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CASE REPORT

S. Singh
I.P. Korah
S.V. Gibikote
N.K. Shyam
A. Nair
A. Korula

Sacral hydatidosis: value of MRI in the diagnosis

S. Singh · I.P. Korah (✉) · S.V. Gibikote
N.K. Shyam
Department of Radiodiagnosis,
Christian Medical College and Hospital,
Vellore, Tamilnadu, India

A. Nair
Department of Surgery,
Christian Medical College and Hospital,
Vellore, Tamilnadu, India

A. Korula
Department of Pathology,
Christian Medical College and Hospital,
Vellore, Tamilnadu, India

Abstract We present a case of primary hydatid disease of the sacrum. The diagnosis was made on MR imaging obtained to evaluate the spine for recurrent disc disease. The patient had previously undergone laminectomy elsewhere for L4–5 radiculopathy.Ultrasound-guided aspiration and visualisation of scolices confirmed the diagnosis. No other site of involvement was found.

Key words Hydatid · Sacral · MRI

Introduction

Hydatid disease is an infestation most commonly caused by the larval stage of tapeworm *Echinococcus granulosus.* While dogs are the definitive hosts, sheep and cattle are the intermediate hosts. Man may replace the usual intermediate host by ingestion of contaminated food [1]. Although the parasite may affect any anatomic location, lung and liver are the most frequently involved organs [2]. Osseous infestation is said to occur in the more highly vascularized areas of the skeleton such as vertebrae, long bone epiphyses, ileum, skull and ribs [3, 4]. Solitary osseous hydatid cyst is very rare and almost always primary [5]. In the spinal column, the hydatid disease usually begins in the vertebral body, with a predilection for the thoracic and lumar spine [6, 7]. Sacral involvement in hydatid disease is rare. To the best of our knowledge, so far four cases of sacral involvement have been reported. In all except one, the diagnosis of hydatid disease was not made prior to surgery [7–10].

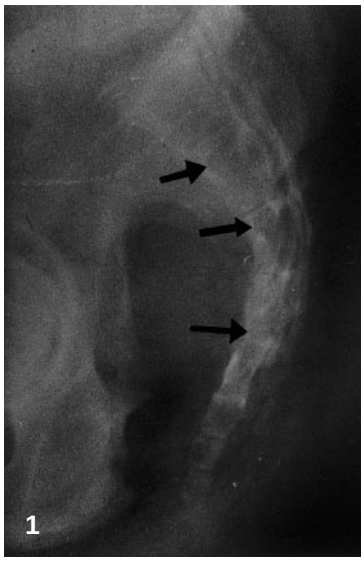
Case report

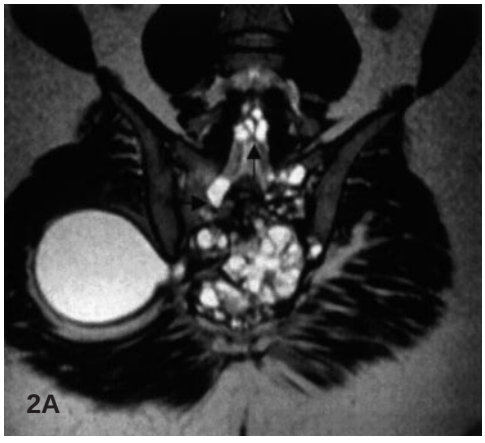
A 29-year-old man had lower limb radiculopathy with bowel and bladder disturbance, for which he had undergone laminectomy at the L4–5 level elsewhere $1^1/_2$ years prior to presenting at our hospital. He complained of increasing radiculopathy of both lower limbs of 1 month's duration. Weakness and sensory impairment of touch and pain of the lower limbs was noticed bilaterally, more on the right side.

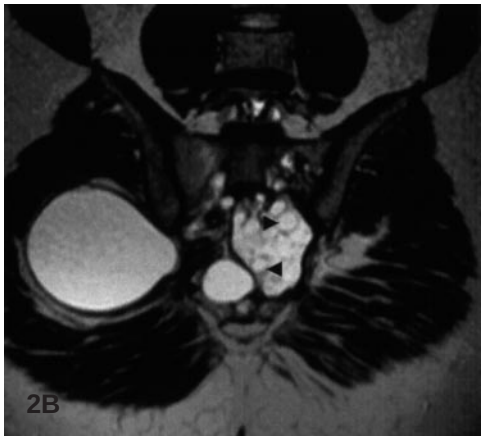
Examination showed a midline surgical scar in the lumbosacral region. There was right paraspinal muscle spasm. Swelling in the right gluteal region was noticed, which was firm in consistency. The right lower limb was oedematous. Anal and bulbocavernous reflexes were absent bilaterally. The remaining examination was unremarkable. Routine laboratory investigations and chest radiograph were normal. Provisional clinical diagnosis was recurrent prolapse of the L4–5 disc.

