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Solitary Nonparasistic Cyst of the Liver in Children : two cases of rapid enlarging cyst caused by minor abdominal injury*)

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ABSTRACT

Two cases of infantile solitary nonparasitic cyst of the liver caused by slight contusion were experienced. These cases showed a similar clinical course with symptoms including enlargement of the liver, abdominal pain, and anemia that probably resulted from intracystic hemorrhage. Abdominal echo examination and CT scanning were found effective for the diagnosis of this diseases. As treatment, it is advisable to first administer conservative treatment for improvement of anemia and inflammation and then perform operation. The surgical treatment should be selected so as to preclude recurrence and entail as little invasion as possible. Partial excision of the liver cyst combined with fenestration seemed to be the safest and most effective procedure.

INTRODUCTION

Congenital solitary nonparasitic cyst of the liver is an uncommon lesion in children[3-5]. These cysts sometimes increase abruptly in size due to infection, or intracystic hemorrhage.

Patients who visit the hospital sometimes have slight fever and other inflammatory signs together with enlarged abdominal mass. In these cases the most important considerations are differential diagnosis from malignant tumor and surgical treatment for benign disease.

We have recently experienced two cases that developed from a minor abdominal injury and showed a clinical course closely resembling this disease. In this report, we discuss the diagnosis and surgical approach for this disease.

CASE REPORT

Case 1. 5-year-old boy

This boy sustained a slight contusion of the abdomen while playing on 30 April 1981. He complained of abdominal pain, but this subsided. On 1 May, the abdominal pain recurred, and increased and decreased alternately. On 9 May, he was admitted to a hospital for slight fever and anemia. His laboratory data indicated inflammation. An abdominal echogram revealed a cystic lesion of the liver, and the following day he was referred to our clinic.

The physical findings at admission showed no abnormalities other than anemia. Physical examination of the abdominal region showed that the spleen was not palpable, the abdomen was flat and soft, and the liver was palpable 2.5 cm below the right costal magin. The abdominal mass was not clearly palpable, but the right epigastric region was slightly tender.

Case 2. 8-year-old girl

This girl fell and sustained a contusion of the right epigastric region while playing in the park on 2 January 1982. Four hours later she started vomiting which was relieved by the administration of a sedative. Her pain intensified from the following day (3 January), and she was referred to and admitted to our clinic.

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On admission, the only physical finding other than the abdominal findings was anemia. The abdominal findings were unremarkable except that the right epigastric region was slightly distended and tender. Laboratory data: Cases 1 and 2 both showed anemia, slightly elevated S-GOT and positive CRP (Table 1). In addition to the above findings, case 2 showed mild leucocytosis and elevation of S-GPT, and case 1 showed mild elevation of LDH.

Table 1. Laboratory data at administration of the case

	Case 1	Case 2
WBC (/mm³)	5900	11200
RBC (/mm³)	310×10^4	213×10^4
Ht (%)	25.0	19.0
Platelet (/mm³)	34.4×10^4	28.0
T. bil (mg/dl)	0.9	0.6
S-GOT (u/liter)	51	71
S-GPT (u/liter)	34	90
LDH (u/liter)	986	421
Amylase (units)	148	98
CRP	+ 2	+ 1

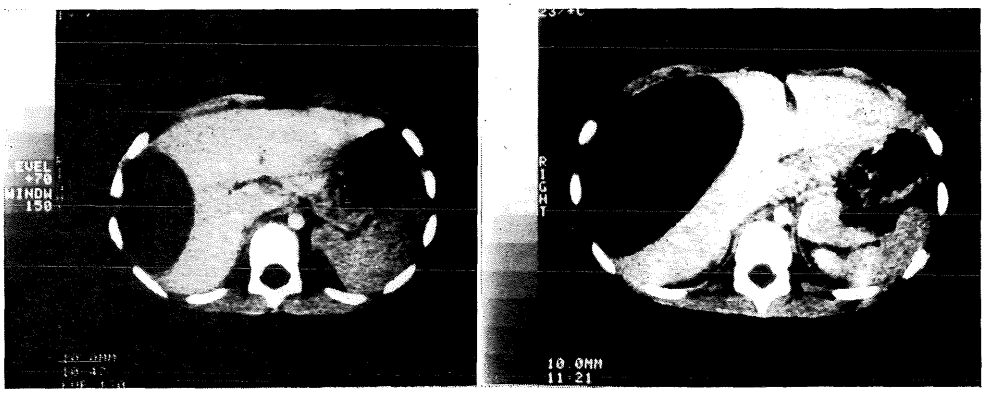
Medical imaging diagnosis: Abdominal plain films showed downward compression of the intestinal tract and hepatomegaly in both cases and right pleural effusion in case 2.

