

Recurrent vertebral hydatid disease: spectrum of MR imaging features

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ABSTRACT

Introduction: This study aimed to describe a spectrum of magnetic resonance (MR) imaging findings in a case series of four patients with recurrent vertebral hydatid disease (HD).

Methods: Four patients with recurrent spinal HD, who were studied with MR imaging at 1.5T or 0.5T MR units, were encountered during a ten-year period. All patients had a history of repeated spinal surgery for hydatid resection.

Results: HD involving the lumbar spine was found in two patients, the thoracolumbar spine in one patient and the lumbosacral in one patient. Skip lesions were seen in one patient. All patients had extensive involvement of the extradural space, soft tissues of the back and posterior vertebral elements. HD involving the vertebral body, intervertebral disk and iliopsoas muscles were noted in three, two and three patients, respectively. Bone and extradural hydatids were typically small, and appeared hypointense on T1-weighted images, with a mildly enhancing rim on post-contrast T1-weighted images. Sacral hydatid was an expansile multicystic process. Muscle hydatids were large, surrounded by a gadolinium-enhancing rim and assumed a variety of patterns – either multilocular or a nonspecific inhomogenous cystic or dumbbell configuration.

Conclusion: MR imaging is a valuable diagnostic tool for follow-up of patients with vertebral HD. Recurrent HD is characterised by extensive involvement of soft tissues of the back and extradural space. Extension into the intervertebral disk and iliopsoas muscles and skip lesions in the extradural space are not uncommon.

Keywords: hydatid cysts, magnetic resonance imaging, spine

INTRODUCTION

Hydatid disease (HD) is caused by the parasitic tapeworm *Taenia echinococcus*. 12 different species of this genus have been identified; however, only *Echinococcus (E.) granulosus* and *Echinococcus multilocularis* have been known to affect humans. *E. granulosus* is a cyclo-zoonotic parasite, with dogs being the definitive hosts, sheep, the intermediate hosts and humans, the incidental intermediate hosts.⁽¹⁻³⁾ The disease has the highest incidence in countries where sheep are raised with the help of dogs, notably the Middle East, Australia, New Zealand and South America.⁽¹⁾ Humans are infected by food or water contaminated by dog faeces that contain eggs of the parasite. The liver is the most common site of involvement, followed by the lungs. Bone HD is rare, involving 0.5%–2% of all cases, while the spine is the most common skeletal manifestation, accounting for 44%–50% of incidences of bone HD.^(1,4-7) Although spinal involvement is rare, it is the most serious manifestation of HD. HD is a relatively silent and slowly progressive disease with nonspecific clinical symptoms and radiologic findings. It is characterised by multiple recurrences that may result in paraplegia.⁽⁷⁻¹²⁾

Magnetic resonance (MR) imaging has revolutionised the management of infectious diseases of the spine due to its early detection and accurate delineation of the extent of disease.⁽¹³⁻¹⁵⁾ However, due to the rarity of spinal HD, few case series using MR imaging have been reported in the radiological literature.⁽¹⁶⁻²²⁾ Cases of recurrent HD have also been rarely described.⁽²³⁻²⁶⁾ In our experience, recurrent HD of the spine is an extensive process, predominantly affecting the extradural space, the paravertebral soft tissues and the soft tissues of the back at the site of previous laminectomies. Involvement of the intervertebral disk has been described in sporadic cases,⁽²⁶⁻²⁸⁾ mainly in those concerning recurrent disease.⁽²⁶⁾ Our study aimed at describing a spectrum of MR imaging features of recurrent vertebral HD in a case series of four patients that we encountered over a ten-year period.

METHODS

During the last ten years, we have encountered four patients (two male and two female) aged 35–69

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Table I. Location of hydatid cysts in the spinal column.

Patient no.	Age (yrs); gender	No. of previous spinal surgeries	Delay from initial diagnosis of SHD*/last operation	History of extraspinal location	Spinal location	Vertebral body	Posterior vertebral elements	Epidural space	Intervertebral disk	Iliopsoas muscles	Multifundis muscles
1	38;M	6	13 yrs/6 yrs	Hepatic hydatid, 16 yrs ago	L3	+	-	+	+	+	-
2	75;F	3	1 yr/13 mths	-	L5-sacrum	+	+	+	+	+	+
3	69;M	2	5 yrs/3 yrs	-	Th9-L2	-	+	+	-	-	+
4	51;F	1	8 yrs	Lung hydatid, 18 yrs ago	L2,L3	+	+	+	-	-	+

+: present; -: not present; SHD: spinal hydatid disease

(mean 58.2 ± 16.9) years, who were suffering from recurrent vertebral HD. All four patients had a previous history of surgery for spinal HD. Surgical procedures included posterior decompression surgery with laminectomy in all patients and additional anterior decompression with bone grafting of the vertebral body in one patient. The number of previous spinal operations for each patient was between one and six, and the time interval between the last surgery and the last MR imaging study ranged from 13 months to six years. Two of the patients had received additional medical treatment with albendazole. Another two patients had undergone additional surgery for hepatic and lung HD 16 and 18 years ago, respectively.

