CASE REPORT

Large hepatic adenoma in a 21-year-old male

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SUMMARY

Hepatic adenoma is an uncommon benign lesion of the liver that occurs more frequently in women in their third and fourth decades. The female/male ratio is up to 11:1. Hepatic adenomas may be single or multiple occasionally reaching sizes up to 20 cm. They are non-cancerous lesions, however they can become malignant. We present a 21-year-old male patient with no medical history who presented with abdominal pain, a palpable abdominal mass, abnormal liver function tests and a 14 kg weight loss in a 2-year period. A CT scan was performed with a 17 cm tumour compressing intrahepatic bile ducts. The patient underwent a right hepatectomy with no complications. Histopathological analysis of the tumour revealed a hepatic adenoma with central necrosis. The patient is asymptomatic at 1-year follow-up.

BACKGROUND

Hepatic adenoma is a rare benign hepatic neoplasm that is of interest for several reasons¹—it is mostly associated with women using oral contraceptives or in men with steroid use or glycogen storage diseases²; it may be difficult to distinguish from other benign or malignant liver tumours³; large tumours have a marked tendency to haemorrhage; and⁴ they may undergo malignant transformation. Our case is exceedingly rare in two aspects: (1) the patient was a healthy young male with an adenoma not associated with steroids or glycogen storage disease and (2) the patient had a large-sized tumour with no signs of intratumoral or intraperitoneal haemorrhage.

CASE PRESENTATION

A 21-year-old male patient with a right radius fracture at 10 years of age with no other medical history had an incidentally discovered liver tumour by ultrasound in the right liver 2 years prior to referral at our centre. He presented to us with moderate epigastrium pain with no other symptoms. Physical examinations show a 65 kg, 178 cm pale man with hepatomegaly and a right upper quadrant palpable mass with an otherwise unremarkable examination.

INVESTIGATIONS

Initial blood work-up showed a complete blood count with haemoglobin 10.7 g/dL, haematocrit 34.6%, platelet count 527×10^3 and white cell count 8.31×10^3 . Serum chemistry showed glucose 83 mg/dL, blood urea nitrogen 5.7 mg/dL and creatinine 0.9 mg/dL. Serum electrolytes showed a Na^+ 138 mEq/L, K^+ 4.2 mEq/L and Cl^- 101 mEq/L. Liver function test were as follows: aspartate

aminotransferase (AST) 18 IU/L, alanine aminotransferase (ALT) 101 IU/L, alkaline phosphatase (ALP) 145 IU/L, lactate dehydrogenase 543 IU/L, total bilirubin 1.6 mg/dL, direct bilirubin 1.1 mg/dL and serum albumin 4 mg/dL. Prothrombin time was 12.8 s and international normalised ratio (INR) 0.91. Tumour markers were alpha fetoprotein 3.3 ng/mL and carcinoembryonic antigen 0.7 ng/mL. Blood type O Rh negative.

An abdominal MRI was performed showing a T2 hyperintense $177 \times 142 \times 173$ mm regularly edge well-defined tumour with extrinsic compression and entrapment of intrahepatic bile ducts in the right liver with the rest of the liver and surrounding structures of normal appearance (figure 1). A percutaneous liver biopsy of the lesion showed mild hepatitis with focal intracytoplasmic cholestasis and regeneration.

DIFFERENTIAL DIAGNOSIS

- Focal nodular hyperplasia
- Liver cell haemangioma
- Well-differentiated hepatocellular carcinoma

TREATMENT

The patient underwent a right hepatectomy ($23 \times 16 \times 14$ cm; figure 2) and right chest tube insertion secondary to right hemidiaphragm perforation during hepatectomy. Patient developed grade B posthepatectomy liver failure¹ managed in the intensive care unit (6 days) and was discharged on postoperative day 16 with no complications. Histopathological analysis showed a 15×12 cm hepatic adenoma described as a solid lesion bounded by a fibrous pseudocapsule (figure 3A), consisting of hepatic cords, arranged in rows and veins simulating central venous with occasionally enlarged sinusoidal spaces in between these hepatic

Figure 1 MRI showing a 17 cm T2 hyperintense tumour in right lobe of the liver.

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Figure 2 Right hepatectomy specimen (23×16×14 cm) with hepatic adenoma.

cords (figure 3B). Tumour cells are hepatocytes without atypia and occasional lipid vacuoles and bile pigment, distributed in a pseudoglandular appearance structures (figure 4A). There was also identified isolated 'naked portal areas' (figure 4B).

