

Medullary Compression Syndrome Revealing Vertebral Medullary Hydatidosis: A Case Report

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Abstract

Case Report

Spinal cord compression syndrome caused by vertebral medullary hydatidosis is a rare but serious condition requiring precise diagnostic and therapeutic approaches. This article examines the crucial role of magnetic resonance imaging (MRI) and computed tomography (CT) in characterizing lesions, assessing their extent, and monitoring postoperative outcomes in patients with this pathology. With a focus on vertebral medullary surgery, the authors explore the challenges associated with this condition, including the complexity of surgical management and the high risk of recurrence. A better understanding of this pathology and the adoption of early preventive strategies are essential to improve clinical outcomes in patients with this syndrome.

Keywords: Spinal Cord Compression Syndrome, Hydatidosis, Magnetic Resonance Imaging, Vertebro-Medullary Surgery.

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INTRODUCTION

Osseous hydatidosis is a rare condition, accounting for 0.5 to 2% of cases despite its endemic nature in Maghreb countries [1]. Spinal locations are the most common, likely due to the rich vascularization of the spine (44% of cases) [1, 5]. It often involves spinal cord affection, justifying the term 'vertebral medullary hydatidosis.' Diagnosis is often delayed due to its lack of specificity and clinical latency characteristic of this condition. It relies on a combination of clinical, laboratory, and radiological findings, with confirmation typically requiring histopathological examination. Imaging plays a crucial role in confirming the diagnosis, assessing lesion extent, and monitoring progression [2]. Here, we present a case of primary vertebral medullary hydatid cyst complicated by paraparesis.

OBSERVATION

A 9-year-old child, with no significant medical history, from a rural background, presented to the emergency department with paraparesis and inability to stand. Despite maintaining general health, a dorso-lumbar CT scan (Figure 1) was requested, revealing cystic formations at the L1-L2 levels, centered on the left pedicles and transverse processes of L1 and L2, with lytic features. These cystic formations were confluent,

hypodense, with thin walls, and enhanced after contrast injection. They filled the spinal canal at the level of L1-L2-L3. A spinal MRI showed a multi-loculated cystic lesion involving the anterior and posterior epidural spaces, as well as the left middle and posterior arches, extending from D12 to L4, resulting in compression and medullary compromise at the L1 level, initially suggestive of a hydatid cyst (Figure 2).

Further investigations, including abdominal ultrasound and thoracic radiography, for visceral localizations, particularly hepatic involvement, were negative.

The patient underwent surgical treatment with partial pericystectomy, removing a multivesicular cyst with curettage of the bone up to the healthy zone, where daughter vesicles were not observed, followed by extensive lavage with hypertonic saline solution. Postoperatively, the patient received medical treatment with albendazole, one tablet daily for four months.

The patient's neurological status showed gradual partial spontaneous recovery, with improvement in muscle strength leading to standing and walking. However, follow-up revealed recurrence of hydatid disease after 20 months.



Figure 1: Abdominal CT scan in axial and sagittal views with bone and chest window settings shows lytic lesions centered on the spinous processes and left pedicles of L1 and L2, with endocanal extension at the level of L1-L2-L3

