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Primary Subcutaneous Hydatid Cyst of the Calf: An Unusual Case Report

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Abstract: Hydatid disease is a public health problem in endemic areas. Although the liver and lung are its most commonly locations, it rarely affects other tissues. A 41-year-old female patient was admitted to our clinic with a palpable, fixed mass over the left calf. On MRI, a subcutaneous oval cystic lesion over the left gastrocnemius muscle, containing round-shaped daughter cysts hypointense and hyperintense on T1- and T2-weighted scans, respectively, was detected. Her serological test for hydatic disease was positive. Her whole body was scanned for any other organ involvement, but scans were all negative. During surgery, a well capsulated cyst was excised en bloc. On histological examination, a hydatid cyst was confirmed. She was treated with albendazol. There were no local or systemic recurrences at 1 month of follow up. Primary subcutaneous tissue involvement is a rarely reported entity in the literature. To the best of our knowledge, this is one of only three cases reported of a primary subcutaneous hydatid cyst detected in the leg.
Keywords: hydatid disease, cyst, calf, leg, primary, subcutaneous

INTRODUCTION

Hydatid disease is a cosmopolitan anthropozoonosis more prevalent in countries where the common intermediate hosts, sheep and cattle, are raised (such as Middle East, Mediterranean region, Central Europe, Australia and South America) [1, 2]. Hydatid cysts (HC) are most frequently found in the liver (75%) and lungs (15%) [2]. the primary subcutaneous tissue echinococcosis is a rarely reported entity in the literature (incidence: 0.2-2%) [3, 4]. Because of an asymptomatic evolution, the diagnosis of hydatidosis remains difficult and the therapeutic management is often complicated. In the present article, we report a rare case involving a primary subcutaneous HC of the calf, with particular emphasis on the diagnostic and therapeutic challenges.

CASE REPORT

A 41-year-old woman was admitted to our clinic for a slowly growing mass that localized in her left leg on the calf. The patient has moderate pain without any daily distraction. She lived in an animal farm in a rural site 15 years previously. She had no prior history of trauma, fever, or weight loss. Her physical examination revealed a 10x6 cm fixed, firm and tender mass in posterolateral and proximal parts of the left leg (Fig. 1).There was no ecchymosis, erythema, increased warmth or lymphadenopathy., Complete blood count and electrolytes was normal,. An ultrasound examination was performed and it showed multiple subcutaneous cystic lesions over the medial head of the left gastrocnemius. MRI was performed for further imaging and showed subcutaneous oval cystic mass approximately 77×40×18mm over the left gastrocnemius muscle, containing round-shaped daughter cysts. The cysts seen hypointense in T1 weighted images and hyperintense in T2 weighted images (Fig. 2). Since the MR images were suggestive of HC, further laboratory and imaging studies were employed to support the diagnosis and to detect the other sites of possible involvement. ELISA (Enzyme-linked immunosorbent assay) test for echinococcosis was positive. Chest X-ray and abdominal ultrasonography did not reveal any abnormality.

