Radiology Case Reports 17 (2022) 2238–2242

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.elsevier.com/locate/radcr

Case Report

Spinal hydatid cyst as cause of neurological injury in a patient from Brazilian amazon region. ☆

Romel Corecha Santos, MD[a,*], *Guilherme José da Nobrega Danda, MD*[b], *Altamir Monteiro Junior, MD*[c], *Ricardo de Amoreira Gepp, MD*[a]

[a] *Neurosurgery Department, Sarah Network of Rehabilitation Hospitals, SMHS 501 Bloco A, Brasília, Federal District, 70335-901, Brazil*

[b] *Internal Medicine Department, Sarah Network of Rehabilitation Hospitals, SMHS 501 Bloco A, Brasília, Federal District, 70335-901, Brazil*

[c] *Pathology Department, Sarah Network of Rehabilitation Hospitals, SMHS 501 Bloco A, Brasília, Federal District, 70335-901, Brazil*

ARTICLE INFO

Article history:
Received 3 March 2022
Revised 25 March 2022
Accepted 25 March 2022

Keywords:
Hydatid cysts
Spine
Echinococcus

ABSTRACT

Echinococcus granulosus infection is the primary cause of spinal hydatidosis. We describe the case of a 22-year-old man from the Brazilian Amazon region with crural spastic paraparesis and back pain. Radiological examinations showed multilocular lesions involving compression of the thoracic spine and rib injury. The patient underwent vertebrectomy with spinal stabilization and thoracoplasty with resectioning of the costal arch. Subsequently, the patient was prescribed oral treatment with albendazole. Marked recovery of the neurological status was achieved. Bone hydatid disease is rare, accounting for 0.5%-0.4% of all hydatid cysts, affecting the spine in 50% of cases. The treatment of choice is surgery accompanied by antiparasitic medication.

Introduction

Hydatid disease, or cystic echinococcosis, caused by the larval stages of cestodes of the genus *Echinococcus*, is a cosmopolitan parasitic infection. Humans are an intermediate host and are at high risk of liver infection (~70%). Bone involvement is reported in the range of 0.5%-4%, affecting the spine in 50% of cases [1].

Spinal hydatidosis can affect the spinal cord and vertebral column. It is associated with varying degrees of morbidity, disability, and mortality, and its behavior is associated with malignant disease ('le cancer blanc') [2].

We present the case of a patient from the Amazon region with spastic leg weakness and limiting back pain. The patient was treated aggressively through 3 surgical procedures and the oral antiparasitic drug, albendazole. This treatment approach resulted in a marked recovery of neurological status.

☆ Competing Interests: The authors have no personal, financial, or institutional interest in any of the drugs, materials, or devices described in this article.

* *Corresponding author.*
E-mail address: romelcorecha@hotmail.com (R.C. Santos).

https://doi.org/10.1016/j.radcr.2022.03.102

Case report

A 22-year-old male from the rural Amazon region, 64 kg, was admitted with paresthesia characterized by numbness in the chest area extending along the lower limbs accompanied by lower limb paraparesis. The patient's clinical condition progressively worsened with significant walking limitation 1 month after symptom onset. He also complained of back pain which occurred predominantly whilst in a sitting position. His medical history included no comorbidities but frequent contact with dogs.

Neurological examination revealed crural spastic paraparesis with lower limb muscle strength grade 4 and hypertonia (Ashworth grade 3). Bilateral hyperreflexia was found in the patellar reflex (+4), ankle jerk (+4), and Achilles tendon clonus. The Babinski sign was also found bilateral.

There were no abnormalities in the patient's blood count prior to surgery. The eosinophil count was 5.3% (350 cells/mm^3). Ultrasonography and computed tomography (CT) of the abdomen did not reveal any abdominal lesions.

Motor evoked potential monitoring demonstrated a conduction defect in corticomotoneuronal rapid pathways to the lower limbs. The somatosensory evoked potential test revealed a central conduction defect of the sensory pathways from the thick myelinated fibers of the lower limbs.

