Concomitant Echinococcal Cyst and Hepatocellular Carcinoma in a Single Liver Lesion

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Both echinococcal cyst and hepatocellular carcinoma are commonly encountered. However, the combination of these two diseases in a liver lesion has never been reported. We present a patient with hepatoma contiguous with an echinococcal cyst. Differentiation between the echinococcal cyst and hepatocellular carcinoma on CT images evoked diagnostic difficulty before operation. **(Mid Taiwan J Med 2004;9:50-4)**

Key words

echinococcal cyst, hepatocellular carcinoma, liver

INTRODUCTION

Echinococcal cyst primarily affects the liver and demonstrates characteristic image findings. Although there are many potential complications, including cyst rupture, biliary communication, portal vein involvement, and abdominal wall invasion [1], no cases of malignant change have been reported. We present a rare case of concomitant echinococcus and hepatocellular carcinoma in a single lesion, and discuss the possible pathogenesis.

CASE REPORT

A 61-year-old man with a history of chronic hepatitis B and C presented with impaired liver function during routine health examination. He denied abdominal discomfort, weight loss, and fatigue. He was a businessman and had traveled to many Asian countries. Physical examination did not reveal significant findings. The laboratory data showed elevated GOT (83.9 IU/L, control data 9-43), r-GT (276.9 U/L, control data 7-50), and α-fetal protein (449 ng/mL, control data 0-30).

We suspected hepatocellular carcinoma, and so performed a dynamic helical CT of the liver with a Picker PQ 5000 scanner (Picker International, Cleveland, OH). After the non-enhanced CT, 100 mL iodinated contrast media (Omnipaque, 350 mg I/mL, Nycomed, Oslo, Norway) was injected at a rate of 3.5 mL per second by a power injector into the superficial vein in the left hand. The slice thickness was 10 mm. The index was 1. Both arterial phase and portovenous phase images were acquired at a delay time of 25 seconds and 75 seconds, respectively. A cystic lesion measuring 9 cm in diameter was found in the inferior portion of the right lobe of the liver. Calcification in the wall and septa of the cyst was found (Fig. A). The cystic portion was not enhanced. Multiple well-enhanced nodules were found adjacent to the cystic portion in the arterial phase images (Figs. B, C). Because the CT features were confusing, diagnostic angiography was performed. Celiac and super-selective hepatic angiography demonstrated that the lesion consisted of a hypovascular cystic part and multiple

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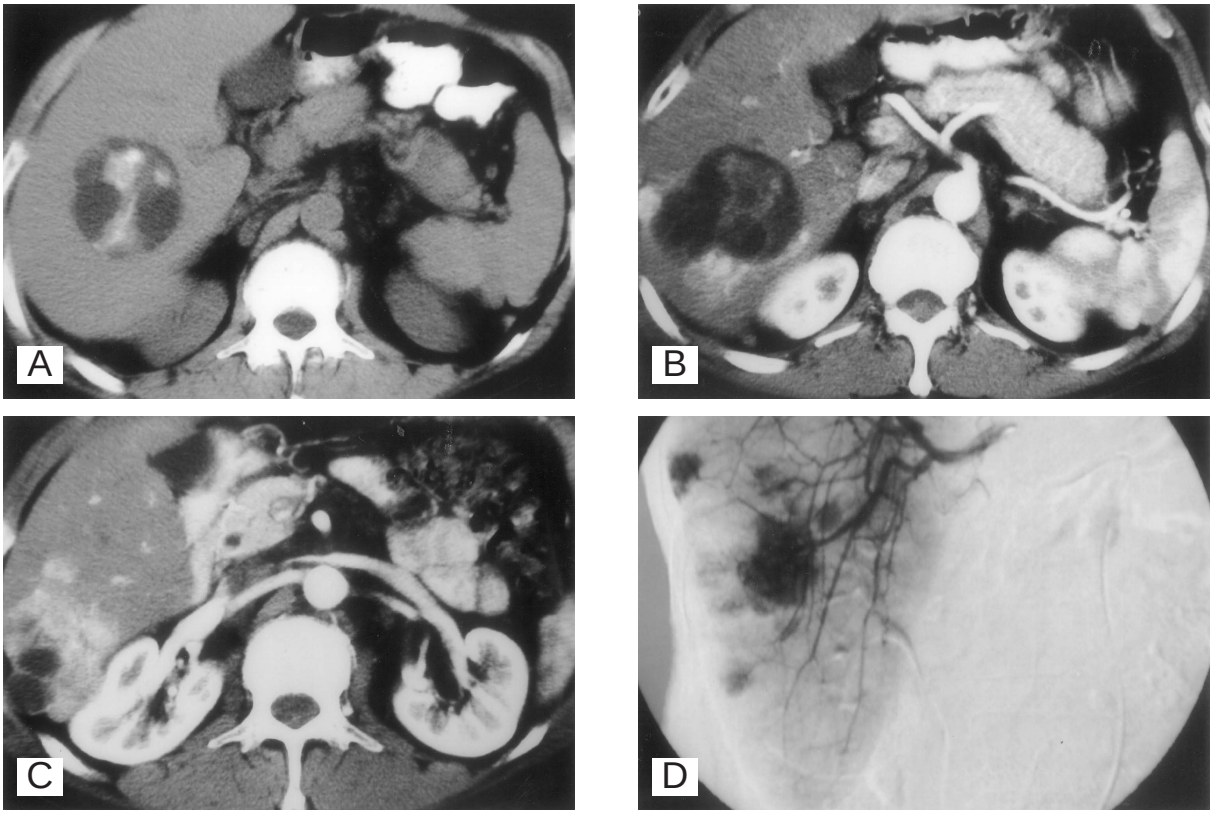

