CASE REPORT

Adult hepatic cavernous haemangioma with highly elevated alpha-fetoprotein

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A 35-year-old Chinese man presented with dull pain in the right hypochondrial region for the previous 2 months. Laboratory examination revealed that his serum alpha-fetoprotein level was 1890 μg/L (reference range, 0-20 μg/L), and computed tomographic scan showed a hypodense lesion in the left liver lobe. At laparotomy, a dark reddish soft tumour (3.0 x 3.5 cm in diameter) was found in the medial segment of the liver (segment III). The tumour became markedly smaller than that before resection, and a vessel-like structure was found on the cut surface. Intra-operative pathology and postoperative histopathology examinations revealed that the tumour was a cavernous haemangioma of the liver. The serum alpha-fetoprotein level decreased to 3.5 μg/L by the fourth postoperative week. Clinicians should be aware that some rare tumours besides hepatocellular carcinoma and endodermal sinus tumours (yolk sac tumour), for example, hepatic haemangioma, can produce alpha-fetoprotein.

Introduction

Alpha-fetoprotein (AFP) is a fetal-specific glycoprotein produced primarily by the fetal liver. Normally, the AFP serum concentration falls rapidly after birth and its synthesis in adult life is repressed.[1-3] Alpha-fetoprotein is one of the best-known carcinofetal proteins, and is useful for the diagnosis of hepatocellular carcinoma (HCC) during follow-up of patients with chronic liver diseases.[4-6] Some reports have indicated that a high serum AFP concentration correlates with a poor prognosis for patients with HCC.[4,5] The serum concentration of AFP also increases in other diseases, such as hepatic cirrhosis, fulminant hepatitis, endodermal sinus tumours (yolk sac tumour) of the testis, ovary, and extragonadal sites.[2,7-11] Hepatic haemangioma is the most common vascular tumour in both adults and children.[2,12,13] This report is of a patient with hepatic haemangioma with a highly elevated AFP level.

Case report

A 35-year-old Chinese man was admitted to the Department of General Surgery, The First Affiliated Hospital of Wenzhou Medical College, Wenzhou, China, in December 2003 with dull pain in the right hypochondrial region for the previous 2 months. At physical examination, no ascites was detected and the abdomen was tender on palpation. The liver and spleen were not palpated, and other systems were normal on examination. The results of laboratory tests performed at admission are shown in the Table.

Ultrasound examination showed a hypodense lesion in the left liver lobe (Fig a). There was no evidence of thromboses in the hepatic or portal veins by Doppler ultrasound. The AFP level was 1890 μg/L (reference range, 0-20 μg/L). Computed tomography (CT) of the abdomen showed that the liver margin was irregular and the left lobe of the liver contained a hypodense lesion resembling primary HCC. Hepatocytes were positive for keratin and AFP, but negative for vimentin. Owing to the young age of the patient, the presence of a hepatic hypodense lesion resembling a tumour and the high AFP level, examination of the testis was also done to exclude a testicular tumour, but there were no abnormal findings.

Exploratory laparotomy performed under epidural and general anaesthesia on 3 December 2003 revealed a dark reddish soft tumour (3.0 x 3.5 cm in diameter) at the medial segment of the left lobe (segment III). After an irregular hepatectomy with 2.0-cm margins, the tumour specimen became markedly smaller than that before resection, because of blood discharged from blood sinus, and a vessel-like structure was found in the cut surface. To exclude another similar lesion, intra-operative ultrasound examination was done but no other lesion was found. Intra-operative pathology and postoperative histopathology examinations revealed that the tumour was a cavernous haemangioma of the liver (Fig b). The serum AFP level decreased to 430 μg/L on the seventh postoperative day and 3.5 μg/L

Key words
alpha-Fetoproteins; Carcinoma, hepatocellular; Hemangioma; Liver neoplasms

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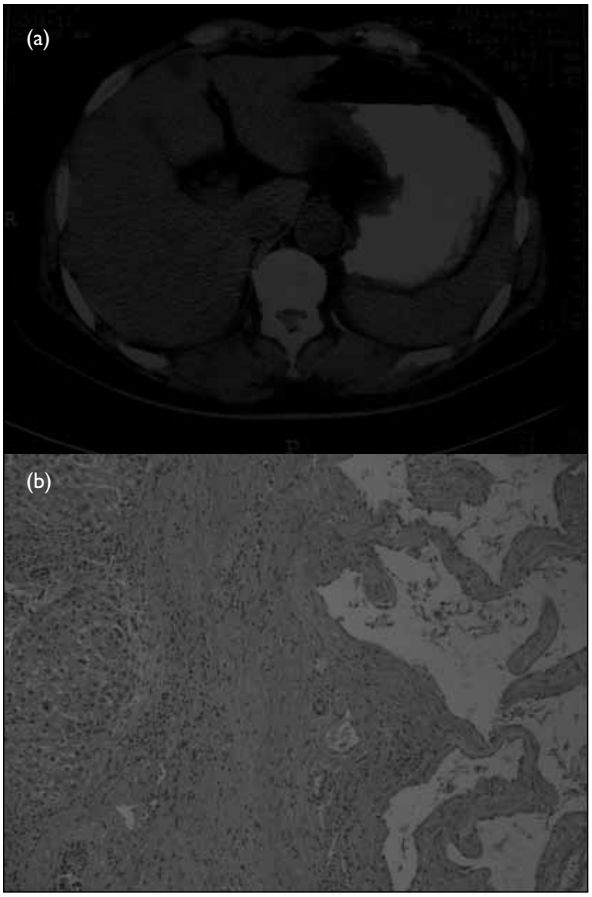

FIG. (a) Computed tomographic scan of the left lobe of the liver showing a hypodense lesion resembling primary hepatocellular carcinoma. (b) Histopathological appearance of cavernous hepatic haemangioma (H&E; original magnification, x 400)

by the fourth postoperative week.

