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Primary spinal extradural hydatid cyst in a child: case report and review of the literature

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Abstract Spinal hydatid cyst is a rare but serious condition. An 8-year-old boy presented with back pain, progressive weakness and numbness in both legs. Magnetic resonance imaging (MRI) of the lumbar region showed a cystic lesion with regular contour located in extradural space. There was cerebrospinal fluid- (CSF-) like signal intensity on T1- and T2-weighted images. The lesion had excessively compressed the dural sac and caudal roots, and expanded to the L3 and L4 neural foramina. The

case was explored with L2, L3, L4 laminectomy and the hydatid cyst was removed totally. The clinical presentation, diagnosis and surgical treatment of this rare case of spinal hydatid disease is discussed, and all available cases of primary extradural hydatid cyst reported in the literature are presented.

Keywords Hydatid cyst · Spine · MRI · Surgery · *Echinococcus granulosus*

Introduction

Hydatid disease is more common in young people who have had contact with dogs, wolves or cats [9, 12]. In a large percentage of cases, *echinococcus granulosus* usually involves the liver and lungs.

Hydatid disease of the bone is found in 1% of all cases, with the spine being involved in 45% of bone infestations. Disease usually spreads to the spine by direct extension from pulmonary, abdominal or pelvic infestation, and most commonly affects the dorsal region [2, 9, 11, 12, 20]. Primary spinal extradural hydatid cyst is extremely rare.

A rare case of primary lumbar extradural hydatid cyst, which causes cauda equina compression, is reported and clinical presentation, diagnosis and surgical treatment are discussed.

Case report

An 8-year-old boy from Eastern Anatolia, who had not suffered from any other disease till then, presented with back pain, progres-

sive weakness, numbness in both legs and difficulty in walking; all symptoms having appeared 7 months earlier.

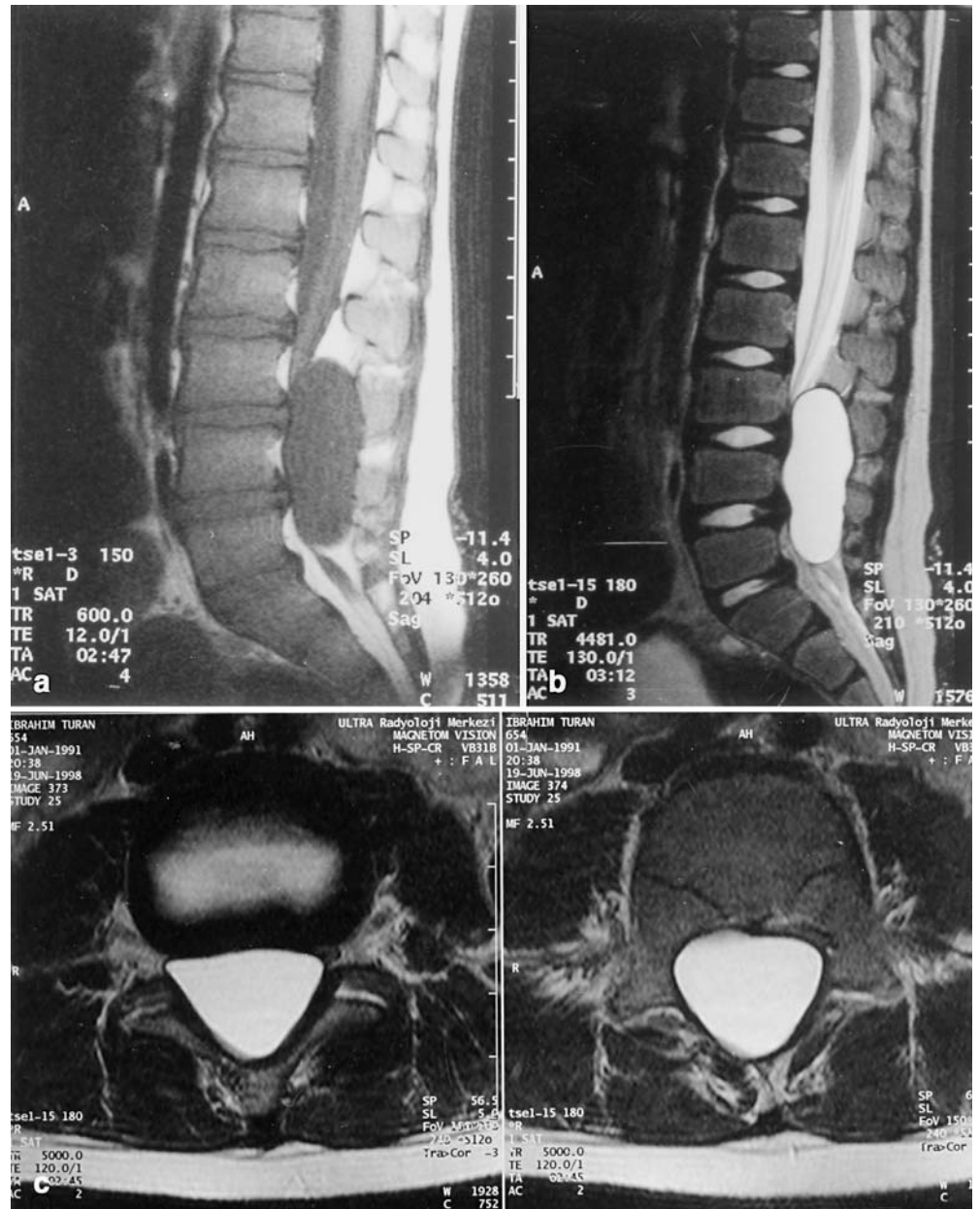
Physical examination showed no abnormality. Neurological examination revealed perianal hypoesthesia and paraparesis prominent on the right and distal muscle groups. There was loss of patella and Achilles reflexes, as well as urinary and anal incontinence.