At the current admissions, clinical symptoms such as back pain and sphincter disturbances were reported by all patients; sciatica was reported by two, iliac fossa pain by two, and paraplegia was noted in two patients (Patient 1 and 2). No patient presented with systemic symptoms. MR imaging of the thoracic spine was performed in one patient, thoracic and lumbar in two, and lumbar and sacrum in one patient, depending on the neurological symptoms. MR imaging studies were performed on different MR imaging systems (1.5 T) for Patients 1 and 2 (Philips NT Intera, Philips Medical Systems, Best, The Netherlands); 1.5 T for Patient 3 (Vision Plus, Siemens, Erlangen, Germany) and 0.5 T for Patient 4 (MR-Max-Plus GF/CGR). A phase array coil was used for imaging Patients 1, 2 and 3. The imaging protocol included unenhanced T1 SE and T2 fast SE sagittal, axial and coronal images and contrast-enhanced T1 SE-weighted sagittal, axial and coronal images. Slice thickness was 3 mm, matrix was at least 220×256 and the field of view was 12–15 cm. All spinal MR imaging studies included whole spine T2 sagittal images for investigation of possible multifocal HD. Previous MR imaging and computed tomography (CT) studies were available in three of the patients. Axial CT images of the affected spinal areas were available in three patients,

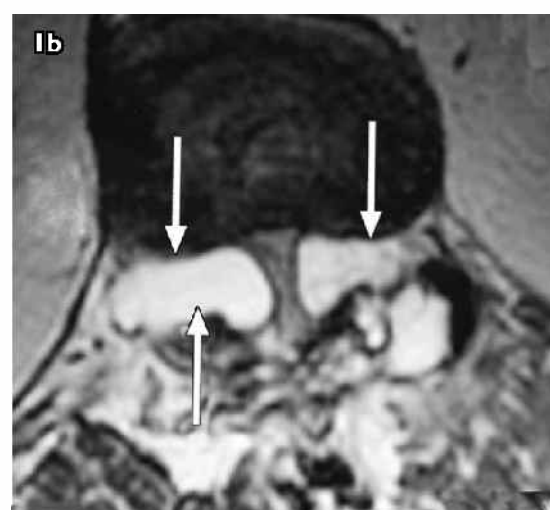


Fig. 1 (a) Sagittal T2-W fast SE image shows multiple hydatid cysts within the extradural space (arrows). (b) Axial T2-W fast SE image shows two of the cysts projecting through the intervertebral foraminae at Th12–L1 level (arrows) and compressing the dural sac.

and these were performed on incremental or spiral CT scanners using slice thickness or collimation of 3 mm, respectively, with coronal and sagittal reconstructions.

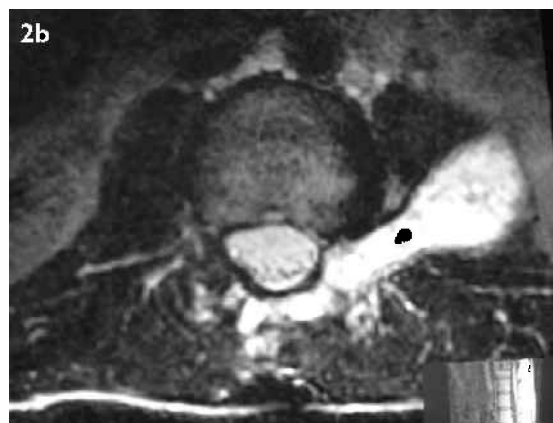
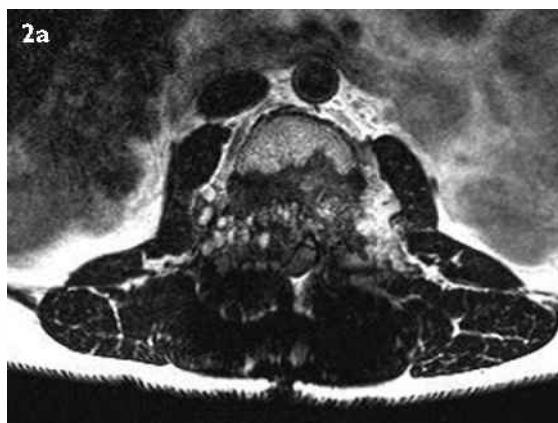


