

Spinal Cord Compression by Vertebral Hydatid Cyst in a Case Report

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Abstract

Background: Spinal hydatidosis is a parasitic infection caused by Echinococcus granulosus. Though rare, it is particularly severe due to its high morbidity and mortality. It is often diagnosed at an advanced stage when neurological complications arise. The absence of specific symptoms and the prolonged clinical latency contribute to delayed diagnosis. Aim: To highlight the diagnostic challenges and management strategies of spinal hydatidosis, emphasizing the role of imaging, surgical intervention, and adjuvant albendazole therapy in improving patient outcomes, while acknowledging the high recurrence rates and the need for preventative measures. Imaging and Diagnosis: Imaging plays a critical role in the diagnosis of spinal hydatidosis. Standard radiography may reveal lacunar, areolar images with irregular septations and no periosteal reaction. MRI is the gold standard, offering clear visualization of the hydatid abscess and its characteristic fluid-like signal, which is useful for assessing locoregional extension. Laboratory tests are limited in their diagnostic value. Treatment: Surgical intervention remains the only definitive treatment, aiming for a complete and wide excision of the lesions, akin to the approach used for malignant tumors. However, total eradication can be challenging, as the surgical field may appear clear, while the bone remains infiltrated with microvesicles that lack distinct boundaries. Adjuvant medical therapy with Albendazole is recommended to improve outcomes. Despite the poor prognosis, early detection and timely intervention can lead to long-term remission or even cure, particularly in localized cases.

Keywords

Spinal Cord Compression, Echinoccocus Granulosis, Vertebral Hydatidosis, Bone Hydatid Cyst

1. Introduction

Hydatid cyst disease results from the development of the larval form of *Echino-coccus granulosus* in humans. Vertebral hydatidosis is a rare condition, accounting for 0.5% to 2% of cases, despite its endemic nature in countries around the Mediterranean basin [1]. Spinal localizations are the most severe and the most common among bone involvement in echinococcosis. While the disease can affect any bone, the spine is the most frequently involved, accounting for 42% of cases. The dorsal region is most commonly affected (56%), followed by the dorsolumbar (11%) and lumbar (27%) regions [2]. Exceptionally, the disease affects the cervical spine or lumbar region. The condition is thought to be related to the vascular richness of the spine.

The diagnosis is often delayed due to the nonspecific and insidious nature of its presentation, typically manifesting as progressive spinal cord compression. This diagnostic challenge has been significantly improved with the advent of magnetic resonance imaging (MRI). Surgery remains the primary treatment, aiming to relieve spinal cord compression and obtain histopathological confirmation. However, the prognosis remains poor due to the high risk of disabling neurological complications and frequent recurrences. The objective of this article is to highlight the diagnostic challenges and management strategies of spinal hydatidosis, emphasizing the role of imaging and surgical intervention.

2. Case Report

A 37-year-old female patient, B.M, with no significant medical history, presented with a one-month history of progressive spinal cord compression syndrome. Clinical examination revealed pyramidal signs, Fränkel grade A flaccid paraplegia, hypoesthesia of legs, and sphincter dysfunction.

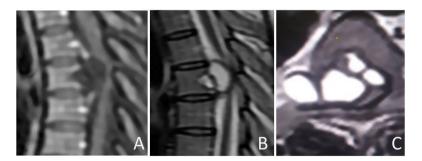


Figure 1. MRI suggests a T1 hyposignal process in the sagittal section (A), T2 hypersignal in sagittal section, axial section (B, C), multi-vesicular, with extension to the body and the posterior arch of T4. This process extends inside the medullary canal, resulting in spinal cord compression.

Spinal imaging was performed, including computed tomography (CT), which revealed an osteolytic lesion at the T4 vertebral body extending into the spinal canal. Magnetic resonance imaging (MRI), performed 2 months later, demonstrated a multi-vesicular cystic lesion with signal characteristics identical to cerebrospinal fluid (CSF), appearing hypointense on T1-weighted sequences and hyperintense on T2-weighted sequences. The lesion involved both the vertebral body and posterior arch of T4, with extension into the vertebral canal, leading to significant spinal cord compression (Figure 1(A), Figure 1(B); sagittal T1, T2, Figure 1(C); T1 axial MRI).

The neurosurgical indication was selected, the risks and benefits of the surgery were explained to the patient who accepted them and she gave her consent to the opportunity to share her case in the context of scientific research.

The surgical procedure began with a midline incision made at the T4 level following pre-incisional skin preparation and fluoroscopic identification. A laminectomy of T3, T4, and T5 exposed a large cyst compressing and surrounding the spinal cord. The cyst was incised, its contents aspirated, and the cyst wall excised, with a sample collected for analysis. The surgical site was irrigated with saline, hemostasis was achieved, and a Redon drain was placed before closure. Post-operative recovery was favorable, with progressive neurological improvement followed by regained sphincter control. Histopathological examination confirmed the diagnosis of a hydatid cyst. Recurrence rates following surgical excision vary widely, depending on the lesion size, the quality of surgical excision, and the medical treatment. Long-term complications primarily include persistent paraplegia, hypoesthesia, sphincter dysfunction, and pain. In our case, after 10 months of treatment with albendazole, the patient is fully recovered (**Figure 2(A)-(C)**; postoperative MRI control, D; intraoperative).

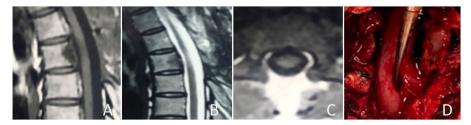


Figure 2. Post-operative MRI showing satisfactory decompression of the dural sac, shriveled hydatid cyst in sagittal view T1 in (A), T2 in (B and C). In (D) intraoperative appearance of the hydatid cyst after laminectomy; the left anterolateral cyst with aspiration of the contents of the cyst and excision of its membrane.

