A sacral hydatid cyst mimicking an anterior sacral meningocele

Case report

Mustapha Hemama, M.D., Ali Lasseini, M.D., Loubna Rifi, M.D., Mahjouba Boutarbourch, M.D., Said Derraz, M.D., Abdessamad El Ouahabi, M.D., and Abdesselam El Khamlichi, M.D.

Service de Neurochirurgie, Hôpital des Spécialités O.N.O., Rabat, Morocco

Hydatid disease is a zoonosis caused by Echinococcus granulosus. It is a progressive disease with serious morbidity risks. Sacral hydatid disease is very uncommon, accounting for < 11% of spinal hydatidosis cases. The diagnosis of a sacral hydatid cyst is sometimes difficult because hydatidosis can simulate other cystic pathologies. The authors report on a 9-year-old boy admitted to their service with a paraparesis that allowed walking without aid. The boy presented with a 2-year history of an evolving incomplete cauda equina syndrome as well as a soft cystic mass in the abdomen extending from the pelvis. Radiological examination revealed an anterior meningocele. A posterior approach with laminectomy from L-5 to S-3 was performed. Three lesions with classic features of a hydatid cyst were observed and removed. The diagnosis of hydatid cyst was confirmed histopathologically. Anthelmintic treatment with albendazole (15 mg/kg/day) was included in the postoperative treatment. The patient’s condition improved after surgery, and he recovered normal mobility.

The unusual site and presentation of hydatid disease in this patient clearly supports the consideration of spinal hydatid disease in the differential diagnosis for any mass in the body, especially in endemic areas.

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History and Examination. A 9-year-old boy was referred to our neurosurgery outpatient clinic by a pediatric surgeon who had been consulted for chronic urinary retention that appeared 3 months prior to presentation. On admission, the patient presented with a 2-year history of progressive lumbago, bilateral sciatica, and paraparesis. Subsequently, a left-sided dropped foot developed, but the boy retained his ability to walk without assistance.

Clinical examination revealed a conscious patient with a Glasgow Coma Scale score of 15. He was afebrile, not pale, anicteric, and acyanotic with no significant lymph node enlargement. Chest and cardiovascular systems were normal. The patient had a soft, cystic nontender mass in the abdomen extending from the pelvis. The kidneys were not ballotable, and there was no renal angle tenderness. The patient had a self-retaining catheter with normal male external genitalia, normal anal sphincter...
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ter tone, and a soft fluctuant mass between the sacrum and rectum. No constipation or saddle paresthesia was observed.

Neurological examination showed a weakness (4/5) of the lower extremities, bilateral hypotonia, hyporeflexia, and mild to moderate flexion contracture of the left ankle. The Babinski response was equivocal. A diagnosis of cauda equina syndrome due to an anterior sacral meningocele was made.

Abdominopelvic ultrasonography scans revealed a presacral 87.5 × 77.3–mm cystic lesion with an intraspinal dumbbell-shaped component resembling an anterior meningocele. On retrograde urethrocystography, the bladder appeared compressed by a huge presacral, retrorectal pelvic mass. The bladder and the rectum were displaced upward.

Magnetic resonance imaging of the pelvis showed a 73 × 84 × 107–mm presacral ovoid and finely septated cystic lesion hypointense on T1-weighted and hyperintense on T2-weighted MR imaging. This mass had widened the S-1 and S-2 foramina and extended into the spinal canal. The intraspinal portion was extending for about 54.6 mm from the L-5/S-1 junction to the S-3 vertebra, compressing the thecal sac (Fig. 1). These findings pointed to an anterior meningocele with intraspinal extension. The patient had a normal CT brain scan, while electromyography recordings of the lower extremities showed bilateral multiple lumbosacral radiculopathies from L-5 to S-2.

Operation. The patient underwent surgery via a posterior approach. Upon an L5–S3 laminectomy, the dural sac was intact but laminated posteriorly by 3 discrete whitish cystic lesions with classic features of hydatid cysts (Fig. 2). Therefore, the surgical field and surrounding regions were irrigated with a chemical sterilizing agent (hypertonic saline 3.5%) to prevent further recurrence. Intraoperative cyst rupture occurred given the difficult localization of the 3 lesions (Fig. 3); however, no anaphylactic reaction developed.

The lesions were removed, revealing a 3 × 2–cm bony defect in the left lateral mass of the sacrum (Fig. 4). This defect was widened to allow extraction of the intrabdominal portion and was subsequently closed with a fascia lata graft to prevent an eventual herniation of pelvic viscera into the spinal canal. Histopathological verification confirmed the diagnosis of hydatid cyst.