Fig. 2 (a) Axial T2-W fast SE image shows a cluster of hydatid cysts with hypointense rim within the vertebral body of L3 and the right pedicle. The cysts extend into the extradural space, the right intervertebral foramina and paravertebral space. There is metal artifact posteriorly. (b) (MR imaging performed ten years ago) Axial T2-W fast SE image at the same level as Fig. 2a shows a large, oval, unilocular hydatid in the extradural space. The hydatid cyst involves the adjacent lamina and also extends into the left psoas muscle through the left intervertebral foramen, assuming a dumbbell configuration. (c) Coronal T1-W SE image taken two years later shows extension of the hypointense hydatids into the L3-L4 intervertebral disk (arrow) and into the psoas muscles, mainly the left one.

RESULTS

Table I shows the individual patient data and the anatomic structures of the spine affected by HD, according to MR imaging findings. The distribution of vertebral HD included the lumbar, thoracolumbar or lumbosacral spine. Involvement of contiguous vertebrae was seen in two patients, whereas one patient exhibited skip lesions affecting the lumbar and thoracic spine (Fig. 1). All patients showed multiple hydatid cysts within the extradural space; three patients presented with involvement of the soft tissues of the back at the site of previous laminectomies and of the posterior vertebral elements, whereas the vertebral body was affected in three patients (Table I). The cysts encroached the extradural space, causing compression of the spinal cord or cauda equina.

Patient 1 exhibited involvement of the L3 vertebral body by a cluster of small hydatid cysts < 1 cm in diameter, which had remained unchanged from his previous MR imaging study performed one year ago; these cysts were outlined by a hypointense rim, which enhanced mildly after gadolinium administration (Figs. 2a & 2b). Involvement

of the sacrum and L5 vertebra was seen in Patient 2. Sacral hydatid was a multicystic expansile process that extended to the right sacroiliac joint (Fig. 3b). Hydatids located in the neural arch or in the vertebral body caused expansion and destruction of the osseous structures in all patients (Fig. 4). Intervertebral disk involvement was seen in one patient (Fig. 2c). In Patients 1 and 2, the cysts extended from the extradural space into the adjacent iliopsoas muscles through the intervertebral foraminae; both patients had a history of multiple recurrent HD. Hydatids in the muscles exhibited a variety of morphologic features, either multilocular cysts containing daughter cysts, daughter cysts that showed higher signal intensity than the parent cyst on T2-weighted fast SE images (Fig. 3a) or that were inhomogenous and predominantly hyperintense on T2-weighted fast SE images (Fig. 3c). The outer rim of all muscle hydatids exhibited mild enhancement by intravenously administered gadolinium. It is interesting to note that a previous CT image of the patient with sacral hydatid showed herniation of a hydatid from the iliopsoas muscle into the subcutaneous tissues of the back through the Petit's triangle (Fig. 3d).

DISCUSSION

At initial presentation, spinal HD usually starts from the vertebral bodies and subsequently extends into the posterior vertebral elements, extradural and paravertebral space.^(1,6,29) Primary intradural HD without

