



CASE REPORT

Intrathoracic Extrapleural Hydatid Cyst with Intraspinal Extension Mimicking a Dumbbell Tumour

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Abstract

Hydatid cystic disease or Echinococcosis is endemic in the Mediterranean countries, Middle and Central Asia, South America, Iceland, Australia, New Zealand, and northern and eastern Africa. However, it is unusual in northern Europe and America. Echinococcosis Granulosus or E. Multilocularis causes this zoonotic disease. Humans are accidental hosts of this zoonosis, and transmission occurs through ingesting parasite eggs in contaminated food. Here we present a 22-year-old male who had an Intrathoracic, extra pleural and extradural hydatid cyst, which mimicked a dumbbell tumour and was successfully managed with simultaneous posterior approach laminectomy with thoracotomy.

Keywords: Hydatid cyst, Echinococcosis, Dumbbell Tumor, Spinal canal, Intervertebral foramen

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Introduction

Mediastinal hydatid cyst with spinal canal extension is a very rare form of Echinococcosis that can mimic various mediastinal cystic lesions. Spinal hydatidosis occurs in less than 1% of cases of Echinococcosis. This case highlights a mediastinal hydatid cyst extending into the spinal canal, mimicking a dumbbell tumour. Humans are accidental hosts of this zoonosis. Transmission occurs through ingestion of parasite eggs in contaminated food. *Echinococcus Granulosus* or *E. Multilocularis* may cause it. *E. Granulosus* is more common, benign and encapsulated, whereas *E. Multilocularis* is a rare, non-encapsulated, and malignant form. Hydatid disease is commonly harboured in the lungs and liver, but it may also occur in other sites. The rarity of such presentations emphasizes the need to include hydatid disease in differential diagnoses, especially in endemic areas. If it is not managed early, it can lead to severe neurologic compromise. The mainstay of treatment is surgical excision with anthelmintic medication [1].

Case Presentation

Here we present a 22-year-old male who had an Intrathoracic, extra pleural and extradural hydatid cyst, which mimicked a dumbbell tumour and was successfully managed with simultaneous posterior approach laminectomy with thoracotomy. A previously healthy, 22-year-old male patient without a family history of any neurological diseases presented with complaints of radicular pain over the upper back radiating to the right anterolateral chest wall for three months, associated with tingling and occasional numbness. There was no history of bowel and bladder

incontinence, trauma, weight loss, or fever. Physical examination revealed that the straight leg raise test was pain-free, all four limbs were normal concerning bulk, tone, power, reflexes and sensations, and chest expansion was 5 cm. without any noticeable paraspinal fullness, vertebral tenderness or muscle spasm. Chest X-ray showed a paravertebral opacity close to the right side's 4th and 5th thoracic vertebrae (Figure 1). Hence, it was decided to do further radiological investigations. A contrast-enhanced computerized tomography (CECT) scan of the thoracic region was done, and it showed an intrathoracic dumbbell-shaped mass. For better visualization and to define its spinal relation, Magnetic resonance imaging (MRI) was done, and it revealed a well-defined abnormal signal intensity cystic lesion 51mm x 53 mm x 22 mm seen along the 5th rib in the right paravertebral location, causing scalloping of rib medially and extending into the right T4-T5 neural foramina without compression of the spinal cord (Figure 2 A & B). The lesion was hyper-intense on T2 and short tau inversion recovery (STIR) and hypo-intense on the T1 weighted image of MRI. Whole spine screening revealed no other lesion, and the patient was planned for laminectomy followed by right posterolateral thoracotomy, keeping a possible diagnosis of dumbbell schwannoma due to the presence of radicular symptoms. Under general anaesthesia and in a prone position, T5 right hemilaminectomy was started. Still, on the table, we found a cystic lesion protruding into the canal without any attachment to the nerve root. On careful aspiration, it revealed clear fluid and a pearly white membrane popping out of the cavity, which led to a suspicion of a hydatid

cyst on gross appearance. The intraoperative frozen section confirmed this diagnosis. As all daughter cysts could not be retrieved through this route and due to intrathoracic extension, we thoroughly washed and packed this cavity with 3% hypertonic saline for scolicial action. After completion of this procedure, we proceeded with right posterolateral thoracotomy through the 4th space and found the pleura and lung to be intact, and the cyst was totally extrapleural (Figure 3A). The pleura opened, residual daughter cysts were removed, and communication with the spinal cavity was packed with 3 per cent hypertonic saline (Figure 3 B & C).

