A viable residual spinal hydatid cyst cured with albendazole

Case report

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Hydatid disease is an uncommon zoonosis in sheep-breeding countries.6,9,10 During its evolution the cestode, Echinococcus granulosus, forms a lesion, hydatid cyst, which is predominantly found in the liver, but can also be located in various sites of the human body. Primary hydatid disease of the bone is quite rare and occurs in 0.5 to 4% of patients.1,6,9,11 Vertebral involvement is found in 44% of these cases.14 Spinal hydatid cysts enlarge 1 to 5 cm yearly and frequently cause neural compression.1,9,10,17

Although the treatment of choice is surgical removal of the cyst without inducing any spillage, it may not be possible to perform this in patients with multiple and fragile cysts. In such cases, the neural structures should be adequately decompressed and albendazole should be administered promptly.

The authors describe the case of a 13-year-old girl who was admitted with a history of back pain and acute-onset lower-extremity weakness. Magnetic resonance imaging scans demonstrated severe spinal cord compression caused by multiple cysts involving T-4 and the mediastinum. The patient underwent surgery, and the cysts were removed, except for one cyst that was hardly exposed. Following histopathological confirmation of spinal hydatid disease, she was treated with albendazole for 1 year. One year postoperatively, the residual cyst had gradually shrunk and had almost disappeared.

Although a single case is not sufficiently promising, we believe that administration of albendazole is efficient to prevent recurrences in cases in which it is not possible to obtain total removal of the cysts without inducing spillage.

Key Words • hydatid disease • hydatid cyst • spinal cord compression • albendazole

Case Report

Presentation. This 13-year-old girl was admitted with a history of progressive back pain and lower-extremity weakness. She had a 2-week history of back pain and had developed bilateral lower-extremity weakness 2 days previously. She was unable to walk without assistance.

Examination. Her initial neurological examination revealed bilateral lower-extremity weakness, bilateral Babinski signs, and impaired sensation below T-4. Magnetic resonance imaging demonstrated multiple cysts involving the vertebral body and the pedicle of T-4, extending in to the spinal canal and mediastinum and compressing the spinal cord (Fig. 1). A diagnosis of hydatid disease was considered, albendazole was administered orally, and the patient underwent surgery.

Operation. The cysts were exposed by a right T-4 hemilaminectomy and T4-5 partial facetectomy, dissected from the spinal cord, nerve roots and surrounding bone, and removed totally. One residual cyst, located deep in front of the spinal cord and extending into mediastinum, was left in place. Although it did not make contact with other cysts, it had connections with the adjacent aortic wall. Because of the operative approach used, it did not seem possible to remove this particular cyst without inducing spillage. Furthermore, because satisfactory neural decompression had been achieved, this residual cyst was not resected (Fig. 2). The area was irrigated with hydrogen peroxide. The diagnosis of spinal hydatid disease was established by histopathological examination of the cyst’s contents.

Postoperative Course. The postoperative course was uneventful. On the 3rd postoperative day the patient was able to walk without assistance and was discharged on the
11th postoperative day. Albendazole (200 mg twice daily) was administered orally for 1 year. By 3 months after the surgery, the patient had made a full clinical recovery. She underwent follow-up MR imaging every 3 months for 1 year. The residual cyst progressively shrank and by her last follow-up visit 1 year later, it had almost disappeared (Fig. 3). The patient was considered to be cured, and albendazole therapy was terminated. The patient is now examined for evidence of recurrence every 6 months.

The patient’s kyphosis in the area in which the cysts were located, which had developed due to the additional effects of the infection and surgical treatment, was stabilized with a Milwaukee brace, and surgical correction will be considered after the patient becomes 18 years of age.

**Discussion**

Vertebral hydatid disease arises with the arrival of the echinococcal embryo to the vertebrae. Necrosis occurs at the initial site of infection. The cysts enlarge by diverticular growing along the intratrabecular space and by infiltrating and destroying the bone.\(^\text{10}\) The extrasosseous stage begins with the resorption of the cancellous bone and perforation of the cortex and the periosteum. It is followed by paraspinal extension. At this stage the cysts start to behave as soft-tissue cysts, enlarging freely into the extradural and paraspinal areas; thus, neural compression is a common consequence.\(^\text{1,5,10}\)

The choice of treatment in spinal hydatid disease is surgery in all cases. Unfortunately, unlike the hydatid cysts located in the other sites in the human body, vertebral cysts tend to be multiple and invasive.\(^\text{1,8-11}\) Furthermore they are usually in close contact with the spinal cord and nerve roots. Therefore, removal of these spinal cysts without damaging the neural structures and perforating the cyst wall is not without risk.\(^\text{5,10}\) Spillage of the cyst fluid may cause severe anaphylaxis and dissemination.\(^\text{5}\) If total removal of the cyst cannot be achieved or spillage has occurred, albendazole or mebendazole should be administered. Although their effectiveness in hydatid disease is not yet well established, albendazole and mebendazole are the most frequently used pharmacological adjuncts.\(^\text{2,10,13,15,16}\)

Because mebendazole is poorly absorbed orally, high doses are required, which may induce neutropenia.\(^\text{16}\) Albendazole is a broad-spectrum antihelminthic agent with good oral absorption that blocks glucose uptake, depletes the glycogen stores, and thus immobilizes and kills the parasite but may be hepatotoxic.\(^\text{17}\) Successful treatment of hydatid dis-
ease with albendazole has been reported previously.\textsuperscript{7,17} In a study reported by Golematis, et al.,\textsuperscript{3} albendazole decreased the size of the large cysts and cured the smaller ones in 44 cases. In a multicenter study organized by the World Health Organization the efficacy of albendazole in hydatid disease was demonstrated.\textsuperscript{2} It was reported that the best way to evaluate the efficacy of albendazole is with follow-up computerized tomography or MR evaluation for 12 months.\textsuperscript{2} The disappearance or gradual shrinkage of the cysts, calcification of the cyst wall, or maintenance of cyst size for 1 year are all considered to be evidence of effective medical therapy.\textsuperscript{2}

Although there are also different treatment regimens, such as 10 mg/kg three times daily for 4 months or 10 mg/kg daily in 28 day cycles, our patient received oral albendazole 200 mg twice daily for 1 year while undergoing close follow up of her hepatic functions.\textsuperscript{2,14} The progressive shrinkage of the residual cyst demonstrated on follow-up MR imaging proves the efficacy of albendazole in our case.

**Conclusions**

The treatment of choice in spinal hydatid disease is total removal of the unruptured cysts.\textsuperscript{6,10,12} Because of the nature of the cysts, however, total removal without causing spillage is unlikely, and recurrence is the rule.\textsuperscript{2,10,18} In such cases, we recommend the administration of albendazole for at least 1 year after adequate neural decompression has been achieved. If a cure is achieved following 1 year of albendazole administration, periodic control MR images should be obtained to safeguard against recurrences.

**References**


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