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## Case report

# Recurrent costovertebral hydatidosis with epidural extension ☆,☆☆

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## ABSTRACT

Osseous echinococcosis is a relatively rare entity and that of the rib is even rare. Few cases of costal echinococcosis have been reported in the literature so far. We report a case of a recurrent costovertebral hydatidosis with epidural extension in a 49-year-old man who presented with paraparesis and back pain. MRI of the dorsal spine was performed. The imaging features were suggestive of echinococcosis involving the rib and vertebrae with epidural extension. This diagnosis was confirmed histopathologically after surgical treatment. The evolution was marked by the reappearance of the same symptoms due to recurrence. The prognosis of costovertebral hydatidosis is gloomy despite radical surgical treatment due to the frequency of recurrences.

This case highlights the role of MRI for the diagnosis and follow-up of patients after treatment.

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## Introduction

Osseous hydatidosis is a rare disease. The diagnosis is made by the combination of clinical, radiological, and laboratory findings.

The role of imaging is to appropriate surgical treatment to prevent the dissemination of the parasite that may be a fatal complication and in long-term follow-up due to the high rate of recurrence.

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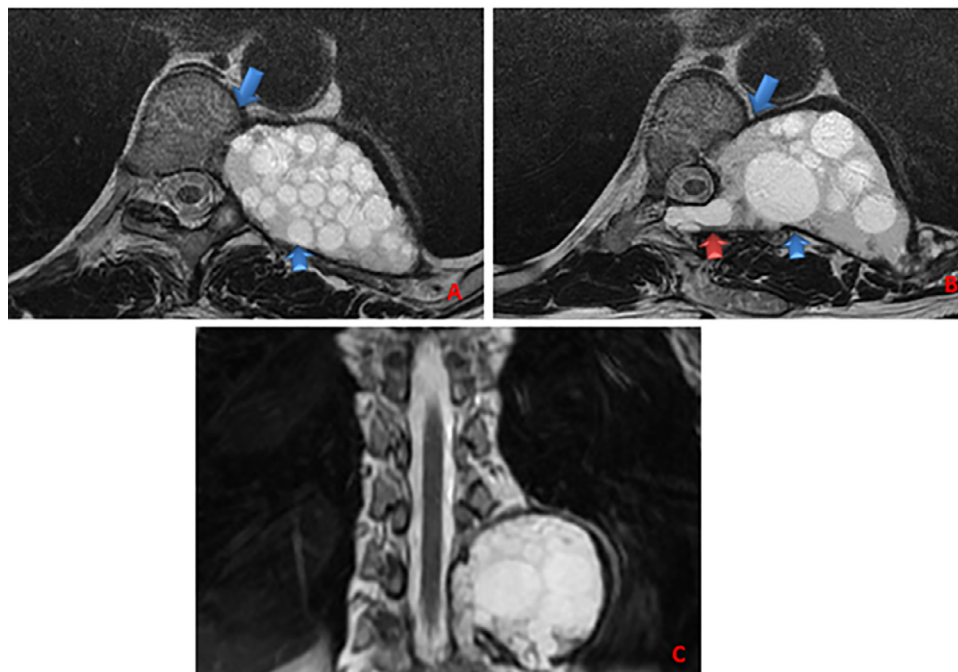
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**Fig. 1 – Multivesicular Costovertebral Hydatidosis • Preoperative MRI T2 weighted images • (A,B) osteolysis of the 8th right rib, transverse process, and vertebral body (blue arrow) with the presence of hydatid vesicles in the intervertebral foramen (red arrow). (C) Localization of lesions in the spine.**

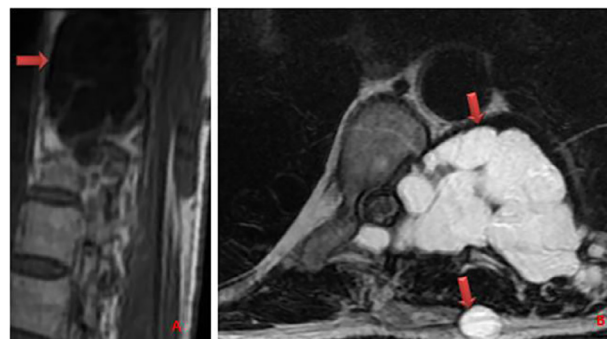
### Clinical presentation and imaging findings

