

Alveolar echinococcosis in the head of pancreas

A case report

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Abstract

Rationale: Pancreatic alveolar echinococcosis (AE) is an exceptionally rare disease; no more than 10 cases have been published to date. It is characterized as extensive local tissue infringement and destruction; thus, extended surgical resection, such as duodenopancreatectomy, is often needed to obtain a negative resection margin so as to improve the long-term outcome and prognosis.

Patient concerns: A middle-aged Tibetan man was admitted due to a 16-year history of recurrent pain in the right upper abdomen. Magnetic resonance imaging scan showed a cystic lesion in the VI segment of his liver and several cystic lesions in the head of pancreas.

Diagnoses: Pancreatic AE.

Interventions: The patient adopted a preserved surgery of partial cystectomy and completely removing the content of the cyst and then soaking by hypertonic saline combined with adjuvant medical therapy of albendazole.

Outcomes: The patient was monitored on a regular basis at the outpatient department; the patient is still alive and has already survived 8 years till now.

Lessons: A preserved surgery combined with adjuvant medical therapy of albendazole can also contribute to a good survival outcome for AE located at the head of pancreas.

Abbreviations: AE = alveolar echinococcosis, MRI = magnetic resonance imaging.

Keywords: albendazole treatment, alveolar echinococcosis, long-term survival, pancreas, preserved surgery

1. Introduction

Echinococcosis, caused by the infection of *Echinococcus granulosus*, *Echinococcus multilocularis*, *Echinococcus vogeli*, and *Echinococcus oligarthruscoccosis*, is an endemic, chronic,

and sometimes fatal parasitic infectious disease.^[1–3] Because of the vast capillary network of liver and lung, the 2 organs work as the primary filtering beds of echinococcus, therefore almost 95% of echinococcosis infections are found in these 2 organs.^[4,5] Pancreatic echinococcosis is an unusual parasitic infestation with a reported proportion of 0.1% to 2% of all echinococcosis, among them, cases of pancreatic alveolar echinococcosis (AE) are exceptionally rare, which has been depicted as result of locoregional invasion or metastasis.^[6] The presenting symptom is variable and imaging features also lack of specificity, thus, establishing a definite preoperative diagnosis seems very difficult. Pancreatic AE has a character of widely invading the surrounding organs and tissues^[7]; thus, extended surgical resection, such as duodenopancreatectomy or distal pancreatectomy or even total pancreatectomy, is often needed to obtain a free margin.

Herein, we presented an infrequent long-term survivor of AE located at the head of pancreas; the patient was treated with a preserved surgery combined with preoperative and postoperative adjuvant medical therapy of albendazole. Our aim was to help recognize the clinical characters and treatment options of this disease.

2. Case presentation

A middle-aged Tibetan man from remote mountainous areas presented with a 16-year history of recurrent pain in the right upper abdomen was admitted to our hospital. Physical examination of the abdomen showed no abnormalities. He denied the history of pancreatitis and pancreatic carcinoma. He had a habit of raw meats ingestion. Laboratory evaluation demonstrated echinococcus antibody was positive, but other values including blood routine examination, liver functions, and

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Rong-Xing Zhou and Hai-Jie Hu contributed equally to this study and were the co-first authors; Rong-Xing Zhou and Hai-Jie Hu contributed to the data acquisition and analysis and drafted the manuscript. Yong Jiang, Wen-Jie Ma collected the data. Fu-Yu Li contributed to the study design and revision of the manuscript. All authors read and approved the final manuscript.

The patient gave informed consent before we wrote the case report.

This article does not contain any studies with human participants or animals performed by any of the authors.

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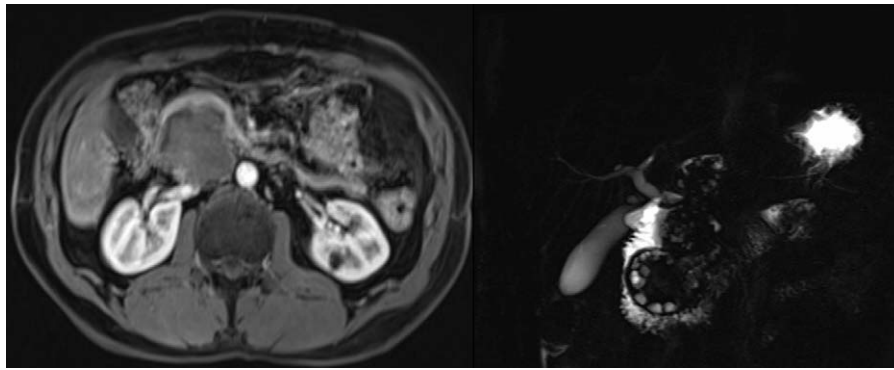


Figure 1. Magnetic resonance imaging scan showed several cystic lesions in the head of pancreas.