Abdominal ultrasonography scans and magnetic resonance (MR) images of the cranium and cervical region were normal. MRI of the lumbar region showed an intracanalicular cystic lesion with a regular contour and extradural location, with dimensions of 4.9 cm craniocaudally and 2.5 cm axially. There was cerebrospinal fluid- (CSF-) like signal intensity on T1- and T2-weighted images. The lesion had excessively compressed the dural sac and caudal roots and had expanded to the L3 and L4 neural foramina without any bone involvement (Fig. 1). Initially, the lesion was considered to be a hydatid cyst, but serological tests (specific ELISA/Western blot) proved negative.

The case was explored with L2, L3, L4 laminectomy. The lesion was removed totally, with a fibrous pseudocapsule that tightly adhered to the dural sac and bone, and the cavity was irrigated with hypertonic saline. Histopathological examination revealed a hydatid cyst.

Albendazole treatment was applied in the early postoperative stage. Paraparesis was improved almost completely in later controls extending over 3 years, and postoperative MRI scans showed that the hydatid cyst had been totally removed (Fig. 2). Anal incontinence was improved, while urinary incontinence persisted, but in a decreasing manner.

Fig. 1 A,B Preoperative sagittal T1- and T2-weighted spinal magnetic resonance (MR) images showing cystic lesion located in extradural space. C Preoperative axial T2-weighted spinal MR image



Discussion

Primary hydatid infestation of the spine without any other systemic involvement can be explained through the direct porto-vertebral venous shunt theory: in rare instances, the disease begins from the extradural area, suggesting that the parasite's embryo is possibly being carried through the porto-vertebral venous shunts [9].

Braithwaite and Lees classified spinal hydatid cysts into five radiological types: intramedullary, intradural-extramedullary, extradural, hydatid cyst of vertebrae and paravertebral lesions extending to spinal structures [8].

The last two groups affect the spinal neural structures by extension and these are more common, but the first three groups are primary infestations and extremely rare.

Spinal hydatid cysts are usually situated in the dorsal region and generate medullary or radicular symptoms according to their location [1, 3, 6, 13, 14, 18]. In the present case the hydatid cyst was extradural and located in the lumbar region.

All available cases of primary spinal extradural hydatid cysts are presented in Table 1. Primary vertebral cysts originating from the vertebral bone and reaching the extradural space after having destroyed the bone are excluded. In the English language literature there are only

Fig. 2A,B Postoperative MR image showing that the hydatid cyst was totally removed

