PRIMARY HYDATID DISEASE IN LUMBAR MUSCLES

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The authors report a case of primary hydatid disease in the lumbar muscles of a 40-year-old male patient. The rarity of this disease in our regions and the low incidence of this location make primary diagnosis difficult. The tumor had been treated elsewhere five years previously by means of simple excision. Recurrence of the lesion was diagnosed five years after the first surgery. Wide excision of the cyst and pericyst with a 3.5-cm security margin was performed. Six years after the last surgery, no recurrence has been detected.

Keywords : hydatid cyst ; lumbar muscles ; muscular tumor.
Mots-clés : kyste hydatique ; muscles lombaires ; tumeur musculaire.

INTRODUCTION

Hydatid disease is a zoonotic infection caused by larval forms of small tapeworms of *Echinococcus granulosus*. This parasite inhabits the small intestine of carnivores such as dogs and wolves, definitive hosts in the cycle (6, 7). Man becomes infested by ingesting ova from the feces of these carnivorous animals (7). The disease is ubiquitously distributed with increased occurrence in the Mediterranean countries, East Africa, South America, Russia and Australia (5, 6, 7).

Because of their role as capillary filter stations, the liver and lung are primarily affected in this disease (1, 6, 7). Musculoskeletal involvement is found in only 1-4% of cases (6).

CASE REPORT

A 45-year-old man, with a previous history of a hydatid cyst in the left lumbar longissimus muscle treated by simple excision in another hospital, presented a recurrence of the lumbar tumor five years after the first surgery. On examination, the non-tender mass was 3 × 3 cm, adherent to underlying tissues, and there was no pain or neurological signs. CT scan and magnetic resonance imaging revealed multiple cysts subcutaneously and in the longissimus muscle at the L4-L5 level with no bone alterations. T1-weighted images (fig. 1) showed multiple cysts with their matrix of slightly lower intensity than the muscle, with a hypointense heterogeneous pattern of signal intensity in daughter cysts. The intramuscular

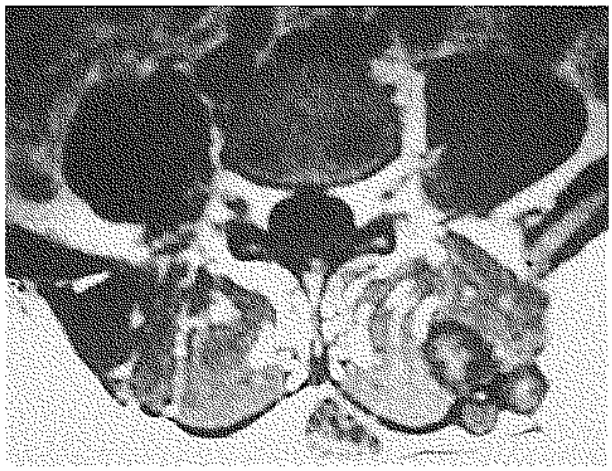

Fig. 1. — MRI findings : T1-weighted image shows multiple cysts with matrix of slightly lower intensity than muscle, with a hypointense heterogeneous signal intensity pattern in daughter cysts. The intramuscular cysts show the "rim sign" : a low signal intensity surrounding the cyst and representing the pericyst.

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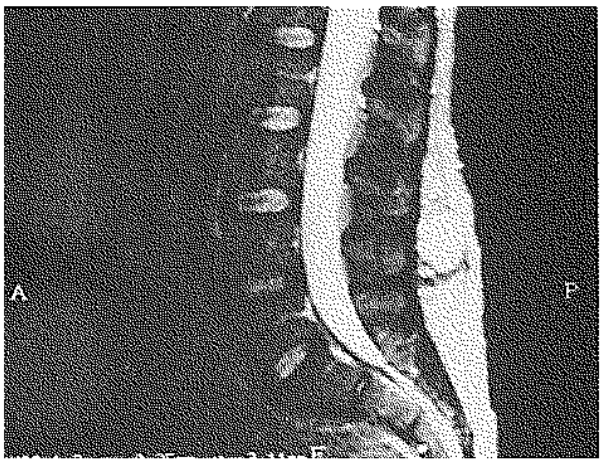

Fig. 2. — On this T2-weighted image the mass shows a significant increase in signal intensity, and the daughter cysts are isointense with the matrix.

cysts showed the "rim sign" : a low signal intensity surrounding the cyst, representing the pericyst. On T2-weighted images (fig. 2) the mass showed a significant increase in signal intensity, and daughter cysts were isointense with the matrix. The lesion was labelled as multiple hydatid cysts. No alterations in electroneurography-electromyography were found. No evidence of hepatic or pulmonary involvement was observed. The serologic test for hydatidosis (indirect hemagglutination test) was positive at 1/1280 dilution. The results of routine laboratory tests were within normal range.

Wide excision with a 3.5-cm safety margin was performed, followed by 20% saline solution washing. The subcutaneous cysts and the longissimus muscle cysts had a diameter of 2-3 and 3-4 cm, respectively. The cysts were surrounded by a thick fibrous capsule, the pericyst, and on opening, cystic cavities were found to contain clear fluid and hydatid vesicles with scoleces. Microscopic study confirmed the diagnosis of hydatid disease. No antiparasite drug was administered.

