

## Case Report

# PRIMARY SPINAL EXTRADURAL HYDATID CYST CAUSING SPINAL CORD COMPRESSION: A CASE REPORT

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## ABSTRACT

**Background:** Hydatid disease of the spine is caused by the parasite *Echinococcus granulosus*, a helminth belonging to the cestode group. Usually located in lungs or liver, hydatidosis of the bone occurs in only 0.5-4% of all hydatid locations, and half of these cases are located in the spine. We report a case of a spinal primary location of a hydatid cyst operated at our department.

**Case presentation:** The patient is a 52 years old lady who was presenting a year before she consulted dorsalgia followed by a weakness of the lower limbs and urinary retention, spine MRI performed objectified an epidural cystic formation. The patient was operated and the lesion was removed without complications. The pathology study revealed the hydatid nature of the cyst.

**Conclusion:** The spinal hydatid cyst is a rare condition leading to major complications once there is spinal cord compression; early management and total removal of the cyst without rupture are the factors with the best clinical outcome and less recurrence.

**Keywords:** *Hydatid Disease, Spinal Cord Compression, Intraspinal Cyst*

## INTRODUCTION

The spinal hydatid disease is a rare parasitic affection, causing in most cases a spinal cord compression and present a threat for the neurologic status, this affection put the surgeon face to many challenges in diagnosis (nonspecific symptoms or images), and in the management (poor response to medical treatment, per operative cysts spillage). We report a case of a primary location of a spinal hydatid cyst.

## CASE

The patient is a 52 years old lady without past medical history who presented 40 days before she consults a back pain which evolved rapidly to weakness of the lower limbs. The clinical exam at the admission found a patient with paraplegia, abolishing of the knee and achilles reflex, a bilateral Babinski sign, and a left hypoesthesia at the tenth thoracic vertebra (Th10). Laboratory examinations found a *Hyperleukocytosis* to  $16 \times 10^3$  U/L with 73 % of granulocytes.

Initially a spine computer tomography (CT) was performed objectifying an osteolytic process of Th10, Th11 and Th12 involving the posterior arches (figure 1); the spine MRI objectified a cystic formation of 30x 17 mm with low intensity in T1 weighted images, high intensity T2 weighted images, and an enhancement after injection of gadolinium of the cyst's wall as well the posterior arches of the osteolytic vertebrae which delimitate a necrotic area (figure 2). Chest X-rays and abdominal ultrasound then a thoracoabdominal CT were performed without hydatidosis signs. The patient underwent surgery; the total removal of the cyst and its capsule was achieved through the laminectomy of Th10, Th11 and Th12.

Pathologist study confirmed the hydatid nature of the cyst.

In post operative the patient presented a relief of her back pain but without improvement of the motor deficit so she was oriented to physical medicine. Five months later the patient presents the same neurological status but there was no recurrence on spinal MRI (figure 3).

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**Figure 1: preoperative CT showing osteolysis on the Th 10 vertebrae (the arrows)**



**Figure 2: preoperative T1 weighted imaging spinal MRI. A: coronal slide B: sagittal slide and C: axial slide; showing the cyst exerting a major spinal cord compression at the level of Th 10 vertebrae (the arrows)**

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**Figure 3:** postoperative spinal MRI, A: T2 weighted imaging sagittal slide, B: T2 weighted imaging axial slide, and C: T1 weighted imaging axial slide; removal of the cyst with its capsule through Th10,11, and 12 laminectomy and a clear spinal cord release (the arrows).

### DISCUSSION

Hydatid disease is caused by a parasite *Echinococcus granulosus*, a helminth belonging to the cestode group, humans are accidental intermediate hosts. According to WHO cystic echinococcosis is globally distributed in most pastoral and rangeland areas of the world, with highly endemic areas in the eastern part of the Mediterranean region, northern Africa, southern and eastern Europe, at the southern tip of South America, in Central Asia, Siberia and western China.