In both cases abdominal echo examination revealed intensified sharp echo waves in the margin of the right lobe of the liver and the existence of a large cystic tumor of mixed hypoechogenic and echogenic substance. Liver scintigraphy (^{99}Tc) showed SOL. Whole body CT centered around the posterior region of the right lobe of the liver in case 1 and over the entire right lobe in case 2 showed a sharply defined cystic lesion in which the capsule was undergoing a smooth change to normal liver, suggesting a possible complication of intracystic hemorrhage or cystic inflammation (Fig. 1).

Celiac angiography of case 1 showed the lobe of the liver and, in its periphery, an inflammatory hyperplasia of minute vessels surrounded by a thick hypervascular zone. Case 2 showed stretching of the right hepatic artery and a hypovascular area (Fig. 2).

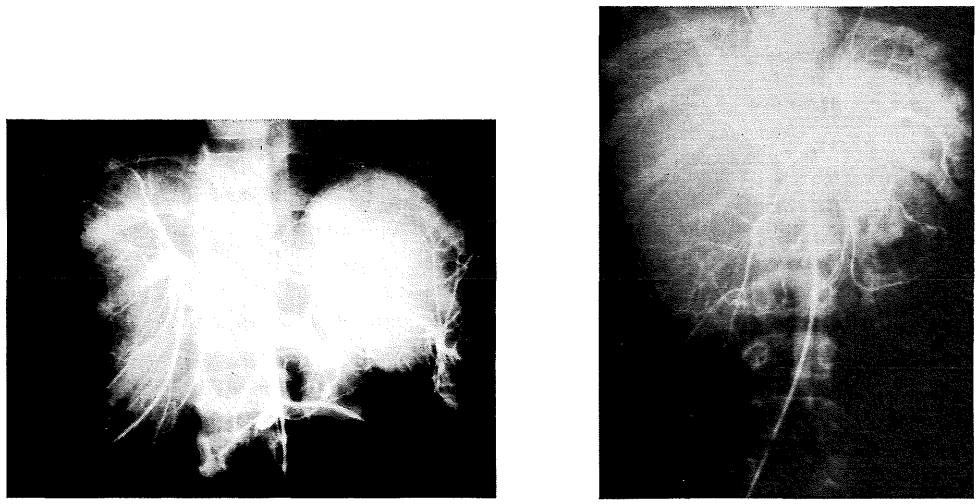
Operative findings and methods: In case 1, a cystic lesion 5.0 cm × 2.0 cm × 5.0 cm in size was present on in the posterior area of the right lobe of the liver, with adhesion of its surface to the retroperitoneum and the diaphragm due to inflammation. Cholangiography performed during the operation showed no communication between the cyst and the biliary system. The cyst contained a mass of coagulated blood. In case 2, adhesion of the gastrocolic omentum to the right lobe of the liver and a giant cyst 7.5 cm × 8 cm × 7 cm in size extending over the anterior and posterior areas of the right lobe of the liver were found. The cyst contained old bloody serous fluid. Partial excision and fenestration of the cystic wall were performed in both cases (Fig. 3).

Pathological findings and prognosis: Cases 1 and 2 showed calcification, lack of distinct



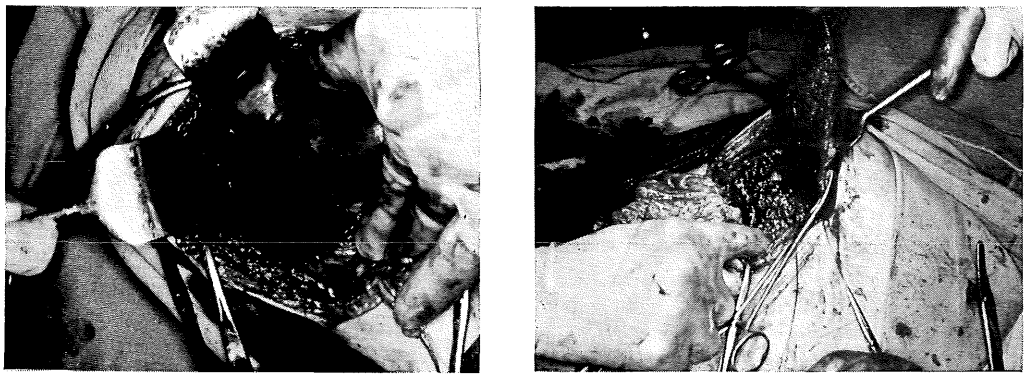
(a) (b)

Fig. 1(a) A sharply defined low density area is seen in the posterior region of the right lobe (Case 1).
Gig. 1(b) A low density area is found beyond the right lobe (Case 2).



