapid passage, while repeated muscle contractions and the biochemical environment rich in lactic acid could inhibit its implantation and growth (4).

Epidemiologically, hydatidosis can affect all ages, with a peak frequency in young adults. A slight female predominance is sometimes reported, possibly related to greater involvement of women in agricultural activities and contact with dogs in some endemic rural areas (Rafaoui *et al,* 2016). Patient was an adolescent girl from a rural background, fits this epidemiological profile. The clinical presentation of muscular HC was often misleading. Development is slow and insidious, with the lesion remaining asymptomatic for a long time. Revealing signs are usually progressive swelling, functional discomfort, or pain, as in our case, occurring when the cyst reaches a considerable size (5). Complications can arise: compression of adjacent neurovascular structures (2), bacterial superinfection (El Malki *et al,* 2007), fissuring or spontaneous or traumatic rupture which can lead to a local inflammatory reaction, or even a potentially fatal anaphylactic shock (Guthrie *et al,* 1996).

Diagnosis relies heavily on imaging. Standard radiography is of little value except in cases of old calcified cysts or to rule out associated bone involvement (Alouini Mekki *et al,* 2005). Ultrasound is a key examination, inexpensive and non-invasive. It typically shows an anechoic fluid collection (Gharbi type I), sometimes with membrane detachment or the presence of daughter vesicles (types II, III), but atypical appearances (pseudo-tumoral, solid, or mixed) can exist, making diagnosis difficult (Veller *et al,* 1988). Computed Tomography (CT) can specify relationships and look for parietal calcifications (Ben M'Rad *et al,* 1998). MRI is often considered the most effective examination for studying soft tissue localizations. It specifies the cystic nature, morphology (possible multiloculated appearance), extent, and relationships with surrounding structures (vessels, nerves), which is essential for surgical planning (Memis *et al,* 1999). Peripheral enhancement after Gadolinium injection may be observed. In the present case, ultrasound strongly suggested the diagnosis, which was confirmed by surgery and histopathology.

Biological tests are of limited value. Hypereosinophilia is inconsistent. Hydatid serology (ELISA, IHA) has limited sensitivity for extrahepatic and extrapulmonary localizations, particularly muscular ones, and is often negative in the absence of fissuring (El Moussaoui *et al,* 1997). However, it is useful for post-treatment follow-up: a progressive negativation indicated treatment effectiveness, while a re-increase in titers may suggest recurrence (Argy *et al,* 2018).

The differential diagnosis for muscular HC is soft tissue tumors, whether benign (lipoma, synovial cyst, encapsulated hematoma, cold abscess) or malignant (sarcomas), origin from an endemic area should systematically raise the suspicion of hydatidosis when faced with an unexplained soft tissue mass (Mayerson *et al,* 2014).

Treatment of choice for muscular hydatid cyst is surgical (Alaoui *et al,* 2016). The aim is the complete excision of the cyst without rupturing its wall (pericystectomy) to avoid parasitic seeding and local recurrence (Ait lahcen *et al,* 2017). Surgery is the treatment of choice for pulmonary hydatid patients and Staging surgery is a must for patient with bilateral pulmonary cyst or pulmonary and liver cysts (El-Sayed *et al,* 2020). But, if total pericystectomy is not possible due to adhesions to noble structures, cystectomy after inactivation of the parasitic content can be done, but with a higher risk of recurrence. The use of scolicidal solutions (hypertonic saline, hydrogen peroxide) must be cautious in muscle due to the risk of tissue necrosis.

Perioperative medical treatment with benzimidazole derivatives (mainly Albendazole) is widely recommended (Keshmiri *et al,* 2001). Administered preoperatively, it aims

to reduce the viability of scolices and intracystic pressure, reducing the risk of dissemination in case of intraoperative rupture. Postoperatively, it helps treat possible microscopic contamination and prevent recurrences. Medical treatment alone is reserved for inoperable patients, multiple cysts, or in cases of recurrence not amenable to surgery (McManus *et al*, 2012). Percutaneous treatment (PAIR: Puncture, Aspiration, Injection, Re-aspirat-ion), well-established for certain liver cysts (Thaunat and Priollet, 2004), is generally not recommended for muscular hydatidosis due to the risks of fistulization and dissemination into tissue planes (Ormeci *et al*, 2007).