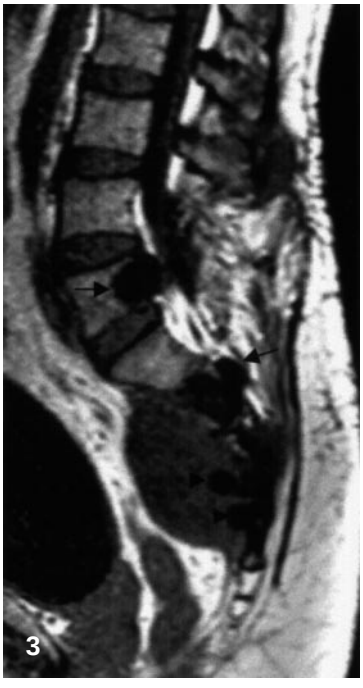
The radiographs of the lumbosacral spine showed multiple, ill-defined, expansive lytic lesions with mild marginal sclerosis in the sacrum (Fig. 1). Laminectomy defect at the L4 and L5 levels was seen, consistent with the postoperative status (not shown).

MR imaging revealed numerous cystic lesions of varying sizes in the bony sacrum, sacral spinal canal, sacral neural foraminae and adjacent presacral and gluteal soft tissues (Fig.









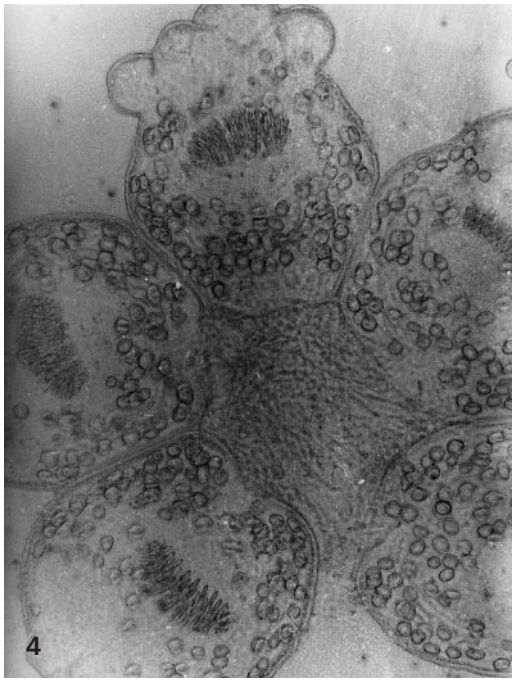


Fig. 1 Lateral radiograph of the lumbosacral spine, showing multiple, rounded, lytic lesions with marginal sclerosis in the sacrum (*arrows*)

Fig. 2A, B T2-weighted (TR/TE: 4000/120) coronal images. showing multiple, cystic lesions of varying sizes in the bony sacrum, neural foraminae and spinal canal (*arrows*) and a large cystic lesion in the right gluteal region. Daughter cysts are visualised (*arrowheads*). The capsule is seen as low signal intensity

Fig. 3 T1-weighted (TR/TE: 418/19) sagittal image, showing heterogeneous low signal intensity sacral lesion with hypointense daughter cysts (*arrowheads*) and cystic lesions in the sacral spinal canal and L5 vertebral body (*arrows*)

Fig. 4 Photomicrograph, shows solices of *Echinococcus granulosus*

2). In addition, a small cystic lesion in the L5 vertebral body and right L5 pedicle was also noted. The cysts varied in size from 5 mm to a right gluteal cyst that measured about 10×9×5 cm. The gluteal and presacral cysts showed multiple, smaller, rounded cysts within. These smaller cystic lesions were of slightly higher signal intensity on heavily T2-weighted images (Fig. 2), and hypointense on T1-weighted images compared to the large, parent cysts (Fig. 3). The capsule of the parent cyst was hypointense on both T1- and T2-weighted sequences. There was no evidence of pericystic oedema.

CT confirmed the multiple expansive osteolytic lesions seen on the radiographs and showed faint peripheral calcification of the cystic lesions in the soft tissues (not shown). Ultrasound-guided aspiration was then performed under steroid cover, and the right gluteal cyst was aspirated in its entirety without complications. Microscopic evaluation of the aspirated fluid showed multiple scolices of *Echinococcus granulosus* (Fig. 4).

