Case Report

Paraplegia due to a large solitary hydatid cyst of the lumbar spine: a case report and review of the literature

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Summary

We are reporting a rare cause of paraplegia in a 40 years old male. Hydatid disease of the spine is found to be the cause of cord compression in this patient. Patient presented with a gradual onset of LL weakness over a period of 2 years. Magnetic resonance imaging (MRI) of the spine raised a suspicion of cystic lesion in the lumbar region and the diagnosis only confirmed after a tissue biopsy. This patient treated with Albendazole and Praziquantel combined with extensive surgery to excise the cyst, decompress the canal and fix the spine. The management was very late and the lesion was quite extensive.

Keywords: Hydatid cyst, paraplegia, lumbar spine

Introduction

Although paraplegia secondary to spine infection is very common in clinical practice, hydatid cyst of spine is extremely rare(1,2,3). The disease is endemic in rural areas where patients are in close contact with dogs. Only few cases were reported in the literature and most of them were in the thoracic area(4,5,6). We are reporting this case because hydatid cyst of the spine is very rare especially in the lumbar region(7,8,9,10). Moreover, it’s a primary lesion without any evidence of previous hepatic or pulmonary affection. Diagnosis is usually late unless a high index of suspicion by the treating doctors is available. Treatment is
mainly surgical to excise the lesion and to decompress the spinal canal.

Case report
A 40 years old male presented to our department with inability to walk for the last 2 years. He was a farmer for the last 20 years. His condition started gradually with low back pain and bilateral lower limb heaviness more in the right side. Gradually he could not walk and function of both sphincters was lost. There was no history of any systemic illness. Physical examination revealed complete paraplegia, lax anal tone and sensation loss up to level of L1. Routine blood count was normal apart from mild eosinophilia. X-ray and MRI scan showed a destructive cystic lesion involving L2 vertebra and extending anteriorly and posteriorly involving the spinal canal from T12 to L3 (Fig 1).

Fig 1: T2 sagittal view of lumbosacral spine showing a lobulated cystic lesion involving the second lumbar vertebra and extending anteriorly and posteriorly to involve the spinal canal.

The swelling was hypointense in T1 and hyperintense in T2. The lesion appeared to be lobulated and a provisional diagnosis of a lymphoma was made. A percutaneous image guided biopsy was taken under local anaesthesia. The pathology showed a laminated hyaline membrane with some degenerate scolices (Fig 2 and 3). A diagnosis of hydatid disease was then made based on the histological result. Culture and sensitivity was done to exclude other co-infection and it was negative. Surgical management was decided and posterior decompression and transpedicular fixation were carried out. The patient was treated with albendazole (400 mg/twice a day) and praziquantel (40 mg/kg per day) twice a week for 4 weeks. This regimen was repeated for three courses with a 2-week interval between each one. Postoperatively, pain decreased significantly when evaluated using visual analogue scale from 9 to only 2; the patient could sit comfortably, but there was no neurological improvement.

Fig 2: Laminated membrane with two degenerate scolices at the top of the figure. The black granular material in the membrane is due to early calcification (H%Ex100).
Fig 3: On the left is the laminated membrane of the hydatid cyst. On the right is part of the bone (H&Ex40).

Discussion
Echinococcosis or hydatid disease is a zoonosis caused by the larvae of a cestode of the genus echinococcus. Most commonly the infection in humans is due to E. granulosus and rarely caused by E. multilocularis. So far the only form of the disease reported in Sudan is due to E. granulosus. The disease is prevalent in camels, sheep and goats and the definitive host is the dog. All strains typed by polymerase chain reaction (PCR) in both man and animals are the camel strain in Sudan. Humans are infected through ingestion of food contaminated with feces of infected dogs. The life cycle of the parasite starts by ingestion of metacestode by dogs where the parasite matures into an adult parasite. Dogs pass the eggs in their feces. Humans are intermediate hosts and get infected when they ingest food contaminated with eggs. They develop cysts in different organs. Primary hydatidosis is common in the liver, spleen, and lungs. Musculoskeletal involvement is secondary and uncommon, with an incidence of less than 2.5%. It affects the pelvis and sacrum, metaphyses of the long bones, skull, spine, and ribs in decreasing order of incidence. Spinal involvement is rare, with an incidence of less than 1%.

Primary spinal hydatid cyst is even rarer and only few cases were reported in the literature. The disease is endemic in rural areas were patients are in close contact with dogs. Spinal hydatid cyst usually affects the dorsal spine. It usually presents as cord compression with neurological deficit, but the presentation can vary from mild discomfort to complete paraplegia. Diagnosis is mainly through an MRI scan which shows the characteristic hypointense cyst wall on T1 weighted image and homogenous hyperintense cyst contents in T2 image. Eosinophilia in paraplegic patients should also raise the suspicion of hydatid disease and schistosomiasis of the spine especially in endemic areas, but definitive diagnosis can only be reached by histopathological examination of the lesion, or positive serological test which is less specific and sensitive in spine when compared to abdominal hydatid disease. Treatment is surgical excision of the cyst to decompress the cord in combination with antihelminthic drugs. Usually, posterior spinal decompression through laminectomy and debridement of paravertebral lesions is the initial surgery, but complete clearance is difficult because of invasive diffuse spread within the bone and canal. Often, spillage of fluid caused by cyst rupture leads to subsequent recurrence.

Although complete excision is difficult particularly in the spine and the recurrence rate is high, retention of spinal stability is a major long term concern. Effectiveness of these agents, both in terms of the dose and duration of treatment and role of combination therapy is controversial. Albendazole has better intestinal absorption, a longer half life of 8.5 hours and a higher cyst concentration than the other drugs. Praziquantel combined with albendazole increased its concentration.
The three most widely used drugs are albendazole, mebendazole and praziquantel. In conclusion, hydatid disease of the spine, although very rare, should be considered in the differential diagnosis of cord compression especially in areas endemic for the parasite. This may facilitate early diagnosis and treatment and prevent permanent neurological damage. Although complete excision is recommended it can rarely be achieved particularly if the disease is extensive.

References
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Hydatid cyst Yasir N Gashi