Primary muscular hydatidosis mimicking soft tissue tumour: a report of five cases

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ABSTRACT

Primary muscular hydatidosis is rare and usually presents as an asymptomatic, slowly growing mass mimicking a soft tissue tumour. Adequate preoperative planning and wide excision is recommended, as incomplete excision may lead to anaphylactic reactions and local recurrence. It should be considered in the differential diagnosis of soft tissue tumours especially in regions endemic for the parasite.

Key words: debridement; drug therapy; echinococcosis

INTRODUCTION

Hydatidosis is a zoonotic infection caused by larvae of *Echinococcus granulosus*, which is endemic in sheep-raising regions such as the Mediterranean and the Gulf states.1–3 Larvae penetrate the intestinal mucosa and enter the portal blood stream; 75% of cases are carried to the liver, 15% to the lungs, and the remainder to the rest of the body (musculoskeletal involvement accounts for only 1 to 4%1–4 of cases). 20% of those infected develop multiple cysts.5 Primary involvement of skeletal muscle is rare and usually involves the pectoralis major, sartorius, biceps brachii, gracilis, supraspinatus, and biceps femoris.6–11

Hydatidosis usually presents as a slowly growing soft tissue mass, mimicking a tumour. Inappropriate biopsy may lead to cyst rupture and a risk of anaphylaxis and recurrence.7,12,13 Detailed radiological studies should be performed to achieve a correct diagnosis. Muscular hydatidosis should be considered in the differential diagnosis of soft tissue lesions in endemic areas.

CASE REPORTS

Case 1

In May 1990, a 33-year-old man presented with a 3-year history of a slowly growing, soft, non-tender mass on
the anterior aspect of his right leg. Laboratory tests and radiography were unremarkable. Ultrasonography revealed a cystic structure of 6x6x5 cm in the tibialis anterior. During surgery the cyst was ruptured, despite careful technique, and the wound was washed out with hypertonic saline. Microscopy revealed a fibrous laminar and inner germinal layer consistent with hydatid disease. The patient had a recurrence at the same location 8 months later. The intradermal Casoni test and serologic test (complement fixation test of Weinberg) were positive; all other laboratory investigations were normal. Both thoracic and abdominal CT were unremarkable. At operation a yellowish mass was dissected from the femoral vein and excised en bloc with the gracilis, with a wide margin from vastus medialis. One of the main branches of the femoral nerve supplying the vastus medialis was also excised. The femoral vein was damaged during dissection and repaired. The cavity was irrigated with hypertonic saline solution. Microscopically, the specimen showed areas of mixed inflammatory cells, multinucleated giant cells, and epithelioid histiocytes. Cuticular membrane with a lamellar appearance was identified. The patient was given 4 courses of albendazole over 4 months. At the 24-month follow-up she had no major functional deficit or evidence of recurrence.

Case 2
In September 2000, a 37-year-old man presented with a one-year history of a painless mass in his left gluteus maximus. His range of movement was not affected. Computed tomography (CT) and magnetic resonance imaging (MRI) revealed a hypodense, cystic, nodular mass measuring 4x6x7 cm. T1-weighted images revealed a low signal intensity mass with a low intensity rim (Fig. 1a). Post-gadolinium T1-weighted images showed considerable enhancement in the capsule and septa but no enhancement in the centre (Fig. 1b). Laboratory tests and CT of the thorax and abdomen were all normal. A large multilobulated mass with a fibrous capsule was excised. Histopathology showed the cyst wall had a basophilic lamella, scoleces, areas of fibrosis, necrosis, and a reactive pericystic structure with histiocytic cells and degeneration of the adjacent muscle fibres. He was given 4 courses of albendazole (10 mg/kg in one intake for 30 days) with 15-day intermissions between each course. He had no evidence of recurrence at the 41-month follow-up.

