

The Peculiarity of the Primary Hydatid Cyst in Soft Tissues Located in Unusual Anatomical Areas: A Report of 4 Cases

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ABSTRACT

The primary location of hydatidosis in the soft tissues is a rare situation, and the clinical picture is not very specific and consists of a painless swelling. The diagnosis is based on a combination of clinical, biological, radiological and anatomopathological findings. The biological work-up is essentially based on serological examinations using different immunological techniques; a negative serology does not however allow the diagnosis of hydatidosis to be excluded. Imaging is the most important step in the diagnosis, namely ultrasound and Computed Tomography (CT), but especially Magnetic Resonance Imaging (MRI) which is the most useful imaging tool in hydatid pathology of

the soft tissues, through their results allow a better approach of this pathology, thus facilitating its management. Surgery remains the reference treatment, but a medical-surgical approach is now the rule. We have described in our study the cases of isolated hydatid cysts of the soft parts operated, with anatomopathological confirmation, by following the evolution of the patients since their consultation until the cure.

Keywords: Primary, Hydatid cyst, Unusual anatomical areas, Soft tissues

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INTRODUCTION

Hydatidosis, or cystic echinococcosis, a name suggested by the World Health Organization (WHO) (Bousofara M, *et al.*, 2005), is an anthrozoosis caused by a parasitic taeniid-type tapeworm, *Echinococcus granulosus*, which is commonly transmitted from infected dogs, through a hand-to-mouth transfer of viable tapeworm eggs released in dog faeces (Tsukada H, *et al.*, 2000). Hydatid cysts can form in any organ or tissue, but it's most commonly found in the liver (75%) and lungs (15%) (Memis A, *et al.*, 1999). Soft-tissue hydatidosis is a very rare location of the parasite, even in endemic regions, this location doesn't exceed 3% (Garcia-Diez AI, *et al.*, 2000). Clinical manifestations of soft-tissue hydatidosis are widely variable, depending on cyst location and its size, it ranges from a painless non-inflammatory swelling, to presentations that can mimic soft-tissue abscess or tumors (Cissé AM, *et al.*, 2002; Daali M and Hssaida R, 2000).

Biological assessment using immunological tests is an essential step, however, negative serological tests doesn't exclude the diagnosis of hydatidosis. Radiological exploration is an important diagnostic tool that allows a better approach in soft-tissue hydatidosis management using ultrasound, CT and especially MRI which is a performing imaging tool in case of soft-tissue involvement (El Haddad S, *et al.*, 2011). Surgery is considered as the standard treatment for hydatid cyst, but a combination of medical and surgical management seems to be more often used (Örmeci N, *et al.*, 2007). Prophylactic approach is an important tool that should be considered at every step of the epidemiological chain, especially in endemic regions (Agoumi A, 2003). We report in this article a case series of 4 patients presenting soft-tissue hydatidosis.

CASE PRESENTATION

Our work consisted on a descriptive retrospective study of 4 patients presenting soft-tissue hydatidosis, being followed in Trauma

and Orthopedics department, in Mohammed VI University Hospital in Oujda, over a period of 6 years extending from November 2014 to September 2021.

We have included in our study, patients presenting soft-tissue hydatidosis with histological confirmation. Collecting epidemiological, clinical, radiological, and histological data was possible by accessing medical records from Trauma and Orthopedics departments (Medical Records Department), and Radiology department (Radiological information system and Carestream/PACS system for image acquisition). Radiological evaluation of soft-tissue hydatidosis in our patients was based on X-ray, ultrasound, CT-scan and MRI findings.

Medical information was collected and analyzed using a data collection sheet including epidemiological, clinical, radiological, pathological, therapeutic and evolutionary data from every patient.

RESULTS

In our series, there is a higher incidence among females (75% of patients were females), and most of them came from rural areas (75% of patients). The main clinical symptom in our series correspond to a slow growing soft-tissue mass. The average delay for seeking a medical advice was about 45 months, with extremes extending from 1 to 10 years.

Clinical examination of our patients revealed a soft-tissue mass as the most predominant symptom, and variably associating other symptoms like pain and movement restriction (*Figure 1*) (*Table 1*).

Biological assessment has shown a normal blood count in all our patients, and a normal level of C-Reactive Protein (CRP), except for one patient. Serological tests were positive in only 2 patients.

