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Primary Muscular Hydatid Disease of the Thigh Complicated by Iliofemoral Deep Vein Thrombosis

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ABSTRACT

Primary involvement of skeletal muscle by cystic echinococcosis is exceptional, even in endemic regions. We describe a 44-year-old man with no relevant medical history who presented with a progressively enlarging, painful mass of the anteromedial left thigh. The mass was complicated by ipsilateral deep vein thrombosis extending from the femoral vein to the external iliac vein. Magnetic resonance imaging showed a thin-walled, multivesicular, honeycomb-like lesion confined to the muscles of the thigh and measuring 26 × 14 × 8 cm, without bone lysis. Hydatid serology was positive, and screening of the liver and lungs was negative. The cyst was removed en bloc through a direct anterior approach without rupture of its wall, with the surrounding soft tissues protected by drapes soaked in hypertonic saline. Albendazole was given as adjuvant medical treatment. This case illustrates that a large intramuscular cystic mass can compress the deep veins and cause thrombosis, and that careful complete excision combined with albendazole is the cornerstone of management.

Keywords

Muscular hydatidosis, Thigh, Deep vein thrombosis.

Introduction

Cystic echinococcosis is a zoonosis caused by the larval stage of Echinococcus granulosus and remains endemic in sheep-farming areas around the Mediterranean basin, including Morocco [1-4]. The liver and the lungs account for most of the lesions. Skeletal muscle is involved in only about 1–4% of cases, and a primary muscular cyst, arising without any visceral focus, is rarer still [5-7]. Several features of muscle tissue, such as repeated contraction and local lactic acid production, are thought to hinder the development of the parasite there [8].

We report a very large primary hydatid cyst of the thigh that was complicated by iliofemoral deep vein thrombosis and treated by en bloc excision without rupture, followed by albendazole.

Case Report

A 44-year-old man with no notable medical history presented with a progressively enlarging mass on the anteromedial aspect of the left thigh, causing mechanical pain. Examination revealed a tense, mobile, tender mass, with dilated superficial collateral veins over it that pointed to deep venous obstruction. Imaging confirmed deep vein thrombosis involving the entire femoral vein and part of the external iliac vein on the same side.

Magnetic resonance imaging (Figures 1 and 2) revealed a large, thin-walled, multivesicular lesion with a honeycomb-like appearance, located strictly within the muscle compartments of the left thigh and measuring 26 × 14 × 8 cm. There was no bone lysis. The appearance suggested a primary muscular hydatid cyst. Hydatid serology was positive. Screening for other sites, in particular the liver and the lungs, found no other lesion.

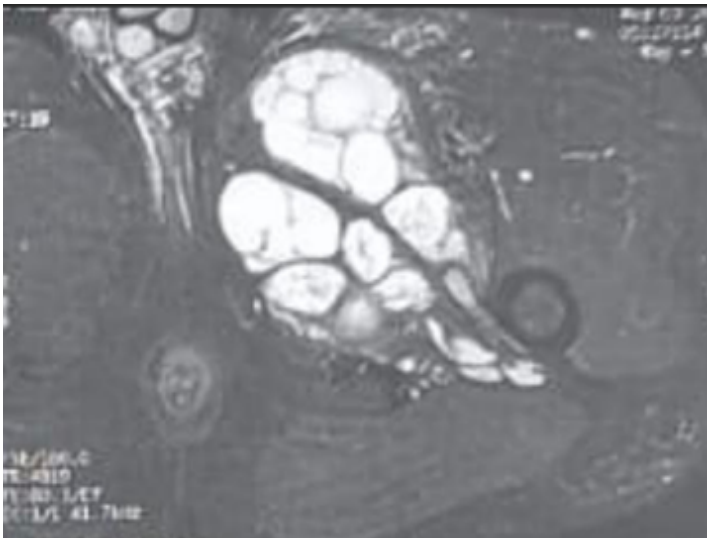


Figure 1: Axial MRI: intramuscular multivesicular cystic lesion with characteristic multiloculated content.