Case 3
In May 2004, a 63-year-old woman presented with an 8-year history of a tender, firm, erythematous mass on the inner aspect of her left upper thigh, which had become painful 5 months earlier. She had undergone excision of a lesion in the same location 28 years previously but records of any pathological examination of the lesion were unavailable. Both T1- and T2-weighted MRI images revealed a multilocular lesion measuring 20x12x15 cm with a low signal intensity rim. Both the Casoni and Weinberg tests were positive; all other laboratory investigations were normal. Both thoracic and abdominal CT were unremarkable. At operation a yellowish mass was dissected from the femoral vein and excised en bloc with the gracilis, with a wide margin from vastus medialis. One of the main branches of the femoral nerve supplying the vastus medialis was also excised. The femoral vein was damaged during dissection and repaired. The cavity was irrigated with hypertonic saline solution. Microscopically, the specimen showed areas of mixed inflammatory cells, multinucleated giant cells, and epithelioid histiocytes. Cuticular membrane with a lamellar appearance was identified. The patient was given 4 courses of albendazole over 4 months. At the 24-month follow-up she had no major functional deficit or evidence of recurrence.

Case 4
In October 2002, a 75-year-old woman presented
with a 5-month history of severe pain and swelling in the posterior aspect of her right thigh. She had undergone excision of a lesion 30 years previously but reports of any pathological examination of the lesion were unavailable. Ultrasonography and MRI showed a multivesicular cyst measuring 20x6.5x5 cm confined to the biceps femoris. It was hyperintense on T2-weighted images and had an incomplete rim with a low signal on both T1- and T2-weighted images. T1-weighted images showed internal cysts within the mother cyst, with low signal intensities (Fig. 2). Cranial, thoracic, abdominal, and pelvic CT did not provide any evidence of organ involvement. The lesion was excised en bloc along with a portion of the biceps femoris, semitendinosus, and semimembranous muscles. A collection of pus was also debrided. The cavity was irrigated with hypertonic saline solution. No micro-organisms could be cultured, but the symptoms resolved with empirical antibiotic therapy. Histopathological examination of the removed tissue showed a cyst with cuticular membrane and scoleces, which are typical of hydatid disease. She was given 4 courses of albendazole courses and showed no signs of recurrence during 32 months of follow-up.

Case 5
In April 2003, a 34-year-old woman presented with a 5-month history of Echinococcus granulosus infestation in the posteromedial aspect of the right crus after a tru-cut biopsy performed in another institution. MRI revealed a cystic mass measuring 4x3x2 cm confined to the gastrocnemius, which was hyperintense on T2-weighted images with an incomplete, hypointense rim (Fig. 3a). Post-contrast images revealed enhancement at the periphery in T1-weighted images (Fig. 3b). Cranial, thoracic, and abdominal CT did not show any organ involvement. The lesion was excised along with a portion of the gastrocnemius. Pathological examination found a cystic structure with a cuticular membrane, scoleces, and infiltration of eosinophils. She received 3 courses of albendazole and was followed up for 38 months without recurrence.

DISCUSSION

Hydatidosis usually involves the liver and lungs because of their blood-filtering functions but can also affect other organs and the musculoskeletal system.
Multiple cysts may occur in up to 20% of those infected; cysts may lodge in the musculoskeletal system via vascular seeding into highly vascularised areas such as the vertebrae, long bone epiphyses, ilia, skull, and ribs.\textsuperscript{4,14-16} Primary muscular hydatid cysts are rare (accounting for 3% of all patients with hydatidosis\textsuperscript{9}) because of the contractility and high lactic acid levels in muscle.\textsuperscript{7,12,17,18} Involvement of the musculature of the chest wall, pectoralis major, sartorius, biceps brachi, supraspinatus, and biceps femoris have been reported.\textsuperscript{7-10} 13 cases of muscular hydatidosis have been reported in the paravertebral pelvic gluteus and lower extremities.\textsuperscript{2} Tibialis anterior involvement has not been reported before. Muscular hydatidosis usually occurs as isolated lesions without hepatic or pulmonary lesions.\textsuperscript{12} None of our 5 cases had involvement of the liver, lungs or any other internal organs based on CT, ultrasonography, and radiography of their thoracic and abdominal cavities (Table).

Hydatidosis mimics primary tumours of the musculoskeletal system and is often misdiagnosed, because of the lack of specific clinical symptoms and radiological signs.\textsuperscript{4} Only 2 of 13 such cases had positive serological tests.\textsuperscript{7} Seronegativity does not exclude the diagnosis, but seropositivity may be helpful in recurrent cases.

