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Imaging diagnosis and management of primary spinal hydatid disease: a case series

Neha Singh[1*], Tushar Anand[1] and Deepak Kumar Singh[2]

Abstract

Background: Primary spinal hydatid disease (HD) is uncommon disease with significant morbidity. The diagnosis of this entity is not simple, unless the patient comes from an endemic area or has a history of HD elsewhere. Only few case reports and case series of this entity are available in the published literature. We report a series of three cases of primary spinal HD who had characteristic MR appearance and were managed successfully.

Case presentation: We report a series of 3 cases, two presenting with paraparesis and one with right lower limb weakness and hesitancy of micturition. MRI demonstrated multiloculated cystic lesion involving thoracic spine in two patients and lumbar spine in third patient. All the three patients were showing intraspinal extension with compression of neural structures. MRI features were characteristic of hydatid disease. All the three patients were managed surgically with gross total excision of the cysts followed by albendazole chemotherapy. Histopathology was consistent with hydatid cyst. Patients with dorsal spine disease showed improvement in motor weakness, but third one with lumbar spinal disease had to use foot splint for walking. None of the patient showed features of recurrence on follow-up.

Conclusion: Radiologist should be familiar with the typical and atypical manifestations of the disease and should keep it in the differential diagnosis of cystic spinal lesions. Radical surgical excision along with postoperative adjuvant albendazole chemotherapy is the mainstay of treatment.

Keywords: Spinal hydatid, Hydatid disease, Primary spinal hydatid, Echinococcus

Background

The zoonotic infection of hydatid disease (HD) is caused by the larva of *Echinococcus* tapeworm with the two main species being *E. granulosus* and *E. multilocularis* which are endemic to the temperate climate [1]. The definitive host is usually a dog, and the intermediate host is usually sheep. Humans can become intermediate hosts on coming in contact with a definitive host and develop infection [2]. Most commonly involved organs in primary HD are liver and lungs constituting 60–70% and 10–15% of cases, respectively [3]. Bony involvement is a rare complication and occurs by hematogenous route seen in 0.5–4% of cases [4]. Involvement of musculoskeletal system is secondary and infrequent with an estimated incidence of ~ 2.5%. The bones affected in decreasing order of incidence are pelvis, sacrum, metaphysis of the long bones, skull, spine and ribs. Involvement of spine is seen in less than 1% cases which is equivalent to almost 50% of the 0.5–4% of cases affecting the bone [1]. Thoracic spine is the most frequently involved site (52%) followed by lumbar (37%), sacral and cervical spine [5]. The spinal hydatidosis is a rare cause of neurological symptoms and does not always have typical imaging appearance. Sometimes it may be difficult to differentiate it from tubercular spondylitis or chronic osteomyelitis. So, in endemic areas like India, a differential of spinal HD should be kept in a case of compressive myelopathy if imaging suggests cystic lesions involving the spine [1, 6]. Herein, we report a series of 3 cases of spinal HD, two involving thoracic

*Correspondence: neha.singh.dr@gmail.com

[1] Department of Radiodiagnosis and Imaging, Dr. Ram Manohar Lohia Institute of Medical Sciences, Lucknow 226010, India
Full list of author information is available at the end of the article

and one involving lumbar spine. All the three cases had characteristic imaging appearance on MRI and were histopathologically proven postoperatively. The aim of this article is to share our experiences in the diagnosis and management of spinal hydatid disease and to provide an outlook through review of the literature. All the three patients provided informed consent for the study.

Case presentation

During last 12 years, we diagnosed and successfully managed three patients of primary spinal hydatid disease. Our case series included one man and two women with age range of 30–53 years. All the three patients presented with progressively increasing lower limb weakness. One patient had low backache with hesitancy of micturition and one had frequency of micturition. Two patients had spastic paraparesis. One showed exaggerated deep tendon reflexes with the absence of superficial abdominal and cremasteric reflexes. One patient had bony tenderness at low back region with weakness of dorsiflexion of foot and perianal hypoesthesia (Table 1).

