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Case Report

Hydatid Cyst, Solitary Primitive with Atypical and Unusual Muscular Localization: Report of 02 Cases

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Abstract: ***Introduction:*** Hydatidosis is a cosmopolitan parasitic infection caused by the larval stage of Echinococcus granulosus. While the liver and lungs are the most affected organs, involvement of other sites is less frequent. Hydatidosis developing within the subcutaneous or muscular tissue is an exceedingly rare occurrence. ***Materials and Methods:*** **Case 1:** A 56-year-old female presented with a left basithoracic tumor infiltrating between the internal and external oblique muscles on MRI. Hydatid serology were negative, but the diagnosis of muscular hydatidosis was strongly suspected based on clinical and radiological evidence. The patient underwent total pericystectomy, with no recurrence at one-year follow-up. **Case 2:** A 85-year-old female was referred to our hospital for an asymptomatic soft tissue tumor located in the root of the right thigh, progressing for 3 years and measuring 11cm. Ultrasound and MRI, revealed an encapsulated lesion exhibiting both cystic and tissue components with multiple compartments. The patient underwent complete surgical excision, and histopathological-exam confirmed the diagnosis. No postoperative medical treatment was administered, and there was no recurrence after a 4-year follow-up. ***Discussion:*** The frequency of muscular hydatid cysts varies from 1 to 5.4% of all hydatid locations. Muscular hydatid cysts pose challenges in detection through serological tests, but a positive result provides diagnostic certainty. Ultrasound is the imaging modality of choice, MRI is recommended for cases where uncertainty persists and for local-regional evaluation. The primary treatment for muscular hydatidosis is surgical. The use of medical treatment with Albendazole remains highly controversial. Long-term patient follow-up is crucial to detect any local or distant recurrence. ***Conclusion:*** In the presence of any subcutaneous or muscular mass in a patient residing or having resided in an endemic area, the diagnosis of muscular hydatid cyst should be considered, despite its rarity. Imaging and serology should be performed and surgery remains the main treatment.

Keywords: Hydatid, Cyst, intramuscular, thigh, ecchinococcosus.

INTRODUCTION

Hydatidosis is a worldwide parasitic infection caused by the larval stage of Echinococcus granulosus. It is particularly prevalent in endemic regions, notably in countries around the Mediterranean basin, North Africa, and Latin America [1, 2]. In Tunisia, its prevalence is estimated at approximately 10.4% [3].

The definitive host for the parasite is the dog, while intermediate hosts are represented by goats. Humans, on the other hand, serve as accidental dead-end hosts by inadvertently ingesting contaminated food.

The liver and lungs are the most commonly affected organs, with other organs being less frequently involved. Cases of hydatidosis developing in subcutaneous or muscular tissues are rare.

CASE REPORT 1

An 85-year-old female patient, with an unremarkable medical history, hailing from a rural area with a history of dog contact, was referred to our attention due to a soft tissue tumor in the root of her right thigh. The tumor had been growing for three years and

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was causing significant discomfort during daily activities and dressing.

Clinical examination revealed a freely mobile mass measuring 11cm in its largest dimension in the supero-external quadrant of the right thigh. The mass was non-tender, lacked adherence to the deep tissue plane, and did not exhibit signs of inflammation (Figure 1). There were no palpable lymph nodes, and the rest of the physical examination was unremarkable. Standard X-rays showed no abnormalities in the bone structure, and further evaluation using ultrasound and MRI indicated an encapsulated lesion with a combination of cystic and solid components containing multiple locules (Figure 2). Hydatid serology yielded negative results.

The patient underwent complete surgical excision (Figure 3) with uncomplicated postoperative recovery. Histological examination confirmed the diagnosis of hydatidosis, and no postoperative medical treatment was necessary. Follow-up over a period of four years did not reveal any local or visceral recurrence.