Thoracic spine CT and magnetic resonance imaging (MRI) revealed multiloculated cysts extending into the entire length of the second right costal arch, predominantly intrathoracic. There were also lytic lesions affecting the body and posterior arch elements of the T2 vertebra and the T1 and T3 posterior elements. Paraspinal soft tissue and intracanal extradural lesions extending T1/T2 and T3/T4 were observed causing stenosis and compression of the spinal cord with edema were also observed (Fig. 1).

Two-stage T2 vertebrectomy and T1 and T3 posterior element decompression followed by posterior cervicothoracic arthrodesis were performed. Pedicle screw systems were used for dorsal stabilization (C5-T6) with vertebral body replacement achieved by titanium cage implantation (T1-T3). Right thoracoplasty with T2 and T3 rib resection was also performed (Fig. 2). Extension of the lesions did not allow for complete excision of those along the anterior chest wall. The patient also developed surgical wound dehiscence, which was treated using a muscular transposition procedure.

Pathologic evaluation of the lesion obtained from surgical specimens showed the larval form of Echinococcus (Fig. 3).

Postoperative albendazole at a dose of 400 mg every 12 hours was prescribed for 11 days with proposed maintenance for 12 months.

This approach resulted in significant neurological recovery, and reasonable control of residual lesions was observed in imaging studies.

At 6 months post-surgery the patient is in outpatient follow up and is tolerating albendazole with no side effects. This approach has resulted in significant neurological recovery. Furthermore, reasonable control of residual lesions was observed in imaging studies.

Discussion

Spinal hydatidosis accounts for approximately 50% of hydatid diseases with bone involvement. *Echinococcus granulosus* is the causative agent in most cases and is responsible for cystic lesions, while *E. multilocularis* is the primary agent of visceral disease (alveolar echinococcosis) [3].

In tropical America, numerous wild animals may be intermediate hosts for *E. granulosus,* of which *Agouti paca* (paca) is the most common. The definitive hosts are domestic and wild canids. The only known natural host in the wild environment is *Speothos venaticus* (bush dog). Human beings participate in the cycle as intermediate hosts both in the wild and in the domestic environment [3]. The Brazilian Amazon reality suggests that domestic and peridomiciliary transmission is fundamental for maintaining this neotropical endemic [4].

In most cases, as noted in this report, spinal hydatidosis affects the thoracic spine (45%-50%), followed by the lumbar spine (20%-39%). [5] In this scenario, the diagnosis is usually delayed, with advanced impairment of the anatomic structures.

In many cases, the exact primary implantation site of the parasite and the primary affected spinal structures remain uncertain. It is suggested that the origin can be either the vertebral bone (with secondary involvement of the paravertebral structures or intraspinal space), the paravertebral region, or intraspinal structures [5].

Intradural extramedullary spinal hydatidosis is relatively rare [5] The intervertebral disc involvement remains intact despite the progression of the lesion towards the periosteum and ligaments [6,7].

The clinical spectrum of spinal hydatidosis is broad and depends on the level and degree of spinal cord compression, ranging from paraparesis (61%-73%), bladder dysfunction (11.1%-32%), sensory deficit (24%) to radicular pain (27%-60%) [8,9,10].

The gold standard imaging method is MRI, which demonstrates multilocular hypointense lesions on T1-weighted images and hyperintensity on T2-weighted images. CT is essential for the evaluation of bone erosion and mechanical instability [11]. Serological diagnosis can be used for preoperative diagnosis in suspected cases [3].

Surgery remains the treatment of choice and is based on 2 pillars: surgical decompression, and stabilization of a compromised spine. Radical surgery is recommended because it improves survival and functional performance but does not prevent disease progression [12].

Therapy with benzimidazole derivatives is effective against visceral diseases. However, evidence for the effectiveness of benzimidazoles in bone hydatid disease is scarce, and the contribution of these drugs to healing and preventing recurrence is the subject of discussion [12]. Albendazole is preferred over mebendazole because of its superior efficacy and pharmacokinetic properties [12]. Successful chemotherapy with albendazole for 1 year for combating residual cysts (post-surgery) has been reported [12]. The dosage recommended by the World Health Organization (WHO) to albendazole is 10-15 mg/(kg•day) [12].