Prolonged follow-up (clinical, imaging, and serological) was essential to detect possible local recurrence or the appearance of other localizations (Rask and Lattig, 1970). Serological monitoring is recommended every 3 to 6 months for at least 2 to 3 years.

Conclusion

Muscular hydatid cyst is a rare manifestation of the hydatid disease, with the quadriceps muscle being a particularly exceptional location. Although benign, this pathology can pose diagnostic challenges by mimicking a soft tissue tumor and cause complications through compression or rupture. Diagnosis of a patient from an endemic area presenting with an unexplained muscle mass, imaging (ultrasound and MRI) is essential, and confirmed by histopathological analysis. Treatme nt is by complete surgical excision (pericystectomy), often combined with perioperative medical treatment with Albendazole to prevent recurrences. Serology has a limited role in diagnosis, but useful for post-therapeutic follow-up. Prevention, based on hygiene, veterinary control, and health education of populations at risk, remains the most effective measure to combat this parasitosis.

Ethical approval: Ethical approval was not required for a single retrospective case report involving standard clinical practice according to institutional guidelines. Informed consent to participate in this case report and for the procedures performed was obtained from the legal guardians of the patient. The study was conducted in accordance with the ethical principles of the Declaration of Helsinki. *(Note: J'ai légèrement généralisé la partie sur l'approbation éthique pour éviter de nommer l'institution ici, afin de maintenir l'anonymat du fichier principal. Le nom de l'institution sera sur la Title Page).*

Publication consent: The written informed consent for publication of this case and any images was obtained from the patient's legal guardians. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Conflict of interest: Author declared that he neither has any conflict of interest nor received any funds.

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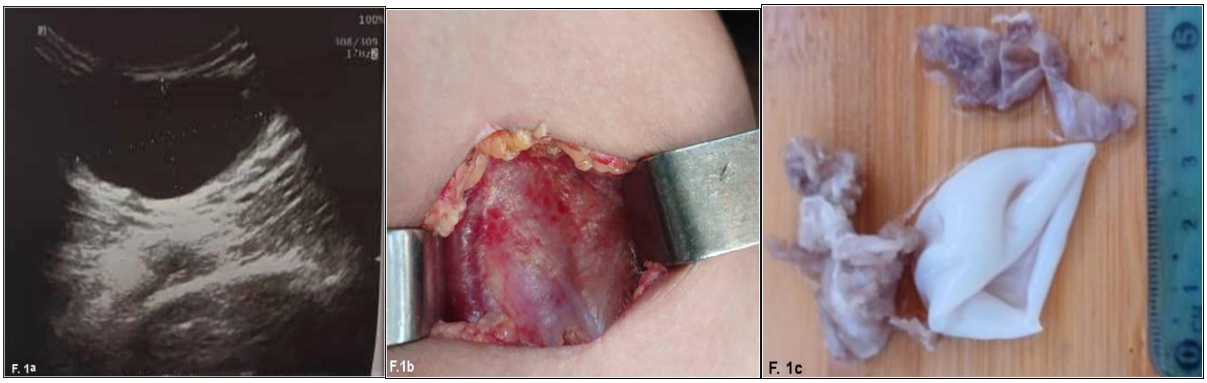
Explanation of figures

Fig. 1: (A) Ultrasound of left thigh showed anechoic intramuscular cystic lesion. (B) Intraoperative view showed pericystectomy of hydatid cyst. (C) Gross surgical specimen after fixation, showed opened cyst and wall.

