

VERTEBRAL HYDATIDOSIS: CASE REPORT

Hidatidosis vertebral: Reporte de caso

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ABSTRACT

Introduction: Vertebral hydatid cyst is a rare disease with an incidence rate of 0.2-1%. In the spinal cord it can cause pain and severe disability due to compression and instability. Magnetic resonance imaging (MRI) shows characteristic lesions in T1 and T2. The combination of medical anthelmintic treatment and cyst excision surgery is the most used strategy.

Clinical Case: A 47-year-old woman with a history of surgery for hepatic hydatidosis associated with intraoperative rupture, with a 7-month history of disease characterized by oppressive back pain, in belt-like pattern, of increasing intensity up to 10/10, associated with paresthesia in the lower limbs and urinary retention. The MRI revealed a multicystic process in the D5 vertebral body with invasion of the posterior mediastinum and the spinal canal. The diagnosis was confirmed serologically with IgM and IgG. A D5 corpectomy and excision of the cystic lesion was performed using a D5-D6 costo-transversectomy; in addition, a D3-D4 and D6-D7 transpedicular fixation and a D5 body replacement with cylindrical mesh were performed. The evolution was favorable without evidence of recurrence one month after surgery.

Conclusion: Vertebral hydatidosis is a rare pathology that requires surgical management combined with prolonged medical treatment with albendazole. Surgery is complex because the presence of multicystic lesions makes extensive surgical resection without rupture of the cyst membrane difficult. Close monitoring of these patients allows detection of recurrence and favors its early management.

Keywords: Echinococcosis, Spinal Cord, Back Pain, Albendazole, Surgical Mesh (Source: MeSH NLM)

RESUMEN

Introducción: El quiste hidatídico vertebral es una enfermedad rara siendo su tasa de incidencia de 0.2-1%. A nivel de la médula espinal puede causar dolor y severa discapacidad debido a compresión e inestabilidad. La resonancia magnética (RMN) muestra lesiones características en T1 y T2. La combinación del tratamiento médico antihelmíntico y la cirugía para exéresis del quiste es la estrategia más comúnmente usada.

Caso Clínico: Mujer de 47 años con antecedente de cirugía por hidatidosis hepática asociada a ruptura intraoperatoria, con enfermedad de 7 meses de evolución caracterizado por dolor dorsal opresivo, en cinturón y de intensidad creciente hasta 10/10, asociado a parestesias en miembros inferiores y retención urinaria. La RMN evidenció un proceso multiquístico en cuerpo vertebral D5 con invasión de mediastino posterior y canal espinal. El diagnóstico se confirmó serológicamente con IgM e IgG. Se realizó una corpectomía D5 y exéresis de la lesión quística mediante una costo-transversectomía D5-D6; además, se realizó una fijación transpedicular D3-D4 y D6-D7 y un remplazo corporal de D5 con malla cilíndrica. La evolución fue favorable sin evidencia de recurrencia al mes de la cirugía.

Conclusión: La hidatidosis vertebral es una patología rara que requiere un manejo quirúrgico combinado con un tratamiento médico prolongado con albendazol. La cirugía es compleja debido a que la presencia de lesiones multiquísticas hace difícil una resección quirúrgica extensa sin ruptura de la membrana del quiste. El seguimiento estrecho de estos pacientes permite detectar la recurrencia y favorece el manejo temprano de la misma.

Palabras Clave: Equinococosis, Médula Espinal, Dolor de espalda, Albendazol, Mallas Quirúrgicas. (Fuente: DeCS Bireme)

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Hydatic cyst is a disease caused by the *Echinococcus granulosus* parasite. Involvement of the vertebral system is rare, with an incidence rate of 0.2-1%. The use of an intracystic catheter to irrigate with chlorhexidine solution is

a treatment option, but it is preferred to be aggressive by totally excising the cyst without rupturing it, in addition to achieving adequate decompression of the spinal cord, irrigation with scholicial drugs and stabilization of the spine. ¹

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When located in the spinal cord, it can cause pain and severe disability due to neurological symptoms and instability. The combination of anthelmintic medical treatment and surgery to excise the cyst is the most used strategy. Medical treatment makes surgical treatment more effective, maintaining the result over time. Subtotal or total resection, including decompression of neural structures and adequate reconstruction of the spine, are the goals of surgical treatment.²

cyst operated on in our hospital, in which the total resection of the cyst was achieved with subsequent reconstruction and stabilization of the spine.

