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Intradural spinal hydatid cysts

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Abstract Spinal hydatid cysts are very rare and comprise only 1% of all bony involvement. Intradural hydatid cysts are extremely rare compared to other types of spinal hydatid cysts. We report the case of a 19-year-old man with lumbar intradural hydatid cysts. He complained of paraparesis and urinary hesitancy. Myelography revealed a block of the contrast medium at the L4 level and multiple round radiolucent lesions rostrally. At surgery, multiple hydatid intradural cysts were extirpated. The patient's neurologic deficits improved postoperatively,

but recurred 6 weeks later. Reoperation led to partial improvement of deficits. Primary intradural hydatid cysts are extremely rare. It is difficult to explain an isolated intradural location of multiple cysts. In our patient, the cysts were multiple and the patient's cerebral CT scan was normal; he had undergone no previous lumbar puncture. So we can hypothesize that intradural spinal cysts may be primarily multiple.

Key words Hydatid cyst · Intradural · Spinal cord compression

Introduction

Hydatid disease is an infestation of humans by the larva of dog tape worm *Echinococcus granulosus*. This disease is rare in the United States and western Europe, but prevalent in the Mediterranean, eastern Europe, Asia, Africa and South American countries.

Liver and lung are the most commonly involved sites in hydatid disease. Spinal hydatid cysts are rare and comprise only 1% of all reported bony involvement [11]. Intradural hydatid cysts are extremely rare compared to other types of spinal hydatid cysts. The first published case was reported by Carrea and Murphy [3]; the second was reported by Ley and Marti [9] and the third was reported by Sharma et al. [14]. In 1991 Medjek et al. [10] pointed out that only 14 cases (including their case) of intradural hydatid cysts had been reported in the literature. In this paper we present a case of intradural hydatid cysts involving the cauda equina.

Case report

A 19-year-old man was admitted to the department of neurology in August 1991 with low back pain radiating to both legs, and urinary hesitancy that had started 3 months previously. He complained of having suffered neurogenic claudication for the preceding 2 weeks. Physical examination revealed paraparesis most prominent distally, hypoaesthesia below the L1 segment and loss of cremasteric reflexes. X-ray examinations of the chest and the spine revealed no abnormalities. An iohexol myelography showed a total block of the contrast medium at the L4 level. Additionally, there were three round radiolucent lesions, 2–3 cm in diameter, one at the L3 and two at the L1 level (Fig. 1). These lesions were thought to be neurofibromatosis because of their multiplicity and round shape. CT-myelography showed intradurally located cystic lesions without any bony involvement, mimicking arachnoid cysts (Fig. 2). MRI could not be performed, because it was not available in our hospital at that time.

On August 15, 1991, the patient was operated with lumbar laminectomies at three levels (L2–4). The bony laminae were normal. The dura mater was distended and pulsation was absent. The dura was opened under the operation microscope, and multiple cysts measuring 5–30 mm in diameter were found floating among

Fig. 1 Myelography showing a total block of the contrast medium at the L4 level and round radiolucent lesions

Fig. 2 CT-myelography showing intradurally located cystic lesions without any bony involvement, mimicking arachnoid cysts

the lumbar roots. They were not attached to the dura mater or roots except for tiny arachnoid junctions in some. The cysts were removed; however, some very thin-walled cysts were ruptured during removal. We extended the laminectomies rostrally to the T12 level in order to remove small cysts at the L1 level. However, no cysts were found at these levels. We suspect that the cysts had changed their locations with the position of the patient.

During the postoperative period, the pain disappeared and the lower extremity weakness slightly improved. The histopathologic examinations confirmed the operative diagnosis of hydatid cysts. Abdominal ultrasonography focusing on intra-abdominal hydatid disease revealed no abnormalities. The patient received mebendazole 12 g/day postoperatively.

Six weeks after the operation the patient was readmitted to the hospital with worsening of the lower extremity weakness and urinary incontinence. Urgent myelography showed a distal block of contrast medium at the L3 level and multiple cysts at the rostral levels.

An emergency reoperation was performed and residual cysts were removed. The area was irrigated with hypertonic saline. The neurologic signs improved partially. During a 15-month follow-up period the patient's neurologic deficits did not change. He remained non-ambulatory with moderate flaccid paraparesis and neurogenic bladder.

Discussion

Infection of humans by *Echinococcus* is initiated by ingestion of the eggs of the adult worm situated in the intestinal tract of a dog that has become infected by eating parasite-infested animal viscera. Embryos liberated in the human duodenum penetrate its wall and pass into the liver or the circulatory system, which distributes them throughout the body [4]. Most spinal lesions are hydatid cysts primarily affecting the body of vertebrae as a result of portovertebral shunts [11, 13, 15]. Ley and Marti [9], to explain the intradural location of the cysts secondary to pulmonary location as seen in their patient, postulated that the parasite embryo circulating in the bloodstream enters through an intercostal artery.