An X-ray was performed in all patients showing variable results (*Figure 2*) (*Table 2*)-



Figure 1: (a) Swelling in anterior aspect of the thigh (b) Swelling of the right buttock

Table 1: Common clinical symptoms seen in our series

Clinical symptoms	No. of patients
Soft-tissue tumefaction	4
Pain	1
Restricted movements	1
Altered performance status	0

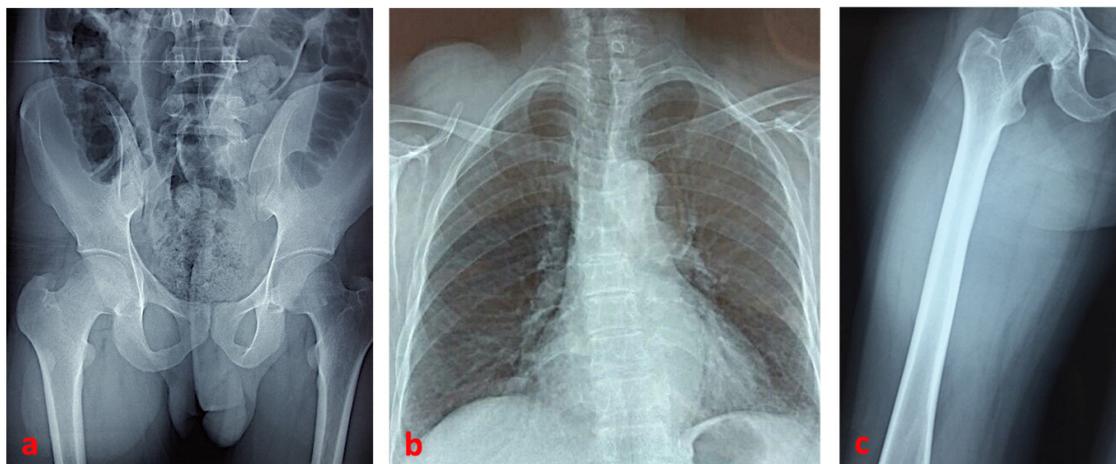


Figure 2: (a) Pelvic x-ray showing a rounded opacity on the right gluteal region (b) Chest x-ray showing a mass in the right supraclavicular region (c) X-ray of the right thigh showing a mass

Table 2: Characteristics of the soft-tissue mass

Characteristics of the mass	Patient 1	Patient 2	Patient 3	Patient 4
Location	Gluteal region	Gluteal region	Thigh	Subclavicular
Edges	Ill-defined	Well-defined	Well-defined	Well-defined
Consistency	Soft	Soft	Soft	Firm
Tenderness	Painful	Painless	Painless	Painless
Mobility	Mobile	Fixed	Mobile	Mobile
Local inflammation	Absent	Absent	Absent	Absent

- Patient 1-No evident lesion.
- Patient 2-No evident lesion.
- Patient 3-Round, well-defined homogenous radio-opacity on the upper 2/3rd of the thigh with no calcifications.
- Patient 4-Round, well-defined homogenous radio-opacity on the right subclavicular region, without any osteolytic lesions.

Two patients underwent a soft-tissue ultrasound, showing stage III hydatid cyst of GHARBI classification. An MRI was performed on all our patients (Figures 3-5), to reveal the exact location of the mass, its size, borders, shape, signal and to evaluate its relationship to adjacent structures. A chest x-ray with abdominal ultrasound were performed in all of our patients

showing no secondary location of hydatid cyst.

Gross aspect of hydatid cyst during surgery had a typical appearance, the mass showed numerous vesicles and pseudo-membranes, histological examination of the mass revealed a lamellated layer confirming the diagnosis of hydatid cyst (Table 3) (Figure 6).

In post-operative phase only one patient has presented vomiting 6 months after surgery, a reason why a thoraco-abdominopelvic CT scan was performed showing no signs of local recurrence, no other complications have been reported in our series and none of our patients had a recurrence so far (Figure 7).

In our series, there was a correlation between radiological, histological and surgical findings.