tumor markers were normal. Serological tests of hepatitis B and C were also negative. Ultrasonography showed a 3-cm hepatic cystic lesion and a 4.2-cm cystic lesion in the uncinata process of pancreas. Magnetic resonance imaging scan showed a cystic lesion in the VI segment of his liver and several cystic lesions in the head of pancreas (Fig. 1). All were with mixed signals in both T1WI and T2WI. The left renal vein and the inferior vena cava were shown to be compressed. The main pancreatic duct was not dilated. The diagnosis of hepatic hydatid disease and pancreatic cystadenoma was initially made, and the patient was started with 2 weeks of preoperative albendazole treatment (400 mg PO bid). Then, the patient underwent surgical exploration. Intraoperative exploration revealed that a 2.5 × 3.0 cm hard mass was found in right posterior inferior segment of liver and was adhered with surrounding tissues. Another cystic lesion was located in the uncinata process of pancreas measured at 8 × 8 cm. There was a clear boundary between pancreatic tissues and the lesion. The pancreatic cystic lesion, with a thin capsule of 0.6 cm, contained yellowish-white material. Total pericystectomy was performed for liver lesion. Partial cystectomy was performed for pancreatic lesion and hypertonic saline (15%) was drainage into the cyst for a minimum duration of 10 min. Histologic examination revealed diagnosis of hepatic and pancreatic alveolar echinococcosis (AE) with scolex seen in the fibrous tissues (Fig. 2). The postoperative course was uneventful and the patient was discharged on the

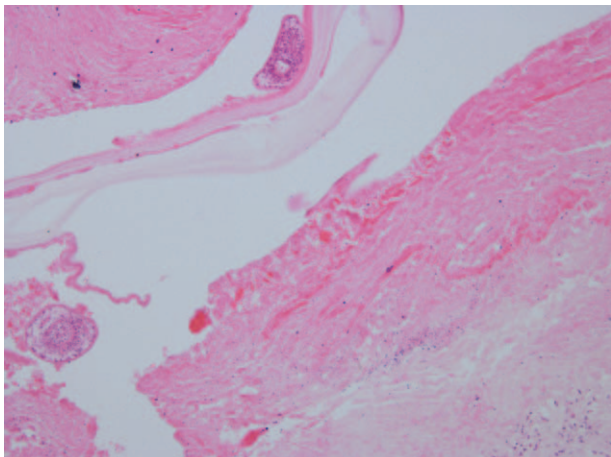


Figure 2. Histologic examination revealed diagnosis of pancreatic alveolar echinococcosis.

5th of postoperative day and then continued 2-year albendazole therapy. He was well followed up at our outpatient department with serologic tests and imaging procedures, and no recurrence was occurred 8 years after the surgery.

3. Discussion

The echinococcosis can be infected through accidental ingestion of echinococcus eggs coming from the feces of infected dogs.^[8] The eggs of the parasite can penetrate the intestinal wall, enter the portal system, and then reach the liver, it was reported liver (50–70%) and lung (20–30%) are the most affected organs.^[9] Other potential infected organs could include the spleen, the central nervous system, heart, thyroid gland, and the bones.^[3,10,11] However, pancreatic echinococcosis is extremely rare; no more than 70 cases were reported in the English literature currently. Head (57%), body (24–34%), and tail (16–19%) of the pancreas are all likely to be involved. Among them, cases of pancreatic AE are exceptionally rare; no more than 10 cases were recorded in previous literature. Due to the different anatomic location, different size, and the variable relationship with the adjacent organs, the clinical manifestation of the pancreatic AE is variable, some patients may present as obstructive jaundice, cholangitis, recurrent acute pancreatitis, pancreatic abscess, pancreatic fistula, or duodenal obstruction.^[12]

Characterized as extensive infringement and destruction, pancreatic AE masquerades as a life-threatening malignant disease. Many previous studies adopted an extensive surgery for pancreatic AE, resulting in high postoperative morbidity. In our current study, we reported a rare case of AE located at the head of pancreas. Based on our experience in the treatment of liver AE, we adopted a preserved surgery combined with albendazole treatment. The patient has already survived 8 years after surgery. He was well followed up at our outpatient department with serologic tests and imaging procedures with no recurrence. Thus, we believe patient diagnosed as pancreatic AE can also benefit from a preserved surgery and albendazole treatment without altering the normal physiological structure.

4. Conclusion

For patients with pancreatic lesions who have a history of raw meats ingestion or traveling or migration to epidemic area, the diagnosis of pancreatic AE cannot be ruled out and should be kept as a differential diagnosis. For cases require surgical treatment, partial cystectomy combined with completely remov-

ing of the cyst content is also an effective treatment without altering the normal physiological structure. Considering that there is a lack of surgical radicality, adjuvant medical therapy with benzimidazoles or albendazole is a supplementary treatment so as to reduce the recurrent rate and improve the long-term outcome.

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