

A CASE OF SACRAL HYDATID CYST AND REVIEW OF LITERATURE

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ABSTRACT

Hydatid cyst of bone constitutes only 0.5-2% of all hydatidoses. The thoracic spine is the most common site of spinal hydatidoses. Primary hydatid cyst of the sacral spinal canal is rare. A 19-year-old boy had cauda equina syndrome with pelvic pain 15 days ago, the pelvic radiography showed a lytic image depending on the left sacral wing. MRI showed an intra-pelvic cystic image invading the sacrum T1 hypointense and T2W hyperintense. The Hydatid serology was positive. Surgical treatment consisted of a wide drainage of hydatid cavity dug in the left sacral wing, and by which it communicated intra pelvic, with removal of the entire cyst by gentle aspiration, abundant rinsing with hypertonic saline, release of sacred roots encompassed in a puddle of fibrosis hydatid. The evolution was good with recovery of perineal sensation and anal tone. The sacroiliac joint was considered stable and did not require synthesis or reconstruction.

KEYWORDS: Hydatid cyst – Sacrum.

INTRODUCTION

Hydatid cysts is caused by *Echinococcus granulosus*, its commonest site is liver and lungs. Skeletal hydatidosis occurs in 0.5 to 2% cases, half of which infest the spine.^[1-3] The commonest site is thoracic and the sacral involvement is rare.^[2] We report a case of sacral hydatid cyst.

CASE REPORT

A boy aged 19 years presented with cauda equine syndrome with pelvic pain 15 days ago, the pelvic radiography showed a lytic image depending on the left sacral wing.



Figure 1: A lytic image depends on the left sacral wing.

We complete by an MRI which showed intra-pelvic cystic picture invading the sacrum hypointense T1 and T2 hyperintense.

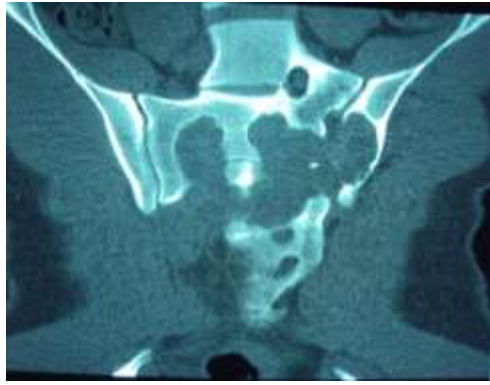


Figure 2: CT lytic image on the left sacral wing.



MRI: intra-pelvic cystic image invading the sacrum T1 hypointense and T2W hyperintense

Hydatid cyst of bone was suspected and we completed by an hydatid serology which was positive.

Surgical treatment consisted of a wide drainage of hydatid cavity dug in the left sacral wing, and by which it communicated intra pelvic, with removal of the entire cyst by gentle aspiration, abundant rinsing with hypertonic saline, release of sacred roots encompassed in a puddle of fibrosis hydatid.



Figure 5: A wide drainage of hydatid cavity dug in the left sacral wing.



Figure 6: Removal of the entire cyst by gentle aspiration, abundant rinsing with hypertonic saline.



Figure 7: Release sacral roots encompassed in a puddle of fibrosis hydatid.

The evolution was good with recovery of perineal sensation and anal tone. The sacroiliac joint was considered stable and did not require synthesis or reconstruction.

DISCUSSION

Epidemiology

Hydatid cysts predominantly occur in liver and lungs. Involvement of other organs is uncommon. Skeletal involvement occurs in 0.5 to 2% of all cases, half of which are in the spine.^[1-4] A neurosurgeon encounters hydatid cyst in brain and spine. Hydatid cyst of brain is more common than spine. In a series of 29 cases of intracranial and spinal hydatid cyst, no sacral lesion was seen.^[5] In a series of 25 cases of vertebral hydatidosis the cyst was located in the cervical vertebrae in three, the thoracic vertebrae in 11, the lumbar in five, and the sacrum in six cases.^[6] In another series of 13 cases of vertebral hydatidosis, sacrum was involved in only one case.^[7] Approximately 90% of spinal hydatidosis cases are located extradurally, most commonly in the vertebral body. Intradural location is very rare and appears like an arachnoid cyst.^[8]