A 49-year-old man presented with progressive dorsal pain on the left side of the back associated with progressing paraparesis over 3 months'. He grew up on a farm surrounded by sheep and dogs. No history of trauma had been reported. Clinical examination showed a localized bulge in the left side, which was mobile on palpation. No motor or sensory deficit was noted. Laboratory tests were normal. The patient underwent a magnetic resonance imaging examination (MRI) that shows a well-defined, multiloculated lesion hypointense on T1 and hyperintense on T2 weighted images along the posterior aspect of the left T8, T9, and T10 ribs with epidural extension. It was also responsible for destructing the left pedicle and transverse process of the D9 vertebra.

These imaging findings suggested primary echinococcosis of the rib (Fig. 1).

No other sites of involvement had been discovered on CT screening, especially on the liver, and spleen.

The patient underwent a carefully complete excision of the cyst using a posterior approach. A T9 hemilaminectomy associated with a left costo-transversectomy was also performed. Histopathological and parasitological results confirmed the diagnosis of hydatid disease caused by *Echinococcus granulosus*. The patient received oral albendazole for 3 months'. Immediate postoperative recovery was inconclusive, but 6 months' later the patient was admitted with recurrence at the same level generating increasing paraplegia and further surgery was indicated (Fig. 2). This procedure could not be accomplished due to cardiac complications. He is currently re-



**Fig. 2 – Recurrence • Post-operative MRI • (A) T1 Sagittal, (B) T2 Axial images showing recurrent intraspinal and extraspinal cysts (arrows).**

ceiving medical treatment (Albendazole and corticosteroids) and is followed closely in the Infectious Disease Clinic.

### Discussion

Bone hydatidosis is a rare disease occurring in only 1%-4%. The involvement of the thoracic cage is uncommon [1]. The exact incidence of rib echinococcosis is not well established. Costal hydatid cysts can be primary (intraosseous) or secondary (extraosseous form). Secondary hydatid cysts happen because of spontaneous or traumatic rupture of a pulmonary or mediastinal hydatid cyst [2].

Then, we assume that our patient had a primary hydatid cyst of the rib due to the nonexistence of other sites of involvement.

The intraosseous form may be further classified into a solitary costal form and a costovertebral form. In our patient, it's a costovertebral form because of the extension of the disease into the adjacent vertebra and epidural space [2].

The diagnosis of rib hydatidosis is based on clinical, serologic, imaging, and surgical means [3].

Computed tomography examination is interesting to precise the location, severity of bone destruction, and expansion in the adjacent soft tissues. The typical presentation is multiple cystic images of different sizes with liquid density (10 UH–20 UH). It's also the best modality to evaluate the degree of osteolysis that may cause rupture of the compact bone [4]. However, MRI is the modality of choice to appreciate the disease's extent and the degree of medullar compression.

It appears typically as a multiloculated lesion hypointense on T1-weighted images, hyperintense on T2-weighted images, lack of calcification, and no enhancement (other than complication such as rupture or superinfection of the cyst) [5]. The signal of the daughter cysts is comparable to that of water.

CT exhibits the bone lesion better but MRI is superior in revealing compression of neural structures. The complementary use of CT and MRI is, therefore, recommended [6]. Differential diagnosis can involve mainly giant cell tumor, osteolytic metastases, plasmacytomas, aneurysmal bone cyst, and cystic neurofibromas [7].

Surgery is the keystone of the treatment.

It consists of a wide resection of the affected bones and surrounding muscles; this resection is not always possible.

Medical treatment by albendazole is highly advocated in association with surgery in pre- and post-operative to prevent recurrence or alone at high doses in inoperable patients to reduce the risk of spread [8].

That been said, Recurrences are frequent and that was the case of our patient. It's a major problem that occurs in 30%–100% [9].

Considering this high rate of recurrence, bone hydatidosis has a poor prognosis and has been compared to malignant disease. Follow-up is recommended every 3 months' in the first year and then annually.

- Surgical treatment is the keystone of treatment
- Recurrences are frequent.

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### Availability of data and material

Data available within the article.

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### Code availability

(N/A).

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### Patient consent

Informed written consent was obtained from the patient.

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### Keypoints

- Costovertebral localization is rare in the hydatid disease, it locally behaves like a bone malignancy.
- MRI is the best modality to appreciate the medullar extension.