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Emergency Radiology

Bone Hydatidosis: A Rare Entity Difficult to Diagnose That Should Not be Overlooked

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Case Report

Bone hydatidosis is a rare entity accounting for 0.2 to 4% of all cases reported in the literature, half of which concern the spine. It follows the haematogenous dissemination of the echinococcus multilocularis or granulosis parasite. It is often secondary to liver or lung involvement, but more rarely primary. The clinic is non-specific, with insidious and progressive development of the disease, which explains why it is so rare among children. The average age at which bone hydatidosis is discovered is 52, following complications such as neurological disorders or pathological fractures. Diagnosis can be difficult in non-endemic regions because it is not systematically evoked. It is therefore usually carried out during or after surgery. Surgical treatment is the gold standard, in association with adjunctive medical treatment. We report here a rare case of primary involvement of bone hydatidosis.

Keywords: Bone Hydatidosis, Echinococcus Granulosus/Multilocularis, Primary Bone Involvement, Haematogenous Dissemination, Surgical Treatment.

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INTRODUCTION

Abstract

Hydatidosis or echinococcosis is a pathology secondary to parasitic infection by echinococcus granulosis or multilocularis.

Humans are accidental intermediate hosts following ingestion of parasite eggs. Bone involvement is unusual and rare, secondary to haematogenous dissemination with a history of hepatic or pulmonary hydatid cysts.

More rarely, bone involvement is the patient's first known site of infection. The absence of an endemic context further complicates the positive diagnosis, which may not be the first to be evoked, if at all.

A large number of differential diagnoses may then be evoked. The Mediterranean basin and North Africa are endemic areas for the disease. We report the rare case of bone hydatidosis first discovered.

OBSERVATION:

We report the case of a 35-year-old man who presented with chronic back pain for 3 months, complicated by paraparesis for 1 week and paraplegia for 6 hours.

A CT scan of the thoracic spine revealed a left paravertebral cystic formation adjacent to D7 and D8, with enlargement of the foramen conjugated and endocrinal extension responsible for compressing the medullary cord.

These formations are enhanced peripherally after injection of contrast medium. Anteriorly, there is collapsed lung parenchyma.

Bone window, reveals vertebral condensation associated with osteolytic lesions and breakage of the bone cortex. Spinal bone hydatidosis with endocanal extension has been suggested. The patient reported contact with dogs and hydatid serology came back positive.

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figure 2: axial and coronal bone window CT images showing vertebral condensation and lysis

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DISCUSSION

Bone hydatidosis is a rare disease location, representing between 0.2 and 4% of all hydatid sites [1]. It most often occurs in people living in endemic countries with a history of liver or lung disease. More rarely, it is a primary manifestation of the disease.

Humans are accidental hosts infested by tapeworm eggs released in the faeces of definitive hosts such as dogs (multilocularis echinococcosis) or foxes (granulosis echinococcosis), or following consumption of uncooked offal from infested intermediate hosts, the most common being sheep [2].

The embryonic form then passes through the intestinal mucosa into the microcirculation and then into the portal system to be intercepted at the first hepatic or second pulmonary relay, where cysts form [2].

The cyst wall comprises three layers, two produced by the parasite (an inner proligeral layer and an intermediate acellular layer), and one outer granulomatous and adventitial layer formed by the host [3].

The bone site is often secondary to haematogenous dissemination with a primary hepatic or pulmonary site in the patient's history; more rarely, it represents a primary site of the disease [4].

Other ubiquitous secondary sites have been reported, including the brain, spleen, kidney, pancreas, peritoneum and pelvis [5].

In bony locations, vertebral and costal involvement is the most common, with sub-periosteal, trans-ligament development, preserving the intervertebral disc [6-8].

Vertebral involvement is most often dorsal, followed by lumbar, sacral and cervical [3]. Peripheral bone involvement is rarer and occurs from the bone metaphysis with secondary extension to the diaphysis and epiphysis [9].

In bone hydatidosis, the outer layer of the cyst, formed by granulation tissue and an adventitia, is absent, which explains the usually multi-loculated appearance and the serology, which is more frequently contributory compared with other locations [10, 11].

Progression is insidious and slow, which explains the late diagnosis, most often in adulthood, with an average age of 52 years [12].

Clinical symptoms include non-inflammatory soft tissue swelling or tumefaction mimicking a cold abscess or tumour [9].

A spinal syndrome or neurological disorders such as paraparesis or paraplegia may occur in cases of spinal cord compression or associated endo canal extension [8, 13].

Osseous hydatidosis is diagnosed during or after surgery, with an additional diagnostic delay of around 6 months due to its rarity [14].

However, on radiological appearance, it should be suspected in the presence of a history of hepatic or pulmonary hydatidosis in endemic countries, and based on hydatid serology, which is most often profitable in bony locations [11, 14].

On radiological examination, the standard radiograph shows an osteolytic lesion with more or less peripheral otosclerosis, with no periosteal reaction.

When a periosteal reaction is present, it is generally secondary to a pathological fracture. Cortical lysis with extension to the adjacent soft tissues is possible and is responsible for the swelling seen clinically.

A CT scan can assess the extent of the lesion, using millimetric helical acquisition with multi-planar reconstruction.

It will reveal a multi-loculated lesion responsible for more or less significant bone lysis, preserving the intervertebral discs, and will specify the extent of the involvement as well as the intra-canal extension.

Endo-canal extension through the foramina of conjugation or secondary to vertebral bone lysis determines the prognosis.

MRI will enable precise assessment of spinal cord and nerve root involvement. Abdominal ultrasound and chest X-rays can be used to look for hepatic and thoracic locations, associated with bone involvement in 45% of cases [8].

There are many differential diagnoses of bone hydatidosis (spondylodiscitis, plasmacytoma, metastasis, giant cell tumour, aneurysmal cyst, essential cyst, fibrous dysplasia, haemophiliac pseudotumours, chondromyxoid fibroma, osteomyelitis) [15]. Hence the importance of anamnestic correlation and serology.

Puncture of the cyst for diagnostic purposes is contraindicated as it exposes the patient to an increased risk of dissemination, sensitisation and anaphylactic shock [4, 9].

Surgical treatment is the treatment of choice and must meet the surgical criteria for a locally malignant lesion. Healthy resection margins of 1 to 2 cm are recommended to avoid recurrence, which averages 80% [4, 9, 16].

Neurological damage has a poor prognosis because it persists postoperatively [9]. Surinfection of the surgical site is the most feared post-surgical complication [4].

Medical treatment with albendazole alone or in combination with praziquantel for 6 to 9 months is indicated as a complement to surgical treatment to reduce the risk of recurrence or postoperative dissemination [17].

CONCLUSION

Bone hydatidosis is a rare and unusual condition, which results in an additional diagnostic delay of around 6 months. Anamnestic, radiological and serological data are vital in making the diagnosis. The prognosis depends on the degree of neurological involvement and the quality of surgical resection to avoid recurrence.

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