MANAGEMENT OF GIANT HEPATIC HEMANGIOMA IN ATYPICAL LOCALIZATION; REPORT OF A CASE AND LITERATURE REVIEW

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Hemangiomas are the most common benign primary hepatic neoplasms, often being incidentally discovered. In most of the cases they are small and asymptomatic. It is widely accepted that clinical intervention is indicated only for symptomatic hemangiomas.

We present a case of an asymptomatic giant hemangioma managed by enucleation due to its atypical localization. The hemangioma, originally located in segment 5, was now described in Computer Tomography (CT) Imaging as separating the gallbladder from the liver parenchyma. A careful evaluation of images revealed proximity to the portal vein (PV), right hepatic artery (RHA), right hepatic duct (RHD) and right branch of the portal vein (RBPV). Thus, in the case of an emergent operation, surgical maneuvers in the area of the altered hepatic anatomy and proximity to the hemangioma itself, would in fact increase the risk endangering the patient’s life. After patient’s consent, a surgical enucleation en block with the gall-bladder was performed. It is of great importance that specifically selected, asymptomatic patients diagnosed with a giant hemangioma, with the above mentioned or similar localization should be considered for surgical treatment.

Key words: giant hemangioma, liver benign neoplasm, cholecystectomy, tumor enucleation, liver surgery

Hemangiomas are the most common benign tumors of the liver, with prevalence ranged from 3% to 20% based upon autopsy series (1, 2, 3). Most hepatic hemangiomas are found in women within the fourth and fifth decade of life, however they can occur at any age. The majority of hepatic hemangiomas is asymptomatic and is diagnosed while screening for other causes. Current evidence indicates that hemangiomas have no malignant potential. Giant hemangiomas are defined as tumors larger than 4 cm and symptoms rarely appear unless the tumor exceeds the size of 4 cm (4, 5).

The symptoms of giant hemangiomas range from abdominal fullness or pain to even hemoperitoneum due to tumor rupture (5). Treatment options range from close observation through interferon administration, to liver resection or transplantation. Conservative management, which limits to observation, is preferred for vast majority of patients. Operative treatment of liver hemangioma is indicated in symptomatic cases or in a controversial diagnosis where malignancy cannot be fully eliminated (6, 7).

We report a case of a cavernous hepatic hemangioma of unique and difficult localization treated with surgical enucleation in an asymptomatic patient.

CASE REPORT

A 56-year-old male incidentally diagnosed in 2000 upon abdomen ultrasound and Computer Tomography (CT) with a hepatic hemangioma. The lesion was described as well-defined, lobulated with peripheral enhancement 5 cm x 3.5 cm, located in segment 5 of the liver. No symptoms occurred at the time of diagnosis. On subsequent presentations slow growth of the tumor was noted. Latest CT-scan
revealed that due to its localization, the hemangioma separated the gall-bladder from the liver parenchyma (fig. 1). Thus, the actual size of the tumor evaluated on ultrasound and CT-scan was 10.5 cm x 9.7 cm x 10.2 cm – which was double the size at the moment of diagnosis. The hemangioma's size and mass as well as localization influenced the adjacent hilum structures, such as the right hepatic artery and portal vein (fig. 2). This meant, that in the case of an emergent operation, the altered anatomy and proximity to the vascular mass, in fact would endanger the patient’s life. The threat of uncontrolled hemorrhage during surgery carried out without proper preparation had to be taken under consideration for future treatment. The patient’s consent for the surgical procedure was obtained. On admission, October 2013, the patient’s laboratory values and serum tumor markers were all within normal limits. An enucleation en block with a cholecystectomy was performed by dissecting the lesion from the surrounding hepatic parenchyma along the plane of the tumor capsule (fig. 3 and 4). While dissecting the hemangioma from the duodenohepatic ligament it was first separated from the portal vein trunk. The superficial layer adhering to liver parenchyma had to be dissected from the right branch of the portal vein, right hepatic artery and right hepatic duct. Blood supply to the hemangioma originated directly from right hepatic artery. Neither Pringle’s maneuver nor selective right hepatic artery inflow control was applied during enucleation. After the procedure a cholangiography check through the cystic duct was performed due to the proximity of the right bile duct.