CLINICAL CASE

History and examination: A 47-year-old female patient, from Lima, with a history of liver hydatid cyst surgery with intraoperative rupture (10 years ago), left salpingo-oophorectomy, cholecystectomy and umbilical hernioplasty

We present the case of a patient with a vertebral hydatid



Fig 1. (A) X-ray of the thoracic spine in anteroposterior view showing osteolytic lesions in D5 (arrow). (B) Chest CT with contrast in coronal view in mediastinal window showing expansive process of D5 with extension to the left side. (C) Chest CT with contrast in sagittal view in mediastinal window, with expansive process in D5 with expansion towards posterior mediastinum and medullary canal (arrow). (D) MRI of the thoracic spine with contrast in T2 sequence showing a multicystic lesion in the D5 vertebral body with extension to the left side. (E) MRI of the thoracic spine with contrast in sagittal view in T2 sequence where an expansive multicystic process is evidenced in D5 (arrow).

who attended the emergency room due to a 7-month disease characterized by oppressive dorsal pain of increasing intensity from 8/10 to 10/10, associated with lower limb (LL) paresthesia and urinary retention. On examination, the patient was awake on the Glasgow Coma Scale (GCS) 15, without motor deficit, increased reflexes in the LL, functional limitation due to back pain, hypoesthesia from dermatome D4, sphincter disorder, Dandy D5 (+), Ramond Dorsal (+).

Magnetic Resonance Imaging (MRI) with contrast of the dorsal and lumbar spine showed an expansive process of the D5 vertebral body, isointense on T1 with multiple intralesional septa and hyperintense lesions on T2 with multiple hypointense septa, compatible with a multicystic lesion that produced lysis of the vertebral body and was it extended to the left lateral part of the vertebral body and invaded 40% of the medullary canal. (Figure 1). IgM and IgG serology studies were positive for hydatidosis. Treatment with albendazole at a dose of 400mg orally every 12 hours in the 2 weeks prior to surgery was started, showing seroconversion of IgG, both quantitatively and qualitatively.

Surgical treatment: A D5 corpectomy and resection of the D5 vertebral tumor were performed, replacement of the vertebral body with a cylindrical titanium mesh plus D3-D4 and D6-D7 transpedicular fixation. The procedure consisted of performing a midline approach, with extensive dissection until the transverse processes from D3 to D7 were fully evident on the right side, and on the left side up to the ribs at those same levels. The bilateral D3-D4 and D6-D7 transpedicular screws were placed, and a D5 laminectomy was performed. Then a D5 and D6 costo-transversectomy was performed, which allowed the D5 corpectomy to be performed, where intact whitish membrane cysts were evidenced, but due to the large number of cysts and their location, an intraoperative rupture occurred. This event could be managed, and the lesion was completely removed, including the part that was in the posterior mediastinum (Figure 2).

After that, a cylindrical titanium mesh was placed as a replacement for the D5 vertebral body, which was positioned slightly laterally to the left due to the risk of

injury to the spinal cord during placement. Finally, the titanium bars and their respective caps were placed, without complications. A computerized tomography (CT) scan of the thoracic spine was performed in the immediate postoperative period, which revealed the screws and bars in proper position, implantation of D5 slightly lateralized to the left, total resection of the lesion and little bilateral pleural effusion (Figure 3)

Clinical evolution: In the postoperative period, the patient wakes up with predominantly left paraparesis (strength in the right LL 2/5 and in the left LL 1/5), with pain in a belt pattern in the D5 territory, recovery of the sensory deficit, reflexes preserved in the LL, but still with sphincter disorder. On the 2nd postoperative day, she presented with hypovolemic shock and severe anemia at 6.5g / dl; Chest tomography revealed a hemothorax, which was treated by transfusion and chest drainage for 3 days, achieving clinical improvement. After 3 weeks of medical management and intensive physical therapy, the patient presented improvement in muscle strength (3/5 on the left LL and 4/5 on the right LL), with sphincter control and no sensory deficit. Tomography of the thoracic spine without contrast showed absence of vertebral tumor for which she was discharged with anthelmintic treatment (Figure 4).