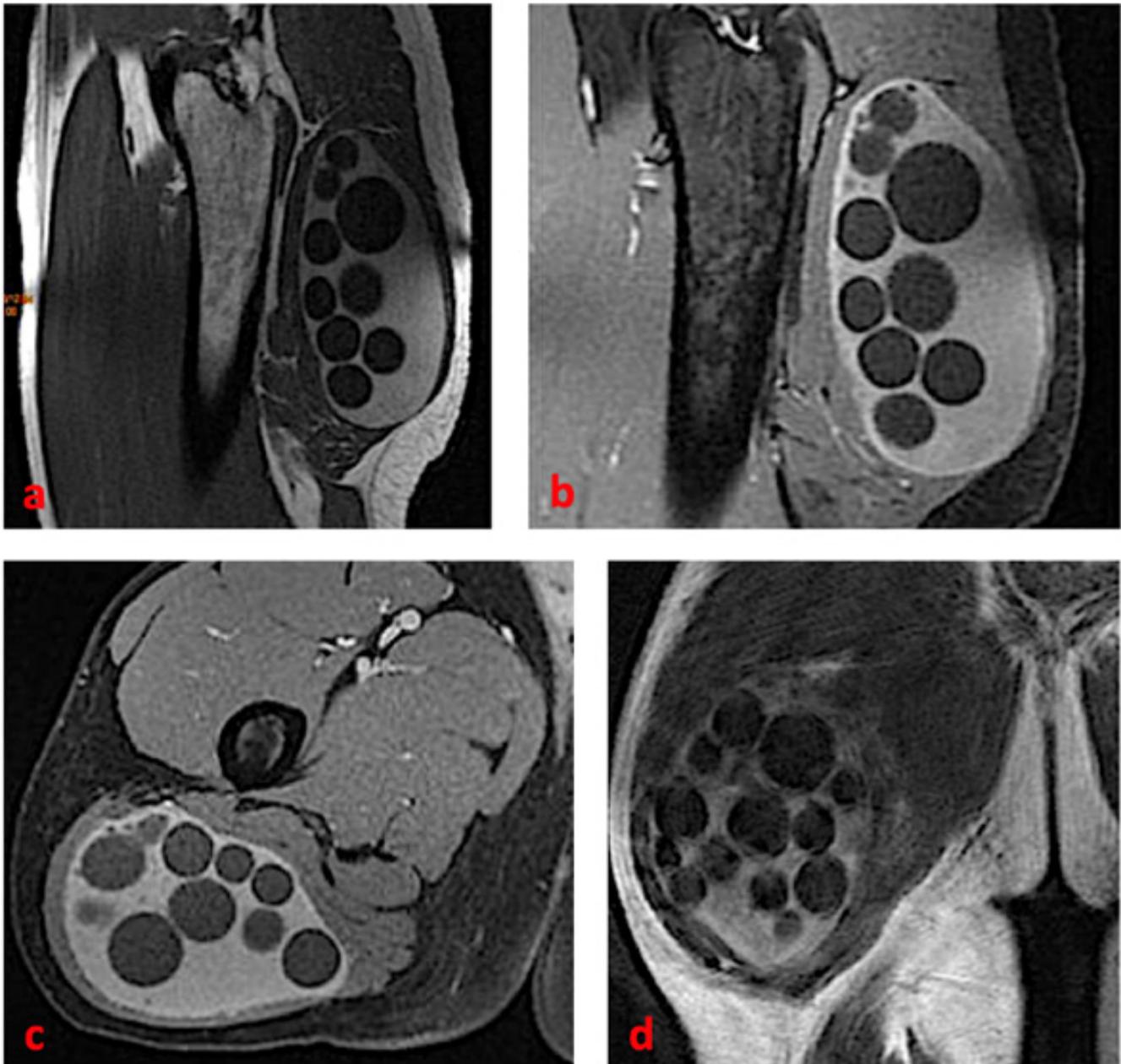


Figure 3: (a) Sagittal T1-weighted MRI before and after Gadolinium injection (b) Axial T1-weighted MRI after injecting Gadolinium (c) Coronal T1 fat sat MRI (d) Showing a multivesicular mass in the gluteus maximus muscle, compatible with type III hydatid cyst