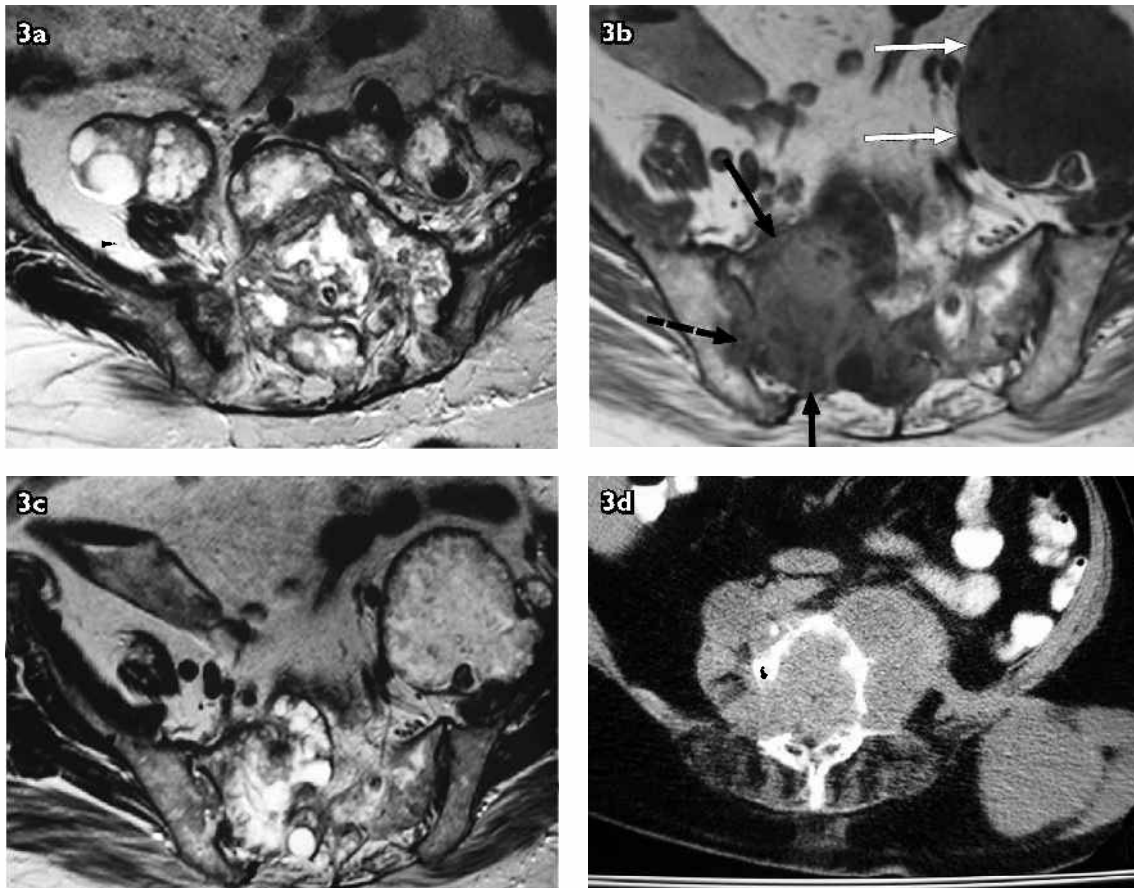


Fig. 3 (a) Axial T2-W fast SE image shows a destructive expansile multicystic process of the sacrum that encroaches the spinal canal and extends to the paravertebral soft tissues. A multilocular hydatid cyst with hypointense rim and daughter cysts is seen in the right psoas muscle. The daughter cysts exhibit higher signal intensity than the mother cyst. (b) Unenhanced axial T1-WV SE image shows a hypointense expansile lesion of the right sacral ala (black arrows) extending to the ipsilateral sacroiliac joint (broken arrow). There is a hypointense hydatid cyst (white arrows) in the left iliacus muscle. (c) Axial T2-W fast SE image shows the hydatid cyst in the left iliacus muscle appearing hyperintense and inhomogenous and surrounded by a hypointense rim. (d) Axial CT image at a higher level through L5, performed nine months before MR imaging, shows the hydatid disease as an osteolytic process that invades the body of L5 and extends to the left psoas muscle. A hydatid cyst herniates through the Petit's triangle into the subcutaneous fat of the lower back; this cyst was subsequently resected.

vertebral involvement is exceedingly rare.^(19,22) Sener et al have postulated that spinal involvement can be caused by the atypical passage of embryos to the inferior vena cava through portocaval anastomoses, and subsequently toward the retroperitoneum via the epidural venous plexus.⁽²⁴⁾ The development of hydatid cysts within the vertebrae presents some peculiar features compared to soft tissues; the pericyst, i.e. the response of the host tissue to the parasite, is missing and the typical bulky spherical hydatid cyst is not formed.⁽³⁾ Instead, the hydatid cysts proliferate by exogenous vesiculation, penetrate the bone trabeculae in the direction of least resistance⁽²³⁾ and incite a slow destruction and expansion of the vertebra.⁽²⁹⁾ When the cortex is breached, the cyst invades the extradural space, causing neurologic deficits, whereas the dura always remains intact. The intervertebral disks are usually not involved due to the low infective potential of the disease.^(4,13,26) As seen in almost all our patients, the disease affects mostly the

thoracic or lumbar spine,⁽⁷⁾ followed by the cervical spine, and rarely, the sacrum,^(20,21) as seen in one of our patients.