Clinical features

As there is no host reaction the cyst can grow to enormous size and remain asymptomatic. The initial symptom is back pain when cyst invades spinal canal after erosion of bony cortex. As whole vertebral body is

destroyed, gibbus deformity does not result. The neurological symptoms are those of extradural tumor or disc disease.^[1]

Hydatid disease of sacrum is not easily detected. In two cases report, sacral hydatid cyst was not suspected and the symptoms were considered due to lumbar disc disease. In the first case, The diagnosis was delayed up to 12 years as pelvis was not imagined, which could have shown a cystic lesion in presacral space.^[9] In the second case, a small cystic lesion was presumed as tarlov's cyst.

The other differential diagnosis of cystic lesion of sacrum includes developmental cysts (epidermoid, dermoid, teratoma, neurenteric and retrorectal cystic hamartoma) anterior sacral meningocele, necrotic sacral chordoma, schwannoma, arachnoid cyst, and aneurysmal bone cyst.^[6]

Imaging

Though no specific imaging features point to hydatid cyst, the presence of the following appearances should consider hydatid cyst at top of suspicion, particularly in an endemic country.^[6]

- Lytic lesion with dense calcified rim
- No disc space involvement
- Multiloculated cyst with signal of parent similar to muscle on T1W
- Signal of daughter cyst similar to water

- Daughter cyst in overflowing or adjacent to parent cyst
- Both parent and daughter cyst with higher signal on T2W with rose or wheel shape formation
- Decrease in hyper intensity and increase in hypo intensity with time as cyst succumbs.

Treatment

Neither surgery nor medical therapy is generally effective for bone, especially spinal hydatidosis. The initial treatment of choice is surgical excision for neural decompression and establishing diagnosis. The operative finding of a large cyst with multiple chitinous small cysts is virtually diagnostic. After excision local scolicidal agents like hypertonic saline or certimide should be used for the site irrigation for formalin. Inadvertent opening of dura can lead to intradural recurrence and spillage local scolicidal agent; in one case, death occurred due to toxic myelitis and respiratory arrest from intradural spillage of formalin.^[7] Patient may require stabilization after extensive resection. Subcutaneous drainage for prolonged period after surgery is advised due to persistent discharge.

Albendazole is the drug of choice against this disease, when suspected, presurgical use of albendazole in echinococcus infestations reduces risk of recurrence and/or facilitates surgery by reducing intracystic pressure. Treatment with albendazole in E.granulosis infection can result in an apparent cure in as many as 30% of patients, a short term observation showed a 40-50% of patients objective evidence of response. Patients who do not show obvious initial evidence of response may be found to be cured when observed during several years. Though it is a benign disease, due to frequent recurrences and dissemination, the behavior is of malignancy. The overall recurrence rate is 30 to 40%. When recurrence occurs, repeating the excision should be attempted if feasible.

CONCLUSION

Preoperative diagnosis of hydatid disease is essential to minimise the complications of this disease. Even in a rare localisation such as the sacrococcygeal area, hydatid disease must be borne in mind especially in endemic areas or in patients coming from endemic areas not to diagnose this disease at an advanced stage.

A missed diagnosis of hydatid cyst could be devastating. Hence, hydatid cyst should be kept as a differential diagnosis, when encountered with a cystic lesion of sacrum.

In addition, long-term follow-up is mandatory as recurrence is high despite use of scolicidal agents.

CONSENT

The patients have given their informed consent for the case to be published.

Competing Interests

The authors declare no competing interest.

Authors 'Contributions

All authors have read and agreed to the final version of this manuscript and have equally contributed to its content and to the management of the manuscript.

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