Case Report

Primary vertebral echinococcosis: A case of mistaken identity

Ashish Bansal[1], Anjuna Reghunath[1], Mahesh Kumar Mittal[2], Reeta Kanaujia[3], Nishu Raj[4]

From [1]Senior Resident, [2]Professor and Consultant, [3]Associate Professor, [4]Specialist, Department of Radiodiagnosis, VMMC and Safdarjung Hospital, New Delhi, India

ABSTRACT

Spinal hydatid is an uncommon entity that may cause paraplegia due to extensive cord compression. Often, the lesion may be misdiagnosed as spinal tuberculosis, especially in endemic countries. We report such a case of a young female with primary vertebral hydatid disease, who had been on antitubercular treatment for a long time due to misdiagnosis. We discuss the distinguishing diagnostic imaging features of this entity, which would beware radiologists of misdiagnosing this rare but serious condition.

Key words: Echinococcosis, Vertebral hydatid, Vertebral compression

Hydatid disease is a parasitic disease primarily involving the lungs and liver, though it can involve almost any part of the body [1]. Osseous involvement by the disease is very rare, with the spine being the most commonly affected site [2]. Computed tomography (CT) delineates the extent of the osseous lesion while magnetic resonance imaging (MRI) demonstrates neural and cord involvement by the lesion. Knowledge of typical imaging features of spinal hydatid is crucial for prompt and accurate diagnosis as it can be confused with the Pott's spine, especially in endemic countries.

CASE REPORT

A 30-year-old female presented with complaints of back pain and paraplegia. The patient was diagnosed as a case of Pott's spine from outside and was on antitubercular treatment (ATT) for 18 months. She was diagnosed at that time based on a thoracolumbar radiograph done for limb weakness, which showed the collapse of the T12 vertebrae. Despite a course of ATT for 18 months, the patient had progressive weakness of lower limbs and hence had undergone MRI from outside. This MRI revealed T12 vertebral collapse along with multiloculated paravertebral collections and she was continued on ATT, assuming multidrug-resistant tuberculosis of the spine. No vertebral biopsy was performed. After a course of ATT for about 18 months, the patient got referred to our hospital for a contrast-enhanced CT (CECT) scan because of worsening paraplegia and non-response to treatment.

Access this article online	
Received - 07 May 2021 Initial Review - 23 May 2021 Accepted - 25 June 2021	Quick Response code
DOI: 10.32677/IJCR.2021.v07.i07.004	

On clinical examination, the patient had gibbus deformity at the T12-L1 level. Neurological examination revealed motor weakness in the bilateral hip, knee, and foot with a power of 2/5. However, there were no complaints of bowel/bladder incontinence.

CECT was performed for the evaluation of vertebral pathology. CT revealed a large well-defined multiloculated cystic lesion involving the lower dorsal vertebrae and paravertebral region on the right side. The wall of the lesion showed few calcific foci and contrast enhancement, while there was no septal enhancement. The lesion extended medially into the spinal canal through the right neural foramen of the T11 vertebra, causing compression and displacement of the spinal cord, and posterolaterally into the right paraspinal muscle (Fig. 1a and b). There was a complete collapse of the T12 vertebra causing acute kyphotic deformity, lytic destruction of the right half of the T11 vertebra including transverse process, lamina, pedicle, and spinous process along with posterior scalloping of T10 vertebral body. Furthermore, there was no significant bone expansion, sclerosis, osteopenia, or periostitis. T11-T12 and T12-L1 intervertebral discs and adjacent endplates were relatively well preserved. Mildly expansive, multiloculated lytic lesions were also seen in the adjacent right 10th and 11th ribs (Fig. 2a and b). However, the ribs showed no evidence of sclerosis or cortical destruction. Another well-defined, unilocular cystic lesion with enhancing wall was seen arising from the left crus of the diaphragm (Fig. 1a and b). Based on imaging findings, a diagnosis of primary spinal hydatidosis and left diaphragmatic crus hydatid was made at our institute after excluding other lesions in the thorax and abdomen.

