

## CASE REPORT

# Kasaback-Merritt Syndrome Treated by Transarterial Embolisation of Giant Cavernous Haemangioma

EPY Fung, WH Luk, TK Loke, JCS Chan

*Department of Radiology and Organ Imaging, United Christian Hospital, Hong Kong*

### ABSTRACT

*Cavernous haemangiomas of the liver are the most common benign tumours of the liver. With modern imaging modalities, haemangiomas are detected more frequently. This report describes a patient with a giant cavernous haemangioma with consumptive coagulopathy that was successfully treated by transarterial embolisation.*

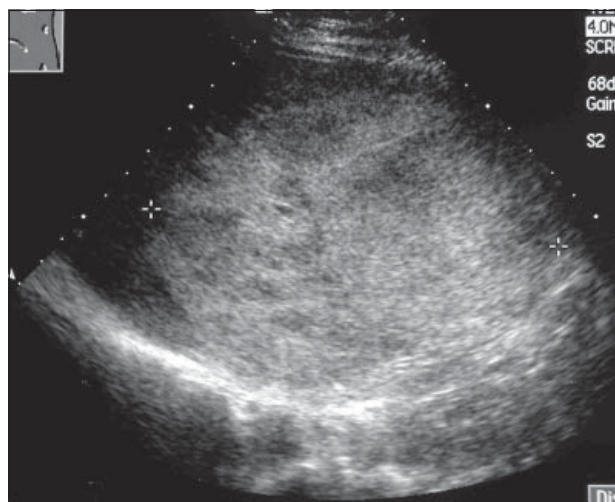
*Key Words: Cavernous hemangioma, Embolization, Treatment*

### INTRODUCTION

Cavernous haemangiomas of the liver are the most common benign tumours of the liver and are found in approximately 2% of liver autopsies. With modern imaging modalities, haemangiomas are detected more frequently.<sup>1</sup> Small haemangiomas are normally clinically silent, and no treatment is needed. However, it is not uncommon for those larger than 4 cm, so called 'giant' haemangiomas to be associated with complications. Patients may present with abdominal distension and pain, rupture of the haemangioma with haemoperitoneum, or disorders of coagulation.<sup>2</sup>

### CASE REPORT

This report is of a previously healthy 49-year-old women who presented to the United Christian Hospital with insidious onset of a right upper quadrant abdominal mass and pain for 1 year. Physical examination was unremarkable apart from hepatomegaly. Liver function test was normal but she had a low platelet count of  $130 \times 10^9/L$  (normal range, 150 to  $450 \times 10^9/L$ ). Ultrasound scan revealed a 13 cm heterogeneous hyperechoic lesion in the left lobe of the liver (Figure 1). Subsequent computed tomography (CT) scan showed a hypodense mass with peripheral nodular enhancement



**Figure 1.** Ultrasound revealed a large heterogeneous hyperechoic lesion in the left lobe of the liver.

after contrast administration and gradual centripetal filling on delayed phases. The CT features were typical for a giant haemangioma (Figures 2a and 2b).

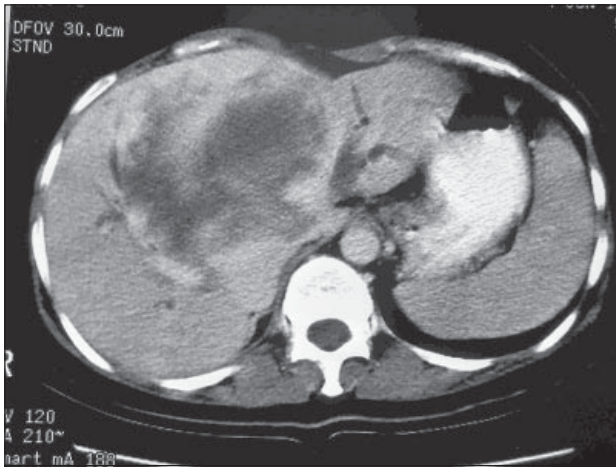
Oral analgesia was given to relieve the abdominal pain. However, the right upper abdominal pain became more intense and was not controlled by oral medication. CT scan of the abdomen showed no evidence of tumour rupture or interval change in size. On this occasion, the patient was found to have deranged liver function in addition to the low platelet count. The platelet count dropped from  $130 \times 10^9/L$  in May 1999 to  $99 \times 10^9/L$  in September 2002, although clinically the patient did not have any bleeding tendency. In view of the consumptive coagulopathy and the increasing abdominal pain,