Fig. 2: Microscopic appearance of HC: (A-C) Muscle (blue arrow) contained a cystic formation with no visible epithelium; a fibrous capsule is present (orange arrow). (D) Evidence of a nuclear eosinophilic layers corresponding to hydatid lamellae, or cuticle (black arrow).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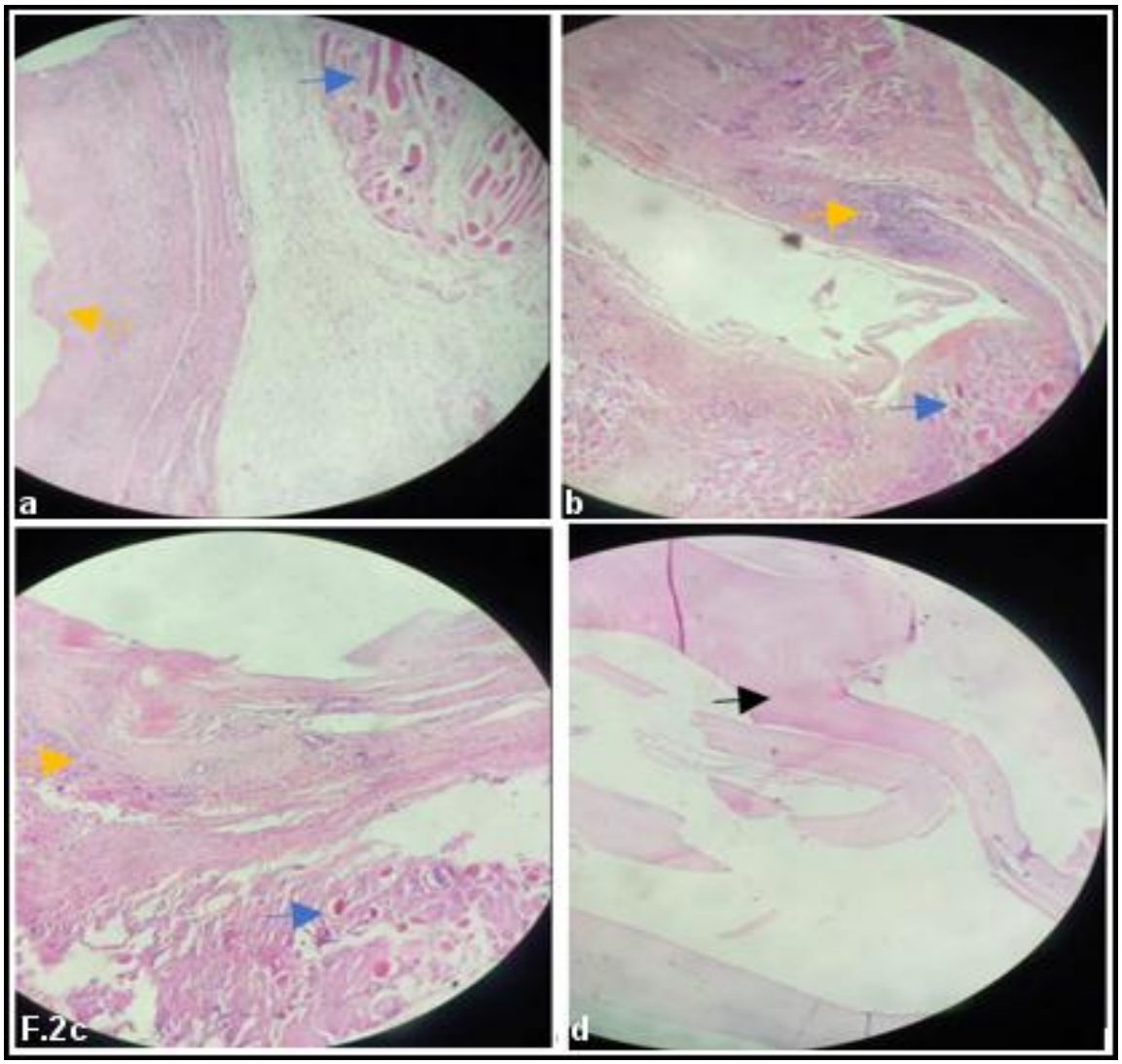
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