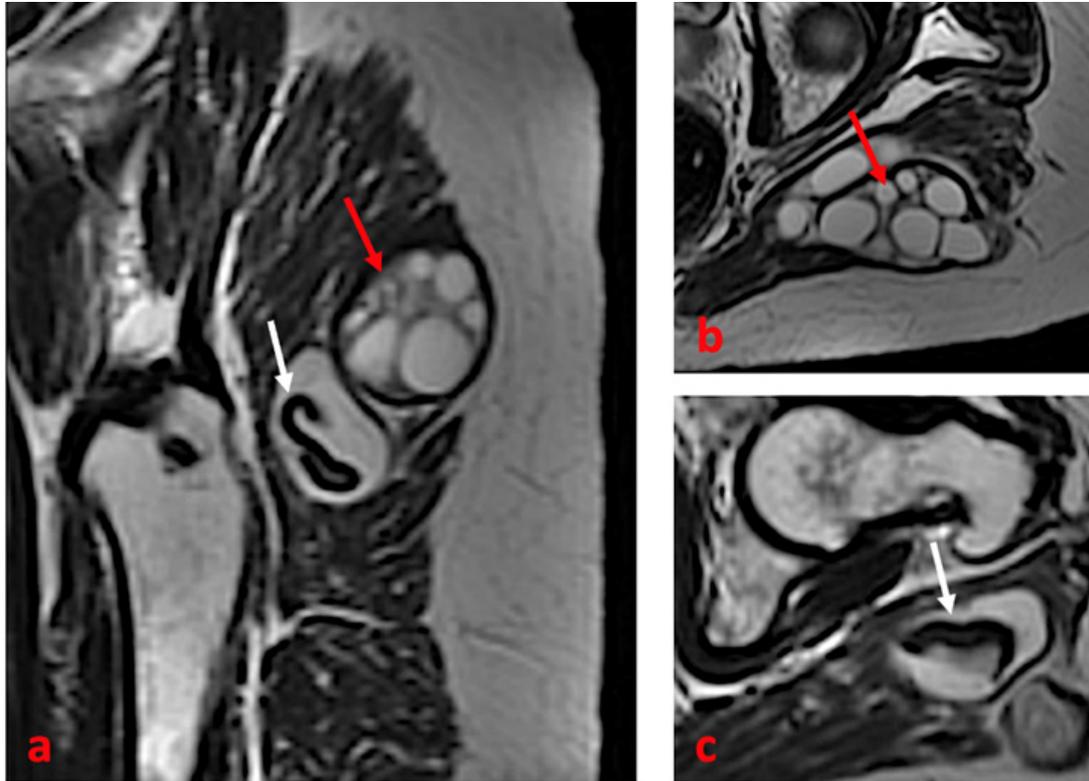


Figure 4: Sagittal (a) and axial (b,c) T2-weighted MRI, showing two contiguous masses

Note: (V): Thin-walled multivesicular mass; (U): Thick-walled cystic formation containing a floating membrane

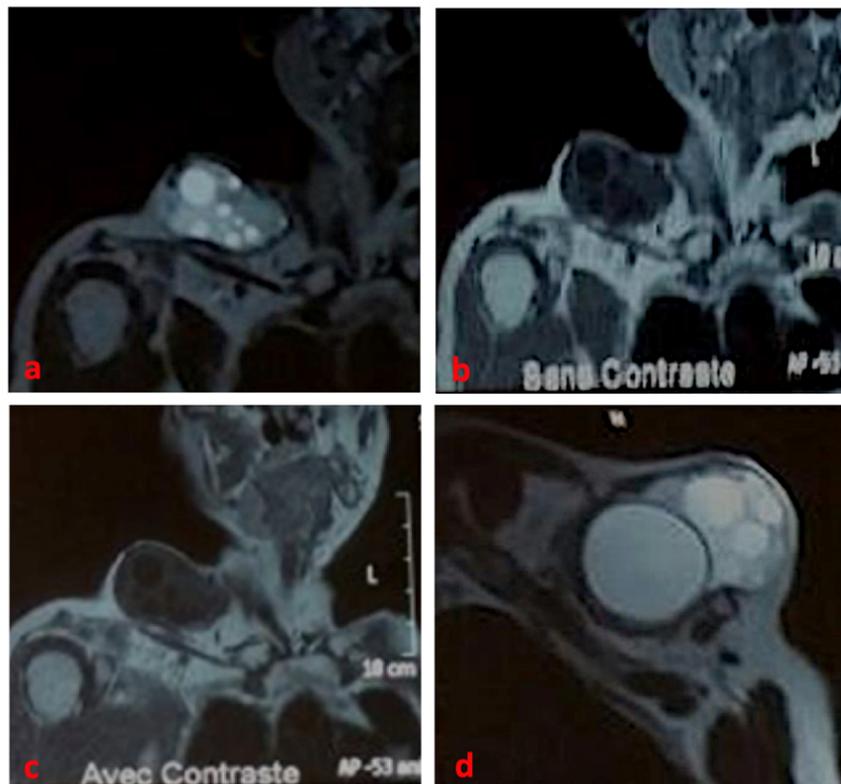


Figure 5: Coronal T2-weighted MRI (a) Coronal T1-weight MRI before (b) Coronal T1-weight MRI after injecting gadolinium (c) Sagittal T2-weighted MRI (d) Showing a multivesicular mass of the right supraclavicular region, compatible with type III hydatid cyst, with no evidence of bone involvement