All the three patients had undergone spinal MRI which revealed multiloculated cystic lesion in the spinal column. The lesion was involving vertebrae in two patients and was extradural in third. In all the three patients, lesions were hypointense on T1WI, hyperintense on STIR and T2WI with T2 hypointense thin and smooth cyst wall. Post-contrast images showed absent or rim enhancement. Extension of the lesion within the spinal canal (with compression of underlying cord or cauda equina) and in extraspinal soft tissue was present in all the three cases (Figs. 1, 2 and 3). Based on these features, possibility of spinal hydatid disease was kept and complete work-up was done to rule out the presence of primary focus elsewhere in the body, but the tests were negative.

All patients were treated surgically with excision of the cysts followed by hypertonic saline wash and decompressive laminectomy. Per-operative findings revealed bunch of multiple pearly white cysts compressing the dural sac (Fig. 1d). In one patient, intraoperative rupture of cyst occurred, and to prevent recurrence, the cavity was washed thoroughly with diluted hydrogen peroxide and hypertonic saline. Finally, anterior vertebral column reconstruction was done using expandable cage and wound was closed in layers over closed suction drain. Postoperative period was uneventful in all the three patients. Histopathology confirmed the diagnosis of hydatid disease, and albendazole was given for a period of 3 months to prevent recurrence. Two patients had shown complete resolution of their symptoms, but in one patient motor weakness persisted. None of our patients had shown features of recurrence on 1- to 2-year follow-up (Table 2).

Discussion

Hydatid disease of bones is uncommon, and the spine is involved in ~ 50% of these cases. It is a rare occurrence, even in rural areas where echinococcus is endemic, but it is the most serious. Primary infestation of the spine occurs through the porto-vertebral shunts, and the center of the vertebral body is the first site to get involved. When larva penetrates cancellous bone in the vertebrae, it causes a multivesicular and diffuse infiltration [7]. In bony HD, pericyst formation does not occur, thus allowing aggressive proliferation along the line of least resistance, particularly the bony canals [4].

Table 1 Clinical presentation of patients

S. no.	Age(y)/sex	Symptoms	Clinical examination
1	30/F	Progressively increasing weakness in both LL*	-MRC# grade 3 spastic paraparesis, -B/L increased Deep Tendon Reflexes, -B/L plantar extensor -Superficial abdominal and cremasteric reflexes absent
2	50/M	Weakness both LL*, frequency of micturition	Spastic paraparesis
3	53/F	Low backache, Right LL weakness, hesitancy of micturition	Bony tenderness at low back region, MRC# grade 2 weakness in dorsiflexion right ankle jerk absent perianal hypoesthesia

