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Case Report

Giant primary muscular hydatid cyst with a secondary bone localization

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ABSTRACT

Primary musculoskeletal hydatidosis is less frequent than hydatidosis of the parenchymal organs. This localization has been little studied and so there is little information in the literature on the subsequent disease evolution. We present a case of primary hydatidosis of the abductor muscle that came to medical attention very late. After complete surgical removal of the huge mass, a secondary bone localization developed, causing a femoral pertrochanteric pathological fracture. The case described is exceptional in view of both the localization and the great size of the primary multi-lobed muscle hydatid cyst. We underline the difficulties of diagnosis and treatment of both the primary muscle localization and the secondary bone recurrence.

1. Introduction

Human hydatidosis is an endemic infection in the Mediterranean Basin, Central Asia, East Africa, the southern part of South America, and in Oceania.[1–3] The disease is caused by *Echinococcus granulosus*, whose larvae develop in cystic form, most often in the liver and lung.[1,4] Involvement of other organs, and particularly of the muscles, is rare, even in endemic areas.[1,5] The case we describe demonstrates that in cases of onset of a cystic mass in endemic areas for this zoonosis, it is essential to take into account the possibility that it might be echinococcosis, even when the lesion presents in a rare localization. In addition, the peculiar rarity of the size of the hydatid cyst we report, as well as the subsequent bone involvement, make it necessary to consider whether treatment associating surgical excision with anthelmintic treatment is indicated in such cases.

2. Case report

A 79-year-old housewife from southern Italy was admitted to the General Surgery Unit of Bari Polyclinic for a voluminous painful mass in the medial region of the left thigh. The mass had started developing three years before. The patient's clinical history was unremarkable except for intermittent episodes of mild fever and pruritus, not present at the time of admission. Clinical examination showed that the mass had a solid consistency, was localized in the abductor region and measured approximately 40 × 10 cm. Blood tests, X-ray of the thigh, cranial and chest–abdomen–pelvic computed tomography (CT) scans were all within the normal range. Magnetic resonance imagining (MRI) demonstrated a multi-lobed mass extending from the flexor to the abductor muscles. The mother cyst showed a low intensity signal in weighted T1 and T2 and the numerous daughter cysts were surrounded by intracystic fluid (Figure 1).

On the suspicion of a hydatid cyst, more detailed questions were put to the patient, who revealed that she used to keep a dog, which had died 5 years before.

A diagnosis of primary muscular hydatidosis was made and surgical excision was performed. Passing between the abductor muscles (long and short) and the sartorius and gracilis muscles, a voluminous cyst, partially infiltrating the great abductor muscle, was individuated. The cyst was completely removed with particular care to prevent accidental dissemination of the daughter cysts (Figures 2 and 3).

Histopathological examination of the mass, measuring 37 × 12 × 10 cm, revealed that the cyst wall consisted of largely lamellar, acellular, eosinophilic matter. The internal surface was covered by a squamous cuboid epithelium, connective tissue, and a mononuclear inflammatory infiltrate. Indirect hemagglutinin tests for Echinococcus were positive at 1:3200, confirming the diagnosis of hydatidosis, classified as primary in view of the absence of other localizations.

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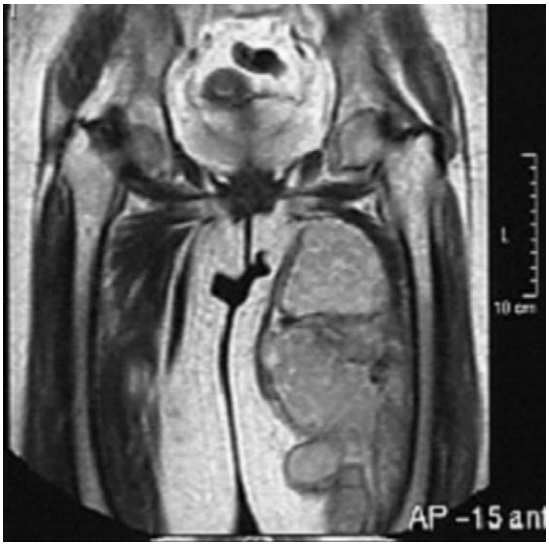

Figure 1. MRI showing the primary muscle hydatidosis localized in the abductor region of the thigh.

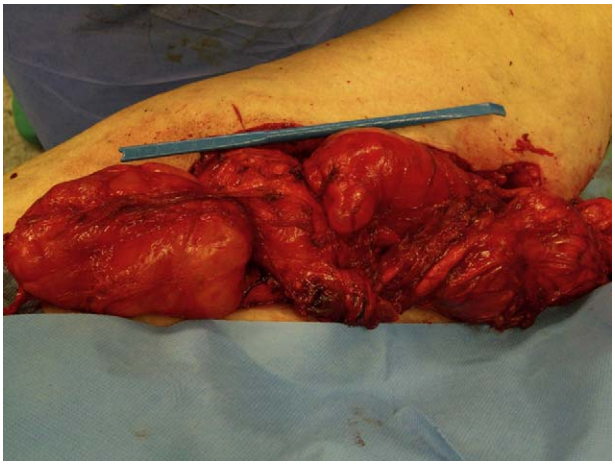

Figure 2. Surgical resection of the giant muscle cyst.