Fig 1: Mass of the the supero-external quadrant of the right thigh

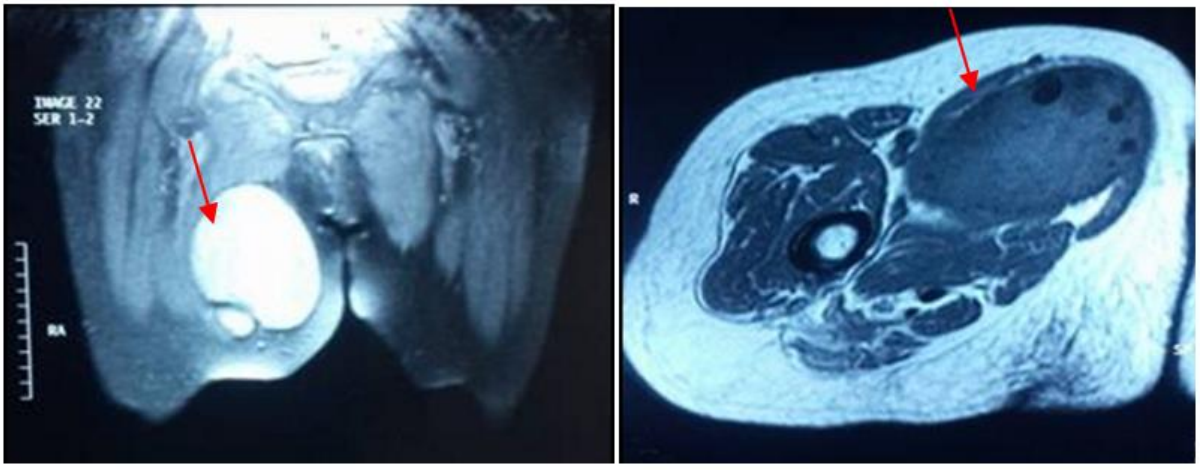

Fig 2: Coronal (A) and axial (B) MRI sections showing an encapsulated process with a dual cystic and solid component containing multiple locules

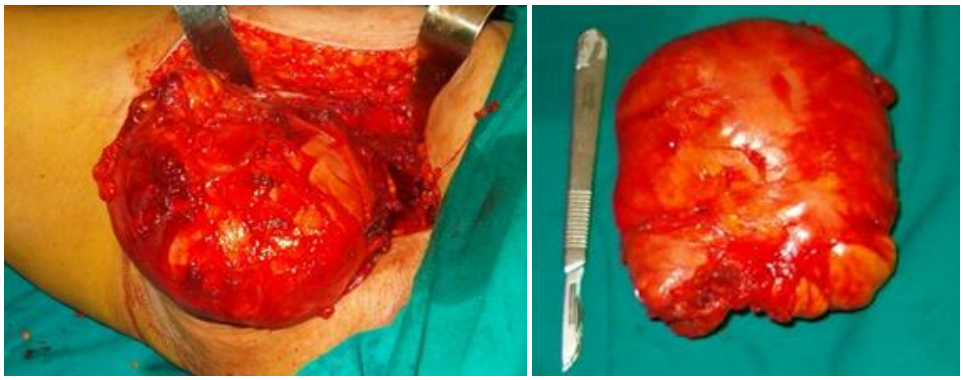

Fig 3: Macroscopic appearance of the resected Hydatid Cyst

CASE REPORT 2

A 56-year-old female patient, with no significant medical history and no contact with dogs, presented with a left basithoracic tumor that had been progressively increasing in size over two years.

Clinical examination showed an immobile mass measuring 4 cm in its largest dimension at the level of the left basithoracic region, related to the 8-10 left ribs. The mass was non-tender and lacked signs of inflammation or respiratory compromise. Ultrasound and MRI revealed a left basithoracic cystic formation infiltrating between the internal and external oblique muscles, demonstrating hypointensity on T1-weighted images and hyperintensity on T2-weighted images (Figure 4). Hydatid serology yielded negative results.

The patient underwent total pericystectomy (Figure 5) with uncomplicated postoperative recovery. Histological examination confirmed the diagnosis of hydatidosis. Follow-up over a period of three years did not reveal any local or visceral recurrence.

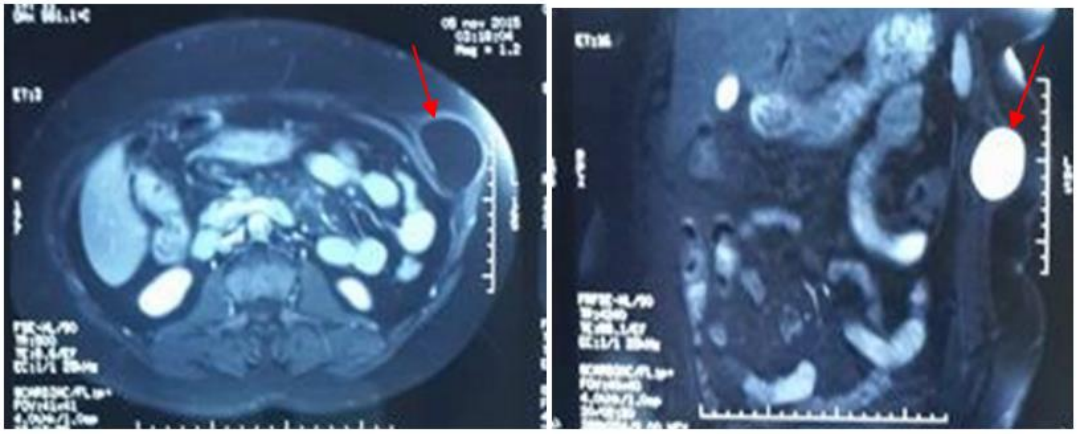

Fig 4: MRI sections suggesting a left basithoracic cystic formation infiltrating between the internal and external oblique muscles with hypointensity on T1-weighted images and hyperintensity on T2-weighted images