Discussion

This report is of a patient for whom abdominal ultrasound examination and CT scan demonstrated a localised hepatic nodule characteristic of hepatic haemangioma. The highly elevated AFP level gave an impression of HCC, but the intra-operative pathology and postoperative histopathology examinations disclosed a liver haemangioma without evidence of chronic hepatitis B.

Only two patients with hepatic haemangioma with highly elevated AFP level have been reported in the Chinese medical literature from January 1994 to March 2005.[14] There was one man and one woman, aged 35 years and 36 years, respectively. Both patients were diagnosed after a routine health examination and were negative for hepatitis B surface antigen. Their preoperative AFP levels were 784 μg/L and 224 μg/L, respectively, and the AFP returned to normal levels within 4 weeks of surgery to remove the

甲胎蛋白水平速高的成人肝海綿狀血管瘤

一名35歲華籍男性在過去兩個月感到右上腹悶痛，化驗檢查發現病人的血清甲胎蛋白水平為1890 μg/L（參考值範圍：0-20 μg/L），電腦斷層造影顯示病人的左肝葉有一低密度病變。剖腹術中在肝臟的內側節（第三肝段）發現一個直徑約為3.0 x 3.5厘米的深紅色柔軟瘤狀物。進行切除後腫瘤明顯變小，並在切割位置出現血管形的物體。術中病理及術後組織病理均顯示腫瘤為肝海綿狀血管瘤。病人術後4星期的血清甲種胎兒蛋白降至3.5 μg/L。醫生應要留意除了肝癌及內胚竇瘤（又稱卵黃瘤），如本病例的肝血管瘤等罕見腫瘤也可以產生甲胎蛋白。

TABLE. Results of the laboratory tests performed on admission

Laboratory test	Laboratory value	Reference range
Alanine aminotransferase (IU/L)	24	0-50
Aspartate aminotransferase (IU/L)	30	5-60
Fasting plasma glucose (mmol/L)	5.7	3.9-6.1
Urea (mmol/L)	1.7	2.9-8.2
Serum creatinine (μmol/L)	51	35-80
Lactate dehydrogenase (IU/mL)	500	240-480
Gamma-glutamyltransferase (U/L)	26	0-49
Alkaline phosphatase (U/L)	372	50-120
Total protein (g/L)	67	66-88
Albumin (g/L)	36	35-52
Globulin (g/L)	31	25-40
Direct bilirubin (μmol/L)	5	0-5
Indirect bilirubin (μmol/L)	9	3-15
Total cholesterol (mmol/L)	2.6	3.4-5.2
Triglyceride (mmol/L)	0.32	0.45-1.47
Low-density lipoprotein (mmol/L)	1.3	2.1-3.1
High-density lipoprotein (mmol/L)	1.03	0.90-1.68
Sodium (mmol/L)	138	136-142
Potassium (mmol/L)	3.9	3.5-5.0
Calcium (mmol/L)	2.2	2.1-2.6
Haemoglobin (g/L)	115	140-175
Haematocrit (proportion of 1.0)	34.8	0.4-0.5
White blood cells (x 10^9/L)	4.7	4.5-11.0
Platelets (x 10^9/L)	215	150-450
Mean corpuscular volume (fL)	83	89-115
Erythrocyte sedimentation rate (mm/h)	12	0-20
Markers of viral hepatitis		
Hepatitis B surface antigen	+	
Antibody to hepatitis B surface antigen	-	
Antibody to hepatitis C virus	-	
Hepatitis B e antigen	-	
Antibody to hepatitis B e antigen	-	

tumour. The tumours were located in segment VI in one patient and segments V and VIII in the other, and were 6.0 cm and 7.0 cm in diameter, respectively. This finding suggests that the production of AFP originates from the hepatic haemangioma, implying that some haemangiomas may have the function of synthesis and secretion of AFP. For the patient in this report, it is likely that the AFP was produced by a few residual primitive embryonic tissues that remained in the haemangioma during development as the patient was hepatitis B–negative with normal liver function. In a study by Seo et al,[12] serum AFP levels were elevated in all five infants with hepatic haemangioendothelioma, and were closely related to the patients' ages, with the younger patients having higher levels.

If an incidental liver lesion with elevated AFP level is found, it is important to differentiate whether or not the liver lesion is HCC. The treatment choice is based on the nature and extent of disease, such that surgery will depend on the volume of the tumour and the surgical risk.

In conclusion, clinicians should be aware that some rare tumours besides HCC and endodermal sinus tumours (yolk sac tumour), for example, hepatic haemangioma, can produce AFP.

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