After putting the chest tube, thoracotomy closure was done, and he was smoothly extubated on the table. The postoperative course was uneventful. The symptoms were relieved entirely following surgery. Histopathological examination of the excised cyst confirmed the diagnosis of a hydatid cyst (Figure 3 D). He was discharged on the 7th postoperative day with healthy wounds. At the discharge, Tablet Albendazole 15mg/kg daily orally in two divided doses was started, and planned to be continued with periodic assessments. Follow-up visit at the three months has revealed no loco-regional recurrence and normal liver function enzyme status.

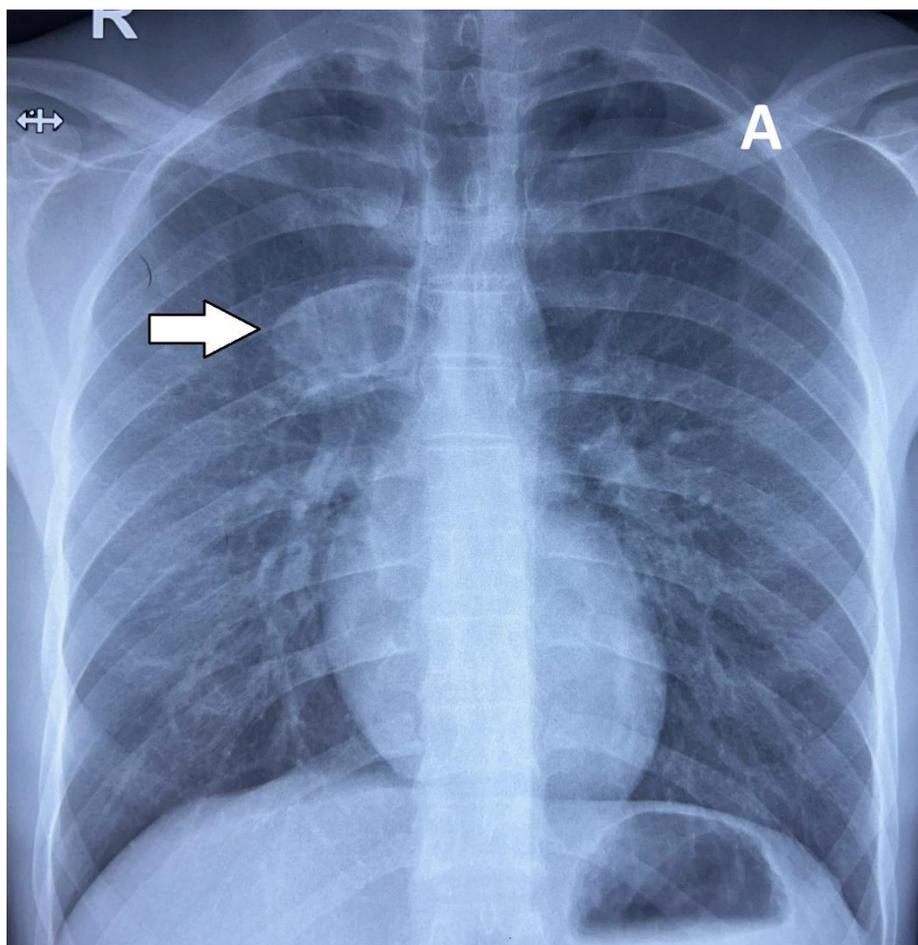


Figure 1. Preoperative Chest X-ray PA view showing paravertebral opacity (marked by arrow).

