Short-Course Combination Therapy with Albendazole and Praziquantel Chemotherapy in Recurrent Complicated Case of Vertebral Hydatidosis

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Abstract

Hydatid disease is the most common helminthic disease in human beings; however, spinal hydatid cyst accounts for 1% of all cases. We report a case of spinal hydatid disease that relapsed after surgery in a patient treated with albendazole for 6 months, and also a combination therapy with albendazole and praziquantel without further surgical intervention.

Keywords: Vertebra; Hydatidosis; Albendazole; Disease

Introduction

Hydatid disease is the most common helminthic disease in human beings, but spinal hydatid cyst accounts for 1% of all cases. Musculoskeletal involvement is commonly misdiagnosed as either a malignancy or pyogenic infection, due to nonspecific radiological appearance. Spinal hydatid disease frequently results in severe, acute-onset neurological deficits. Management of spinal hydatidosis is surgical. However, there is an increasing role for adjuvant chemotherapy. Of course, the selection of drug(s) and the duration of medical treatment is still controversial.

We report a case of spinal hydatid disease that relapsed after surgery in a patient receiving albendazole for 6 months, and also a combination therapy with albendazole and praziquantel without further surgical intervention.

Materials and Methods

A 47-year-old man was admitted in neurosurgery ward, complaining of low back pain and paresthesia in the left lower extremity when resting and walking. He denied any history of bites, trauma or previous back pain. MRI finding before surgery showed a cystic lesion involving T12-L3 (Figure 1).

The patient was working in a medical library before admission. During the hospitalization period, the patient was afebrile, without leukocytosis and his liver function tests were normal. A spinal surgery was done for the patient showing a cystic lesion containing multiple daughter cyst in T12-L3. After excision of the cyst, irrigation of the surrounding tissue with scolicidal agents was done. Pathologic examination confirmed Hydatid cyst. Early post-operative treatment with albendazole 400mg bid was recommended for 6 months (each lasting 3 weeks, being interrupted by 1 week off therapy). Meanwhile, the patient was asymptomatic. After 6 months, the patient developed low back pain, paresthesia, and severe pain in both lower extremities.

MRI findings suggested only the scar of the previous surgery. So, analgesics and dexamethasone were recommended. Due to the severity of symptoms, the patient was unable to work and had to stay at home. The 5th MRI of the spine was done after progression of symptoms, revealing multiple cystic lesions in epidural space (Figure 2). The patient was advised to reoperate, but he preferred combination medical ther-
apy with albendazole 400 mg bid, and praziquantel 600 mg TID. After one course combination of chemotherapy (albendazole and praziquantel for one week followed by 3 weeks albendazole), the patient’s symptoms were relieved and MRI finding showed resorption of cystic lesions in the epidural space (Figure 3). The patient received 3 courses of combination therapy as mentioned above. Due to LFT rising to 1.5 times of baseline between the second and third courses, chemotherapy was discontinued for 1 month. After the first course of combination therapy, the patient resumed his work, and after 6 months follow-up with physical findings and MRI, the patient was in good condition without any evidence of relapse.

Fig 1: Lumbar mass in sagital MRI before surgery.

Fig 2: Recurrence of spinal hydatidosis; 6 months after surgery.

Fig 3: Lumbar MRI of patients after combination therapy.

Discussion

Surgical excision of vertebral hydatidosis remains the treatment of choice.\textsuperscript{7} High rates of postoperative recurrence need adjuvant chemotherapy, paraplegia being the most serious complication of vertebral hydatidosis, and cysts invading the spinal canal and causing direct compression of spinal cord.\textsuperscript{8} Chemotherapy appears to be effective in the prevention of late recurrence after spinal hydatidosis surgery but the selection of the drug(s)\textsuperscript{1} and the duration of the medical treatment are still controversial. Postoperative chemotherapy with albendazole for at least 1 month up to 1 year is reported.\textsuperscript{5,9} In one report, postoperative chemotherapy in cervical hydatidosis continued for 9 years.\textsuperscript{7} Although the efficacy of post-operative albendazole treatment is reported in some cases of vertebral hydatidosis,\textsuperscript{6,7} failure of albendazole therapy is reported in other cases.\textsuperscript{11,12} A few reports of the combination therapy with albendazole and praziquantel are seen.\textsuperscript{13,15} In contrast to monotherapy, combination therapy produced a significant reduction in the number and viability of cysts and decrease in the treatment course.\textsuperscript{14,17} However, failure of long term combination therapy with albendazole and praziquantel was reported in one case.\textsuperscript{17} But in our case, the patient made an uneventful recovery after chemotherapy with albendazole 400 mg bid. and praziquantel 600 mg TID for one week. According to clinical and paraclinical findings, plasma and CSF levels of albendazole and praziquantel in patients with neurocysticercosis
showed effective CSF level for each drug.\(^{18}\) In vivo studies of praziquantel treatment in experimental hydatidosis showed reduction of treatment course to 1 and 2 month.\(^{19}\) Praziquantel is a highly effective scolicidal agent both in vivo and in vitro.\(^{20,21}\) The probable role for praziquantel might be preventing en- cystment of protoscoleces following perioperative spillage in human hydatidosis.\(^{22}\) Hydatid disease should be considered in the different diagnosis of spinal mass. Surgery accompanied by early postoperative chemotherapy is the treatment of choice in the management of complicated and relapsing hydatid disease. The role of praziquantel is not clearly defined in the literature and further studies are required to prove the merits of combination therapy.

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References

22. Taylor DH, Morris DL. Combination chemotherapy is more effective in post spillage prophylaxis for hydatid disease than either albendazole or praziquantel alone. Br J Surg 1989;76:954. [2804596]