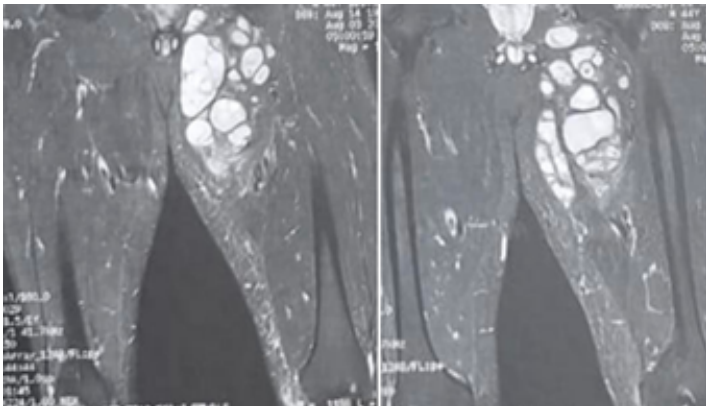


Figure 2: Coronal MRI: extension of the multivesicular cystic lesion within the muscle compartments of the thigh, without bone involvement.

Surgery aimed at removing the cyst in one piece without breaching its wall. A direct anterior approach was used. The surrounding soft tissues were isolated with drapes soaked in hypertonic saline to limit the consequences of any accidental spillage, and the cyst was excised en bloc with its capsule intact. Albendazole was administered as medical treatment.

Discussion

Primary muscular cystic echinococcosis, in the absence of hepatic or pulmonary disease, is exceptional and accounts for fewer than 5% of extrahepatic and extrapulmonary locations, even in endemic areas [6,7]. The oncosphere has to pass through the hepatic and pulmonary filters before it can settle in muscle, which explains why primary forms are so uncommon [5]. The thigh is among the most frequently reported muscular sites, probably because of its bulk and rich blood supply [8-11].

Muscular cysts usually grow slowly and silently, so a painless swelling is the typical presentation and diagnosis is often delayed [9,11]. Our patient was unusual in two respects: the cyst was exceptionally large (26 cm), and it was revealed by a vascular complication. Extrinsic compression of the femoral and iliac veins by the mass is the most likely mechanism of the thrombosis. Vascular compromise by a muscular hydatid cyst has been described before, for example with arterial insufficiency [12], but venous thrombosis is rarely reported and should prompt a search for a compressive mass.

Diagnosis relies on imaging and serology. Ultrasonography is the first-line examination, and MRI is the most informative modality: it shows the multivesicular structure, the low-signal rim on T2-weighted images, and the relationship of the lesion to vessels, nerves and bone [2,8,10]. Serology supports the diagnosis, but a negative result does not exclude it, particularly for extrahepatic cysts [1]. The differential diagnosis includes soft-tissue sarcoma, abscess, organized hematoma, synovial cyst and lipoma [5,8].
Needle biopsy is classically avoided because of the risk of dissemination and anaphylactic reaction [1,2,13]. Complete surgical excision, preferably as a total pericystectomy performed without rupture, remains the reference treatment, with protection of the operative field by scolicidal-soaked packs [1,9,14]. Incomplete excision or intraoperative spillage exposes the patient to local recurrence and secondary echinococcosis. In our patient, the size of the lesion and the venous thrombosis called for meticulous dissection, and the direct anterior approach gave access to the entire lesion.

Albendazole is the benzimidazole of choice and is recommended as a perioperative adjunct, started before surgery and continued afterwards, to lower the risk of recurrence from unrecognized spillage [1,2,15]. Its use alongside surgery is generally accepted for extrahepatic cystic echinococcosis, and combination regimens with praziquantel have been proposed for complicated cases [13-16].

This report has the limitations inherent to a single case. Long-term clinical, imaging and serological surveillance is needed to detect recurrence.

Conclusion

Primary muscular hydatid disease should be considered in any large cystic intramuscular mass in an endemic area, including when it presents with deep vein thrombosis. MRI is key to the diagnosis. En bloc excision without rupture of the cyst wall, with protection of the soft tissues using hypertonic saline, together with albendazole, offers the best protection against recurrence.

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