Six years after the last surgery, no recurrence has been detected.

DISCUSSION

Hydatid disease of the soft tissues is very infrequent. Owing to its dangerous character, careful treatment must be carried out to avoid anaphylactic shock and the liberation of contaminant larval forms that could lead to systemic complications and recurrence. Thus, correct initial diagnosis is necessary to perform curative surgery in a single intervention.

Cystic echinococcosis has hepatic (65%) or pulmonary (15%) manifestations, and musculoskeletal involvement is found in only 1-4% of cases (6). In fact, musculoskeletal lesions of cystic echinococcosis occur most commonly as isolated findings and without concomitant hepatic or pulmonary involvement (1, 6) ; this was also true in our patient Nevertheless, involvement of other organs, such as the liver and lung, should be ruled out with appropriate diagnostic methods (sonography and chest radiography, respectively) (6). In the literature, most cases of muscular hydatid disease are associated with skeletal lesions (6) ; this was not true in our patient.

Several patterns of disease have been recognized using various imaging techniques. These include the unilocular cyst, the multivesicular lesion and the atypical complex or solid lesion (1, 5). The multivesicular lesion is characteristic but not pathognomonic of hydatid disease, and it presents multiple daughter cysts inside the mother cyst (1, 5).

Endovesicular daughter cysts, present in our case, are reported as exceptions in musculoskeletal cystic echinococcosis by some authors (2, 6, 9). The magnetic resonance (MRI) signal intensity pattern of the daughter cysts depends upon their contents and may also vary depending on whether the cysts are dead or alive, because they disintegrate at death so that the production of hydatid fluid ceases (5, 6). The presence of bacterial infection or abundant intracystic debris and inflammatory changes may alter the typical cystic morphology transforming it into a complex or solid lesion mimicking a tumor (5). Our case presented a T1-weighted hypointense signal intensity pattern in daughter cysts, corresponding to the bibliographic references (2). However, the daughter cysts were typically indistinguishable from the matrix on T2-weighted MR images (2). The "rim sign", which was present in our case on the T1-weighted MR images, as a low signal intensity surrounding

the cyst and representing the pericyst may be useful to differentiate hydatid cysts from other lesions. However, this sign is not specific for hydatid disease and may be present in other lesions that contain a fibrous capsule (2, 5). Other diagnostic means such as fine needle aspiration cytology could be dangerous because of anaphylactic reactions and should be avoided (1). However, some authors (2) think this risk is probably lower than originally thought.

Serological tests and eosinophil count are not positive for echinococcosis in all cases (1, 5), thus they may be helpful but not definitive in differential diagnosis.

Adjuvant administration of benzimidazole derivatives preoperatively and for about 3 months postoperatively is advocated by some authors (2). However, clear recommendations are lacking in the guidelines for the treatment of echinococcosis published by the World Health Organization (6). Moreover, some authors report that treatment of soft tissue hydatid disease with mebendazole did not reduce the size of the lesion, and surgical excision was still necessary (5). Surgery constitutes the treatment of choice. The high recurrence rate requires broad excision of the cyst and pericyst with a safety margin. Chemotherapy, albendazole 15 mg/kg/day or mebendazole 50 mg/kg/day for three cycles of 28 days with surveillance for hepatic toxicity, must be administered in all cases in which it is not possible to perform radical surgery (3, 4, 8).

In conclusion, the rarity of the disease in our region and the low incidence of this location make preoperative diagnosis of hydatid disease difficult to achieve in the lumbar area. Previous knowledge of this disease and patient history, MRI findings, serological tests and eosinophil count may help to establish the primary diagnosis, to avoid biopsy and to prevent anaphylactic complications and recurrence.

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SAMENVATTING

F. GARCÍA-ALVAREZ, J. TORCAL, J. C. SALINAS, A. GÜEMES, A. C. NAVARRO, R. LOZANO. Primaire hydatide kyste in de lumbale spieren.

De auteurs beschrijven een hydatide kyste in de lumbale spieren bij een 40-jarige patiënt. De zeldzaamheid van deze aandoening in onze regionen en de zeldzame lokalisatie maakten een diagnose moeilijk. De primaire tumor werd elders behandeld en het recidief werd 5 jaar later vastgesteld. De kyste werd met een ruime marge van 3.5 cm geëxiseerd. Tot 6 jaar later werd geen recidief vastgesteld.

RÉSUMÉ

F. GARCÍA-ALVAREZ, J. TORCAL, J. C. SALINAS, A. GÜEMES, A. C. NAVARRO, R. LOZANO. Kyste hydatique au niveau des muscles lombaires : présentation d'un cas.

Les auteurs présentent un cas de kyste hydatique primaire observé au niveau de la musculature lombaire

chez un patient âgé de 40 ans. La rareté de cette pathologie dans nos régions s'ajoutant à la rareté de cette localisation musculaire rend le diagnostic difficile. La tumeur avait été traitée ailleurs, 5 ans plus tôt, par excision simple et la récidive a été diagnostiquée 5 ans après cette première intervention. Le patient a été traité par excision large du kyste avec une marge de sécurité de 3,5 cm. Aucune récidive n'a été décelée avec six ans de recul.