Fig 5: Macroscopic aspect of the Hydatid Cyst after total pericystectomy

DISCUSSION

Echinococcosis is classified as an anthropozoonosis, where the adult worm typically resides in the intestine of the definitive host (DH). Thousands of eggs are released daily, and upon contact with the intermediate host, they release oncospheres into the host's intestinal mucosa, subsequently entering the circulation. The liver and lungs act as a double physiological barrier, reducing the frequency of extrahepatic and extrapulmonary localizations. Oncospheres develop into protoscoleces, which, when the DH consumes infected meat, are reintroduced into the intestinal mucosa and mature into adult worms [4, 5]. Human-to-human transmission has not been confirmed [5].

Certain professions involving close contact with animals, such as tanners, shepherds, and veterinarians, carry a higher risk of infection [6].

Uncommon localizations have been reported, encompassing bone (1-3%), pleura or peritoneum (4-7%), spleen or kidneys (2.5%), and exceedingly rare occurrences in the thyroid, pancreas, ovaries, subcutaneous, and muscular tissues [2].

The prevalence of muscular hydatidosis varies among studies, ranging from 1% to 5%. Muscular locations have been documented in the diaphragm, pectoralis major, biceps brachii, sartorius, and psoas muscles [7].

Muscular tissue often displays resistance to hydatidosis due to its tendency to encapsulate and contain the larvae, as well as its contractile activity and high lactic acid content [7, 8].

In both of our cases, the solitary and unusual subcutaneous muscular localization of the cyst is of interest. Such localization accounts for approximately 2-3% of hydatid cysts in endemic regions [9]. The physiopathology of these localizations is challenging to explain, but the hypothesis of lymphatic dissemination appears plausible due to the subcutaneous tissue's low vascularity [10].

Cysts require high oxygen levels for growth, whereas muscles have low oxygen content and high lactic acid levels, hindering cyst development [8-11]. However, proximal muscles in the lower limbs possess a greater vascular mass and vascularity [12]. Nonetheless, compression of adjacent structures may occur, and cyst evolution can lead to fistulization to the skin, resulting in the emergence of daughter vesicles.

Clinically, muscular hydatidosis often presents as a slow-growing, painless, and mobile mass with normal overlying skin [13]. Patients typically seek medical attention when a palpable mass has been present for at least three months and usually measures more than 3 cm in its largest dimension upon initial assessment [14, 15].

Standard radiography may reveal intra-cystic calcification and rule out bone involvement in aged cysts [16, 17].

Regarding subcutaneous hydatid cysts, ultrasound shows multivesicular cysts in 61% of cases and univesicular cysts in 33% [13].

Due to the high rate of false negatives, serology offers limited diagnostic value [18]. It is rarely positive in this location, but a negative result does not exclude the diagnosis [19].

MRI is considered the gold standard for evaluating hydatid cysts in soft tissues. It aids in establishing the diagnosis of soft tissue hydatidosis, determining morphology, topography, and relationships with neighboring vascular and neural structures, which are critical for surgical planning [20].

Surgical treatment is recommended, and precautions should be taken to avoid opening the cyst during dissection. The use of gauze soaked in hypertonic saline solution can prevent local scolex dissemination [19, 21, 22]. Currently, percutaneous techniques such as puncture, aspiration, injection, and reaspiration (PAIR) serve as an alternative to surgical excision for selected patients. Monobloc excision with total pericystectomy is the ideal procedure but may not always be feasible, especially in cases where the cyst is large, deep, or in contact with vascular and neural elements.

Medical treatment with Albendazole is recommended in combination with surgery, either pre- or post-operatively, to prevent recurrence [23], or it can be used alone at high doses in inoperable cases to minimize the risk of dissemination [24].

Muscular hydatid cysts resemble slow-growing soft tissue tumors and can mimic myositis or calcified hematoma symptoms [8], making the diagnosis sometimes challenging. Due to unclear radiological characteristics and differential diagnosis, complete lesion excision has been the preferred approach for definitive diagnosis [11].

CONCLUSION

Although extremely rare, the subcutaneous muscular localization of hydatid cysts can be encountered and should be considered in the differential diagnosis of any soft tissue mass with normal overlying

skin. Ultrasound is a valuable tool for assessing the lesion's texture. Diagnostic and surgical precautions for this location do not differ significantly from other locations, and therefore, all cystic lesions should be thoroughly examined with imaging to prevent potential complications.

Authors contribution

El Ayachi Montassar: original draft writing
Khairi Saibi: Data analysis, Paper validation
Emna Ben brahim: Writing paper
Mouna Ounaies: Contributor
Said Baccari: Supervision

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