*Correspondence: Dr WH Luk, Department of Radiology and Organ Imaging, United Christian Hospital, 130, Hip Wo Street, Kwun Tong, Hong Kong.*

*Tel: (852) 2379 4159; Fax: (852) 2379 4139;*

*E-mail: lukwinghang@netvigator.com*

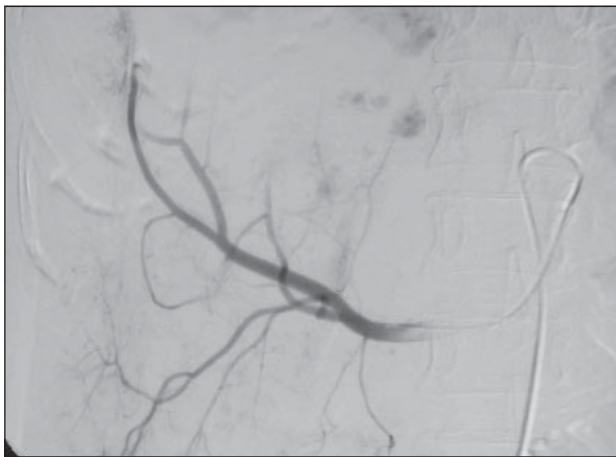
Submitted: 31 March 2003; Accepted: 18 June 2003.



**Figure 2.** Contrast computed tomography scan was typical for a giant cavernous haemangioma.



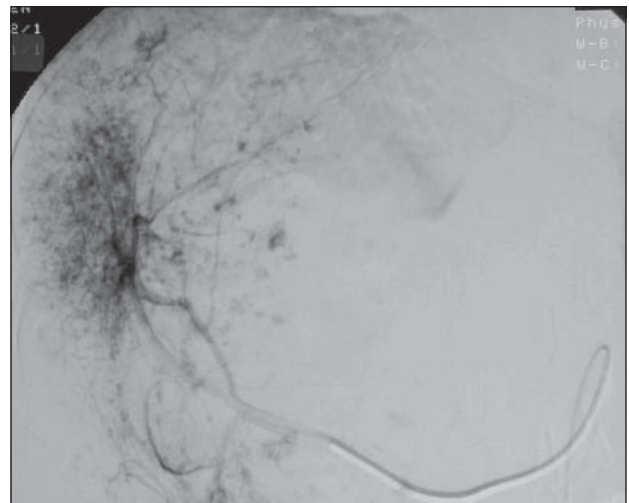
**Figure 4.** There was no significant change in size and vascularity of the haemangioma after embolisation.



**Figure 3.** The right hepatic artery was embolised with Ivalon.

transarterial embolisation was offered as treatment. Selective embolisation of the supplying branches of the right hepatic artery was performed with PolyVinyl-alcohol (PVA) particles (350 microns). The left hepatic artery was not embolised on the first occasion as the problem of consumption coagulopathy was expected to improve with embolisation of the right hepatic artery (Figure 3).

Follow up CT of the abdomen was done 1 month after the procedure but there was no significant change in size and vascularity of the haemangioma (Figure 4). The platelet count remained low at  $103 \times 10^9/L$ . Since the low platelet count did not improve with single right hepatic artery embolisation, a decision was made to embolise both hepatic arteries. Despite the presence of previous PVA particles, there was still significant contrast pooling in the right hepatic artery. On the second procedure (Figure 5), further embolisation was performed, initially using Gelfoam to reduce the blood



**Figure 5.** Significant pooling of contrast in the right hepatic artery after the first embolisation.

supply and, subsequently, coil placement in the right hepatic artery, which was completely and permanently occluded (Figure 6). The left hepatic artery was then embolised with Embosphere 500 microns (PVA was not available at that time). The post-embolisation angiogram showed small residual nodular vascularity supplied by the proximal tributaries of the left hepatic artery. No immediate complications were observed. There was no post-embolisation syndrome such as fever or leucocytosis. Only mild abdominal pain was encountered. The platelet count had returned to normal at  $151 \times 10^9/L$  at the follow up visit and the abdominal pain was relieved after the procedure. The patient has been asymptomatic for 6 months to date.

