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Solitary Subcutaneous Echinococcosis Cyst of the Ankle, Unusual Localization: A Case Report and Literature Review

Badarou Chaibou*, Espoir Amour Mokoko-Loukou, Smail Abdouli, Zakariyaou Harouna Ide, Mohammed Elidrissi Abdelhalim ElIbrahimi, Abdelmajid Elmrini

Service of traumatology and orthopedic B of CHU Hassan II, Fes Morocco

***Corresponding author:** Badarou Chaibou
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Abstract **Case Report**

Subcutaneous echinococcosis cyst is uncommon even in endemic regions. Ankle area localization of echinococcosis cyst is the first case reporting in the literature. It's a case of a 48-year-old male, breeder and farmer, Arab of rural origin, presents with incipient painless ankle swelling dating 8 month prior to his consultation without fever and good general conditions. Ultrasonography revealed an echinococcosis cyst aspect. We have managed this case surgically by monobloc resection. The patient recovered well after surgical management without any recidivism. Symptoms of subcutaneous echinococcosis cyst are often discrete with imaging confirming diagnosis. Ultrasonography and/or magnetic resonance imaging help reduce untimely management. Surgical management by monobloc resection with total pericystectomy represents the sole curative treatment. Nonetheless, prevention remains the best tool to control the spread of this endemic.
Keys words: Echinococcosis cyst; ankle; imaging; surgery.

INTRODUCTION

Hydatidosis or *echinococcosis* is a cosmopolitan parasitosis caused by *Echinococcus granulosus*, rarely *Echinococcus multilocularis*, which usually infect the liver or lungs. Diagnosis is based on patient's clinical history, physical examination and relies largely on imaging features. Surgical excision remains the sole potentially curative treatment in the management of echinococcosis

Subcutaneous localization, not to mention cysts in the ankle region remain exceptional even in endemic countries like Morocco. Hence, the purpose of this article is to report a peculiar case of solitary subcutaneous echinococcosis cyst (EC) in the ankle area, to emphasize its pre-operative diagnosis as well highlight certain pitfalls in the management of the affection.

CASE REPORT

Patient, 48-year-old male, breeder and farmer, Arab of morocco rural area origin, presents with incipient painless ankle swelling dating 8 month prior to his consultation without fever and with good general conditions.

Physical examination found a swelling in front of the lateral malleolus of the left ankle. A subcutaneous rounded mass, measuring 6 cm in diameter, renitent, easily mobile without any local inflammatory signs (Figure 1A). Furthermore, local and regional lymph nodes examination did not reveal any swelling. The rest of the physical examination, notably abdominal and chest exams were unremarkable.

Plain ankle X-rays (AP and lateral views) were unremarkable. Ultrasonography revealed a liquid mass with clearly defined edges, measuring 5cm in diameter, containing multiple vesicles resembling grade III EC according to Gharbi classification [1] (Figure 2). There were no sign of invasion of surrounding structures of the ankle region. Further work up (chest X-ray, abdomino-pelvic ultrasound) came back unremarkable for other visceral localizations.

Laboratory test, notably WBC counts and eosinophil levels were normal as well as a negative echinococcosis serology. Surgical treatment consisted of a direct approach with cutaneous and subcutaneous incision done directly on the mass and subsequent careful dissection of underlying structures (Figure 1B). Per operatively, cleavage between the mass and surrounding tissue was evident, which allowed for a pericystectomy with monobloc resection of mass

(Figure 1C). Multiple vesicles resembling EC were discovered post operatively on opening and analysis of surgical specimen (Figure 1D). Diagnosis was confirmed by pathology exam of specimen. The patient had received in post-operative two doses of Albendazole two weeks apart. Post-operative recovery was uneventful with a favorable outcome and no local or visceral recurrence 30 months after surgery.

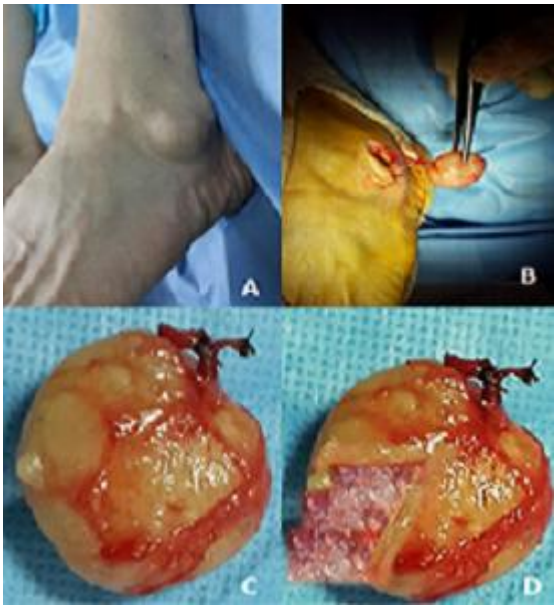

Fig-1: clinical aspect of pre-malleolus swelling (A); per operative image showing monobloc cyst resection (B); post-operative image of intact cyst (C); macroscopic image of the cyst contents (D)