The lesions were considered too extensive for surgery. The patient was started on albendazole for 3 weeks and advised to undergo physiotherapy for strengthening his lower limbs. He was under consideration for surgery should the lesions enlarge. The patient had persistent symptoms, and follow-up MR done

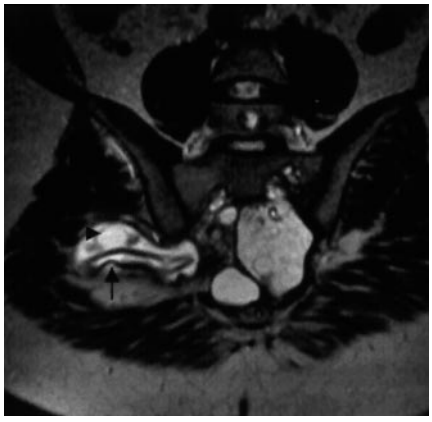


Fig. 5 T2-weighted (TR-TE: 4000/120) coronal image, after 1 month of albendazole therapy, shows previously aspirated, collapsed, right gluteal cyst with linear signal void areas representing the capsule with calcifications (*arrows*). The sacral cysts are unchanged in size

after 1 month showed the cysts had remained the same size, except for the collapsed previously aspirated right gluteal cyst (Fig. 5). A small daughter cyst was also visualised within the collapsed cyst (Fig. 5). Small hydatid cysts in the neural foraminae and spinal canal (Fig. 2B) were considered to be the cause of the patient's symptoms. The patient was put on long-term albendazole treatment.

Discussion

Primary osseous infestation of *Echinococcus granulosus* is rare, occurring in only 0.5–4% of cases [3–6]. In osseous hydatidosis, vertebral involvement accounts for 44% of cases, with a predilection for lower thoracic and lumbar vertebrae [6]. This is attributed to the presence of portovertebral shunts [5, 6]. Five patterns of cystic spinal involvement have been described: primary intramedullary, intradural intraspinal, extradural intraspinal, vertebral and paravertebral [1, 5]. Within bone, the parasite is almost always multilocular due to lack of defensive reaction. The parasite grows slowly in the direction of least resistance, particularly along the intertrabecular spaces, eroding the bone like a benign tumour [1, 6]. As the larvae enlarge, there are dilatations of the bony spaces of spongiosa and resorption of cancellous bone [1]. Enlargement and spread is achieved partly by local pressure erosion of bone, while pressure on the blood vessels within the bone may cause local bone necrosis [1]. Erosion through the bone and periosteum into the adjacent soft tissue proceeds without stimulating subperiosteal new bone formation, which may lead to spontaneous fracture [1, 6].

On radiographs, hydatid disease is initially seen as a small osteolytic lesion with ill-defined margins and no periosteal reaction [11]. As the disease progresses, the lesion takes the form of multiple sharply demarcated, expanding, osteolytic lesions with thin curvilinear septae, giving a soap-bubble or honeycomb appearance [6, 11]. Intervertebral discs, capsules and ligaments are said to be resistant to hydatidosis, and in most cases, as in our case, the disc is preserved [1, 6, 11]. Osteoporosis, bone thickening with coarse trabeculae, spread through the subperiosteal and subligamentous path, soft tissue swelling, and the presence of intralesional calcifications are not features usually associated with vertebral hydatidosis [3, 4, 11].

Prior to MR imaging, CT had a major role in evaluation of osseous involvement. Calcified or uncalcified cysts, separate from bony lesions, which are hardly recognised on conventional radiological examination, are identified, as in our case (not shown).

MR imaging is the most comprehensive of all imaging examinations to evaluate hydatid disease in the spine; and is especially valuable in showing its relationship to neural structures, spinal cord, and extension into soft tissue [3, 12]. Multiple cysts (daughter cysts) within a cyst, which are of varying signal intensities, is the characteristic imaging feature of this disease. The presence of a capsule is helpful in differentiating unilocular hydatid cyst from simple cyst [13]. The capsule is seen as a low-intensity rim, which shows mild enhancement following intravenous gadolinium. The enhancement reflects the vascularity of the pericyst [13]. On T1-weighted images, the daughter cysts are more hypointense than the parent cyst, and on T2-weighted images the daughter cysts are of slightly higher signal intensity than the parent cyst [14], as in our case. The signal intensity may changes with co-existing infection, calcifications, or haemorrhage. Perilesional oedema is not a feature.

Although hydatid disease is not uncommon in our region, it was not seriously considered in the differential diagnosis, because of the relative rarity of osseous and especially sacral hydatid. The patient was referred for MR imaging of the lumbosacral spine to exclude recurrent prolapse of the L4–5 disc. The MR images revealed unsuspected but characteristic findings of hydatid disease. The identification of daughter cysts with characteristic signal intensity permitted a confident diagnosis.

Steroid cover was instituted before ultrasound-guided aspiration, to avoid the remote possibility of anaphylaxis – a complication some authors believe is very infrequent [15]. Tumour and tumour-like lesions of the sacrum have a long differential diagnosis based on morphologic appearances, location of the lesion, and age of the patient. In our case, visualisation of the daughter cysts limited the diagnosis to hydatidosis, which was confirmed by visualisation of scolices of *Echinococcus granulosus* in the aspirated cyst content.

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