## DISCUSSION

Kasaback-Merritt syndrome is a type of haemorrhagic diathesis due to platelet sequestration by a tumour, and



**Figure 6.** Complete occlusion of right hepatic artery by coil placement.

giant liver haemangioma is one of the causes. The management of giant liver haemangioma remains controversial.<sup>3,4</sup> Interventions should only be performed for symptomatic progressively growing haemangiomas, and those at high risk for bleeding.<sup>5</sup> Treatment options have included enucleation, hepatectomy, hepatic artery ligation, and transarterial embolisation. Some centres have demonstrated the usefulness of preoperative embolisation of the haemangioma in order to reduce the amount of blood loss.<sup>6,7</sup>

Percutaneous embolisation avoids the risk of operative intervention, extended hospitalisation, and postoperative recuperation. The identification of aberrant hepatic arteries and their anatomical location in relation to the proposed site for flow interruption is important. The origin of the cystic artery should also be noted to prevent inadvertent embolisation and subsequent necrosis of the gallbladder. Adequate portal vein flow must be confirmed before the interruption of the artery to avoid hepatic necrosis.<sup>8</sup> The left, right, or both hepatic arteries can be occluded. However, the common hepatic artery is usually spared to preserve the gastroduodenal flow and to give vascular access for future embolisation should recanalisation of the occluded vessels occur or gastroduodenal collateral vessels develop. In this patient, the platelet count failed to return to normal after embolisation of the right hepatic artery. As giant haemangioma usually has a blood supply from both hepatic arteries, both should be embolised for a promising outcome.

The choice of embolisation agent depends on the initial angiographic appearance and the experience of the radiologists. A preliminary peripheral embolisation for vascular interstices within the haemangioma can be done with gelfoam particles or PVA. Some authors prefer the use of Ivalon as it is a permanent occlusive agent with established biocompatibility.<sup>9</sup> Once the peripheral circulation is occluded, principal arteries can be embolised with steel coils. For patients with arteriovenous or arterioportal shunting, particulate material should not be used to prevent passage into the systemic portal circulation. Postembolisation side effects of fever, pain, and leukocytosis are common. Serious complications are rare and include infection, hepatic abscess, and sepsis; the risk increases with the size of the lesion. While the size of the tumour does not usually change significantly,<sup>7</sup> there is relief of symptoms of abdominal pain and coagulopathy after successful embolisation, which is more important.

In conclusion, this patient underwent successful embolisation of a hepatic haemangioma complicating Kasabach-Merritt syndrome. Transarterial embolisation is an effective therapeutic option for the management of symptomatic haemangioma as an alternative to major surgery.

## REFERENCES

1. Lise M, Feltrin G, Da Pian PP, et al. Giant cavernous hemangiomas: diagnosis and surgical strategies. *World J Surg* 1992; 16:516-520.
2. Trastek VF, van Heerden JA, Sheedi PF II, Adson MA. Cavernous hemangiomas of the liver: resect or observe? *Am J Surg* 1983;145: 49-53.
3. Bornman PC, Terblanche J, Blumgart RL, Harries Jones EP, Pickard H, Kalvaria I. Giant hepatic hemangiomas: diagnostic and therapeutic dilemmas. *Surgery* 1987;101:445-449.
4. Pietrabissa A, Giulianotti P, Campatelli A, et al. Management and follow-up of 78 giant haemangiomas of the liver. *Br J Surg* 1996;83:915-918.
5. Srivastava DN, Gandhi D, Seith A, Pande GK, Sahni P. Transcatheter arterial embolization in the treatment of symptomatic cavernous hemangiomas of the liver: a prospective study. *Abdom Imaging* 2001;26:510-514.
6. Suzuki H, Nimura Y, Kamiya J, et al. Preoperative transcatheter arterial embolization for giant cavernous hemangioma of the liver with consumption coagulopathy. *Am J Gastroenterol* 1997; 92:688-691.
7. Reading NG, Forbes A, Nunnerley HB, Williams R. Hepatic haemangioma: a critical review of diagnosis and management. *Quarterly J Med* 1988;67:431-445.
8. Deutsch GS, Yeh KA, Bates WB III, Tannehill WB. Embolization for management of hepatic hemangiomas. *Am J Surg* 2001; 67:159-164.
9. Stanley P, Grinnell VS, Stanton RE, Williams KO, Shore NA. Therapeutic embolization of infantile hepatic hemangioma with Polyvinyl Alcohol. *AJR Am J Roentgenol* 1983;141:1047-1051.