The removed specimen was examined histopathologically, which gave the result of “hemangioma cavernosum” (fig. 5). The post-operative period was complicated by moderate bile leakage successfully managed by conservative treatment with percutaneous drainage. Patient was discharged home on day 11 after the surgery. Two months post-op the patient remains well and asymptomatic.

Fig. 1. CT-scan of the lesion one month prior the operation. HA – hemangioma, GB – gall-bladder

Fig. 2. CT-scan one month prior operation, liver hemangioma localized near right branch of portal vein. HA – hemangioma, PV – right branch of portal vein

Fig. 3. Intraoperative image of the lesion. CA – cystic artery, HA – hemangioma, GB – gall-bladder, RHA – right hepatic artery

Fig. 4. Hemangioma bed after the enucleation. CA – ligated cystic artery, RPV – right branch of portal vein, HD – hepatic duct, RHA – right hepatic artery, OC – oxidized cellulose
Management of giant hepatic hemangioma in atypical localization; report of a case and literature review

DISCUSSION

Advances in imaging technology, as well as the widespread application of diagnostic imaging, has resulted in the more frequent detection of hepatic hemangiomas (8). The majority of them are small (<4 cm), asymptomatic and require no intervention. Hemangiomas exceeding 4 cm are called “giant” and symptoms appear with lesion growth. Symptoms presented by giant hemangioma are abdominal fullness or pain, hemorrhage within the hemangioma or within abdominal cavity with associated hemodynamic compromise, jaundice due to compression of the biliary tree, cardiac failure from massive arterial-venous shunting or very rare consumptive coagulopathy (Kasabach-Merritt Syndrome). For patients with giant hemangioma or one causing symptoms several treatment options are established. Radiological procedures (transcatheter arterial embolization, radiofrequency ablation) and drug administration (corticosteroids, interferon) are efficient methods with considerable limitations (9-12).

Liver transplantation for both deceased and living donors, remains an important method of treatment for a very limited group of patients namely those with a ruptured hemangioma (an extremely rare event, with incidence of 1% to 4% (13), Kasabach-Merritt Syndrome (also very rare complication, first described in 1940 in a 2-month-old child (14) and non-resectable symptomatic giant hemangioma (15, 16). The most preferred option for symptomatic giant hepatic hemangiomas remains surgery.

It is widely accepted that even giant, asymptomatic hemangiomas are managed by observation and no clinical intervention is taken unless symptoms occur (17). We present a case of a giant hemangioma (>4 cm at time of diagnosis) managed conservatively by observation for over a decade. During that time the tumor doubled its size but still no abdominal symptoms appeared. The conclusion from numerous reports (17, 18, 19) states that in asymptomatic patients even in the presence of a giant hemangioma, conservative management is reasonable. In the presented case, an unique and difficult localization of the hemangioma provoked the decision of operative treatment. To the author’s knowledge it is the first report on hepatic hemangioma of such localization. A giant hemangioma separating the gallbladder from liver parenchyma and affecting the adjacent hilum structures, such as the right hepatic artery and portal vein was indeed a dangerous situation. The threat, however was not in the risk of rupture, which is very rare (5, 13), but in the eventual future necessity of surgery i.e. cholecystectomy. The effect of the conglomerate altering the anatomy and complete adhesion of the gallbladder wall to the hemangioma could cause bleeding from the gall-bladder-bed, which overall may be very difficult to control. An enucleation of the tumor along with a cholecystectomy carried out by an experienced hepatobiliary team of surgeons seemed optimal strategy for this patient. Although enucleation is a safe procedure with 4% morbidity (20), available studies refer to symptomatic patients with indisputable indication for surgery.

In conclusion, indications for surgical treatment proposed by the author’s report and available in literature are giant hemangiomas with progressive abdominal pain, increase in size and difficulties in excluding a malignancy. Nevertheless, in the strategy on hemangioma management, every possible circumstance should always be taken under consideration. Thus, in unique situations, operating on asymptomatic patients is justified, as presented in the report.

REFERENCES