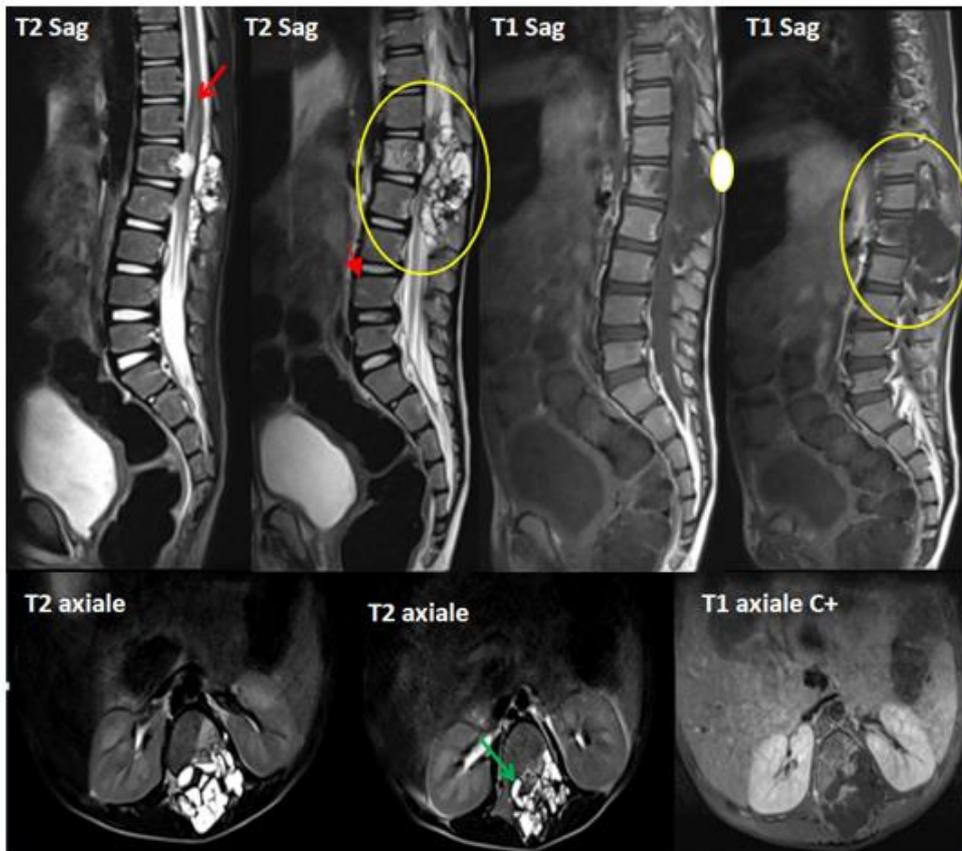


Figure 2: Sagittal and axial spinal MRI images with T2-weighted, T1-weighted sequences, and post-contrast injection demonstrate a multi-loculated cystic lesion involving the anterior and posterior epidural spaces, as well as the left middle and posterior arches, extending from D12 to L4 (yellow circle), resulting in spinal cord compression and compression of the cauda equina roots (green arrow), with medullary compromise at the level of L1 (Red arrow)

DISCUSSION

Osseous hydatidosis is rare even in endemic regions. Spinal involvement is the most common, with the dorsal spine being the most affected, accounting for 80% of spinal locations, while the lumbosacral localization constitutes 18% of cases [2]. It primarily occurs in young individuals [3]. Clinically, the general condition remains preserved for a long time, with neurological symptoms often manifesting as back pain, motor and/or sensory disturbances, and sphincter dysfunction related to spinal cord compression. Our patient presented with bilateral paraparesis and inability to stand upright, with progressive worsening and clinical examination revealing absent osteotendinous reflexes in both lower limbs, with muscle weakness estimated at 3–5. Biological diagnosis relies on immunoelectrophoresis of proteins, which remains the preferred test. It is often coupled with hemagglutination or immunofluorescence [3]. The development of *Echinococcus granulosus* occurs through multivesicular budding from the initial vesicle, without any encystment. Therefore, there is no true osseous cyst. The bone shape remains preserved for a long time, and the disc remains intact initially [2], with involvement of the dural sheath being late. In the absence of superinfection, there is no osteocondensation or periosteal reaction.

Conventional radiological exploration is the first-line examination. It helps to specify the topography, relationships, multiplicity of lesions, and guides further radiological investigations [5]. In the spine, the involvement is generally dorsal (80%), rarely lumbosacral (18%), and exceptionally cervical [2, 3], consistent with our case where the lumbar level was affected. The vertebral body is more frequently affected than the posterior arch, as in our case where the lesion is centered on the left pedicles and transverse processes of L1 and L2, which appear lytic. Lesions may involve multiple contiguous vertebrae. The number of affected vertebrae varies between 1 and 5 according to Tazi [9] and Ouestadi [5], in our case, the number of affected vertebrae is 4. The lacunar image of osteolysis is the most characteristic sign. The usually confluent lacunae are of variable size, poorly defined without periphery condensation, often multilocular, separated by septa [4, 8]. The cortical bone, external morphology of the bone, and intervertebral disc are preserved for a long time. Later, there is extension to neighboring bones: adjacent vertebra, rib, and iliac bone, vertebral body collapse, and disc involvement. Costovertebral involvement strongly suggests hydatid cyst (KH) [2, 7]. Intraspinal involvement is suspected if there is an increase in the interpedicular distance at one level, widening of the neural foramina, or pedicular lysis [5]. Lateral views are suggestive if they reveal "scalopping" of the posterior vertebral wall or involvement of the posterior arch computed tomography (CT) clearly depicts lytic bone lesions, varying in delineation, vertebral collapse, and disc involvement when present. It evaluates the extension of lytic lesions to adjacent ribs, strongly

indicative of hydatid disease, as well as the extension of the cystic process into the soft tissue of the lateral and mediastinal muscles. CT reveals characteristic signs suggestive of the hydatid origin of the cystic lesion, notably the multilocular or multivesicular appearance and the presence of a serpinginous intracystic membrane with a distinct wall. Intracanal extension is more challenging to confirm without intrathecal injection [6] but may be suspected in cases where the spinal cord is displaced or compressed by perimedullary collection.