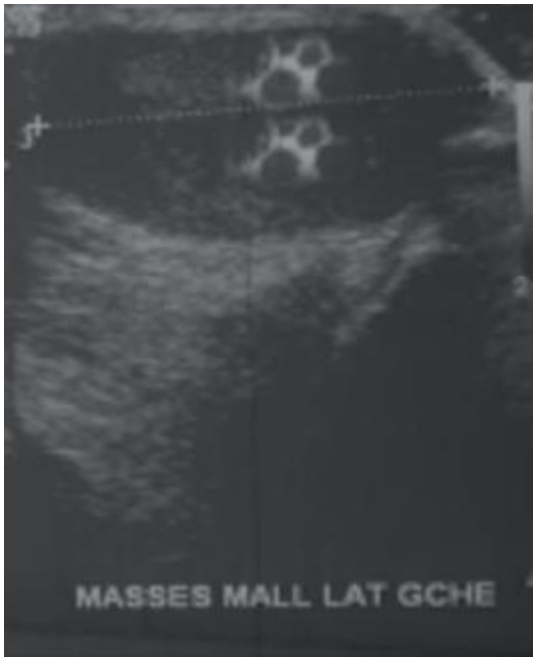

Fig-2: Ankle ultrasound: characteristics of lesion

DISCUSSION

Echinococcosis is an endemic disease that remains a major public health concern in some parts of the world, including the Mediterranean basin, the Middle East and South America where livestock breeding is widespread [2]. It constitutes a cosmopolitan anthropozoonosis common to human and other mammals, caused by the growth and development of larva form of a tapeworm of dogs, *E granulosus*, rarely due to *E multilocularis*. Its definite host is the dog, with humans serving as secondary hosts in course of the cycle. Accidentally ingesting parasite-infested meat (such as the viscera) or directly touching affected dogs affects human beings. Hence, humans become secondary hosts (main host being sheep) in the life cycle of the tapeworm. Once in the GI tract the parasites follow the portal system, from which they spread to rest of the body. This explains the relatively

higher incidence of liver (70%) and lung (10-15%) localizations. More so, these two organs constitute a double physiologic filter to parasite dissemination, thereby making extra hepatic and non-pulmonary localizations less frequent [3].

The particularity highlighted in this case is the solitary nature of the affection as well as unusual localization in the ankle area as subcutaneous EC represents only 2.3 % of all EC in endemic zones [4]. Secondly, it is difficult to explain how larvae can evade liver and lungs filters and form a solitary secondary cyst without visceral localization given that the portal system is the only widely accepted mechanism of dissemination of echinococcosis larva in humans. Lymphatic dissemination may be possible [5].

Clinically, symptoms of subcutaneous EC are variable. Incipient skin swelling, with good general state, in a person of rural origin with a long history of contact with dogs, in endemic areas should suggest EC. Sometimes, it is discovered as a result of compression of adjacent structures [7]. Subcutaneous EC can develop fistula with subsequent discharge of daughter vesicles [8].

Echinococcosis serology is rather less sensitive marker for soft tissue localization due to many false-negatives [9]. However, it is very useful as a monitoring tool only when it is positive before treatment [10]. Standard X-ray could reveal intra-cystic calcifications in inactive cysts. It could help eliminate bone localization.

Ultrasound is a key front-line imaging tool as far as diagnosis is concerned. It allows for positive diagnosis [8] as it characterizes the cyst in terms of its echogenicity, localization and type according to Gharbi classification. According to Z. Orhan *et al*. [6] this imaging technic has up to 100% sensitivity when it shows daughter cysts, vesicles and membranes as was the case in our patient. However, atypical forms of cyst exist where mass may be more echoic, and vesicles and/or membranes or even septa may not be clearly visible.

Magnetic resonance imaging (MRI) represents the first choice technic in cases where ultrasound cannot confirm diagnosis of echinococcosis disease. It may reveal vesicles, membranes, and cystic wall in the form of a surrounding membrane that is relatively hypo-intense in T2 weighted sequence. Peripheral cystic enhancement after gadolinium injection confirms the diagnosis of echinococcosis in soft tissues. MRI also allows for study of accurate links of cyst with surrounding structures [8]. These imaging technics are very essential to surgical planning and useful during follow-up to detect possible local recurrence. In our case, diagnosis was achieved based on ultrasound only.

Treatment of subcutaneous EC is surgical. Surgery must be prudent as there is risk of dissemination of daughter cyst due to per operative opening or cracking of cyst that could lead to fatal anaphylactic shock. Operative fields should be protected with hypertonic saline solution and/or hydrogen peroxide solution during surgery [5, 6, 8]. Monobloc resection with total pericystectomy, an ideal approach, may not often be feasible especially when cyst have close contacts with neighborhood vessels and nerves. Currently, percutaneous treatment either by puncture-aspiration-injection-re-aspiration (PAIR) or percutaneous drainage constitute valid alternatives for surgical excision in selected patients [7].We are not for this opinion because aspiration can be incomplete or cause dissemination. Conservative treatment with Benzimidazole (Albendazole) has little to no place in the management of solitary musculoskeletal EC because of their poor penetration into cystic liquid [5, 6]. However, we also hold the belief that medical management should be reserved for complicated forms, or in the event of per operative rupture or dissemination, or in non-operable patients. It is important to recall some prevention measures of echinococcosis based on:

- Collective prevention measures (information on risks, hygiene of clothing, adequate disposal of animal viscera, etc.);
- Handling of live or dead wild animals (wearing gloves, hand washing, disinfection of wounds).
- In areas at risk, dogs should be dewormed.

CONCLUSION

Isolated subcutaneous EC remains exceptional. Clinical presentation is often insidious. Positive diagnosis can be achieved by ultrasound, sometimes by MRI. Surgical management by monobloc resection with total pericystectomy represents the sole curative treatment. Nonetheless, prevention remains the best tool to control the spread of this endemic.

Abbreviations

CE: cystic echinococcosis; MRI: magnetic resonance imaging; PAIR: puncture-aspiration-injection-re-aspiration.

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Competing interest

The authors declare that they have no competing interests.

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