Correspondence to: Dr. Reeta Kanaujia, Department of Radiology and Imaging, Vardhman Mahavir Medical College and Safdarjung Hospital, New Delhi - 110029, India. E-mail: reetakanaujia@gamil.com

On reviewing the previous radiograph of the patient, concertina collapse of T12 vertebra with relative sparing of adjacent intervertebral discs and endplates was appreciated (Fig. 3). Mild posterior angulation was seen at the thoracolumbar junction, due to the collapse. No obvious paravertebral opacity could be delineated. Her previous MRI also showed the classical "cyst-within-cyst" appearance of the right paravertebral lesion and the "diverticulated cysts" appearance of the vertebral lesion. The typical absence of discitis, perilesional soft tissue, and bone marrow edema points against tuberculosis. MRI also clearly depicted the posterior subligamentous spread, besides the epidural extension and cord compression by the lesion (Fig. 4a-c). However, no signal alteration or atrophy was seen in the cord. A subsequent positive IgG-ELISA serology test for hydatid confirmed our diagnosis.

The patient was started on albendazole and subsequently underwent decompression surgery with T11, T12 laminectomy. However, the patient was then lost to follow-up.

DISCUSSION

Hydatid disease is a parasitic infection caused by larval stages of *Echinococcus granulosus* and *Echinococcus multilocularis,* the former being the most frequent causal organism with a global presence. The man may become an intermediate host by the ingestion of parasite ova [3]. Osseous involvement occurs only in 0.28%–3.1% of cases of hydatid disease with vertebral involvement seen in one-third to one-half of cases [4]. Other sites that may be involved are pelvic bones, femur, tibia, humerus, skull, and ribs. Braithwaite and Lees [5] had classified spinal hydatid as intramedullary, intradural extramedullary, extradural intraspinal, vertebral (most common), and paravertebral. When hydatid disease extends from the spinal canal through the neural foramen into the spinal canal, it is referred to as "dumbbell" formation or configuration [6].

Primary hydatid cysts of the spinal cord, intradural and extradural cysts are very rare. Vertebral hydatid is mostly due to the hematogenous spread of *Echinococcus* embryo to highly vascularized zones of the vertebra. Other sources of spread are direct invasion from extraspinal structures and infection secondary to cerebrospinal fluid seeding from ruptured cysts of cerebral hydatid. Hydatid grows along with intratrabecular spaces by exogenous vesiculation giving the appearance of diverticulated cysts. Concertina collapse (vertebral collapse without the involvement of disc) has not been typically described for spinal hydatid in the literature, although few studies have mentioned the occurrence of the same in spinal tuberculosis [7]. We suspect that the vertebral collapse, in this case, could be due to the chronicity of the lesion, as bone destruction leading to collapse can be seen in advanced cases of spinal hydatid. Local ischemic necrosis and direct local pressure erosion of bone have been attributed to this occurrence [8]. Bony destruction, paraspinal, and adjacent rib involvement are the hallmarks of vertebral hydatid. The patient may present with radiculopathy, back pain, and paraparesis [3,4].

The radiological approach to vertebral hydatidosis includes radiographs, CT, and MRI. Differential diagnoses include various types of osteomyelitis and aggressive tumors like metastasis, a comparison of which is discussed in Table 1 [1,7]. However, typical imaging characteristics of primary vertebral hydatid

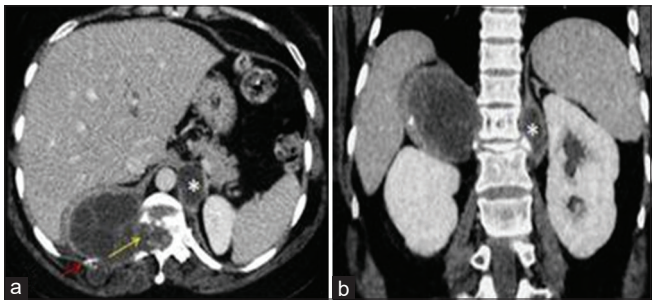

Figure 1: CECT axial (a) and coronal (b) reconstruction reveals multiloculated cystic lesion in the right paravertebral region, with intraspinal extension (yellow arrow). A calcific focus is seen in the wall of the lesion (red arrow). Another cystic lesion (asterisk) is seen in the left crus of diaphragm

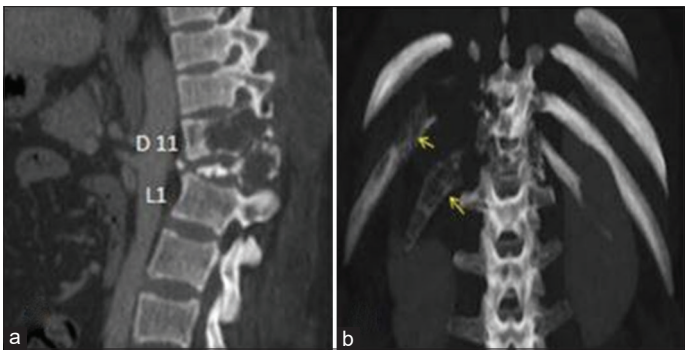

Figure 2: Sagittal (a) and coronal MIP (b) reformats of CT scan of dorsolumbar region in bone window show collapse of T12 and lytic destruction of T11 vertebral body and its right-sided posterior elements. A multiloculated lytic lesion is seen involving right 11th and 12th ribs (yellow arrows), causing mild expansion of the ribs, however, no sclerosis or cortical destruction noted