DISCUSSION

Parasitic infections are quite common in the world in areas with developing economies, with cases being rare in developed countries. Hydatidosis is a parasitic disease caused by the *Echinococcus granulosus* parasite and when it affects the spine, patients may present typical symptoms such as back pain, numbness, weakness in the lower limbs, fecal or urinary incontinence. ³

Besides *Echinococcus granulosus* which is the most common form, the medical literature also mentions that hydatidosis can be caused by *Echinococcus multilocularis*. On the one hand, *Echinococcus granulosus* is known as the dog tapeworm and is transmitted to humans through the oral fecal route, by ingesting the eggs found in the feces of dogs. They usually cause infection of the liver in the form of a hydatid cyst. Contact with sheep is also a risk factor, and

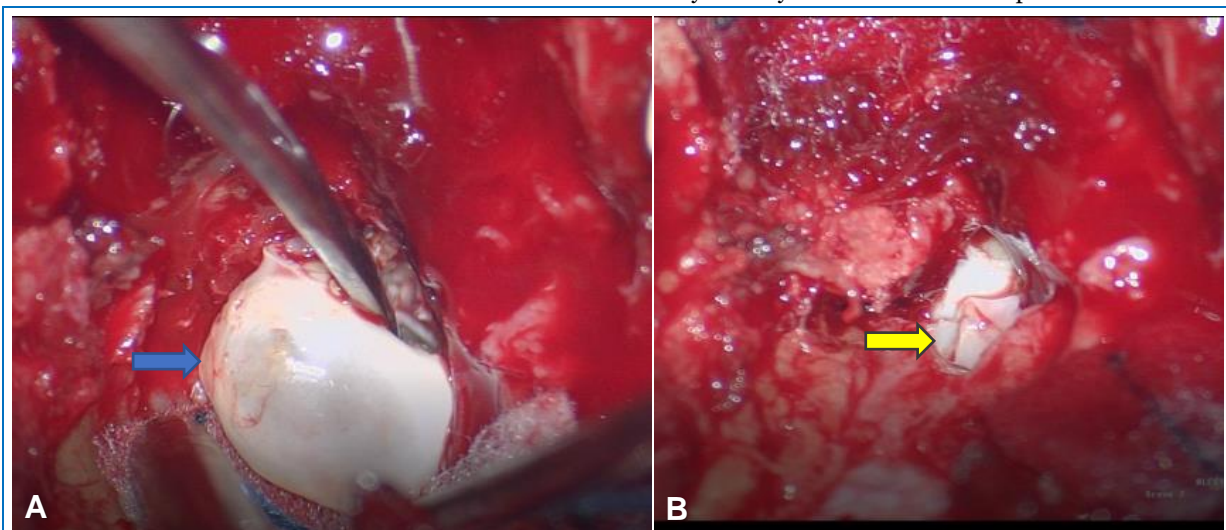


Fig 2. (A) Hydatid cyst with its intact white membrane in the operative bed (blue arrow). **(B)** Rupture of another hydatid cyst in the operative bed (yellow arrow).

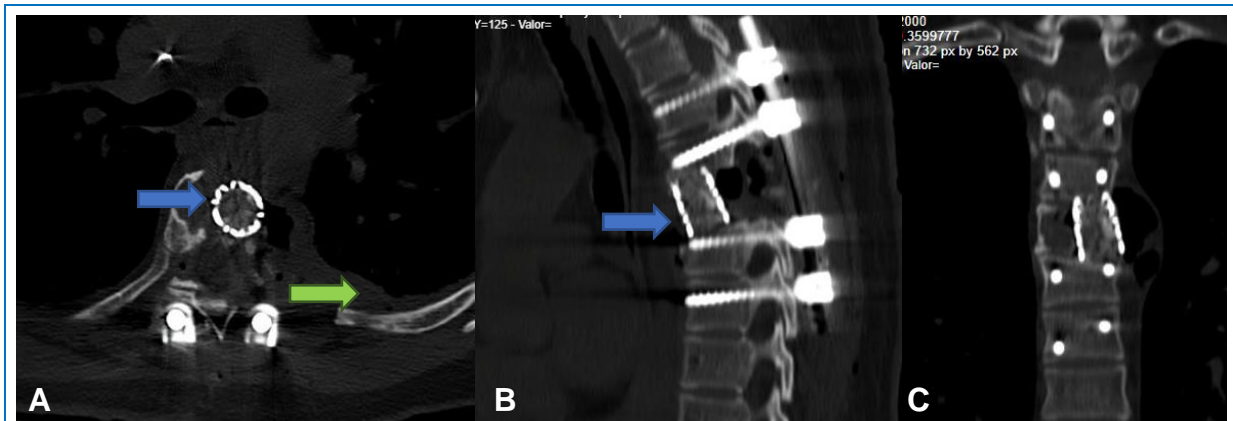


Fig 3. (A) Chest CT without contrast in bone window axial view showing circular titanium mesh in D5 (blue arrow) with scant bilateral pleural effusion (green arrow). **(B)** Non-contrast chest CT in sagittal view in bone window showing the transpedicular screws and the cylindrical mesh (blue arrow). **(C)** Chest CT without contrast in coronal view in bone window.

