Rare disease

Back pain caused by a pseudo-tumorous vertebral collapse: atypical presentation of primary vertebral hydatidosis

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Summary

Hydatidosis, also known as echinococcosis, is a rare but serious parasitic disease in endemic areas. Primary spinal location is extremely rare. This case report describes a rare instance of hydatid cyst that caused severe and progressive low-back pain and neurologic dysfunction. Spine MRI showed a unique vertebral collapse of Th12 body with multicystic lesions filling the spinal canal. In addition, hydatidosis serodiagnostic test was positive at 1/725. Treatment depended on the actual surgical removal of the cysts. Surgery consisted in excision and extirpation of the cysts, associated with decompressive laminectomy. The diagnosis was confirmed on the basis of histological results. No coincidental hydatid visceral involvement was found. Anthelmintic drugs (Albendazole) were promptly given before surgery for a long period. The outcome was satisfactorily marked by total regression of the motor deficit and sphincter disorders.

BACKGROUND

Due to the serious location of spinal hydatid cysts, a better understanding of the clinically challenging disease is intended. As a vertebral collapse rarely reveals hydatidosis, we considered justifiable to report this case so that we can discuss the clinical presentation, diagnosis and surgical treatment of vertebral hydatid cyst.

CASE PRESENTATION

A 54-year-old man, farmer, coming from a geographic distribution with a high prevalence in sheep-raising region, presented with a 6-month history of low-back pain and sciatica in the left side. The pain was associated with difficulty in walking for the last 2 weeks, numbness, weakness in his left lower limb and sensory loss. The patient confessed that he had been suffering from urinary incontinence for 2 months. On physical examination, lumbar spinal movements were painful with spasm of the musculature. The pain radiated to the left buttock. Tenderness on the thoraco-lumbar junction was demonstrated. Neurologic examination revealed 3/5 quadriceps weakness as well as diminished sensation on the bilateral Th11 to L1 dermatomes. His reflexes were decreased. Anal sphincter tone was weak. Bilateral Babinski sign and perianal hypoesthesia were also noted. The liver was not enlarged and no masses were palpable in the abdomen.

INVESTIGATIONS

Laboratory investigations showed an increased erythrocyte sedimentation rate at 58 mm/h and a C reactive-protein at 32 mg/l with a normal white cell count. Eosinophilia was not found.

The antero-posterior radiograph view of the thoracic spine showed paravertebral shadows with rounded

margins at the level of Th12 vertebra, that on the left being larger than that on the right (figure 1A).

The lateral view confirmed that the body of Th12 vertebra was partly collapsed, and an irregular density was visible within it (figure 1B). The intervertebral disk spaces were well preserved.

Thoraco-lumbar MRI demonstrated septated multilocular hydatid cystic lesions involving the body of Th12. We noted a low-intensity signal on T1 weighted images and a high-intensity signal on T2 weighted images, which were not enhanced after gadolinium injection. The posterior arch was preserved (figure 2A,B). MRI also showed an extension into the Th12-level spinal canal and into the paraspinal soft tissue (figure 3A,B). The lesion was initially regarded as a hydatid cyst. In addition, the hydatid serodiagnostic test by specific ELISA was positive at 1/725. Chest x-ray and ultrasound examination of the abdomen did not reveal the primary source of the infestation by *Echinococcus granulosus*.

DIFFERENTIAL DIAGNOSIS

Radiological findings in this disease can be mistaken for a number of other conditions, mainly tuberculous infection and malignant disease of bone. Spinal involvement appearance can suggest a number of different diagnoses such as plasmacytoma, brown tumour of hyperparathyroidism, chondromyxoid fibroma, fibrous dysplasia, giant cell tumour, aneurysmal bone cyst, myelomatosis or lymphoma.

TREATMENT

The patient underwent exploratory surgery. Via a posterior approach, decompressive Th11, Th12 and L1 laminectomies were performed at the same session,

Figure 1 (A) The antero-posterior radiograph view of the thoracic spine: lytic lesion involving the vertebral body of Th12 vertebra and adjacent paravertebral sharp opacity (arrows). (B) Lateral view of the thoracic spine: heterogeneous body of Th12 vertebra which is partly collapsed.

Figure 2 MRI sagittal T2 weighted images (A) and sagittal T1 weighted images (B). Multi vesicular hydatid cyst hyperintense on T2 images and hypointense on T1 weighted images.

after this, a lot of pearly white intradural extramedullary daughter cysts were removed totally with their capsule. The body of Th12 vertebra was exposed and scraped. Management also included total excision of the multiple epidural and paraspinal multilocular cysts. The appearance was characteristic of hydatid disease. Then the cavity was irrigated with 20% hypertonic saline solution. Postoperative course was uneventful. Subsequent pathological examination of the specimen confirmed the

presence of scolices and an amorphous acellular cuticular lesion compatible with hydatid cyst. To prevent any recurrence, oral albendazole 15 mg/kg was prescribed for 6 months.

