CASE REPORT

Hydatid disease drug therapy primarily to reconstruction of a multilevel thoracolumbar vertebral lesion

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Accepted: 30 May 2011

Summary We report, a very unusual case of multilevel vertebral hydatidosis adjacent to the thoracolumbar junction, without concomitant chord compression. Two months after initiating oral antiparasite treatment, the patient underwent resection of the lesion using a posterior approach, medullary decompression, and a T11–L3 instrumented arthrodesis. Arthrodesis via the anterior approach was performed at a later stage. In addition to its diagnostic value, this case raises renewed discussion about single-level lesions given their rarity: their indication for preoperative medullary angiography, their indication for circumferential surgery, the timing of medical treatment, and the strategy to implement for the residual lesions.

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Introduction

Echinococcus infection, or hydatid disease, is the most frequent and the most severe of human cestode infestations in the world. Humans are the accidental hosts of Echinococcus granulosus. Canidae are the final hosts and livestock, generally sheep, are the intermediate hosts. The adult tapeworm lives in the digestive tube of canines where it lays eggs that are evacuated in the feces. These feces contaminate the environment of livestock, which ingest the eggs. These eggs develop into larvae that are ingested by canines that eat livestock. Humans are contaminated by ingesting the eggs when they are in contact with infested dogs, but the parasite’s life cycle cannot be completed in humans.

The preferential locations are the liver (60–70%) and lungs (10–15%). Bone involvement remains rare (0.5–2%), with vertebral locations observed in half of these cases, generally located in a single vertebra [1–4], although multvertebral involvement has been described [5–8]. These cases are marked by neurological complications, relapses, and considerable bone destruction. The severity of this disease is related to the risk of neurological complications, given the medullary compression caused by the cysts and the deformity induced by the bone destruction.

Observation

Clinical observation

A 36-year-old male from Algeria, with no particular medical history, initially consulted for chronic lumbar pain. The
Vertebral hydatid disease, chemotherapy prior to reconstruction surgery

The patient’s history revealed a context of sheep herding. The mechanical type lumbago had been progressing over 10 years and had initially been attributed to the harsh physical conditions of his work. X-rays taken in 2005 had been judged normal. Beginning in August 2007, the symptoms had modified, with intensification of lumbar pain with more of an inflammatory aspect and alteration of the patient’s general condition (weight loss, 6 kg). No neurological deficit was noted, but fatigability when walking was described by the patient.

The clinical examination at admission to the department found spinal stiffness predominating in flexion and right lateral inflexion, associated with pain upon palpation of the T11, T12, and L1 spinous processes. There was also a right paravertebral mass, fluctuating and painful upon palpation. This pain irradiated in a poorly systematized manner at the groin fold and the right thigh. The neurological exam found abolition of the patellar reflex and nonsystematized hypoesthesia of the right foot.

Complementary exams

The X-rays taken at admission showed effacement of the right vertebral pedicles and posterior vertebral scalloping from T12 to L2 (Figs. 1 and 2). A CT scan (Fig. 3) and an MRI (Fig. 4) were taken in an emergency setting, demonstrating a multiloculated mass from T11 to L2, associated with lysis of the right posterior components, and compressive epiduritis. There was also invasion of the right psoas muscle and the right paravertebral muscles. The staging examination (cranial and thoracoabdominal-pelvic CT and MRI) demonstrated no other location. A medullary arteriography was performed before treatment so as to identify the artery of Adamkiewicz for planning the surgical strategy.

With these findings, the diagnoses of tumor, tuberculosis, and hydatidosis were suggested. Arguing in favor of tuberculosis was the patient’s history (his geographic origin, the progressive nature of the disorder, and the inflammatory aspect of the pain) and the frequency, but the imaging findings argued against this hypothesis because of absence of the following signs: discal involvement, vertebral collapse, irregularity of the vertebral plateaux, and Pott abscess. Arguing in favor of a tumor was the patient’s history, but the diagnosis of secondary tumor was ruled out because of the absence of a primary tumor, with the diagnosis of chondrosarcoma possible because of the frequency, the calcifications on the X-ray, and the lobulated aspect and T2 hyperintense signal on MRI, but the absence of cortical destruction argued against this. A giant-cell tumor could be suggested because of the patient’s age and the frequency, but the absence of major involvement and vertebral body collapse as well as the hyperintense T2 signal were arguments against this diagnosis.

Figure 1  Preoperative antero-posterior X-Ray view.

Figure 2  Preoperative lateral X-ray.

Figure 3  Preoperative frontal reconstruction CT scan.
The diagnosis of hydatidosis was confirmed a few days later by positive hydatid blood results.