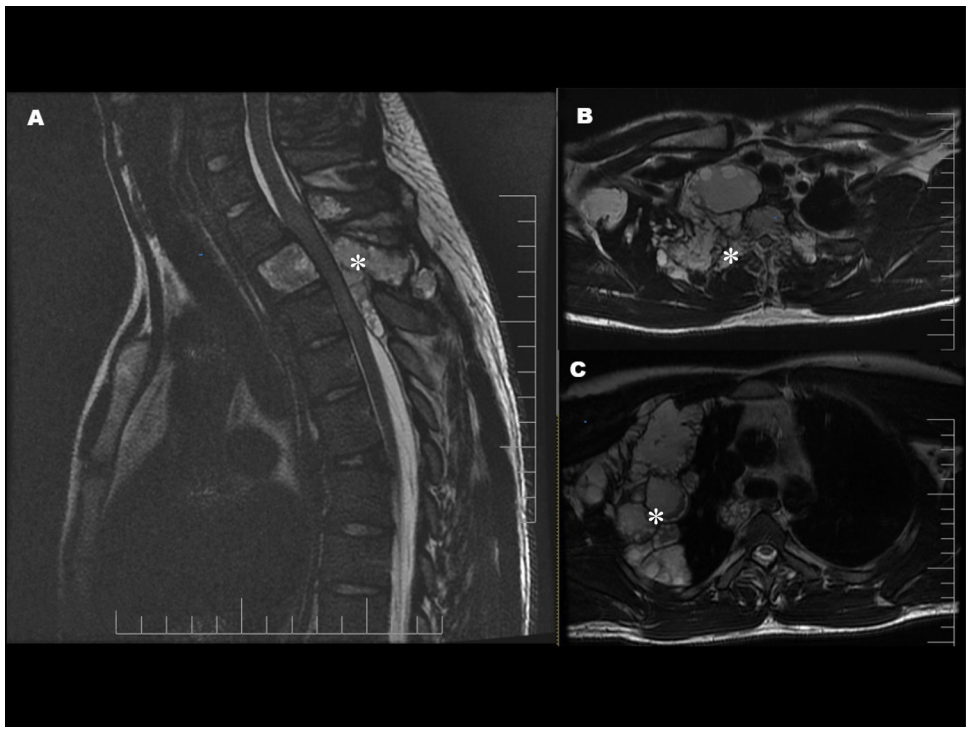

Fig. 1 – (A) MRI T2-weighted image, sagittal view, spinal hydatidosis affecting the vertebral body of T2 (white arrow) and the posterior elements of T1, T2 (white asterisk), and T3. (B) MRI on T2- weighted image, axial view, showing lesion of the anterior (white arrow) and posterior elements (white asterisk) of T2. (C) MRI on T2- weighted image, axial view, showing lesion involving of the second costal arch on the right with the involvement of the soft parts around.