Table 3: Correlation between radiological, surgical and histological findings

Case	MRI	Surgical exploration	Histology
Patient 1	Type II and III hydatid cyst	<ul style="list-style-type: none"> White mass Endogenous daughter cyst 	Hydatid cyst
Patient 2	Type III hydatid cyst	<ul style="list-style-type: none"> Firm mass Endogenous daughter cyst 	Hydatid cyst
Patient 3	Type III hydatid cyst	<ul style="list-style-type: none"> White mass Endogenous daughter cyst 	Hydatid cyst
Patient 4	Type III hydatid cyst	Endogenous daughter cyst	Hydatid cyst

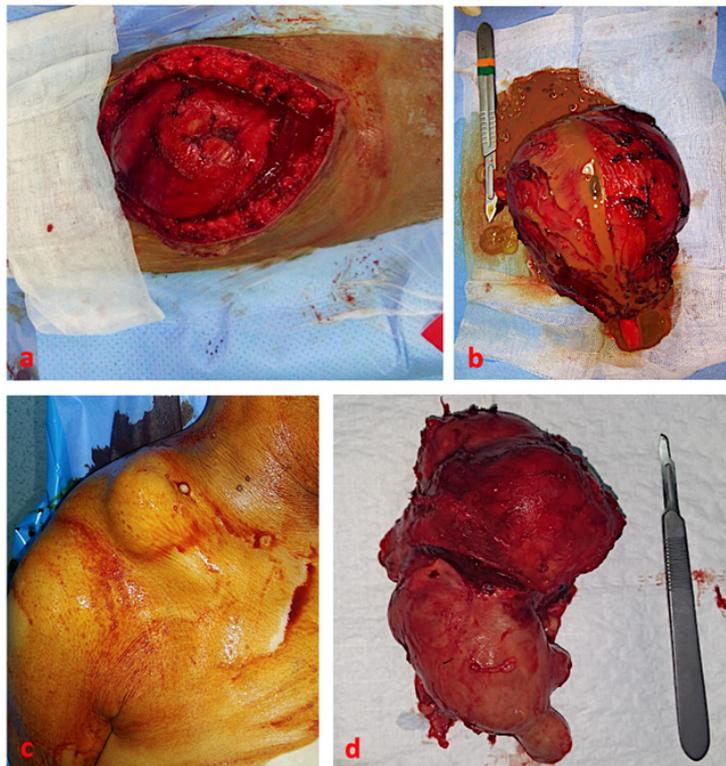


Figure 6: (a) Intraoperative images showing a hydatid cyst in the buttock, before (b) Intraoperative images showing a hydatid cyst in the buttock after resection (c) Right-supraclavicular hydatid cyst, before (d) Right-supraclavicular hydatid cyst after resection

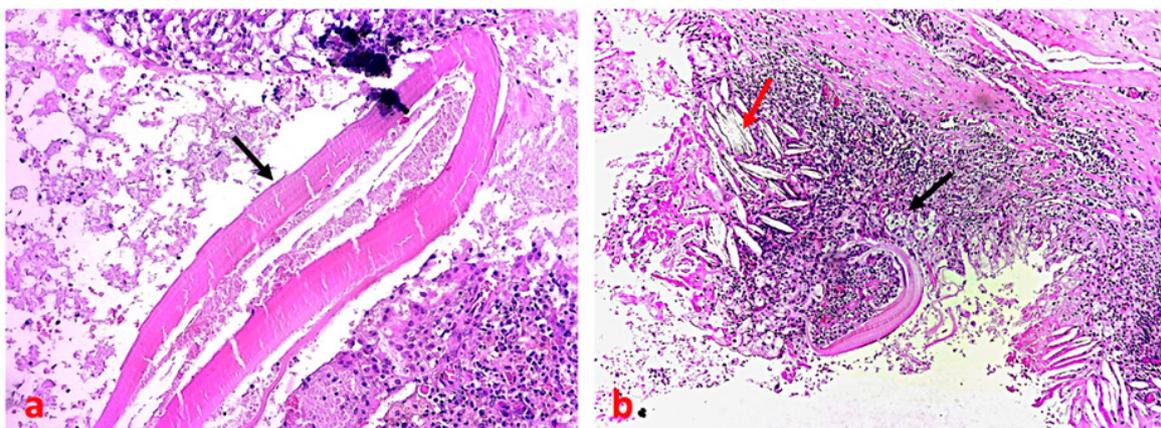


Figure 7: Histopathology images (a) Showing a laminated membrane (N) (b) Laminated membrane surrounded by histiocytic cells (N) and cholesterol crystals, due to host inflammatory reaction against the cyst