**LL* Lower limbs, #*MRC* Medical research council

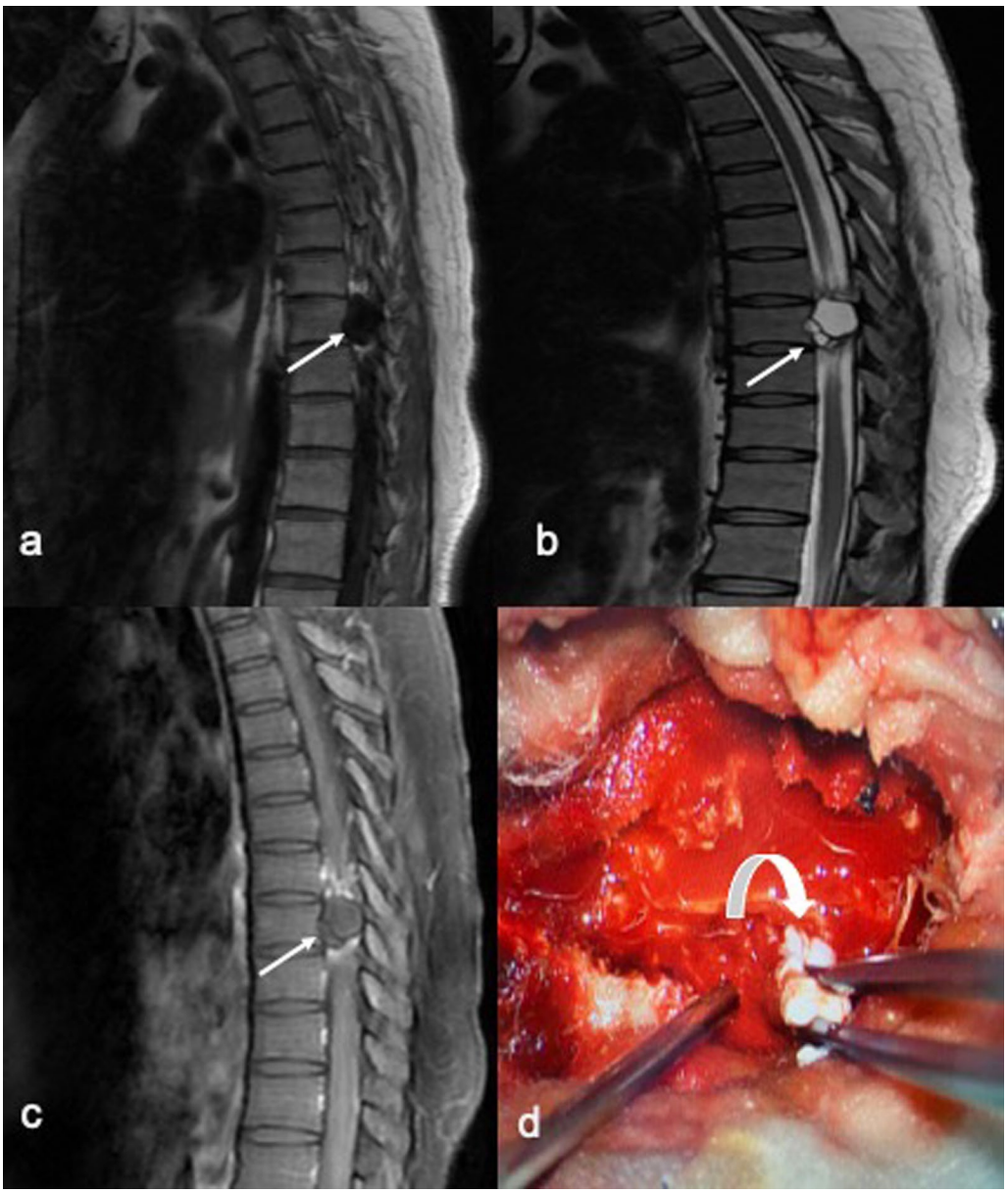

Fig. 1 MRI of dorsal spine in sagittal plane shows a well-defined cystic lesion in the spinal canal [marked with white arrow] **a** hypointense on T1WI and **b** hyperintense on T2WI. **c** Fat-suppressed contrast-enhanced sagittal T1WI shows the absence of enhancement in cyst or its wall. Mild enhancement of adjacent compressed tissue is present. **d** Per-operative image shows bunch of multiple pearly white cysts (curved white arrow) lying in the extradural space

Eventually, daughter vesicles invade the bone and replace the medullary cavity and the disease reaches the cortex and destroys it with subsequent spread of the disease to surrounding tissues [8]. They may grow to very large sizes and may remain asymptomatic for years. While extraosseous hydatids often calcify, intraosseous hydatids rarely do so [9]. In majority of the cases, the disease is confined to the bone and the epidural space [10]. Intradural