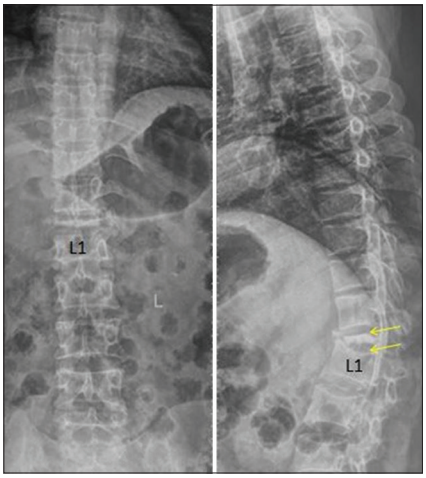

Figure 3: Frontal and lateral radiographs of the dorsolumbar spine show concertina collapse of T12 vertebra with relatively maintained T11-T12 and T12-L1 intervertebral disc spaces (yellow arrows) and preserved inferior and superior end plates of T11 and L1 vertebrae respectively. Mild kyphotic deformity is observed at thoracolumbar junction

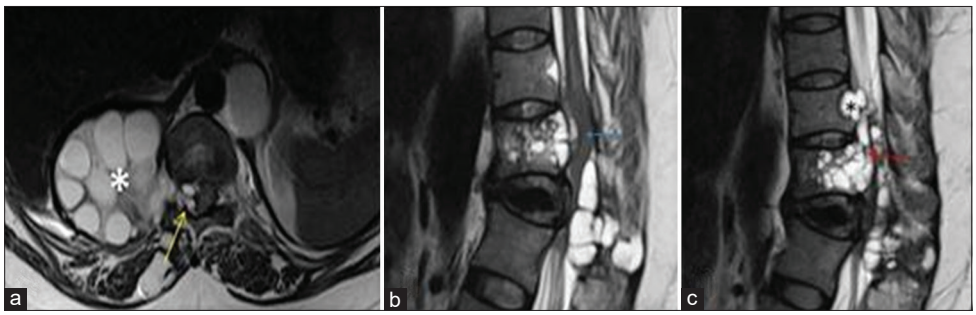

Figure 4: Axial T2-weighted MRI (a) shows typical "cyst-within-cyst" appearance of paravertebral lesion (asterisk) with intraspinal extension (yellow arrow). Sagittal T2-weighted images. (b,c) Cord compression by the lesion (blue arrow) and characteristic diverticulated-cyst appearance (asterisk) of vertebral hydatidosis with posterior subligamentous extension (red arrow). Cord signal appears to be normal

Table 1: Comparison of the clinicoradiological features of spinal lytic lesions

	Pyogenic	Tubercular	Brucellar	Hydatid	Metastasis
Duration of illness	Short	Intermediate	Short/intermediate	Long	Short
Age of presentation	Any age	Children and young adults	Middle aged	Middle aged	Middle aged and the elderly
Spinal and paraspinal involvement	Vertebral body and intervertebral disc, minimal soft-tissue involvement.	Vertebral body and intervertebral disc, extensive soft-tissue involvement. Lack of proteolytic enzymes, unlike pyogenic infection, has been attributed to the preferential subligamentous spread of infection [7].	Vertebral body and intervertebral disc, minimal soft-tissue involvement, and sacroiliitis.	Vertebral body, adjacent bones (like contiguous ribs), and soft tissue are involved, intervertebral discs spared usually. Subligamentous spread is common as growth of hydatid cysts occurs along line of least resistance [5].	Posterior vertebral body, lamina, and pedicles.
Salient imaging features	Destruction of vertebral bodies and disc spaces, marked enhancement of the lesion, epidural abscess	Destruction of vertebral bodies and disc spaces although disc is involved late as compared to pyogenic, rim enhancement of the soft-tissue masses	Intact vertebral architecture despite diffuse vertebral osteomyelitis. Presence of gas in disc.	Multicystic lesions in multiple contiguous vertebral bodies and ribs, usually no vertebral collapse. Discs and endplates are spared. No significant enhancement, marrow edema, and periosteal reaction.	T1-hypointense, T2-hyperintense lesions with heterogeneous enhancement. Posterior convex margin of vertebral body and associated soft tissue may be seen.
Laboratory features	Leukocytosis, raised ESR and C-RP	Raised ESR	Leukocytosis with raised ESR	IgE-ELISA positive for hydatid	Absence of leukocytosis. ESR and CRP not raised.