Postoperative Course. Albendazole was given after surgery at a dose of 15 mg/kg/day for 6 months. Postoperative recovery was very good. The urethral catheter was removed the 4th day after surgery, and the patient became ambulant by the 8th day with near-normal recovery of his limb deficit on discharge. A lumbosacral CT scan with 3D reconstruction showed good spinal anatomy.

Discussion

Spinal involvement occurs in about 50% of the cases of bone hydatid disease. The lesions are most commonly located at the thoracic (52%) and lumbar (37%) levels. More rarely, hydatid cysts are encountered at the cervical and sacral regions (11%).11

Symptoms of spinal hydatid disease are nonpathognomonic and are usually related to compression of the spinal cord.6 Back pain has been observed in 85%, radicular pain in 25%–95%, and paraparesis in 25%–77% of patients.12 Moreover, cases of urine retention,7 sciatica8 and paraplegia2,9,13 have been reported. The disease usually...
spreads from other organs, but primary vertebral hydatid disease without any other systematic involvement can occur with direct paravertebral venous shunts.5

Preoperative diagnosis of a spinal hydatid cyst is difficult. No distinguishing radiological findings exist; simple radiographic findings are nonspecific and may show bone destruction and a soft-tissue mass. Magnetic resonance imaging is the preferred modality in the diagnosis of hydatid cysts since these lesions have a fairly consistent appearance on MR imaging: they appear sausage-like in shape with 2 dome-shaped ends and no debris in the lumen. Occasionally, the cysts appear spherical, have thin walls, and are regular with no septation. On T1-weighted MR images, the cyst wall may be isointense or reveal slightly lower signal than its contents. The T2-weighted MR images show a low-intensity rim surrounding the homogeneous hyperintense cyst contents.17,18 The presence of a markedly hypointense cyst wall on T1- and T2-weighted MR images is characteristic of hydatid disease.12 Differentials, such as arachnoid cyst, cystic meningioma, tuberculosis, and cysticercosis, have been reported.1–3,8,15

Anterior sacral meningocele is a congenital cystic structure containing CSF, which results from a hernia through a defect on the anterior surface of the sacral bone. The diagnosis of anterior sacral meningocele can be suspected on the basis of physical findings, that is, a soft, fluctuant presacral mass over which the rectal mucosa moves freely and which usually becomes tense with coughing or straining. Most symptoms appear because of pressure on surrounding structures: rectum, bladder, female genitals, and sacral nerve roots. Symptoms include constipation, irregular menstruation, frequent urination, infections, nonspecific pain in the lower parts of the back sometimes radiating into the legs, hypalgesia in the perineum zone, paresthesia, reduced tonus of the anal sphincter, and reduced tonus of the detrusor.10

In our case, the patient presented with an incomplete cauda equina syndrome, which is a common presentation of both sacral hydatidosis and anterior meningocele. Furthermore, MR imaging favored a diagnosis of anterior meningocele, probably based on the lack of obvious bony destructions and the unilocular pattern. More important was the fact that the intraspinal lesion was continuous with the pelvic portion through the sacral foramina and had a signal intensity similar to CSF. Abdominal ultrasonography scanning also supported that view. The rare location and the negative history of contact may also explain the difficulties in diagnosis, even if the young age of the patient was an important epidemiological factor.

Although the standard treatment is total surgical removal of the cysts without inducing any spillage, rupture occurs frequently in spinal hydatidosis because the cysts are contained in narrow spaces within bone boundaries. No specific surgical technique has been found to avoid this problem completely, although the operating microscope can be helpful.14,16 Irrigation with scolicidal agents, such as hypertonicsolution, can prevent recurrence by osmotic destruction of the parasites. However, a few cases have indicated that sterilizing hydatid cysts using hypertonic solution carries a risk of resorption with acute hypertonics and massive tissue necrosis.14 Furthermore, adjuvant chemotherapy with albendazole postoperatively is generally used to control the disease locally, avoid systemic spread, and prevent recurrence.16

Conclusions

Although primary spinal hydatid cysts are uncommon, the incidence of these lesions may be higher if we consider those that could not be verified as hydatid cysts but simulated other cystic lesions radiologically and thus were managed conservatively, especially in endemic areas. Hydatid disease should be kept in mind for some bone cystic lesions when planning a surgical approach for cystic spinal lesions diagnosed as anterior sacral meningocele based on preoperative findings.

Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

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Address correspondence to: Mustapha Hemama, M.D., Hôpital des Spécialités O.N.O., Rabat, Morocco. email: mhemama@yahoo.com.