Figure. A 61-year-old man with concomitant echinococcal cyst and hepatocelluar carcinoma. A: Non-enhanced CT scan of the liver shows a round, water density cystic lesion with thick calcified septa. B: Arterial phase image of dynamic helical CT one centimeter caudal to A shows two small well-enhanced nodules in the periphery of the cystic lesion. Note the septa within the cyst. C: Arterial phase image of dynamic helical CT three centimeters caudal to B shows confluence of the multiple enhanced nodules surrounding the cyst. D: Superselective hepatic angiography shows a hypovascualr lesion mixed with multiple variable size enhanced nodules in the inferior-lateral aspect of the right lobe of liver.

hypervascular nodules (Fig. D); no arterioportal shunting was found.

We believed that the leaion was a cystic liver tumor, and so a bi-segmentectomy (S6 and S7) and cholecystectomy were performed. Grossly, the peripheral area of the resected specimen as well as the liver parenchyma was brown, soft, and elastic. There was a cystic space measuring 5 cm in diameter centrally located in the specimen. The cavity was lined with calcification. Multiple pieces of transparent and myxoid material measuring up to 3 cm × 2 cm × 2 cm were noted within the cyst with calcified septa. Outside of and intimately contiguous to the cystic part were multiple grayish-white and well circumscribed nodules measuring up to 1 cm in diameter. Microscopically, these nodules showed grade 3 to grade 4 non-capsulated hepatocellular carcinoma with portal vein invasion. The section margin was free of tumor. The cystic portion revealed an echinococcal cyst lined with external laminated membrane and germinal membrane; scolices were also present. The cyst was walled by broad fibrous tissue. The adjacent liver tissue showed hepatitis with portal fibrosis and piecemeal necrosis.

DISCUSSION

The Echinococcus includes two subtypes: *Echinococcus granulosus* and *Echinoccus multicularis*. Humans are an intermediate host. Liver is one of the target organs. *Echinococcus granulosus* is more commonly encountered and when it involves the liver, it presents as a cystic lesion with ring-like calcification on radiographs. Thus many asymptomactic echinococcal cysts are first discovered incidentally during radiological examination [2]. Involvement of the liver by

Echinococcus granulosus is suggested in plain adiography by the presence of curvilinear and ring-like calcification, and on CT by well defined oval or spherical lesions of near water density (3-30 HU). However, theses findings are not specific and may be seen in abscesses, cystic metastasis, Caroli's disease and congenital cysts. *Echinococcus multicularis* is less common than *Echinococcal granulosus* and is often confused with hepatic malignancy because of its ill-defined margins, mass-like features with areas of necrosis, smaller cavities, and amorphous calcification. The complications of echinococcal cyst include cyst rupture, infection, trans-diaphragmatic thoracic involvement, perforation into hollow viscera, peritoneal seeding, biliary communication, portal vein involvement, and abdominal wall invasion. Involvement of the biliary system and portal vein, or compression of the portal vein and the hepatic vein may induce chronic liver tissue damage [1,3,4].

Cases of hepatic echinococcus can mimic a liver tumor by presenting as a solid mass with or without calcification on sonograms, CT, or MR images [5-9]. These hepatic tumors might have rim calcification or cystic change [3,10-14]. One case of focal nodular hyperplasia contiguous to an echinococcal cyst was reported [15]. It is believed that the hyperemic rim of the echinococcal cyst causes the development of an adjacent focus of FNH [15]. However, no cases of echinococcal cyst coexistent with hepatocellular carcinoma have been reported.