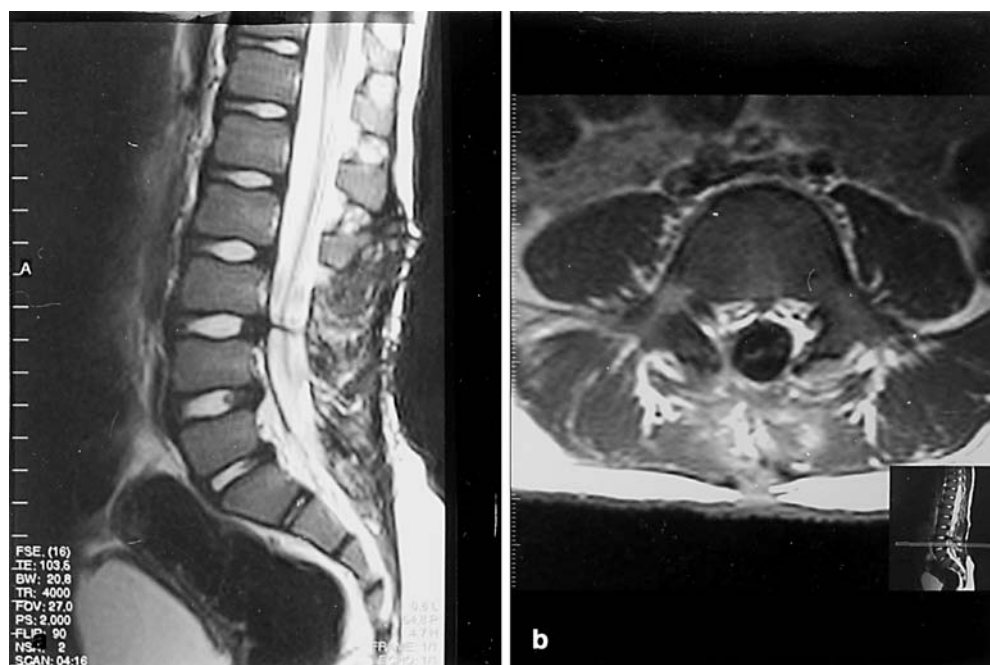


Table 1 Details of all primary spinal extradural hydatid cysts recorded in the literature (*CT* computed tomography, *USG* ultrasonography, *MRI* magnetic resonance imaging)

	No. of patients	Age	Gender	Location	Serology	Neurologic status	Radiology	Treatment	Outcome
Pluchino and Lodrini (1981) [15]	1	56	Male	T10–L2	Casoni(–)	Paraparesis	Myelography	Posterior	Complete recovery
Wani et al. (1989) [18]	1	14	Male	T9–10		Paraparesis	Myelography	Surgery	Complete recovery
Kars et al. (1990) [10]	1	40	Male	C5–6	Casoni(–)	Tetraparesis	CT, USG	Posterior	
Bavbek et al. (1992) [2]	1	40	Male	T5–9		Normal	MRI	Posterior	Normal
Tekkök and Benli (1993) [16]	1	54	Male	L2–5		Cauda equina synd.	CT, MRI	Posterior	No change
Baysefer (1996) [4]	2	21	Male	T5–6		Paraparesis	Myelography	Posterior	Complete recovery
		22	Male	T6		Paraparesis	Myelography	Posterior	Improved
Pandey and Chaudhari (1997) [14]	1	15	Male	S1–2		Cauda equina synd.	MRI	Posterior	Improved
Bayar et al. (1997) [3]	1	30	Female	L5-S1		S1 radicular findings	MRI	Posterior	Improved
Berk et al. (1998) [5]	1	17	Male	T7–9		Paraparesis	CT, MRI	Posterior	
Bouklata et al. (2000) [7]	1	8	Male	T8–11		Paraparesis	MRI	Posterior	
Karadereler (2002) present paper	1	8	Male	L2–5	Elisa (–)	Cauda equina synd.	MRI	Posterior	Improved

12 cases of primary extradural hydatid cysts, including ours [2, 3, 4, 5, 7, 10, 14, 15, 16, 18].

In reports originating from a period prior to computed tomography (CT) and MRI, the possibility of the existence of a single extradural hydatid cyst or of soft tissue involvement without bony origin was viewed with scepticism [13, 19]. Following the widespread usage of CT and MRI, however, several singular cases of this kind are reported, as is evident in Table 1. The table shows that there is a male preponderance (11 male vs 1 female) and that the ages of the patients ranged from 7 to 56 years, with a mean age of 27 years. Neurological signs are essentially paraparesis and signs of cauda equina syndrome and surgery consisted essentially in laminectomy, and led to outcomes that were in general very favourable.

Hydatid cyst can be pre-diagnosed by means of the anamnesis if the patient originates from a region where the disease is endemic, by serological tests, or radiological examinations. It is known that the Casoni-Weinberg test is not very reliable. Specific ELISA/Western blot serology is 80–100% sensitive and 88–96% specific for liver cyst infection, but less sensitive for the involvement

of lung (50–56%) or other organs (25–56%) [11]. In the light of these figures, it is well understood why, in this present case, the serological tests were negative.

Radiological diagnosis and determination of extensions of the hydatid cyst are usually provided by MRI scans [12, 16, 17]. The cyst does not have a solid component and has a tendency to invade anatomical cavities. It does not demonstrate contrast enhancement, but has a CSF-like signal intensity on MR images. The final diagnosis is provided, as in this case, through surgical exploration.

Conclusion

Lumbar extradural hydatid cyst has only occasionally been reported in the literature. Treatment of hydatid cyst is primarily surgical, demanding total removal without rupture. Due to the stated clinical and neuroradiological features, even if the serological tests are negative, hydatid cysts should be considered in the differential diagnosis of cystic lesions of the spine.

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