The symptoms are not specific, medullary or radicular symptoms are present, the signs are closely depending on the cyst localization, the backpain is present in 85 % of cases (Dalila Mrabet *et al.*, 2011), and paraplegia in 25–50% of cases (Long Xin *et al.*, 2015), a cauda equina syndrome has been described in some cases (Selhan Karadereler *et al.*, 2002) . The most common sites of occurrence of hydatid cysts are the liver in 60% of cases and lungs with 20-30 % of all cases (Malay Sarkar *et al.*, 2016), Bone affection is rare (0.5-4%) of which spine involvement is seen in half of the cases (Andreas Neumayr *et al.*, 2013, Akshay Jain *et al.*, 2014, Selhan Karadereler *et al.*, 2002, Dalila Mrabet *et al.*, 2011, Long Xin *et al.*, 2015). It involves most likely the dorsal spine (Andreas Neumayr *et al.*, 2013, AGTW FIENNES *et*

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al., 1982, Selhan Karadereler *et al.*, 2002, Ahmet Karakaslib *et al.*, 2015) which is the case of our patient. In a Review of the Literature by Neumayr *et al.*, they found that only 17.9% (120 cases) of all reviewed spinal hydatidosis cases had a history of extraspinal hydatid disease or were found to have concomitant newly diagnosed extraspinal hydatidosis (Andreas Neumayr *et al.*, 2013). The primary location in the spine can be explained by the presence of the portovertebral shunt theory (AGTW FIENNES *et al.*, 1982, Ahmet Karakaslib *et al.*, 2015, Long Xin *et al.*, 2015).

Braithwaite and Lees classified spinal hydatid cysts into five radiological types: I: intramedullary, II: extra medullar intradural, III: extra dural but intra spinal, IV: vertebral, and V: para vertebral (Braithwaite and Lees, 1981). The three first types are extremely rare (Selhan Karadereler *et al.*, 2002, Ahmet Karakaslib *et al.*, 2015, Long Xin *et al.*, 2015).

Laboratory tests are less helpful for the diagnosis than the liver location: the most common findings include hypereosinophilia, serologic tests are positive in only 40% of extrahepatic lesions in the late stages (Selhan Karadereler *et al.*, 2002, Mrabet *et al.*, 2011, Ahmet Karakaslib *et al.*, 2015, Long Xin *et al.*, 2015). The radiological diagnostic is made by MRI images, it shows the cysts with a liquid component tendency to invade anatomical cavities through the neural foramen, It does not demonstrate contrast enhancement, but has a CSF-like signal intensity (Selhan Karadereler *et al.*, 2002, Ahmet Karakaslib *et al.*, 2015, Long Xin *et al.*, 2015). CT scanning may be more convenient and more advantageous in following the progress of bone lesions associated with this disease. Although plain radiographs can show in the advance stage the bone destruction, the radiological features are not pathognomonic (Long Xin *et al.*, 2015, Braithwaite PA et Lees RF, 1981), destructive changes develop slowly but aggressively.

Depending on the spinal location of the lesions, the differential diagnoses of a bony spinal hydatid disease are: infections (tuberculosis, pyogenic infection, brucellosis), fibrous dysplasia, and tumors especially metastasis. The differential diagnoses of an intraspinal cystic lesion includes dorsal arachnoid diverticula and meningoceles; and the differential diagnoses of intradural cystic lesion includes arachnoid cysts and syringomyelia, other parasitic diseases are rare (*neurocysticercosis, Cysticerci, cysticercosis, E. multilocularis*) (Andreas Neumayr *et al.*, 2013). Surgical treatment of the spinal hydatid disease remains the gold standard; decompression of the spinal cord is a priority then if there is a risk of instability an osteosynthesis can be indicated. In the review of Neumayr *et al.*, posterior decompression by laminectomy is performed in over 90% of all cases (Andreas Neumayr *et al.*, 2013). The bone involvement lead to an inevitable release of the daughters cysts, the management of such cases demand a radical removal of the bone involved then the use of scolicalid agent, 95% ethanol and 20% hypertonic saline is the most frequently used solutions, scolicalid agent are effective to sterilize the operative field when cyst rupture happen but ineffective against intact cysts which indicate chemotherapy (Andreas Neumayr *et al.*, 2013). The efficiency of benzimidazole derivatives to cure or even prevent recurrence in vertebral hydatid disease is frequently debated but the successful medical treatment of a residual cyst (post-surgery) with a one year course of albendazole has been reported (Baykaner MK *et al.*, 2000) the authors recommend different treatment regimens, such as 10 mg/kg three times daily for 4 months or 10 mg/kg daily in 28 day cycles (Andreas Neumayr *et al.*, 2013, AGTW FIENNES *et al.*, 1982, Baykaner MK *et al.*, 2000).

## **CONCLUSION**

The spinal hydatid disease is rare, and the intra spinal location is the less seen, the spinal location is independent to other locations, the clinical exam is not specific neither the imaging and the laboratory tests are not reliable, reporting such cases will help the surgeon to raise the diagnosis of the hydatid cyst and then deal carefully with its capsule to prevent the rupture which is the major incident.

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