3. Discussion

Hydatidosis is a zoonotic infection caused by the larval stage of *Echinococcus granulosus*, a cestode parasite. The disease primarily affects the liver and lungs. Vertebral hydatidosis is a rare manifestation, accounting for only 1% - 2% of all cases, even in endemic regions. It has a latent and insidious evolutionary character. It can remain silent for decades. This character may explain the rarity of diagnosis in children; the disease usually manifests itself in adulthood around 40 by non-specific functional signs or, more frequently, by local complications with the general condition. However, it remains the most frequent and severe form of bone hydatidosis, representing approximately 45% of osseous involvement [3]. Vertebral

hydatidosis is often a primary infection, likely due to the rich vascular supply of the spine and the possibility of paradoxical embolism.

A sudden increase in intra-abdominal pressure can lead to the retrograde flow of blood from the portal venous system into the spinal venous plexus. This reversal of venous circulation allows the hexacanth embryo to bypass the hepatic and pulmonary filters, reaching the vertebral spongy tissue [4].

Vertebral hydatidosis predominantly affects young individuals between 10 and 30 years of age, with a slight male predominance (60%) [5]. Spinal involvement is often primary, whereas spinal cord lesions occur secondary to the migration of hydatid vesicles through the neural foramina or as a result of bone destruction. Faced with the mechanical resistance of bone, the larvae proliferate through exogenous vesiculation, progressively invading the subperiosteal spaces [6].

Clinically, the condition is characterized by a spinal syndrome associated with progressive neurological deterioration, ultimately resulting in chronic spinal cord compression. From a clinical point of view, there is no contracture or segmental rigidity. Gibbosity is rare in contrast to Pott's disease. The ossifluent paravertebral abscess is very common in the dorsal stage where it is most often posterior and not very bulky. It is seen in one in three cases; in the lumbar stage, a psoas abscess can be found on palpation. The progression of the abscess has been very slow and torpid. It has little migration and very rarely fistulates. Neurological disorders are minor in the beginning but can progress slowly to paraplegia whose prognosis remains poor, especially in the dorsal. The frequency of neurological deficits remains high. Computed tomography (CT) typically reveals hydatid osteopathy, presenting as well-defined, hypodense lesions of variable size and shape, with a density lower than that of bone [7]. The prolonged preservation of the overall shape of the vertebra and the thickness of the disc, despite sometimes extensive lesions, is an important element of diagnostic orientation.

Magnetic resonance imaging (MRI) is the diagnostic modality of choice for evaluating spinal cord compression. It provides a detailed assessment of the spinal canal contents, determines the extent of spinal cord involvement, and clarifies the relationship between hydatid vesicles and the dural sheath [8]. It makes it possible to highlight the typical fluid signal of hydatic vesicles, hyper-intense in T2 weighted sequence, hypo-intense in T1 unmodified after gadolinium injection. In spinal locations, MRI is an excellent means to evaluate the height extent of lesions as well as the intra-canal extension of the daughter vesicles. In addition to the typical multi-vesicular appearance, a pseudo-tumor aspect can be observed. Histopathological examination confirms the diagnosis of hydatidosis. Analysis of paraffin-embedded sections stained with hematoxylin-eosin-saffron (HES) reveals the characteristic trilaminar structure of the hydatid cyst wall: an outer adventitial layer (pericyst), a middle cuticular layer that is lamellar, poorly stainable, and acellular, and an inner germinative layer containing nucleated cells. The germinative layer produces daughter vesicles through budding, which eventually harbor scolices. Over time, the proliferation of scolices within these daughter cysts contributes to cyst enlargement [9].

The primary treatment for spinal hydatidosis is surgical intervention, which aims to eradicate both spinal and extraspinal lesions; it aims at the complete excision of hydatid lesions without, however, being able to ward off the risk of recurrence in extensive lesions. This complete eradication is met by microscopic vesicular infiltration of the bone tissue, giving the disease its character of "local malignancy." The surgical strategy requires a true representation of the lesion and its relationships with neighboring organs. This planning makes it possible to predict the extent of the resection, the sacrifices and the reconstruction that it requires.

Multidisciplinary management is sometimes necessary. Bone resection is necessary to move to a healthy area. Reconstruction by graft with osteosynthesis is often necessary since the parasitic lesion does not contradict the use of an inert implant. Possible septic complications are related to the duration and severity of the surgery. During this surgery, irrigation of the operating area with a scolicidal solution is mandatory. Hypertonic saline serum is the most used solution despite the risk of tissue necrosis. The use of oxygenated water is rare due to the risk of gas embolism. Formaldehyde at 1% is virtually abandoned. The combination approach is recommended in most cases.

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. Adjunctive medical therapy with the anthelmintic Albendazole (400 mg twice daily) is essential to reduce the risk of recurrence. However, recurrence rates remain high, ranging from 30% to 40%, with reported mortality rates between 3% and 14% [10]. Prevention is crucial and involves prophylactic measures targeting all levels of the epidemiological chain, including minimizing human exposure and protecting both intermediate (herbivores) and definitive (dogs) hosts from infection.

4. Conclusion

The vertebral localization of hydatid cysts is rare but severe due to its potential neurological complications. Diagnosis is often delayed due to the absence of specific clinical signs, leading to an insidious progression marked by frequent recurrences, which contribute to a poor prognosis. Prevention remains the most effective strategy for disease control. While surgery is essential for spinal cord decompression and lesion removal, recurrence remains a major challenge despite complete excision.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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