DISCUSSION

Human hydatid cyst is an anthroponosis linked to the development in humans of *Echinococcus granulosus* larvae. The parasitic cycle usually involves two hosts, dogs or wild canids as definitive hosts, and an intermediate host most commonly sheep, goats, cattle, equines, suids or camels (Pedrosa I, *et al.*, 2000). Muscle hydatidosis is rather a rare location, even in endemic regions it accounts for 1% to 5% (Mekki RA, *et al.*, 2005; Bennis A and Maazouzi W, 2001).

Patients included in our study were aged from 29 years to 78 years, with a median age of around 55 years. Several studies have reported high prevalence in young adults, as reported in Oğur HU, *et al.*, 2009 study, where the average age reported was 39.4 years.

Several authors have reported that the majority of cases were females (Bellil S, *et al.*, 2009; Habboubi K, *et al.*, 2016), the same observation has been reported in Morocco between 1980 and 1992, where 75.4% of reported cases were females, against 24.6% of males. This is probably linked to agricultural tasks and daily handling of vegetables more often done by women. This predominance of infection in females has also been reported in other countries. In our study we report a higher incidence in women as well, 75% of females against 25% of males.

Most patients reported in literature have rural origins (Hlili F, *et al.*, 2011; Zian D, 1998). In our study 75% of our patients come from rural areas. None of them had a previous history or second location of hydatid cyst, which confirms the primary involvement of soft-tissue. The main risk factor related to hydatidosis is a previous contact with dogs, whether direct or indirect (feces), it is found in 54.8% (DAALI series) and 82.7% (HLILI series) of hydatidosis cases (Hlili F, *et al.*, 2011). In our study 80% of patients have had a previous contact with dogs.

In this study no hyper-eosinophilia has been observed. In Aït-Ammar N, *et al.*, 2016 and Al Alami N, 2020 studies, hyper-eosinophilia was noted in only 10% of patients. CRP was elevated in only one of our patients, which had suggested a cyst infection. All our patients had a serological test using Indirect Hemagglutination Assay (IHA) which confirmed seropositivity in two of them.

Management of muscular hydatidosis is based on a combination of medical and surgical approach, associating cyst excision and drug therapy using Albendazol; antibiotics are usually used in case of cyst superinfection (Zian D, 1998). Some studies report benefits of using formalin, alcohol, silver nitrate (as a scolical agent) and a hypertonic saline solution to prevent recurrence and disease spreading (Aït-Ammar N, *et al.*, 2016). In our series, all patients underwent a conservative surgical excision of the cyst, abundant washing with saline solution and hydrogen peroxide with preservation of adjacent healthy soft-tissue, then, they received an antiparasitic treatment based on Albendazol for 28 days (1 cycle), followed by a 14-day drug-free interval, for a total of 3 cycles; liver function and complete blood count were constantly monitored during treatment.

CONCLUSION

Hydatid disease is a frequent endemic parasitic infection in Morocco as well as in most African countries, primary soft-tissue and specifically muscular location of hydatid cyst is an uncommon situation, occasionally such locations can give rise to many complications due to compression of vascular or nervous structures, which can represent a challenging operative situation, requiring a very careful surgical intervention that aims to remove the mass without bringing any damage to adjacent neurovascular bundles. Interventional radiology could be a useful tool in that situation allowing a percutaneous drainage of the cyst without the need for an invasive procedure.

AUTHORS' CONTRIBUTIONS

Adnane Lachkar, Hicham Yacoubi and Najib Abdeljaouad did the surgery.

Samir Ben Salah, Ayman Benabdellah drafted the documents for the ethics vote. Samir Ben Salah designed the study, developed the questionnaire, treated the data, did the statistics, and then drafted the manuscript, tables and figures. All authors read and approved the final manuscript.

ETHICAL APPROVAL AND CONSENT TO PARTICIPATE

Ethical approval was not needed in view of the retrospective and observational type of our study.

CONSENT FOR PUBLICATION

A written consent signed by all patients was obtained authorizing the use of their medical data in the scientific research and for publication on open access.

AVAILABILITY OF DATA AND MATERIALS

The collection of data necessary to accomplish this work is facilitated by our computerized system of archiving medical records and the evolution of patients and their final results. Due to the sensitivity of the raw data, all data are available from the first author on request.

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