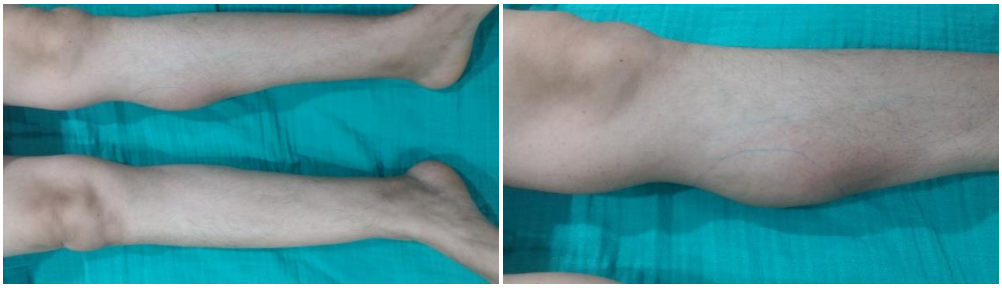

Fig-1: The swelling on the left calf

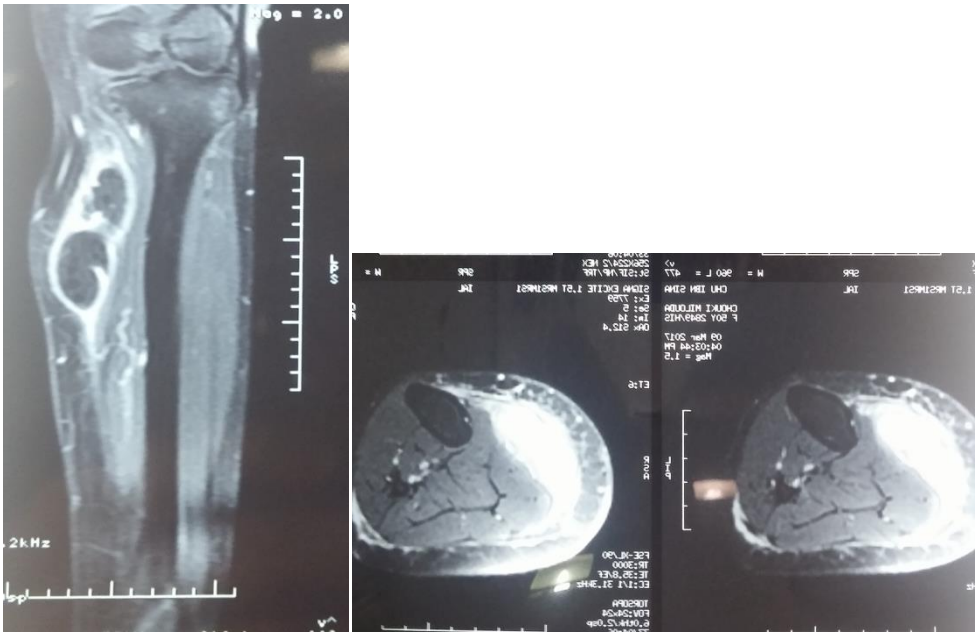

Fig-2: MRI showing subcutaneous oval cystic mass approximately over the left gastrocnemius muscle, containing round-shaped daughter cysts. The cysts seen hypointense in T1 weighted image (left) and hyperintense in T2 weighted image (right)

The mass was operated under general anesthesia. Cyst was removed with 2cm margin of medial gastrocnemius muscle for preventing cyst wall intact (Figs. 3 and 4). Cyst area was irrigated with hypertonic 3% saline after removal of mass to reduce risk of recurrence. Incision was closed primarily after inserting suction drain and patient was discharged after removal of drain on day two. Histological examination of the specimen revealed daughter cysts and fragments of lamellar membrane of the HC. Albendazole therapy, 200mg twice daily, was given for 12 weeks after operation. Clinical, radiological and serologic tests showed no recurrence 4 weeks after operation.