this occurs on farms where sheep have contact with dogs, which are the definitive host. On the other hand, *Echinococcus multilocularis* usually causes lung disease, its definitive host is the fox, so its incidence is higher in places where there are many foxes. ⁴

Echinococcus involvement of the central nervous system (CNS) is rare, with the thoracic spine being the part most frequently affected. When the spine is affected, the most frequent symptoms are nonspecific and result from spinal or root compression, and manifest as radiculopathy or myelopathy. However, it is not uncommon for some patients to have asymptomatic large cysts. ⁴

In the diagnostic process, although plain radiographs show cystic lesions in contiguous vertebral bodies, bone lysis, or spondylitis, CT and MRI are always required to clarify the diagnosis. Ultrasound can help detect abdominal injuries. The CT gives us better data on the bone tissue and allows us to appreciate osteolytic lesions in the vertebral bodies, which do not capture contrast. On MRI, T1 shows an isointense or hypointense cystic lesion with a cystic wall, while T2 shows a hyperintense cyst with a hypointense cyst wall. Diffusion can also be used to rule out an abscess, which restricts diffusion, while the hydatid cyst does not restrict, that is, it is seen as a hypointense lesion. ⁴

The differential diagnosis of spinal echinococcosis includes tuberculosis since both can be found in the same endemic areas. But malignancy, abscesses, cystic lesions such as spinal arachnoid cysts or aneurysmal bone cyst of the spine must also be ruled out. For this, the clinical history, imaging studies, laboratory studies must be seen, but only the surgical exploration and histopathological examination will give the definitive diagnosis. Serology is specific but not sensitive. ⁴

It has been seen that cysts are usually in highly vascular tissues such as the brain, liver, or lung, so bone involvement is rare. In case of bone involvement, the spongy part of the vertebral body is one of the preferred sites. ¹

Surgery is the treatment of choice in spinal hydatidosis, associated with long-term preoperative treatment with anthelmintics such as albendazole since this reduces intracystic pressure. The most reported surgical procedure is decompression through a laminectomy, but the need for vertebral fusion should always be considered depending on the extent of the injury. Most studies use the posterior approach, although some also use the anterior approach. Radical excision is preferred because needle aspiration increases the risk of cyst rupture. The use of schollicidal

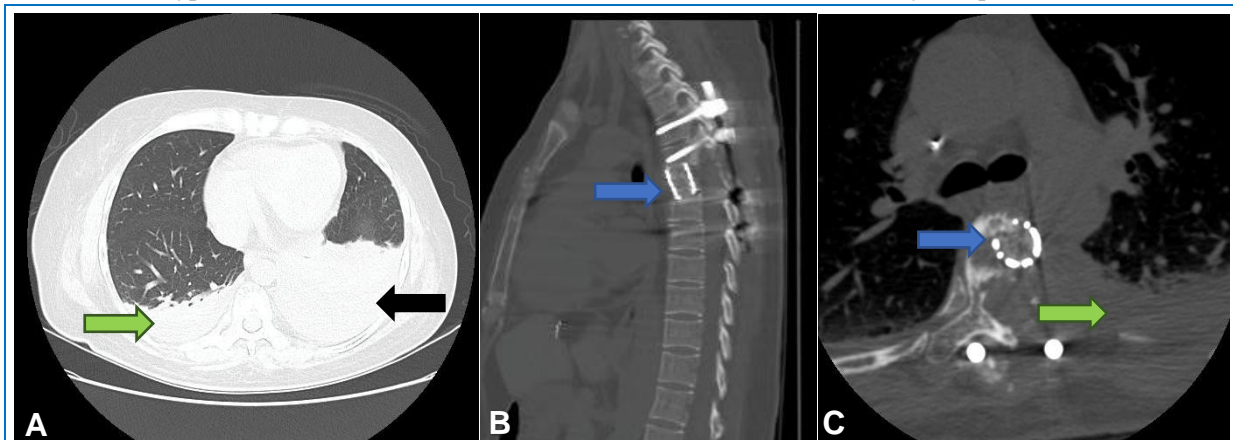


Fig 4. (A) Non-contrast chest CT in axial view (pulmonary window), in which the left hemothorax is evidenced (black arrow) with little right pleural effusion (green arrow). **(B)** Non-contrast chest CT in sagittal view (bone window) showing screws D3 and D4, with circular titanium mesh in D5 (blue arrow), without tumor recurrence. **(C)** Non-contrast chest CT in axial view (mediastinal window), with cylindrical titanium mesh in D5 (blue arrow) with little left pleural effusion (green arrow).