OUTCOME AND FOLLOW-UP

The patient had regular follow-up clinic visits. He remains asymptomatic 12 months following surgery and had returned to study at a local university. His last complete blood count at 1 year posthepatectomy showed a haemoglobin 12.7 mg/dL, haematocrit 37.6%, platelet count 295×10^3 and white cell

count 8.1×10^3 . Liver function test were as follows: AST 21 IU/L, ALT 22 IU/L, ALP 108 IU/L, total bilirubin 0.5 mg/dL, direct bilirubin 0.2 mg/dL and serum albumin 4.7 mg/dL. Prothrombin time was 14.6 s and INR 1.18. An abdominal CT has been reported unremarkable with liver regeneration (figure 5).

DISCUSSION

Hepatic adenoma is a rare benign liver neoplasm that occurs more frequently in young woman taking oral contraceptives. The risk of developing a hepatic adenoma increases with the duration of contraceptive use.^{2–3} This tumour can also occur in men taking anabolic steroids⁴ or may be associated with metabolic diseases, including type 1 glycogen storage disease and iron overload.⁵ The female/male ratio is up to 11:1.⁵ However, it is now clear that hepatic adenomas may also affect men without these risk factors.

Hepatic adenomas may be single or multiple and they may occasionally reach sizes larger than 20 cm, thus clinical presentation varies widely. Pain in the upper abdomen (epigastric or right upper quadrant) is common (25–50% of patients). Liver enzyme changes (elevation of ALP in 23% of cases) and bilirubin elevation may occur in association with expansion of the neoplasm.^{4–8} Large hepatic adenomas (>5 cm) have the potential to bleed and rupture spontaneously and present as acute abdomen in 8–20% of the cases. Spontaneous rupture occurs more often in men, especially steroid users and is a life-threatening condition.^{9–10}

The ultrasonographic sensitivity to diagnose an adenoma is of 30%. On a CT scan, adenomas are seen as a discrete hypodense lesion that shows arterial-phase enhancement. They may show intralesional hypodensity or hyperdensity, depending on the presence or absence of necrosis or haemorrhage. On MRI, adenomas are hypointense to hyperintense T1-weighted images.⁵

The primary diagnostic consideration is to differentiate adenoma from hepatocellular carcinoma and from focal nodular hyperplasia. Malignant transformation of hepatic adenoma is rare but not minimal (4–10% of resected adenomas have hepatocellular carcinomas within). In most cases, adenomas can be

Figure 3 Histologically, it is a solid lesion with fibrous pseudocapsule (A) constituted by hepatocytes arranged in cords with veins in between that resemble central veins and occasionally dilated sinusoid spaces (B).

Figure 4 Tumour cells belong to hepatocytes without atypia and occasional lipid vacuoles with biliary pigment, forming pseudoglandular structures (A) with 'naked portal spaces' (B).

distinguished from focal nodular hyperplasia based on its characteristic pathological features. Adenomas are large, circumscribed, encapsulated lesions with areas of necrosis and haemorrhage visible on the cut surface. The histological pattern is monotonous with regular proliferation of normal appearing hepatocytes often in trabeculae with compressed sinusoids in between with absent reticular fibres and bile ducts.^{8 11}

Surgical resection has been recommended for adenomas with a diameter >5 cm in symptomatic patients. For smaller and asymptomatic adenomas, an expectant approach with discontinuation of contraceptives in women is advocated. The main surgical procedure for hepatic adenomas should be anatomic or segmental resection.^{11–13} In case of a haemorrhage and a surgical emergency, the lesion must be handled as hepatic trauma. When multiple adenomas or giant adenomas are not feasible to

excise, total hepatic vascular isolation or liver transplantation has been successfully performed.¹⁴

Learning points

- ▶ Hepatic adenoma is difficult to diagnose preoperatively by clinical, radiological and histological means.
- ▶ Hepatic adenoma carries a risk of malignant transformation.
- ▶ Surgery is indicated in large (>5 cm) hepatic adenoma due to potential catastrophic haemorrhage.
- ▶ Surgery is indicated in symptomatic hepatic adenoma (compression mostly and haemorrhage).

Contributors GMM: literature search, manuscript review; HEDLS: literature search; PGP: histopathological analysis and literature search.

Competing interests None.

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Figure 5 Abdominal CT on 9 months following surgery with liver regeneration.

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