Primary intradural hydatid cysts are uncommon [12]. Arana-Iniguez [2] commented that in virtually all cases of hydatid disease of the spine, compression of the spinal cord and intraspinal nerve roots is caused by hydatid material from an affected vertebra. Dew [6], in 1928, pointed out the theoretical possibility of primary epidural or intradural lesion. In 1948 Dévé [5] claimed that in the literature there was no well documented case without bony involvement. For the first case, reported by Carrea and Murphy [3], the authors noted that it was debatable whether the cyst originated in the arachnoid membrane or in the white matter near the surface of the spinal cord. Being usually located in the dorsal region, primary intradural cysts may lead to rapid spinal cord compression [1].

Dévé's hypothesis that when a cyst developed in such a small organ as the spinal cord, it would be single and small, occurring in young adults and producing its symptoms over a short period, was supported by the features shown in the two cases of Carrea and Murphy [3] and Ley and Marti [9]. The case reported by Sharma et al. [14] was similar to those cases, except for the multiplicity. According to Sharma, a previous lumbar puncture and pressure changes had presumably led to the rupture of a cyst and caused the multiplicity. Rayport et al. [13] implied that multiple intradural cysts may develop secondary to rupture and dissemination from an intracranial cyst.

Over a period of 16 years, we operated on 503 cases of spinal cord compression, of which 13 (2.6%) were caused by hydatid cysts. Only one of these cases involved intradural hydatid cyst (0.2% of spinal compression cases, 7.7% of all spinal hydatidosis).

The clinical findings of spinal hydatid cyst are no different from the findings of other spinal cord compression diseases. The best diagnostic tool seems to be MRI, because it can give the full image in the vertical axis of the spinal canal, and thus reveal the full spinal extent of the disease [8, 15]. Myelography carries a risk of cyst puncture, which can cause intradural spreading of the disease.

With all the treatment modalities this benign disease has a malignant course, because of its high rate of recur-

rence. Although satisfactory results have been reported with flubendazole and mebendazole treatment, haematogenous spread could not be prevented [7].

Total removal of the cysts without rupture should be the surgical goal in all cases. If cysts rupture during extirpation, the surgical area should be irrigated with hypertonic saline; however, this seems relatively ineffective, since if any cyst ruptures into the intradural space, recurrence is inevitable.

References

1. Akhan O, Dinçer A, Saatçi I, Gülekon N, Besim A (1991) Spinal intradural hydatid cyst in a child. *Br J Radiol* 64: 465–466
2. Arana-Iniguez R (1978) *Echinococcus*. In Vinken PJ, Bruyn GW (eds) *Handbook of clinical neurology*, vol 35. North Holland, Amsterdam, pp 175–208
3. Carrea R, Murphy G (1964) Primary hydatid cyst of the spinal cord. *Acta Neurol Lat Am* 10: 308–312
4. Charles RW, Govender S, Naidoo KS (1988) Echinococcal infection of the spine with neural involvement. *Spine* 13: 47–49
5. Dévé F (1981) L'échinococcose osseuse. In: Sharma A, Kashyap V, Abraham J, Kurian S (eds) *Intradural hydatid cysts of the spinal cord*. *Surg Neurol* 16: 235–237
6. Dew HR (1981) Hydatid disease. In: Sharma A, Kashyap V, Abraham J, Kurian S (eds) *Intradural hydatid cysts of the spinal cord*. *Surg Neurol* 16: 235–237
7. Göçer AI, Tuna M, Ildan F, Bağdatoğlu H, Hacıyakupoğlu S (1994) Cervical intradural extramedullary hydatid cyst. *Türk J Neurosurg* 4: 169–171
8. Kaoutzanis M, Anagnostopoulos D, Apostolou A (1989) Hydatid disease affecting the vertebrae. *Acta Neurochir (Wien)* 98: 60–65
9. Ley A Jr, Marti A (1970) Intramedullary hydatid cyst. *J Neurosurg* 33: 457–459
10. Medjek L, Zenini S, Hammoum S, Hartani M (1991) Hydatidose intradurale rachidienne dorsale. A propos d'un cas. *Ann Radiol* 34: 251–255
11. Özek MM (1994) Complications of central nervous system hydatid disease. *Pediatr Neurosurg* 20: 84–91
12. Pamir MN, Akalan N, Özgen T, Erbenli A (1984) Spinal hydatid cyst. *Surg Neurol* 21: 53–57
13. Rayport M, Wisoff HS, Zaiman H (1964) Vertebral echinococcosis. *J Neurosurg* 21: 647–659
14. Sharma A, Kashyap V, Abraham J, Kurian S (1981) Intradural hydatid cysts of the spinal cord. *Surg Neurol* 16: 235–237
15. Tekkök IH, Benli K (1993) Spinal extradural hydatid disease: report of a case with magnetic resonance characteristics. *Neurosurgery* 33: 320–323