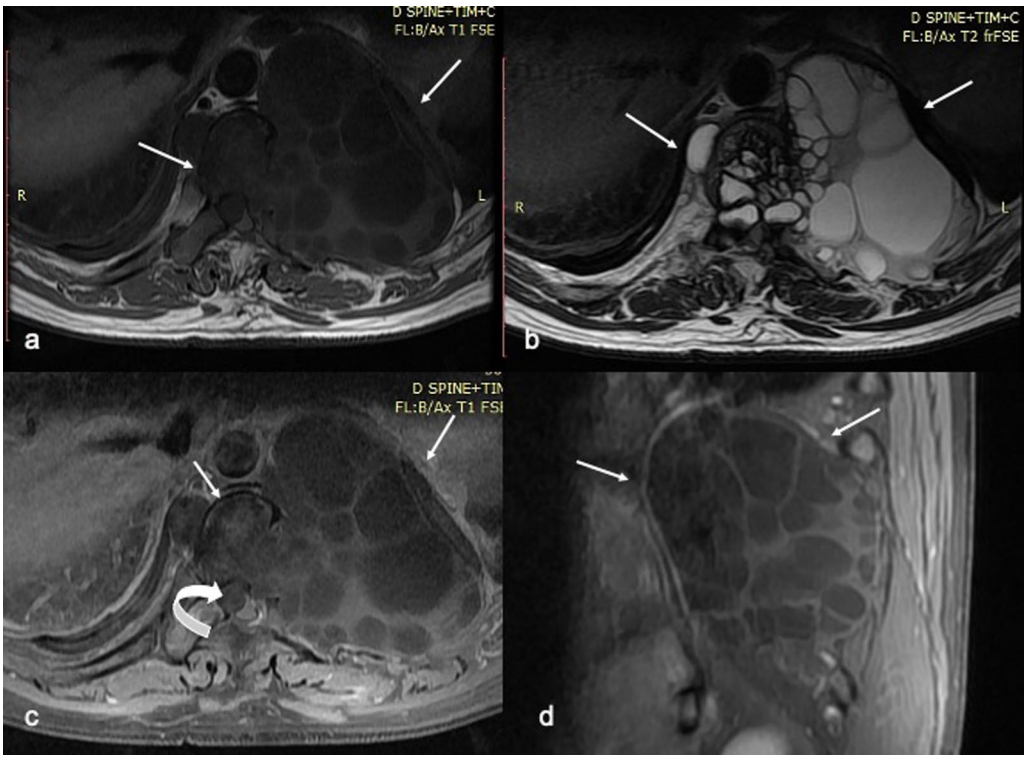

Fig. 2 MRI of dorsal spine in axial plane shows a large multiloculated cystic lesion (white arrow) with extension in adjacent paravertebral soft tissue (left > right) and spinal canal with cord compression (curved white arrow). The lesion is **a** hypointense on T1WI and **b** hyperintense on T2WI. **c** Fat-suppressed contrast-enhanced axial and **d** sagittal T1WI shows patchy rim enhancement. Multiple daughter cysts are clearly visible within the lesion

extramedullary disease has been reported only in 9% of the cases [11, 12]. Neurological complications arise as a result of compressive myelopathy or neuropathy secondary to intradural and extradural disease component.

On radiograph, the disease may mimic non-specific osteomyelitis in its initial phase because of medullary infiltration by the larvae. Bony erosion, osteolysis and destruction occur with progression of time, and in later stages, a multiloculated cystic appearance may be seen [13]. Some typical imaging features of spinal HD include lack of osteoporosis or sclerosis in involved bone, lack of intervertebral disk space involvement and subperiosteal, subligamentous or paraspinal extension of the disease [8]. CT may contribute in better demarcation of the lesion showing bony expansion with honeycomb-like appearance of the spine and may even demonstrate rim calcification [13]. MRI is superior in assessing the lesion extent and neural involvement and can help in ruling out other differentials. The lesions show intermediate to low-signal intensities on T1-weighted and high-signal intensity on T2-weighted images with multilocularity being a key feature [14]. Hyperintensity on T2WI suggests viable cysts, whereas isointense signal to muscle on both T1 and T2WI or T2 hypointense signals and T1 hypointense signals represent dying or dead cysts. Though efficacy of albendazole for primary bony hydatid treatment is questionable, postoperative albendazole therapy seems only way to prevent recurrence [15]. All of our patients received albendazole (15 mg/kg) in divided doses daily for 3 months, and there was no recurrence in any of our cases till latest follow-up. Because of poor bony penetration of albendazole, radical excisional surgery of primary disease should be done at earliest. The aim of surgery should be removal of all cysts along with involved bone and soft tissues. We were able to get good surgical clearance in all our cases. We irrigated the surgical field with hypertonic saline to

