

## Primary Hydatid Cyst of Soft Tissues: Description of 6 Cases

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### Abstract

**Background:** Hydatid cyst (HD) is a major zoonotic infection common in Morocco. The musculoskeletal involvement is rare.

**Material and methods:** A retrospective study was conducted in the Department of pathology at Ibn Sina teaching Hospital in Rabat, Morocco. It compiled 6 cases of primary HD of the soft tissues.

**Result:** The age of the patients ranged from 25 to 70 years. The HD was found in the psoas muscle (4 cases) and in the thigh (2 patients). The average size of the cysts was 12 cm. All patients undergone surgical treatment and chemotherapy using albendazole. Histopathological examination confirmed the diagnosis. Recurrence was observed in one patient.

**Discussion:** HD is a parasitic disease commonly involving lungs and the liver. The soft tissue's involvement is rare (0.5 to 4.7%). The imaging methods used for diagnosis and evaluation of HD are ultrasonography, computed tomography and magnetic resonance imaging. The diagnosis of hydatid cyst is confirmed by histology which shows three layers of hydatid cyst. The inner germinal one, the middle laminated layer and the pericyst associated to a surrounding host reaction. The excision of the cyst is the gold standard treatment.

**Conclusion:** HD should be considered in intramuscular cystic mass. The present study emphasized the importance of its inclusion in the differential diagnosis of soft tissue masses.

**Keywords:** Hydatid cyst; Soft tissues; Histopathology

**Abbreviations:** HD: Hydatid Cysts; USG: Ultrasonography; CT: Computed Tomography; PAIR: Percutaneous Aspiration, Infusion of Scolicidal Agents and Reaspiration; HE: Hematoxylin-Eosin

### Introduction

Echinococcosis (hydatid cyst disease) is a major zoonotic infection caused by the larvae of *E. granulosus* [1]. It is a common disease in the temperate zone's countries, including the eastern part of the Mediterranean region; including Morocco; northern Africa, southern and Eastern Europe, at the southern tip of South America, in Central Asia, Siberia and western China [2]. Due to their physiologic role as capillary filters and vast capillary volume, the liver and lungs are most often affected [3], but several other tissues might be infected, including: the brain, muscles and bones. In fact, the musculoskeletal involvement has been reported in only 1-4% of the cases [1], as the presence of lactic acid in muscles creates an unfavorable condition for hydatid cyst development. The aim of the present study is to emphasize the epidemiological, clinical, radiological and histological features of hydatid cysts of the soft tissues; it consists of a retrospective study from Ibn Sina teaching hospital in Rabat, Morocco, where we report on 6 cases of primary hydatid cysts of the soft tissues.

### Material and Methods

This is a retrospective study conducted in the Department of pathology at the Ibn Sina teaching Hospital in Rabat, Morocco. It compiled 6 cases of primary hydatid cyst of the soft tissues diagnosed and confirmed by histology from January 2014 to June 2019. All the data is collected from the patients' clinical files from the Departments of surgery of the same hospital. The following clinical and paraclinical parameters were studied: demographic variables (age and gender), clinical presentations, radiological findings, histological features, treatment and evolution.

### Results

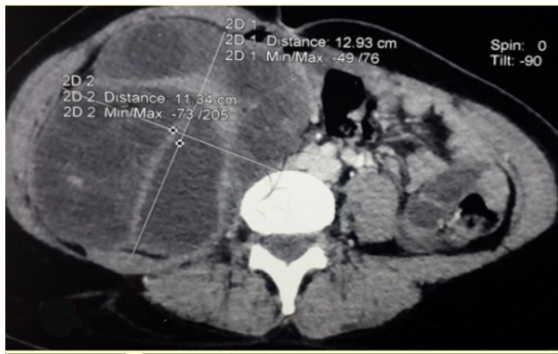
The age of the patients ranged from 25 to 70 years, with a median of 39. There were three men and three women. The hydatid cysts were found in the following sites: the psoas muscle in 4 cases, the anterior region of the thigh in 2 patients. None of the patients had significant co-morbidities, nor suffered from immunodeficiency syndromes. Two patients came from rural areas. One patient had a previous surgery for hydatid disease. In patients with the psoas involvement, the presenting symptoms leading to the diagnosis were pain in all cases, fever in 2 cases and a weight loss in one patient. For the thigh localisation, both patients presented swelling of the right thigh which becomes progressively painful as it increases in volume. The physical examination revealed a palpable mass in the right iliac fossa in 4 patients and in the anterior region of the thigh in 2 cases. Hydatid serologic tests were positive in all cases. Ultrasonography was performed in all patients, and CT scan was done in 5 patients. They showed evidence of hydatid cysts of the psoas (4 cases) and of the quadriceps (2 cases). The average size for the cysts was 12 cm (5-22 cm) Figure 1. Type III was in 5 patients and IV in 1 case. All patients undergone surgical treatment consisting of either partial (5 cases) or total pericystectomy (1 patient). A chemotherapy using albendazole was established in all our patients for 6 months. The diagnosis of hydatid cyst was confirmed in all cases by histopathological examination of resected specimens. Macroscopically, the cysts were filled with clear hydatid fluid surrounded by thin