Soft tissue masses and calcification may be seen in the radiographs of 38% of patients.\textsuperscript{19,20} None of our patients had soft tissue calcification. Ultrasonographical appearances pathognomonic for hydatid cysts include echogenic hydatid sand (the ‘snowflake sign’), unilocular cysts with daughter cysts (‘honeycomb sign’), and cysts with a floating detached laminated membrane (‘waterlily sign’).\textsuperscript{21} In our series, ultrasonography was performed in 2 cases and both revealed multivesicular cysts.

MRI is useful for the differential diagnosis of hydatid disease and other soft tissue malignancies. The lesion usually has a high signal intensity on T2-weighted images and low intensity on T1-weighted images with a low intensity rim on both T1- and T2-weighted images.\textsuperscript{6,17,22} This rim sign is not specific for hydatid disease and can be seen in other lesions with a fibrous capsule or a calcific rim.\textsuperscript{12,14} Peripheral enhancement reflects the vascularity of the pericyst.\textsuperscript{12} The cystic structure with internal cysts was observed in 3 of our patients. Daughter cysts are the most characteristic feature of hydatid disease and imply viability, but 30% of hydatid cysts may lack daughter cysts.\textsuperscript{12}

Incisional biopsy and marginal excision are contraindicated in hydatid disease because the cysts contain foreign proteins and scoleces that may precipitate anaphylaxis.\textsuperscript{7,12,18} Chemotherapy alone (such as benzimidazole) is not usually sufficient in muscular hydatidosis. The curative method is complete and wide excision, similar to oncologic procedures.\textsuperscript{3,13} Total removal of hydatid lesions may be difficult to achieve because of the neighbouring neurovascular structures, which make preoperative evaluation crucial. When a wide excision is infeasible, meticulous drainage and adjuvant hypertonic saline irrigation is preferred.\textsuperscript{19} Intra-operative irrigation with scolecidal agents such as hypertonic saline prevents recurrence from inoculation of scoleces during excision.\textsuperscript{7} Leakage of cyst contents and dissemination of viable scoleces increase the recurrence rates.\textsuperscript{7,12} In our series, the excision was wide in 3 and marginal in 2 cases. Marginal excision was performed because of the proximity of the lesion to vascular structures, and hypertonic saline irrigation was used as an adjuvant treatment. Recurrence occurred in one patient 8 months later and he recovered with further wide excision and albendazole therapy.

Pharmacological complementary treatments (3 or

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**Table**

Characteristics of the 5 cases of primary muscular hydatidosis

<table>
<thead>
<tr>
<th>Patient No.</th>
<th>Sex/age (years)</th>
<th>Location</th>
<th>Serology</th>
<th>Radiology*</th>
<th>Previous Surgery</th>
<th>Surgery</th>
<th>Adjuvant treatment</th>
<th>Albendazole (courses)</th>
<th>Recurrence</th>
<th>Follow-up (months)</th>
</tr>
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<tbody>
<tr>
<td>1</td>
<td>M/33</td>
<td>Tibialis anterior</td>
<td>+ve</td>
<td>US</td>
<td>No</td>
<td>Marginal excision (but cyst ruptured)</td>
<td>Hypertonic saline</td>
<td>4</td>
<td>Yes</td>
<td>54</td>
</tr>
<tr>
<td>2</td>
<td>M/37</td>
<td>Gluteus maximus</td>
<td>-ve</td>
<td>CT, MRI</td>
<td>No</td>
<td>Wide excision</td>
<td>-</td>
<td>4</td>
<td>No</td>
<td>41</td>
</tr>
<tr>
<td>3</td>
<td>F/63</td>
<td>Gracilis</td>
<td>+ve</td>
<td>MRI</td>
<td>Yes</td>
<td>Marginal excision</td>
<td>Wide excision</td>
<td>4</td>
<td>No</td>
<td>24</td>
</tr>
<tr>
<td>4</td>
<td>F/75</td>
<td>Biceps femoris</td>
<td>-ve</td>
<td>US, MRI</td>
<td>Yes</td>
<td>Tru-cut biopsy</td>
<td>-</td>
<td>4</td>
<td>No</td>
<td>32</td>
</tr>
<tr>
<td>5</td>
<td>F/34</td>
<td>Gastrocnemius</td>
<td>-ve</td>
<td>MRI</td>
<td></td>
<td></td>
<td></td>
<td>3</td>
<td>No</td>
<td>38</td>
</tr>
</tbody>
</table>

* US denotes ultrasonography, CT computed tomography, MRI magnetic resonance imaging.
REFERENCES