MRI is the preferred imaging modality for exploring spinal and medullary lesions, especially in cases of spinal cord compression. It allows for precise localization of the lesion (osseous, epidural, intradural, intramedullary) and assessment of extension into the surrounding soft tissues. Additionally, MRI helps identify signs of spinal cord compromise, guides surgical intervention, and plays a crucial role in postoperative monitoring. Hydatid cysts typically appear as hypointense on T1-weighted images and hyperintense on T2-weighted images, usually unaffected by contrast injection. However, enhancement of cyst walls and septa may occur if the cysts are altered [7]. In spinal hydatidosis, the intervertebral disc is often preserved, unlike in infectious spondylodiscitis, which serves as a differential diagnosis. In our patient, analysis of the vertebral disc revealed a normal signal, allowing exclusion of spondylodiscitis. MRI also revealed daughter cysts in the anterior and posterior epidural spaces, as well as in the left middle and posterior arches extending from D12 to L4, causing compression and medullary compromise at the L1 level. However, MRI is less adept at evaluating osseous involvement compared to CT.

Ultrasound can assist in diagnosis when anatomical conditions permit. It can reveal fluid collections and help identify other visceral locations [4]. Differential diagnosis includes bone tumors and tuberculous osteitis [4].

The treatment of spinal hydatidosis relies on surgery, which should be as radical as possible. It is often challenging, occasionally mutilating, and does not guarantee freedom from recurrence. The indication for surgery and its outcomes depend on the extent and location of the lesions, as well as the presence or absence of complications. Medical treatment has not yet proven effective and is reserved for inoperable or poor prognosis cases, as well as adjuvant therapy to surgery [9]. Recurrences are very common, with 30 to 40% of cases experiencing recurrence in spinal locations and a mortality rate ranging from 3 to 14% [1].

CONCLUSION

Vertebral medullary hydatidosis is a rare condition even in endemic regions. It is highly aggressive, with a significant risk of spinal cord compression due to lesion extension and near-constant

recurrence. Diagnosis is often delayed, even in endemic areas. This highlights the need to intensify efforts to improve diagnostic performance, relying on MRI and CT scans to characterize lesions, establish a precise lesion assessment, and monitor postoperative evolution. Its prognosis remains poor, underscoring the importance of prevention efforts.

REFERENCE

1. Abdelmoula Cheikhrouhou, L., Amira, C., Chaabouni, L., Ben Hadj Yahia, C., Montacer Kchir, M., & Zouari, R. (2005). L'hydatidose vertébrale: apport de l'imagerie moderne et actualités thérapeutiques. A propos d'un cas. *Bulletin de la Société de pathologie exotique*, 98(2), 114-117.
2. Amrani, M., Zouaidia, F., Belabbas, M. A., Labrousse, F., Catanzano, G., & Elhachimi, A. (2000). Hydatidose: à propos de quelques localisations inhabituelles. *Med Trop*, 60, 271-272.
3. Brian, J., Richez, P., Belliol, E., Barea, D., Raillat, A., & Salamand, P. (1998). Atteintes ostéo-articulaires d'origine parasitaire: L'échinococcose osseuse. *Journal de radiologie (Paris)*, 79(11), 1351-1357.
4. Bronstein, J. A., & Klotz, F. (2005). Cestodose larvaires. *Encycl Med Chir*. 8-511-A-12, 18 p.
5. Chiraoui, N., Adil, A., & Kadiri, R. (1993). Aspects radiologiques de l'hydatidose vertébro-médullaire. *J Radiol*, 74, 621-628.
6. Rezig, A. L. (2002). Hydatidose osseuse. *Revue du rhumatisme (Ed. française)*, 69(8), 835-841.
7. Gennari, A., Almairac, F., Litrico, S., Albert, C., Marty, P., & Paquis, P. (2016). Spinal cord compression due to a primary vertebral hydatid disease: a rare case report in metropolitan France and a literature review. *Neurochirurgie*, 62(4), 226-228.
8. Marouf, R. (2014). Kyste hydatique à localisation costo vertébrale. *Pan African Medical Journal*, 19(1), 343.
9. Pandey, M., & Chaudhari, M. P. (1997). Primary hydatid cyst of sacral spinal canal: case report. *Neurosurgery*, 40(2), 407-409.