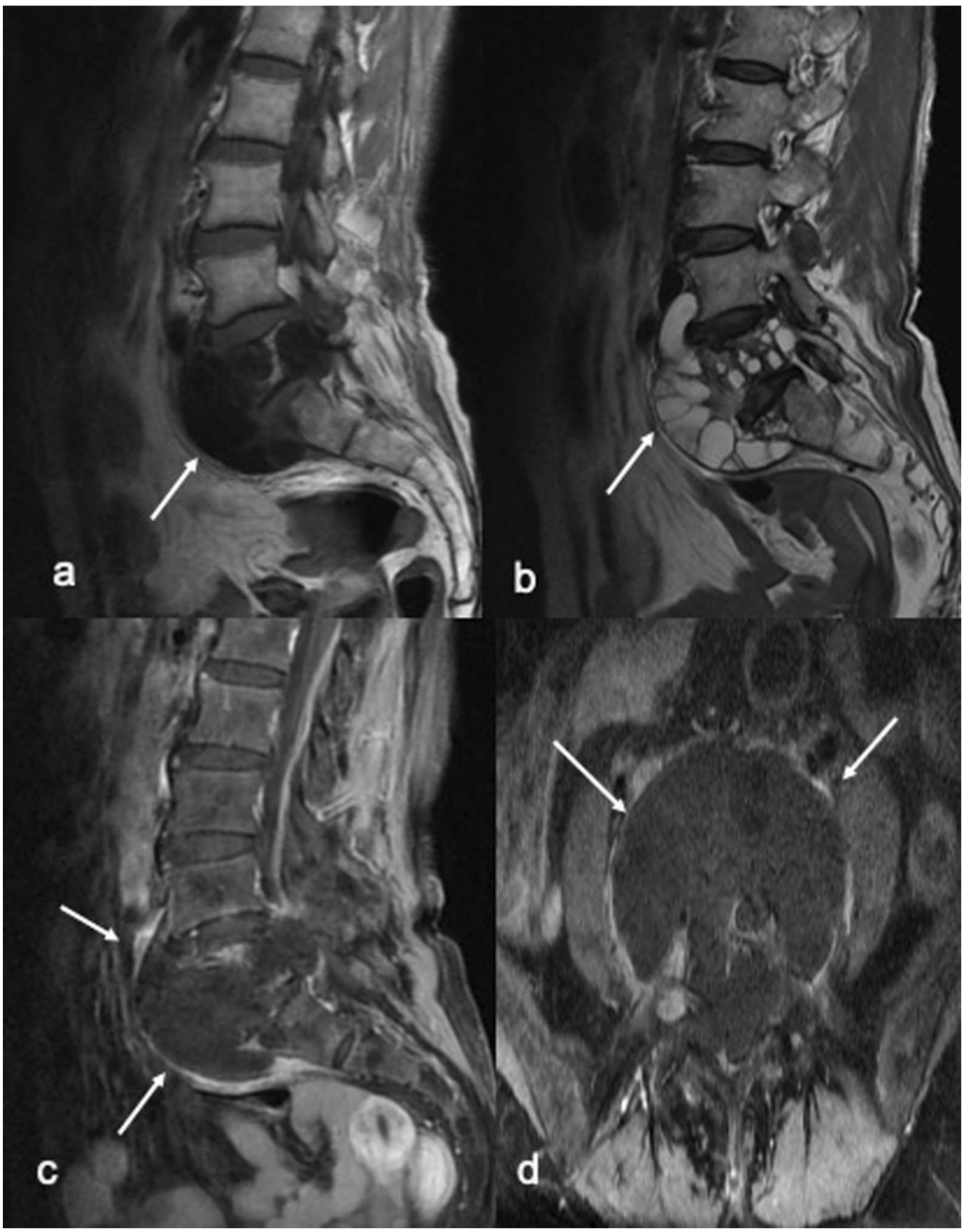

Fig. 3 MRI of lumbosacral spine in sagittal plane shows a large multiloculated cystic lesion involving L5 vertebra with extension in adjacent prevertebral soft tissue and spinal canal. The lesion is **a** hypointense on T1WI and **b** hyperintense on T2WI. **c** Fat-suppressed contrast-enhanced sagittal and **d** axial T1WI shows rim enhancement of adjacent compressed tissue (marked with white arrow)

prevent local recurrence (16). Though there was a theoretical risk of chemical injury, we did not encounter any such problem. The strength of our study is that it confirms the role of diagnostic imaging and radical excision in the management of these rare lesions, and only possible limitation is short-term follow-up to conclude complete remission.

Table 2 Imaging features, management and outcome of patients

S. no.	MRI findings				Imaging differentials	Imaging evaluative for primary hydatid disease elsewhere	Management	Outcome	Recurrence
	Location	Signal characteristics	Compression over cord/ cauda equina	Extraspinal soft tissue involvement					
1	Extradural cystic lesion at D8-9 level	T1-hypo, T2/STIR-hyper with thin smooth hypo-wall, No enhancement	Cord compression present	Absent	Hydatidosis Cysticercosis Cystic tumor	Negative	D8 to D10 laminectomy with cyst excision f/b albendazole therapy	Improved	Absent
2	D9-10 vertebral body multiloculated cystic lesion	T1-hypo, T2/STIR-hyper with thin smooth hypo-wall, patchy rim enhancement	Cord compression present	Present	Hydatid disease	Negative	D8-D10 transpedicular rod and screw fixation with D9 hemilaminectomy and cyst excision	Improved	Absent
3	L5 vertebral body multiloculated cystic lesion	T1-hypo, T2/STIR-hyper with thin smooth hypo-wall, patchy rim enhancement	Cauda equina compression present	Present	Hydatid disease	Negative	L3-4 7 S1 transpedicular rod and screw fixation with L4-5 laminectomy and L5 corpectomy with total cyst excision	Motor weakness persisted	Absent