Treatment of spinal HD comprises removal of the cysts, combined with posterior, anterior or dual decompression surgery that may be supplemented by corpectomy with graft placement, depending on the severity and the anatomic site of involvement.⁽⁹⁻¹¹⁾ Additional posterior stabilisation may be required to avoid spinal instability.⁽¹⁾ Despite surgical removal, hydatid cysts usually recur, resulting in long-term morbidity and possibly, paraplegia. Adjuvant administration of medications such as mebendazole, benzimidazole or albendazole on a long-term basis can improve the prognosis,^(7,11) thus stabilising the disease for a long period of time.

MR imaging is the method of choice for the initial detection of spinal hydatid cysts, preoperative planning and postoperative follow-up.^(3,13-16) Vertebral



Fig. 4 (a) Unenhanced sagittal T1-W SE image shows a hypointense lesion that occupies the left L2-L3 intervertebral foramen (arrow). The lesion enlarges the foramen and scallops the posterior cortex of the vertebral body. There are two smaller cystic lesions within the soft tissues of the back at the level of L4 vertebra (arrowheads). (b) CT image through L3 shows a hypodense oval and multiloculated lesion with rim calcification. The lesion seems to occupy the whole of the spinal canal and erodes the posterior cortex of the vertebral body.

hydatid cysts exhibit typical features on MR imaging; they exhibit an invariably hypointense rim that may enhance mildly after gadolinium administration,^(17,23,24) appear in clusters that resemble a bunch of grapes, cause expansion and destruction of the bony elements involved and extend into the adjacent extradural space and paravertebral soft tissues. The main differential diagnosis is tuberculosis, which may present with multiple cystic lesions within the paravertebral and epidural space, within the vertebral body and rarely, within the disk.^(6,17,27) In contrast to tuberculous abscesses

that show strong peripheral enhancement, hydatids show only mild rim enhancement, whereas HD does not usually have a solid component and rarely involves the intervertebral disk. Complicated and unilocular hydatid cysts may also be confused with necrotic metastatic tumours.⁽³⁰⁾

In our case series, all the patients had recurrent HD. Cysts within the extradural space were larger compared to those in the bones of the patients. Extensive involvement of the posterior extradural space and the soft tissues of the back at the site of previous laminectomies was invariably present, whereas from the extradural space, hydatids extended to the paravertebral space through the intervertebral foraminae, occasionally assuming a dumbbell configuration.⁽¹⁰⁾ According to the literature, recurrent HD is usually extensive when discovered and may be more prone to disk involvement.⁽²⁶⁾ Hydatids in the intervertebral disk share similar MR imaging features with bone hydatids. Karray et al reported at least partial destruction of the disk in 12 of their 13 patients with spinal HD.⁽⁴⁾ In our opinion, disk involvement in recurrent HD may have been underestimated in the medical literature, as most reported cases of spinal HD had been studied without MR imaging, which could have otherwise documented disk disease. Multifocal HD in the spine, as seen in one of our patients, does not seem to be uncommon in recurrent HD.^(11,12,23) Extension of sacral HD into the sacroiliac joints, as seen in Patient 4, is considered extremely rare in both primary and recurrent type HD.^(20,29,30) Although all the patients in our series had severely compromised spinal canal on MR imaging, only two of them were paraplegic. In contrast to bone hydatids, muscle hydatids have features of hydatids in other soft tissues, i.e. they possess an outer pericyst that uptakes gadolinium, can be quite large, and follow the sheath of the iliopsoas muscles down to the iliac fossae.^(27,30) Muscle hydatids may either be multilocular and containing daughter cysts or inhomogenous with a nonspecific appearance that may simulate haematoma, abscess or necrotic soft tissue tumour.^(6,29,31) The former type has been attributed to active, and the latter, to inactive hydatid cysts.⁽³²⁾ Moreover, in our study, one of these cysts herniated through the Petit's triangle in the soft tissues of the back, an imaging finding that has not been previously reported. One drawback of our study is the lack of sequential MR or CT imaging studies; therefore, we were unable to document the changes in the MR imaging features after administration of pharmaceutical treatment.

In conclusion, extensive involvement of the soft tissues of the back and the posterior extradural space with

extension of the hydatids within the iliopsoas muscles is a common finding in recurrent spinal HD, whereas intervertebral disk involvement and skip lesions within the extradural space are seen occasionally.

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