Treatment

Per os albendazole antiparasite treatment (400 mg twice a day while monitoring liver biology) was initiated immediately to reduce the infectious and inflammatory phenomena, aiming to obtain preoperative systemic impregnation in hopes of reducing the risk of systemic dissemination and local recurrence.

Surgery took place after 2 months of antiparasite treatment, scheduled at this time despite the presence of neurological signs on the initial examination because these neurological signs were minimal, particularly the absence of motor involvement; progression with treatment, instructions for strict rest, and brace wear gave favorable results. Posterior segment arthrodesis extending from T11 to L3 was performed through the enlarged posterior approach and included laminectomy and costotransversectomy.

During exposure, substantial mobility of the T12, L1, and L2 posterior vertebral arches was observed. The spine was first stabilized on the left side, then a massive resection removing the right paravertebral muscles from T11 to L3, as well as the T12, L1, L2, and L3 posterior vertebral arches was performed. Dissection of the dural sac and the right T11 to L2 roots allowed us to identify the hydatid lesion, stemming from the right costotransverse joint from T12 extending into the right anterolateral part of the spine opposite T12, L1, and L2 and in front of the L3 transverse process. The cysts, which were extradural, were then injected with hypertonic saline and aspirated to prevent contamination; aspiration produced a thick yellowish fluid corresponding to a long-standing hydatid fluid. Once débridement had been achieved, the spine was instrumented and stabilized on the right side.

Postoperative recovery was uneventful, with no neurological complications, and walking was resumed on the 2nd postoperative day with a thoracolumbosacral brace.

The anatomopathological examination of the operative specimens demonstrated a multilocular cystic formation, substantially reorganized by a foreign body macrophagic reaction. The presence of anhistic membrane fragments confirmed the diagnosis of hydatidosis. No scoleces or pro- ligorous membrane were observed.

The postoperative follow-up CT scan verified that the implants were properly positioned, but small nodules in the right psoas muscle, seemingly long-standing, were identified. Simple monitoring was decided for these residual lesions.

Given the instability of the spine related to the resection of the posterior arches at the thoracolumbar seat of the pathology and its multi-level vertebral extension, to maintain sagittal and frontal balance, a complementary left-sided thoraco-phreno-lumbotomy arthrodesis was performed with intersomatic graft using autologous iliac bone and T11–T12, T12–L1, L1–L2, and L2–L3 intervertebral cages (Figs. 5 and 6). The absence of vertebral body involvement and the patient’s good response to antiparasite treatment, ensuring little risk of recurrence, argued in favor of the intersomatic implants.

Postoperative recovery was uneventful and the duration of oral antiparasite treatment was set at 1 year in the multi-disciplinary meeting. At 20 months of follow-up, the patient remains with no particular signs.

Discussion

An interesting aspect of this case is that no biopsy was done because the ethnic and clinical context, the radiological aspect, and the serology converged sufficiently. In most cases, it is the intraoperative pathognomonic aspect (presence of hydatid vesicles) that corrects any diagnostic
The originality of this case also stems from the circumferential approach, with most authors content to perform posterior decompression surgery and excision [10–12] or more rarely a single anterior surgery [13], although for less extensive lesions. The series are retrospective and only studied a small number of patients with varying involvement, precluding a conclusion on the superiority of any particular strategy. Here, the therapeutic strategy comprised three axes, which required combined posterior and anterior surgery: medullary and radicular decompression, total excision of the lesion, and spinal reconstruction. The multilevel vertebral aspect of the disease was the source of unique problems compared to the many cases of single-vertebra involvement described in the literature: the extent of the release and reconstruction to perform and foraminal multilevel vertebral involvement requiring preoperative medullary arteriography, although it was located on the right.

The recurrence rate in this disease is, unfortunately, high [13], but most authors only begin chemotherapy after surgery, contrary to the case described here. Finally, the strategy adopted can be discussed with regard to the psoas lesions. The risk of recurrence may have been increased by leaving seemingly old lesions, but aggressive surgery in the spinal root area included a risk of iatrogenic neurologic damage. Moreover, albendazole in prolonged adjuvant treatment can sterilize residual lesions [14]. In addition, knowing the potential problems with the anterior approach revisions and the major risk of recurrence of hydatidosis, it seems reasonable to preserve an approach.

**Conclusion**

Adjacent multilevel vertebral involvement in spinal echinococcosis is remarkable for its rarity and its severity. The lesson of this observation to apply, even in cases of single-vertebra involvement, is neoadjuvant chemotherapy, the need for wide resection readily including an anterior approach, and the absolute requirement of reconstruction aiming to maintain vertebral stability in the frontal and sagittal planes.

**Disclosure of interest**

The authors declare that they have no conflicts of interest concerning this article.

**References**


