Hypergiant Hepatic Hemangiomas: Case Series

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Abstract Hemangiomas are the most common benign tumors of the liver. Hepatic hemangiomas originate from the proliferation of vascular endothelial cells, and enlarge by ectasia rather than hyperplasia. They are very variable in size and are classified accordingly and their management in the larger variety is debatable. Hypergiant hepatic hemangiomas are defined as those which are more than 10 cm in size. These are fairly uncommon in clinical practice. The treatment spectrum varies from masterly inactivity to resection depending upon a number of factors. We report a series of 10 cases of hyper giant hepatic hemangiomas studied and reviewed over a period of 5 years. These were evaluated with respect to their age, gender, clinical presentation, investigation findings, treatment offered, and final outcome. Five were treated with resection, four with enucleation, and one was kept under observation. All operated patients had an uneventful post-operative recovery and the one managed conservatively was asymptomatic on follow-up.

Keywords Liver hemangioma · Large liver hemangioma · Giant liver hemangioma · Hypergiant liver hemangioma · Liver resection · Enucleation of liver hemangioma

Introduction

Hepatic hemangiomas are the most common benign hepatic tumors. The incidence of hemangiomas is highest in the third to fifth decade of life and is more common in women. Exposure to high levels of estrogen and progesterone, occurring with multiparity, pregnancy, and oral contraceptive use, are reasons for the increased incidence in women. Studies have revealed that estrogen augments endothelial cell proliferation and organization into capillary-like structures [1]. Wang et al. demonstrated a link between interleukin-17 and hepatic hemangiomas. Patients with hepatic hemangiomas had a significant increase in IL-17 mRNA compared to normal individuals, and it is concluded that IL-17 may mediate angiogenesis [2]. Hemangiomas are classified as small (<4 cm), giant (>4 cm), and hypergiant (>10 cm) and are often solitary focal lesions, but in 40 % of cases, they may be multiple.

Methods

Ten cases of hypergiant liver hemangiomas were reviewed from 2007 to 2012. Inclusion criterion was at least one lesion greater than 10 cm in size. Diagnosis was established on the basis of integrated findings of USG and CECT. MRI was done in one case. Patients were evaluated with respect to age, sex, symptoms, size, number, and location. Indications for operation were intractable abdominal pain and uncertainty of diagnosis. Type of surgery was based on size and location of the hemangioma.

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Table 1 Clinical presentation, location, size, and treatment done

Case No.	Age	Sex	Clinical presentation	Liver segments involved	Size (max diameter in cm)	Treatment
1	51	F	Abdominal pain, lump	II, III	14	Enucleation
2	45	F	Abdominal lump	IV, V	12	Enucleation
3	42	F	Abdominal pain, lump	IV	14	Enucleation (failed attempt at embolization)
4	42	F	Epigastric lump, abdominal pain following liver biopsy	IV, V, VI	14	Enucleation
5	50	M	Incidental finding in a case of carcinoma stomach	II, III	12	Left lateral segmentectomy with radical subtotal gastrectomy
6	40	F	Abdominal pain, lump	II, III	20	Left lateral segmentectomy
7	45	F	Abdominal pain, lump	II, III, IV	22	Left hemi-hepatectomy
8	67	M	Epigastric pain, lump	II, III	10	Left hemi-hepatectomy (histopath showed hepatocellular carcinoma)
9	55	F	Abdominal pain, lump	Right hemi-liver, segment IV, and satellite lesions in segment II and III	35	Right hepatic artery ligation and right partial non-anatomical liver resection
10	40	F	Mild epigastric pain and dyspepsia	VI	12	Observation

Results

The age, sex, clinical presentation, and treatment offered are outlined in Table 1. All the operated patients had an uneventful recovery and were asymptomatic on a median follow-up of 2 years. Enucleation was done in four patients. Left lateral segmentectomy was done in two patients (Fig 1). A left hemi-hepatectomy was done in

the remaining two patients. This included one patient with a large lesion involving segments II, III, and IV, and another patient with a lesion in segments II and III but with a diagnostic uncertainty, in whom the final histopathology was hepatocellular carcinoma. One patient with lesion involving all segments of the right hemi-liver, as well as segment IV and satellite lesions in segments II and III underwent a right hepatic artery

Fig. 1 Cases where resection (left lateral) segmentectomy was done (composite picture)

ligation with partial non-anatomic liver resection. One patient was conserved and kept under observation as he was essentially asymptomatic.

Discussion

Hepatic hemangiomas are often diagnosed incidentally on imaging studies of the abdomen and during surgery. When symptomatic, they present with intermittent right upper quadrant pain which may be due to capsular distension, thrombosis, infarction, and hemorrhage into the lesion. Large hemangiomas can cause compression of adjacent structures, leading to obstructive jaundice and gastric outlet obstruction. Intra-tumoral hemorrhage can occur due to an inadvertent invasive diagnostic procedure like biopsy or trauma which can result in hemoperitoneum or hemobilia. Rupture is also known to occur spontaneously, especially in giant variety. Disseminated intravascular coagulopathy can also occur (Kasabach-Merritt syndrome) [3].

The main imaging modalities are abdominal ultrasonography (USG), contrast-enhanced computed tomography (CECT), magnetic resonance imaging (MRI), hepatic arteriography, digital subtraction angiography, and nuclear medicine studies [4]. MRI is highly specific and sensitive. Hemangiomas have low signal intensity on T1-weighted images and high signal intensity on T2-weighted images with a characteristic “light bulb” sign. When gadolinium is used as an intravenous contrast agent, hemangiomas enhance in a fashion similar to that seen on dynamic CT. The sensitivity for detection using this is above 90 % [5].

Surgery is indicated for persistent symptoms of a progressively growing tumor, uncertain diagnosis, and impending complications, and the options can be enucleation or resection. Enucleation helps in preserving the liver parenchyma, resulting in fewer complications while resection is indicated when the diagnosis is uncertain. Transarterial embolization (TAE) and radiofrequency ablation (RFA) are also described [6].

It has been proposed that asymptomatic patients with hemangiomas less than 5 cm require no intervention therapy. Iwatsuki et al. emphasized that large hemangiomas (10 cm in diameter) may rupture or bleed and should be resected. However, in a recent study, Schnelldorfer et al. reported that clinical observation is preferred in most patients even with giant hemangioma [7]. Size of the lesion is not the criterion for surgery.

The size and location of the lesion influence the decision to perform either a segmental resection or an enucleation. However, for larger lesions, enucleation would be more difficult resulting in greater blood loss and lobar resection maybe the preferred option.

The use of preoperative arterial embolization in patients with consumption coagulopathy was related to intravascular coagulation in the hemangioma. RFA using both percutaneous and laparoscopic methods have been performed successfully to improve abdominal pain in small numbers with symptomatic hepatic hemangioma. Hepatic irradiation has been reported to produce complete regression of hepatic hemangioma with minimal morbidity. Liver transplantation is described as a rescue treatment in children with hepatic vascular malformations leading to hemodynamic insufficiency, and when conventional therapy is unsuccessful and multiorgan failure develops [8].

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