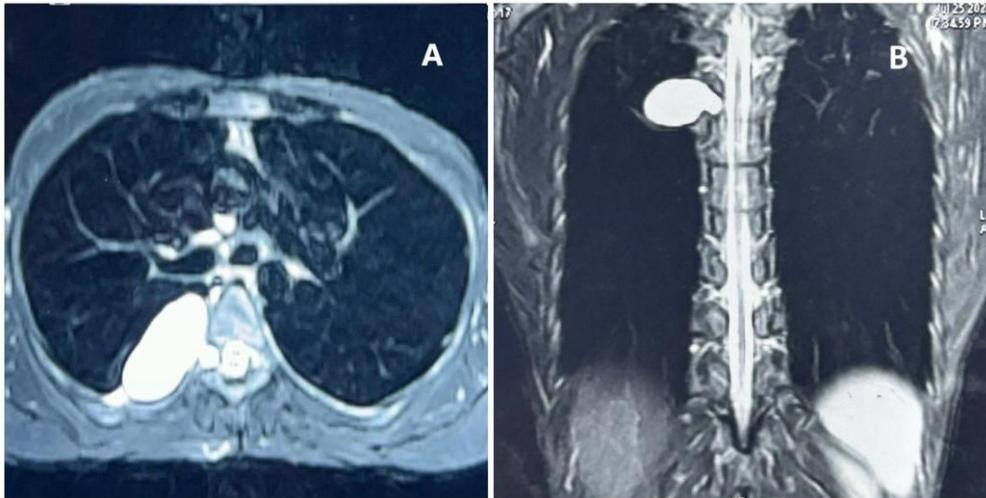


Figure 2. **A:** Coronal T2-weighted Magnetic resonance imaging showing well-defined abnormal signal intensity cystic lesion 51mm x 53 mm x 22 mm seen along 5th rib in right paravertebral location extending into right T4-T5 neural foramina without spinal cord compression. **B:** Axial T2-weighted Magnetic resonance imaging showing well-defined abnormal signal intensity cystic lesion 51mm x 53 mm x 22 mm seen along 5th rib in right paravertebral location extending into right T4-T5 Neural foramina without spinal cord compression.

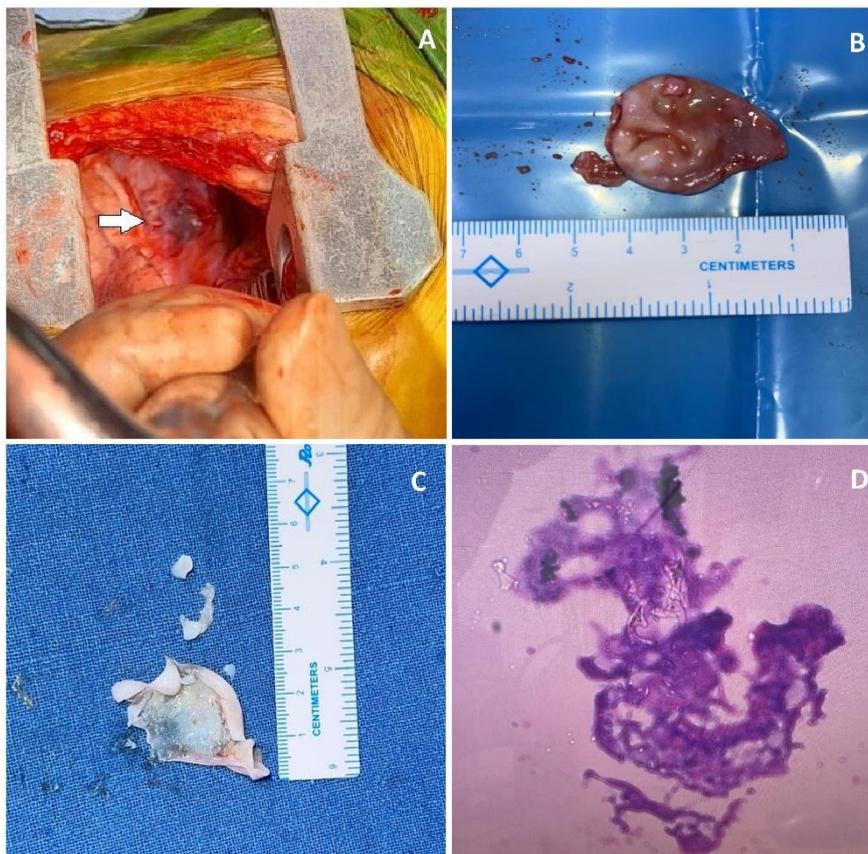


Figure 3. **A:** Intraoperative image showing intact pleura and extrapleural cyst. **B** and **C:** Excised specimen showing daughter cysts with laminated membrane. **D:** Histopathology shows occasional hooklets embedded in fibro collagenous tissue, which suggests a hydatid cyst.