OUTCOME AND FOLLOW-UP

The patient recovered from surgery free of new deficits. His low-back pain disappeared after the operation. He was able to walk without assistance at the time of discharge.

Figure 3 T2-weighted MRI in sagittal (A) and axial (B) slices: Multilobar cystic mass extending in the paraspinal soft tissue and in the epidural space compressing the spinal cord (arrow).

Second-month follow-up neurologic examination was normal. Response and toxicity related to therapy were closely monitored by biochemical and radiological follow-up. The hydatid cysts had totally disappeared in the fourth month postoperative MRI. Seven months later; the patient remained completely asymptomatic, free of any sign of recurrence and the hydatid serodiagnostic test by ELISA was at 1/80.

DISCUSSION

Hydatid disease is caused by an infection with tapeworm larvae of *Echinococcus granulosus*. The liver is the most commonly involved site (75%), followed by the lungs. Bone involvement is rare (0.2–4%), affecting the spine in almost half of cases. Incidence ranges from 1.1% in Australia to as high as 14% in Tunisia. Thoracic vertebrae are the most commonly involved, usually between Th4 and Th10 (80% of cases).¹ The lumbar spine is involved in 18% of cases. Cervical spine is the least commonly involved area.^{1–5}

Primary spinal hydatid disease is rarely encountered. It is thought to occur through portovertebral shunts.² The center of the vertebral body is the first site to be involved and subsequently to extend asymmetrically to the entire vertebra or to the posterior elements,⁶ then to the extradural or paravertebral spaces.^{2,7} Five types of spinal hydatidosis have been distinguished: primary intramedullary hydatid cysts; intradural extramedullary hydatid cysts; extradural intraspinal hydatid cysts; vertebral hydatidosis and paravertebral hydatidosis.^{8,9}

In very advanced stages, compression fractures occur as does disk involvement.

Usually, clinical manifestations are not typical. Back pain is present in 85% of cases. Pain results from a compressive radiculopathy or from erosion of the vertebral corpus.^{2–5} Laboratory evaluation may be helpful for diagnosis: the most common findings include hyper-eosinophilia.⁶

In addition, serologic tests are positive in 40% of extra-hepatic lesions which limits their use in the diagnosis or in the follow-up of primary bone disease.²

The radiological changes in spinal hydatid disease may be considered in relation to the stage at which they are observed. In the early stages, there are no changes in the general shape or size of the body of the vertebra, nor is there any change in the intervertebral spaces; there is no bony or periosteal reaction and no calcification in the surrounding area. In the late stages, destructive changes develop slowly but aggressively and may be seen in the vertebral bodies. However, the absence of collapse of the vertebral bodies, the escape of the intervertebral disks and the presence of eburnation are the most characteristic findings.²

Very rare previous reports of univertebral collapse presenting as pseudo-tumour form revealing primary hydatidosis have been disclosed in the literature. To the best of the authors' knowledge, 40 cases have been published to date. El Quessar *et al*,⁹ in a series of eight cases of spinal hydatidosis, reported only one vertebral collapse. Maamar *et al*¹⁰ described a case of a woman with bilateral sciatic pain and a third lumbar vertebral collapse shown on x-ray. CT scan of lumbar spine showed lysis of the L3 body mimicking an osteolytic tumour. MRI confirmed the collapse of L3 with multiple cysts in the vertebral body and surrounding tissues suggesting hydatidosis.¹⁰ Two adjacent vertebral bodies can be affected like in the report by Mahi *et al*¹¹ where total destruction of the body and of the posterior arch of L2 and L3 vertebrae and of L2-L3 disk space was noted.

Several vertebrae may be affected by hydatid cysts. Semlali *et al*¹² reported the case of a young male admitted for quadriplegia secondary to cervical hydatid disease with extension towards the retropharynx. Cervical axial CT scan showed a space-occupying multilocular mass causing multiple collapses of the bodies of C1, C2, C3 and C4 vertebrae. Basak *et al*¹³ presented also a case of a patient with hydatid disease in Th11-L1 vertebral bodies and involvement of the bilateral psoas muscles.

Learning points

- ▶ Hydatid cyst should be considered in the differential diagnosis of low-back pain and collapsed vertebrae, especially when treating patients from areas where *Echinococcus granulosus* is prevalent. It is also very important to take an accurate occupational history.
- ▶ The severity of vertebral echinococcosis is related to the neurological complications and therapeutic problems especially in advanced stages.
- ▶ Management problems and pitfalls must be discussed as well as current therapeutic alternatives, results and outcome.
- ▶ The success in the treatment of vertebral hydatidosis represents a challenge due to its invasive nature; consequently, its poor prognosis has been compared with that of malignant spinal tumour.
- ▶ Prophylaxis is more effective than treatment. Elimination of the hydatid disease can only be achieved by breaking the life cycle of the parasite in its host through the implementation of active nationwide preventive measures.

Competing interests None.

Patient consent Obtained.

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