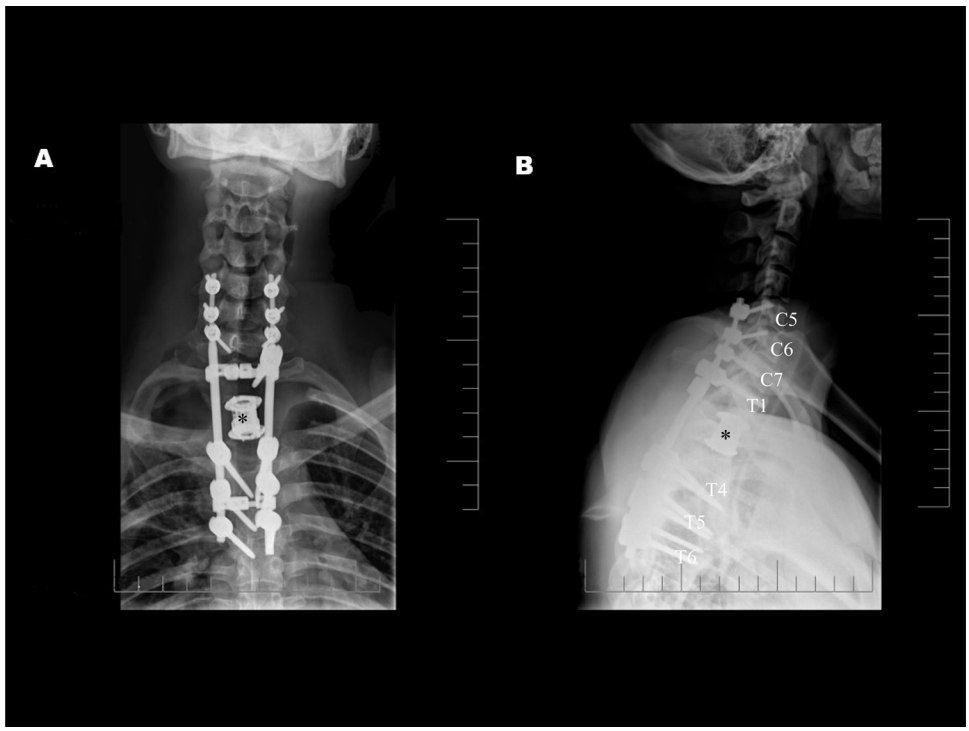

Fig. 2 – (A) Anterior view and (B) Lateral view. Postoperative control demonstrating spinal instrumentation, extending from C5 to T6. Pedicle screw systems were used for dorsal stabilization (C5-T6) with vertebral body replacement achieved by titanium cage implantation (T1-T3) (black asterisk).

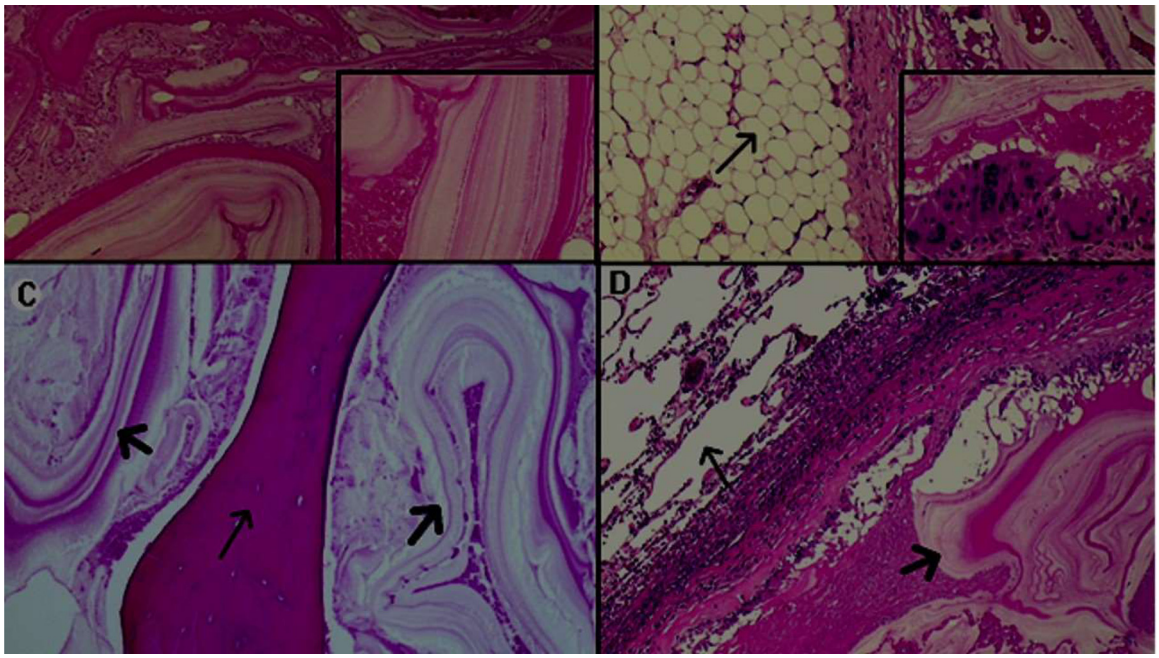

Fig. 3 – (A) Necrotic parasitic remains of larval stages of Echinococcus granulosus (hydatid disease) are represented by an external laminated, acellular, cuticular membrane. (B) Soft tissue commitment (thin arrow – adipose tissue; thick arrows – parasitic remains). (C) Bone commitment (thin arrow – necrotic trabecular bone; thick arrows - parasitic remains). (D) Lung commitment.