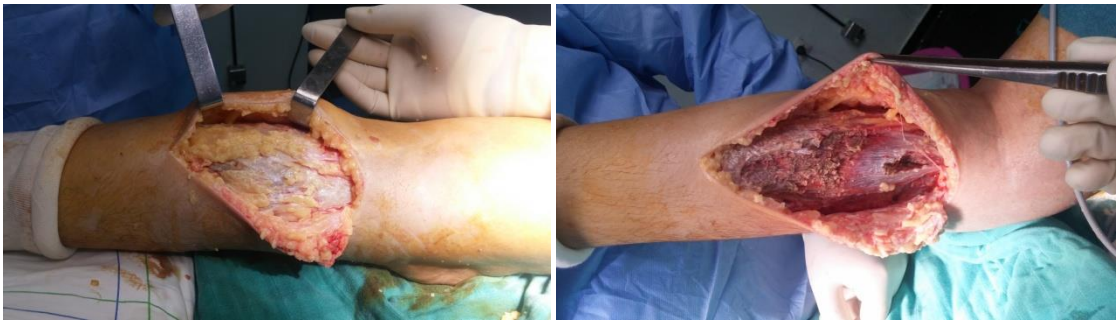

Fig-3: Peroperative image of the cyst

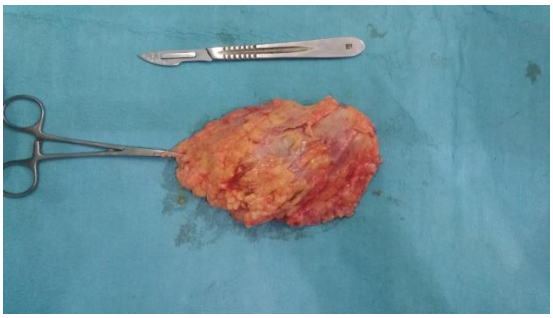

Fig-4: Hydatid cyst excision en bloc

DISCUSSION

The exact mechanism of the subcutaneous location of HC is unclear. Humans are infected directly with parasite eggs released in definitive hosts feces or indirectly by contamination via water, food, or arthropods. Eggs release embryos in the small intestine.

Embryos penetrate the bowel wall and pass to the liver via blood [1,3,5]. In our case, there were no other sites of infection other than subcutaneous tissue. This might have occurred by dissemination through the lymphatic system, bypassing the liver [3, 5] which is the most commonly infected organ (>65%), the second most commonly infected organ is the lung (25%) [1,6]. The cyst is less common in other organs such as the central nervous system, heart, bone (1-4%), spleen (<2%), pancreas (0.2-2%), peritoneal cavity (13%), kidney (3%), adrenal gland, ovary, breast, omentum, retroperitoneum, mediastinum, muscles, pelvic organs, and salivary glands [1-6].

Subcutaneous involvement is usually reported to be caused by iatrogenic spillage of cyst contents to the subcutaneous tissue [2]. Primary subcutaneous involvement is a rare entity in the literature (Incidence: 0.2-2%) [3, 4]. The most common location reported in the literature is the thigh (27%) [2,3,4,7,8].

A slowly growing mass just under the intact skin is the main complaint for subcutaneous HC [4]. The cyst enlarges progressively; thus, the clinical symptoms and signs may appear according to tolerability of the organ involved. Clinical manifestations differ according to the cyst size, location, and condition of the cyst itself [1, 3]. Nevertheless, allergic reactions may occur in case of the cyst rupture [1]. Hydatid disease has a mortality rate of 4% [6].

In endemic areas, the HC should always be taken in consideration when confronted to cystic lesions, among other diseases included in differential diagnosis; those are abscesses, hematomas, mycoses, benign cysts, benign or malignant neoplasms, tuberculosis, and aneurysms [1-3].

Radiological assessment combined with immunohistochemical techniques helps in making correct diagnosis [1-3,6]. MR imaging, computed tomography (CT) scan, USG, and sometimes plain radiography are valuable radiologic tools for assessment of cysts in all organs [1-3,6,9]. MR imaging is superior to others for cutaneous imaging [2]. A pathology-based classification with radiological correlation has been described by Lewall in 1998 [5]. The cyst in our case had daughter cysts, representing a type II cyst.

ELISA and indirect hemagglutination are useful tests for serum screening [1, 3]. It is important to remember that intact, unruptured cysts do not release proteins and do not cause immunological reactions; hence, serology screening can be falsely negative in this condition [1].

Surgery with total resection, if applicable, is the main therapy. During resection, the wall should be kept intact. If not, dissemination of the disease and anaphylaxis may occur. Pregnancy, multiple cysts, unsuitable medical condition, and patient's avoidance are main contraindications for surgery. In such circumstances, puncture, aspiration, injection, reaspiration (PAIR) and medical treatment are methods of choice [1, 5, 10].

The medical treatment is based on Albendazole (10-15 mg/kg/day) which efficiency has been confirmed by many clinical trials [1,11,12]. A complete cure can be achieved in one-third of patients. In considerable percent of patients (30-50%), regression of cyst size is achieved [1,8,11,12].

Minimal treatment period is 3 months. In our case, after diagnosis, the patient used albendazole for 3 months. Serological test was negative, and treatment was stopped with ongoing outpatient follow-ups.

CONCLUSION

Primary subcutaneous hydatid cyst is a rare condition, it should be taken into consideration for differential diagnosis when observing soft tissue mass of the extremities in patients from areas endemic of Echinococcus granulosus. En bloc resection is the treatment of choice. After resection, treatment with benzimidazole regimens is mandatory to prevent recurrences.

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