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**Received** July 30, 2019; **Accepted** September 18, 2019; **Published** September 25, 2019

**Citation:** Derqaoui S\*, Irahim M, EL Ouazzani H, Zouaidia F, Jahid A, et al. (2019) Primary Hydatid Cyst of Soft Tissues: Description of 6 Cases. J Cytol Histol 10: 545.

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**Figure 1:** Contrast-enhanced computed tomography of abdomen unenhanced hypodense mass of the right psoas with well-defined borders and internal septa.

membrane in 3 cases and by fibrous thick layer over the inner germinal layer in 3 cases. The following histological features were found in all cases: thick, acellular, laminated layer with characteristic acidophilic staining, cellular germinal layer and a host produced granulomatous reaction surrounding the cyst. Brood capsules or protoscolices were found in only 2 cases. After a follow-up period of 4 years, recurrence was observed in one patient (thigh localisation).

## Discussion

Hydatid cyst is a parasitic disease, prevalent in farming and livestock areas, where preemptive healthcare measures are rather low. The infection is spread to the main hosts, namely dogs and other canidae, when they eat the infected internal organs of the intermediate hosts (sheep, goats, swine...). Humans represent intermediate hosts as blind end in parasite life cycle when they occasionally ingest eggs through contaminated food or water]. The parasitic larvae (scolices) attach to the human intestines by utilizing their hooks, and bypass the intestine barrier [4]. The liver is the first line of defense in the human body and therefore becomes the first organ to be involved. The disease commonly involves the lungs and liver. Larvae may reach other organs, such as the spleen, kidneys, skin, muscle tissue, retroperitoneum, bones, heart and the brain [5]. The involvement of soft tissue is rare (0.5 to 4.7%) and the main localizations are skeletal muscles of the neck and the proximal limbs. In fact, muscle function may be an impediment, both mechanically and environmentally (lactic acid concentration) for implantation and development of the larvae [6].

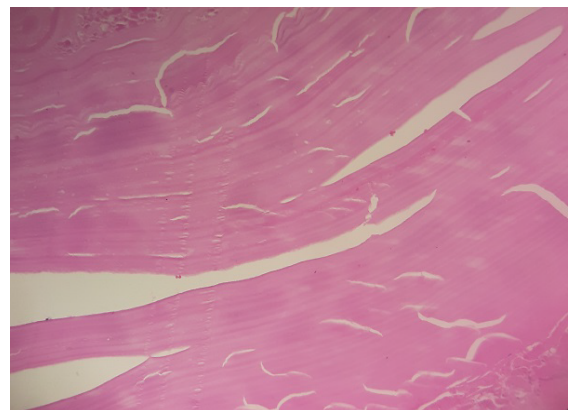
Clinically, the main presenting symptom is the presence of a mass; in addition to local symptoms related to compression of adjacent structures. General symptoms related to immune reactions or to complications (anaphylactic shock, infection of the cyst...) may occur [7]. Because of low sensitivity and incomplete specificity, the use of serology is restricted to case confirmation, especially among symptomatic patients because the capsule isolates the parasite from the host's immune system. Serology may also be of use for patient follow-up as an indicator of relapse or recurrence [8].

At imaging, hydatid cysts of tissues may appear as unilocular or multilocular cystic lesion or complex solid lesion depending upon growth of the cyst. Differential diagnosis includes abscess, synovial cyst, chronic hematoma, and necrotic soft tissue tumor. The imaging methods used for diagnosis and evaluation of the extent of HD are ultrasonography (USG), computed tomography (CT) and magnetic resonance imaging (MRI). USG is screening modality of choice and is also used to monitor the efficacy of treatment. In fact, it clearly