Conclusion

Spinal HD is uncommon disease with significant morbidity. The diagnosis of this entity is not simple, unless the patient comes from an endemic area or has a history of HD elsewhere. It may be misdiagnosed as spondylitis at initial radiological evaluation. Additional imaging with CT and especially MRI may be crucial in making the correct diagnosis. Radiologist should be familiar with the atypical manifestations of the disease and should keep it in the differential diagnosis of cystic spinal lesions. Radical surgical excision along with postoperative adjuvant albendazole chemotherapy is the mainstay of treatment. Our study supports the standard diagnostic and treatment modalities already published in scientific literature.

Abbreviations
HD: Hydatid disease; MRC: Medical research council.

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Author contributions
NS contributed to conceptualization and manuscript writing, and TA was involved in data collection and image formation. DS helped in manuscript writing and proof reading of the manuscript. All authors read and approved the final manuscript.

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Availability of data and materials
The datasets used during the current study can be made available from the corresponding author on reasonable request.

Declarations

Ethics approval and Consent to participate
Not applicable. Written consent taken

Consent for publication
Taken in writing.

Competing interests
The authors declare that they have no competing interests.

Author details
[1]Department of Radiodiagnosis and Imaging, Dr. Ram Manohar Lohia Institute of Medical Sciences, Lucknow 226010, India. [2]Department of Neurosurgery, Dr. Ram Manohar Lohia Institute of Medical Sciences, Lucknow, India.

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