agents during the intraoperative period reduces the risk of dissemination in abdominal and pelvic surgeries, and it is also used in spinal surgeries for the same purpose, with no studies supporting its use. ^{4,5}

The resection of the cyst is a problem since the simple puncture is not a solution. Spinal hydatidosis has a high rate of morbidity, recurrence, and mortality. The excision of the cysts without rupture of its wall is vital because the rupture of this brings with its dissemination and chronic recurrence, including in the subcutaneous tissue. If the dura is compromised, dural excision and duroplasty are indicated. Excision is much more difficult when it is not a single cyst but a multicystic lesion that is difficult to access, as was the case in our patient. ¹

Brett et al mention in their article that surgical treatment must be accompanied by albendazole therapy for at least 6 months. Only 30 to 40% of patients achieve a cure for their disease. The recurrence rate is high, with up to 48% recurrence in the first 24 months, for which close follow-up with images is required. ³

In our case, the patient had a multicystic lesion that involved the entire D5 vertebral body, with a high probability of intraoperative rupture, which brought with it a high probability of recurrence. The patient had a total resection of the lesion, but due to the difficult access to the lesion, she presented with a neurological deficit which partially recovered in time with the support of physical therapy, without presenting recurrence one month after surgery. However, due to the history and rupture of the cysts during surgery, close imaging controls should be continued. Likewise, it is recommended to continue studying this type of neurosurgical pathology to create a more suitable treatment protocol in this disease whose vertebral involvement is rare in the population.

CONCLUSION

Vertebral hydatidosis is a rare pathology that requires surgical management in addition to medical management based on prolonged treatment with albendazole. Surgical management is complex because the presence of multicystic lesions makes extensive surgical resection without rupture of the cyst membrane difficult. Close follow-up of these patients allows the detection of recurrence and favors its early management.

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Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Authors Contributions

Conception and design: All authors. *Drafting the article:* Vargas. *Critically revising the article:* Basurco, Laos. *Reviewed submitted version of manuscript:* Vargas. *Approved the final version of the manuscript on behalf of all authors:* Vargas.

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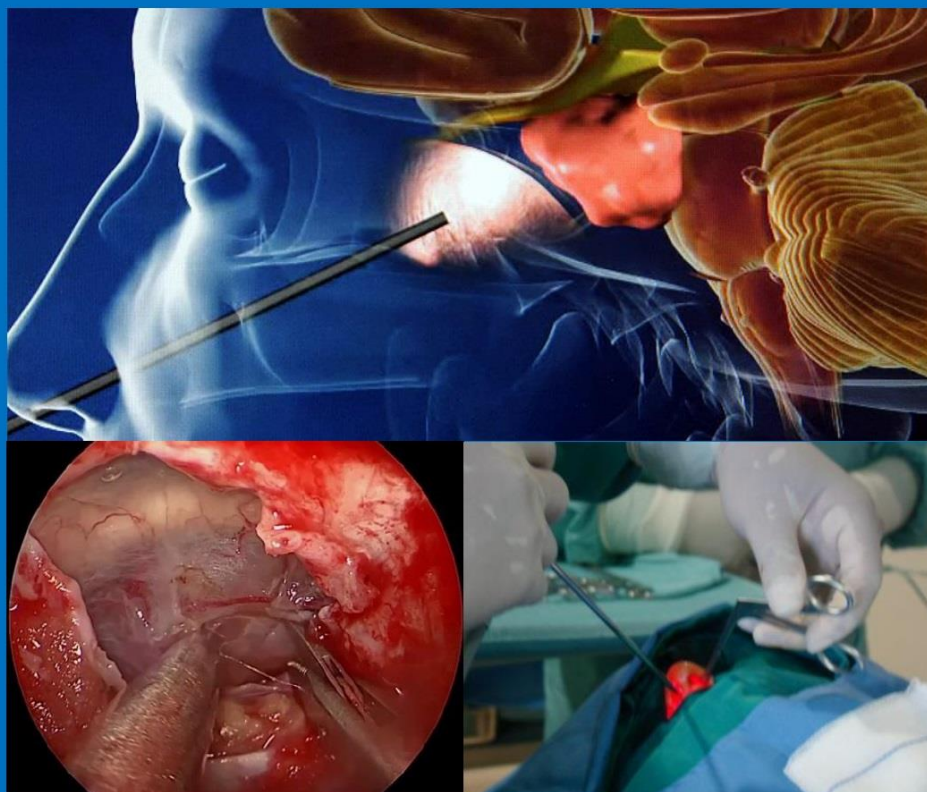
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