disease help in making an accurate diagnosis. These include lack of bone expansion, osteopenia, sclerosis and periostitis in affected bones, sparing of intervertebral discs, absence of perilesional edema, subperiosteal and subligamentous spread of disease, a paraspinal extension of the lesion, and the involvement of contiguous ribs; all of which were seen in our case [3,5]. Besides, our case was unique due to synchronous hydatid disease of the left diaphragmatic crus, which is hitherto unreported. The serological diagnosis of spinal hydatid includes the IgG ELISA test, but it may be negative in chronic cases [9]. Casoni and latex tests are unreliable in osseous hydatid. Imaging is considered diagnostic of the condition and treatment may be started based on imaging.

Treatment options include surgical, medical, and percutaneous methods. Surgery involving simple decompression with laminectomy and fixation is the treatment of choice [10]. Albendazole plays a role in reducing recurrence after surgery [10]. Percutaneous treatment with CT-guided PAIR approach (puncture of cyst, aspiration of contents, injection of scolicidal agent, and reaspiration of the contents) is particularly useful in patients with an extensive disease where radical removal is impossible. Various agents that can be used in PAIR include 3–7.2% hypertonic saline, povidone-iodine, cetrimide, formalin, hydrogen peroxide, and silver nitrate. However, since recurrence is common, patients should undergo post-surgical routine imaging follow-up [11].

CONCLUSION

Primary vertebral echinococcosis is a rare differential diagnosis for the vertebral collapse in a country endemic for spinal tuberculosis. However, clinical suspicion and familiarity with the imaging characteristics of spinal hydatid disease can help the astute radiologist in arriving at an early and accurate diagnosis. The absence of bone marrow and soft-tissue edema and sparing of intervertebral discs could be useful pointers toward diagnosing

this unusual condition, thus significantly decreasing the morbidity from delay in providing appropriate treatment.

REFERENCES

1. Jain S, Verma A, Rohra S. A rare case to primary spinal hydatid cyst presenting with neurodeficit. J Orthop Traumatol Rehabil 2017;9:134-8.
2. Charles RW, Govender S, Naidoo KS. Echinococcal infection of the spine with neural involvement. Spine (Phila Pa 1976) 1988;13:47-9.
3. Pedrosa I, Saiz A, Arrazola J, Ferreirós J, Pedrosa CS. Hydatid disease: Radiologic and pathologic features and complications. Radiographics 2000;20:795-817.
4. Tsitouridis I, Dimitriadis AS. CT and MRI in vertebral hydatid disease. Eur Radiol 1997;7:1207-10.
5. Braithwaite PA, Lees RF. Vertebral hydatid disease: Radiological assessment. Radiology 1981;140:763-6.
6. Jagadale RR, Yashi K, Reddy U, Agrawal A. Dumbell hydatid disease of spine with posterior paraspinal muscle involvement. Rom Neurosurg 2015;3:363-7.
7. Garg RK, Somvanshi DS. Spinal tuberculosis: A review. J Spinal Cord Med 2011;34:440-54.
8. Alijani B, Emamhadi M, Yousefzadeh-Chabok S, Behzadnia H, Dehghani S. Hydatid disease of the cervical spine mimicking traumatic burst fracture. Iran J Neurosurg 2016;1:20-3.
9. Pamir MN, Ozduman K, Elmaci I. Spinal hydatid disease. Spinal Cord 2002;40:153-60.
10. Bhake A, Agrawal A. Hydatid disease of the spine. J Neurosci Rural Pract 2010;1:61-2.
11. Neumayr A, Tamarozzi F, Goblirsch S, Blum J, Brunetti E. Spinal cystic echinococcosis-a systematic analysis and review of the literature: Part 2. Treatment, follow-up and outcome. PLoS Negl Trop Dis 2013;7:e2458.

Funding: None; Conflicts of Interest: None Stated.

How to cite this article: Bansal A, Reghunath A, Mittal MK, Kanaujia R, Raj N. Primary vertebral echinococcosis: A case of mistaken identity. Indian J Case Reports. 2021;7(7):278-281.