It is not known if there is a direct relationship between the echinococcal cyst and the hepatomas seen in this case. But it is well-known that any factor causing chronic liver cell damage makes the DNA of the hepatocyte more susceptible to genetic alterations. Thus, as indicated above, chronic liver disease of any type is a risk factor and might lead to the development of liver cell carcinoma [16]. It is therefore postulated that the echinococcal cyst induced chronic liver parenchymal damage and increased the risk of heptocellular carcinoma in this case. Although chronic hepatitis B and C virus infections, which were noted in this case, might be another factor for developing hepatocellular carcinoma, evidence of liver cirrhosis was not obvious in this patient. Moreover, the multiple small hepatoma foci occurred locally in the lower portion of the right lobe of liver and intimately surrounded the echinococcal cyst.

In conclusion, echinococcal cyst is a benign disease but it may induce chronic liver parenchyma damage. Therefore, we assume that this condition may increase the risk of hepatocelluar carcinoma. Although rarely encountered, when images of a liver lesion show features of both calcified cystic parts and well-enhanced nodules, the possibility of concomitant echinococcal cyst and hepatocellular carcinoma should be included in the differential diagnosis. It is important to carefully correlate the diagnosis with the medical history, clinical presentation and laboratory data.

REFERENCES

1. Pedrosa I, Saiz A, Arrazola J, et al. Hydatid disease: radiologic and pathologic features and complications. [Review] *Radiographics* 2000;20:795-817.
2. Markell EK, Voge M, John DT. The cestodes. In: Selma Ozmat, ed. Medical parasitology. Philadelphia: Saunders, 1992:226-60.
3. Mitchell AW, Higgins JN, Tate J, et al. Ring calcification in primary liver cell carcinoma mimicking hydatid disease. *Gut* 1994;35:567-8.
4. Slim J, Sarra S, Kamel A, et al. Hepatic hydatid cyst with fat-fluid level and Budd-Chiari syndrome. *Ann Saudi Med* 2001;21:225-7.
5. Muhlhauser J, Kreft B, Kalff JC. Atypical Echinococus cysticus (granulosus) of the liver. Radiologe 1993;33: 466-8. (German)
6. Kamei T, Futatsuya R, Seto H, et al. A case of liver echinococcosis mimicking a solid liver tumor. *Radiat Med* 1991;9:101-4.
7. Grosser G, Hauenstein KH, Henke W. Echinococcus cysticus of the liver-sonographically a solid tumor. *Radiology* 1985;25:442-4. (German)
8. Sansot M, Le Treut Y, Burger G, et al. Semiology of pseudotumoral forms of hydatid cyst of the liver. Apropos of 7 cases]. *Sem Hop* 1983;59:2979-84. (French)
9. Barriga P, Cruz F, Lepe V, et al. An ultrasonographically solid, tumor-like appearance of echinococcal cyst in the liver. *J Ultrasound Med*

1983;2:123-5.
10. Siebenkotten V, Wenning M, Malms J, et al. Cystic liver tumor resembling echinococcosis. *Z Gastroenterol* 1999;37:379-83. (German)
11. Singh Y, Winick AB, Tabbara SO. Multiloculated cystic liver lesions: radiologic-pathologic differential diagnosis. *Radiographics* 1997;17:219-24.
12. De Backer AI, De Schepper AM, Michielsen PP, et al. Hepatobiliary cystadenoma with mesenchymal stsroma mimicking hydatid cyst. Report of a case. *Acta Gastroenterol Belg* 2000;63:317-20.
13. Flemming P, Frerker M, Klempnauer J, et al. Benign schwannoma of liver with cystic changes misinterpreteted as hydatid disease. *Hepatogastroenterology* 1998;45:1764-6.
14. Rutledge JN, Pratt MC, Taupmann RE. Biliary cystadenoma mistaken for an echnococcal cyst. *South Med J* 1983;76:1575-7.
15. Komisarof JA, Olthoff K, Siegelman ES, et al. Focal nodular hyperplasia contiguous with an echinococcal cyst. *Am J Gastroenterol* 2000;95:1078-81.
16. Isselbacher KJ, Dienstag JL. Tumors of the liver and biliary tract. In: Fauci AS, Braunwald E, Jsselbacher KJ, et al, eds. Harrison's principles of internal medicine. New York: McGraw-Hill, 1998:578-81.

包囊蟲病合併肝細胞癌

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包囊蟲病及肝細胞癌都很常見，但是在肝臟這兩種病灶同時合併在一起的情形卻不曾被報告過。包囊蟲病主要侵犯肝臟，而且有特別的影像學表現，雖然包囊蟲病有許多的併發症，包括囊泡破裂、與膽道相通、侵犯門靜脈或腹壁等，但是尚未有癌化的病例被報告過。我們報告一肝細胞癌直接與包囊蟲病的囊泡相連在一起的病例，影像學上同時包含了肝細胞癌與包囊蟲病的特徵，造成開刀前診斷上的困難。造成此種罕見合併症的可能致病機轉在文中也將被討論到。（中台灣醫誌 **2004;9:50-4**）

關鍵詞

包囊蟲病，肝細胞癌，肝癌

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