(a) (b)

Fig. 2(a) Stretched blood vessels in the liver and small vessels with hypervascular zone show existence of inflammation (Case 1).
Fig. 2(b) Stretching and hypovascular zone exist in the right hepatic artery (Case 2).



(a) (b)

Fig. 3(a): Case 1, 3 (b): Case 2
During the cyst has been resected to the liver edge (Fig. 3(b)), and the free margin oversewn with a continuous suture of dexon (Fig. 3(a)).

epithelium, and lack of neoplastic change. Both cases obtained early relief and were discharged from the hospital. Tests done at the outpatient clinic have shown no abnormalities.

DISCUSSION

Liver cyst is a comparatively rare, asymptomatic disease, and few cases have been reported. However, reports of these cases have been increasing recently with the progress of medical imaging[3,4]. This disease occurs mainly in those patients with age ranging from forties to sixties, with the peak age in the forties[4]. Cases occurring in infancy are concentrated among those with six months to under three years of age[5]. Reports of infantile cases are rare. The origin of this disease is not known, but the most commonly accepted view at present attributes it to displasia of the liver in the embryonic stage. Others ascribe it to biliary stagnation and inflammation due to obstruction of the bile duct[4-6]. On the other hand, infantile liver cysts are, according to Edmondson[1], mostly cystic mesenchymal hamartoma or solitary nonparasitic cysts. The former are characterized by irregular thickness

of the wall and are polycystic or partially noncystic. The latter are characterized by their smooth surface, their wall, and by the fact that they are solitary and may be very large. Histologically, the former are mesenchymal or have developed through hyperproliferation of connective tissues that have differentiated to varying degrees; cyst formation is marked and the inside is covered with pavement cells. The latter are reportedly usually large and solitary; the inside is often covered with smooth, columnar or cuboidal epithelium and flattened epithelium; and the wall is comparatively thin and composed of differentiated connective tissue. In view of these facts, the cases reported here can be considered to be cases of solitary nonparasitic cyst affected by hemorrhage and inflammation due to injury. Solitary nonparasitic cysts generally present no specific symptoms and are often discovered when, as in the cases of this report, intracystic hemorrhage develops, when a mass grows abruptly in size, and when pain and anemia are present. On the other hand, abnormalities rarely occur in laboratory findings, and, if found, are often very mild. As a matter of fact differential diagnosis from malignant tumor is important in these cases. Abdominal echo examination and abdominal CT scanning can be effective diagnostic methods for this purpose. Diagnosis can be easily made, especially by abdominal echo examination based on the quality of ultrasonic waves. The characteristic cystic pattern is one in which the margin of the echo pattern is even, internal echo is absent, and echo of the posterior wall is increased. The limit of detectability is said to be 1 cm in diameter as against 2 cm in the case of tumor[2]. Diagnosis by body CT scanning is more often used in differentiation from liver tumor. Liver cyst is observed as a round, circumscribed, low-density area, but differentiation of liver cyst from other diseases is difficult by CT. When differentiation of liver cyst from other diseases is difficult, hemangioma and other malignant diseases of the liver can be ruled out by the combined use of angiography.

Surgical treatment is the only method of treating this disease. Various techniques are employed according to the size and site of the cyst, but enucleation of the cyst or partial excision of the liver is performed if the cyst is comparatively small. If the cyst is large or involves important blood vessels such as the portal vein, only partial removal should be performed since it is a benign disease. The problem is treatment following partial removal. Besides fenestration, methods include anastomosis to the jejunum, incineration, external marsupialization of the residual cyst, etc. Internal fistulization with the jejunum without removing the cystic wall is, in our opinion, inappropriate, as it will cause retrograde infection due to low intracystic pressure. The most suitable method seems to be removal of the part of the cystic wall exposed to the surface of the liver if it is confirmed that the cyst contains no large quantity of bile and there is no communication with the bile duct.

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