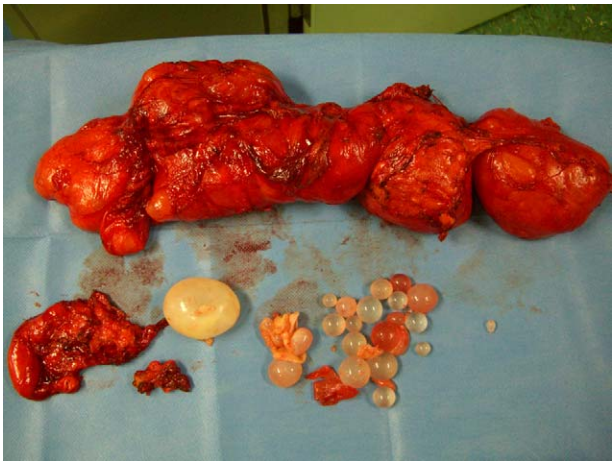

Figure 3. Macroscopic aspect of the hydatid cyst after the surgical excision.

At the 12-month follow-up, the patient showed no signs of reactivation of the disease. However, after 14 months she suffered a trauma of the left thigh that resulted in a pertrochanteric fracture of the femur. Radiography demonstrated the presence of a cystic lesion, revealing a pathological origin of the fracture (Figure 4).

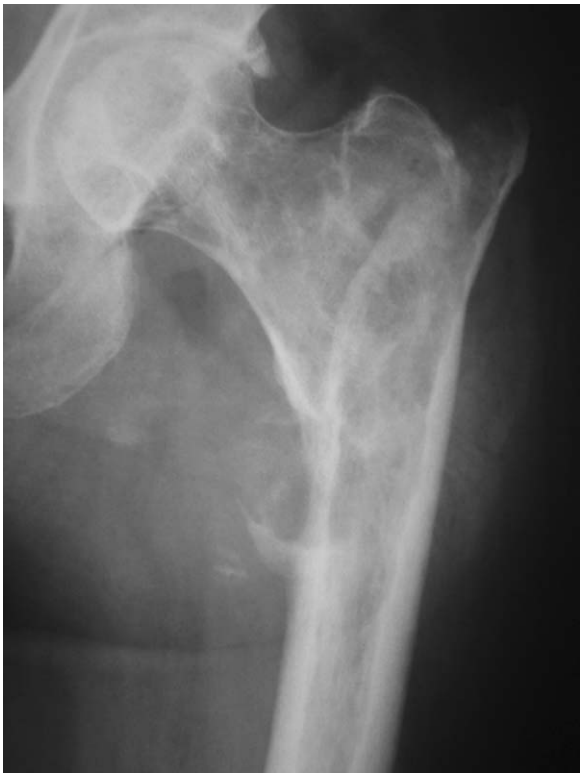

Figure 4. X-ray of the left femur showing the pertrochanteric fracture and the secondary hydatid bone cyst.

Although blood tests were still normal, a secondary localization of the hydatidosis was suspected and so ample local curettage was performed intraoperatively, before placing a femoral prosthesis. Microbiological, serological and microscopic tests of the cyst fluid revealed the presence of bone tissue with a hyaline, germinative membrane, lymphocytes and monocytes, confirming the diagnosis of secondary bone hydatidosis. The patient was prescribed albendazole therapy at a dosage of 10 mg/kg/day, to be taken for 6 months. After surgery and up until the last follow-up at 20 months from the first diagnosis of hydatidosis, no infectious spread or septic complications have been observed, or any other secondary localizations. The patient gave her consent to the reporting of her case.

3. Discussion

We describe a unique case of a giant multi-lobed hydatid cyst localized in the striped muscle of the thigh, complicated after surgical excision by disease recurrence in the adjacent femoral bone.

Cystic echinococcosis is a cosmopolitan parasitosis that is endemic in zones where there is widespread sheep farming.[6] Regions with the highest prevalence are the Mediterranean Basin, the Balkans, the Middle-East and northern Africa, as well as the southern part of South America, Central Asia, Mongolia, Xinjiang and Tibet in China, and East Africa. In Italy, the disease is present in the south and the islands.[7] This parasitic anthropozoonosis is caused by invasion of the tissue by the larvae of *Echinococcus granulosus*, whose intermediate host is the sheep and final host the dog.[8] The larvae develop in cystic form, usually in the liver (60–70%) and lungs, and ultimately cause symptoms due to local compression, as well as systemic symptoms of allergic type. Some less frequent localizations have been reported in the literature, such as the spleen, soft tissues, breast, heart, and extradural