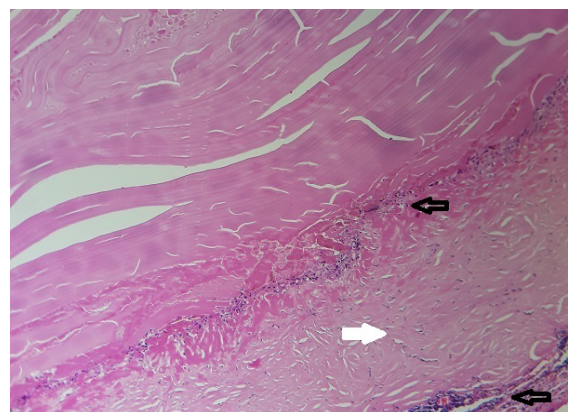
demonstrates the hydatid sand, floating membranes, daughter cysts and vesicles inside the cyst. CT has high sensitivity and specificity for HD. CT is an important diagnostic modality in detecting cyst wall or septal calcification, demonstrating internal cystic structure posterior to calcification and assessing complications [9].

The diagnosis of HD is confirmed by histology which shows three layers of hydatid cyst. The inner germinal one is thin and translucent on gross. The embryonic tapeworms, scolices, develop from an out pouching of the germinal layer and form hydatid sand, settling into the dependent parts of cyst. The middle laminated membrane (Figure 2A) is white 2 mm thick, avascular, eosinophilic, refractile and chitinous; strongly PAS+ and GMS+. It is selectively permeable to nutrients but not to bacteria. The outer layer or pericyst is a rigid protective layer with a few millimeters thickness, representing response of the host to the parasite. It is a dense fibrovascular tissue with chronic inflammatory cells and variable calcification (Figure 2B). The cyst fluid is crystal clear, as it is transudate of serum containing proteins. The surrounding host reaction, which is composed of the inflammatory fibrous tissue, forms a dense pseudocapsule around the cyst and might show foreign body type giant cell reaction (Figure 3) [10].

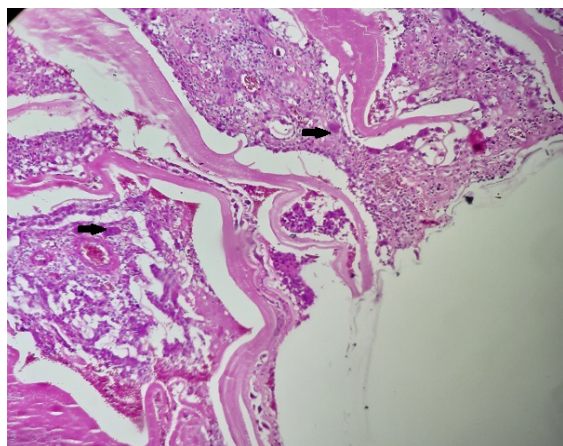
Incisional biopsy and marginal excision are contraindicated if hydatid disease is suspected, because of the dissemination's risk and



**Figure 2A:** Hematoxyline eosine stained histology section: showing lamellated membrane of the cyst.



**Figure 2B:** Hematoxyline eosine stained histology section: showing chronic inflammatory response with lymphoplasmacytes (black arrows) and fibrosis (white arrow).



**Figure 3:** Hematoxyline eosine stained histology section: Cyst wall showing foreign body type giant cell reaction (20X): giant multinucleated cells (black arrows).

possibility of allergic reaction [11]. The total excision of the cyst to avoid its rupture and spillage is the gold standard treatment. Preoperative planning is important to achieve curative management without complications as the cyst might develop close to the neurovascular structures [12]. Percutaneous aspiration, infusion of scolicalid agents and reaspiration (PAIR), under imaging (USG or CT) guidance can be used as alternative to surgery in inoperable cases [11]. Supplementary chemotherapy with antihelminthics for skeletal muscle hydatid disease is controversial and no evidence based recommendations can be made. Albendazole remains the gold standard drug administered in adjuvant therapy [13]. Six months of oral albendazole 15 mg/kg/day is recommended along with surgical treatment [14].

## Conclusion

Hydatid cyst should be considered in intramuscular cystic mass. The importance of its inclusion in the differential diagnosis of soft tissue masses is emphasized in the present study. Thus the preoperative diagnosis remains essential to improve the management of hydatid cysts.

## Declarations

**Ethics approval and consent to participate:** Not applicable

**Consent for publication:** All authors read and approved the final manuscript

**Availability of data and material:** Not Applicable since data were collected from the files sent routinely to the department with the specimens

**Funding:** Not applicable since no funding source was needed

**Acknowledgements:** Not applicable

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**Competing interests:** The authors declare that they have no competing interests.

**Authors' contributions:** All authors read and approved the final manuscript.

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