This report demonstrates that the treatment of spinal hydatidosis with a surgical approach and antiparasitic drug therapy is beneficial in decreasing recurrence risk and even preventing intraoperative cyst rupture [12].

In a systematic analysis, Neumayr noted 48% disease recurrence in reviewed cases with vertebral bone involvement [12]. In advanced cases, surgery is advocated as palliative, and recurrence is inexorable [12].

Kafaji et al. found recurrence of spinal hydatidosis in 89% (n = 36) of patients treated surgically with an average 2.5-year follow-up (1–8 years). The authors considered this to be related to the subtotal excision due to anatomical difficulties. [9]

Despite advances in diagnostic techniques and surgical treatments, spinal hydatidosis remains associated with high morbidity, disability, and mortality. Currently, the long-term surgical outcomes of spinal hydatidosis are insufficient, and further studies are required to better understand this disease. Long-term follow-up of the patient is mandatory due to subsequent recurrences.

Conclusions

Bone involvement may be an initial manifestation of hydatid disease. In endemic regions, it is crucial to consider this possible diagnosis in patients with a vertebral mass of unknown origin or spinal cord compression syndrome so that the proper course of treatment may be administered quickly to improve the chances of favorable recovery.

Patient consent statement

Written consent for publication of this case was obtained from the patient and is available upon request.

REFERENCES

[1] Ozdemir HM, Ogun TC, Tasbas B. A lasting solution is hard to achieve in primary hydatid disease of the spine: long-term results and an overview. Spine 2004;29:932–7 Phila Pa 1976.
[2] Dévé F. L'Echinococcose osseuse. Montevideo: Monteverde y Cia; 1948.
[3] . In: R Focaccia, editor. Tratado de Infectologia. São Paulo, Atheneu, Inc; 1997. p. 1713. 1420.
[4] Soares MCP, Moreira-Silva CA, Alves MM, Nunes HM, do Amaral IA, Móia LJMP, et al. Polycystic echinococcosis in the eastern brazilian Amazon: an update. Rev. Soc. Bras. Med. Trop 2004;37(2):75–83.
[5] Neumayr A, Tamarozzi F, Goblirsch S, Blum J, Brunetti E. Spinal cystic echinococcosis – a systematic analysis and review of the literature: part 1. epidemiology and anatomy. PloS Negl Trop Dis 2013;7(9):e2450.
[6] Baysefer A, Gonul E, Canakci Z, Erdogan E, Aydogan N, Kayali H. Hydatid disease of the spine. Spinal Cord 1996;34:297–300.

[7] Tsitouridis I, Dimitriadis AS. CT and MRI in vertebral hydatid disease. Eur Radiol 1997;7:1207–10.
[8] Herrera A, Martínez AA, Rodríguez J, hydatidosis Spinal. Spine 2005;30(21):2439–44.
[9] Kafaji A, Al-Zain T, Lemcke J, Al-Zain F. Spinal manifestation of hydatid disease: a case series of 36 patients. World Neurosurg 2013;80(5):620–6.
[10] Turgut M. Hydatid disease of the spine: a survey study from Turkey. Infection 1997;25(4):221–6.
[11] Gennari A, Almairac F, Litrico S, Albert C, Marty P, Paquis P. Spinal cord compression due to a primary vertebral hydatid disease: A rare case report in metropolitan France and a literature review. Neurochirurgie 2016;62(4):226–8. doi:10.1016/j.neuchi.2016.03.001.
[12] Neumayr A, Tamarozzi F, Goblirsch S, Blum J, Brunetti EFlisser A, editor. Spinal cystic echinococcosis – a systematic analysis and review of the literature: part 2. treatment, follow-up and outcome. PLoS Neglected Tropical Diseases 2013;7(9):e2458.