Discussion

Most dumbbell tumors are benign in nature and of neurogenic origin in 90% cases as schwannoma, neurofibroma, ganglioneuroma, and neuroblastoma with schwannomas. Spinal hydatidosis occurs in less than 01% of cases of *Echinococcosis*. Among these, completely extradural hydatid cyst very rarely occurs. Benzagmout *et al.* reported the first transthoracic excision of a para-spinal hydatid cyst [2]. In 90% of cases, these cysts are extradural and usually affect the dorsal vertebra [3]. Spinal involvement can be secondary to liver or lung hydatid or rarely primary, as in our case. Primary paravertebral hydatid without any other foci is explained by porto-vertebral venous shunts theory [4]. Spinal hydatidosis can present with paraparesis (62%), radicular pain (55%), numbness or sensory loss (36%), paraplegia (26%) and sphincter disturbance (30%). Imaging and serological tests can be used in diagnosis. *Echinococcus* IgG has high specificity and low sensitivity. Computed tomography can be used for any lytic lesions or vertebral involvement. MRI is the gold standard in radiological investigation. Braithwaite and Lees classified spinal hydatid cysts into five radiological types: intramedullary, intradural-extramedullary, extradural, hydatid osseous cyst of vertebrae, and paravertebral lesions extending to spinal structures. Advanced imaging, particularly MRI, demonstrates the cyst's nature and intraspinal impact. Hydatid cyst shows cerebrospinal fluid-like intensity in MRI. CT scans complemented MRI findings by detailing bone involvement. Hydatid disease should have a high degree of suspicion in endemic areas. Multiloculated cysts with hypointense T1 weighted MRI

and hyperintense T2 weighted MRI favoured our diagnosis towards hydatid cyst [5]. The correlation between imaging findings and intraoperative discoveries offers an area for further exploration. A hydatid cyst can occur in different sizes and shapes, which may mimic benign and even malignant neoplasms and may create diagnostic challenges in those cases. Especially in the musculoskeletal system and in this region, differentiating these lesions from malignancies such as schwannomas or neuroblastomas is a bit tricky [6].

For managing thoracic dumbbell tumors ultimate aim is to remove both the intraspinal and the extraspinal tumor mass. A combined approach firstly, a posterior laminectomy by a neurosurgeon, followed by a postero-lateral thoracotomy method avoids the risk of bleeding from remnant tumor tissue and compression of the spinal cord. Also, leakage of cerebrospinal fluid and damage to the spinal cord can be easily visualized before thoracotomy by this approach. Hence the standard surgical approach often involved decompression (laminectomy) and resection of intraspinal extended mass with or without spinal stabilization followed by thoracic excision. As the standard management of Hydatid disease is the surgical removal of the cyst along with all daughter cysts coupled with anthelmintic therapy. There are isolated case reports of similar type of presentation [7,8]. Albendazole and Mebendazole both can be used with. However, Albendazole is the preferred drug of choice and is crucial to prevent recurrence, as observed in similar cases [9]. It should be kept in mind that both of these drugs are teratogenic and embryotoxic, and both may cause alterations in liver function and

haematological adverse reactions. The complete blood picture and liver enzyme titre should be assessed monthly during follow-up.

Conclusion

The rarity of such presentations emphasizes the need to include hydatid disease in differential diagnoses, especially in endemic areas. The surgical approach is definitive management and may be individualized based on imaging. Our case has unusual clinical and radiological presentation, so we used simultaneous combined both posterior and anterior approaches with successful outcomes.

Most tumors are benign, neurogenic tumors, with schwannoma, neurofibroma, ganglioneuroma, and neuroblastoma with schwannomas accounting for 90% of all dumbbell tumors

Statements and Declarations

Conflicts of interest

The authors declare that they do not have conflict of interest.

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