รายงานผู้ป่วย

Solitary giant sessile polypoid hemangioma of the liver

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Shuangshoti S. Solitary giant sessile polypoid hemangioma of the liver. Chula Med J 1993 Feb; 37(2): 127-132

A case is reported of solitary giant sessile polypoid hemangioma of the liver in a 53-year-old woman who had also chronic rheumatic endocarditis with mitral stenosis. The patient died from bleeding tendency in an opened cadiac surgery with replacement of the deformed mitral leaflets. At postmortem examination, a sessile polypoid hemangioma, $10.0 \times 10.5 \times 11.0$ cm, was found attached to the superior border of the left lobe of the liver. A hepatic hemangioma of more than 4 cm in diameter is usually symptomatic. The current lesion, however, was regarded as symptomless because the patient had no symptom or sign throughout her life although the hemangioma was gigantic. Bleeding tendency of the patient was thought to be unrelated to the hemangioma but probably was associated with severe centrilobular necrosis of the liver cells resulting in prolonged prothrombin time and hemorrhagic diathesis.

Key words: Hemangioma, Liver, Rheumatic heart disease, Bleeding tendency.

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Received for publication. May 27, 1992.

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สำรวย ช่วงโชติ. โพลีปอยด์ ฮีแมงจิโอมา เดี่ยวขนาดยักษ์ของดับ. จุฬาลงกรณ์เวชสาร 2586 กุมภาพันธ์; 37(2): 127-132

ผู้เขียนได้รายงานผู้ป่วย 1 ราย เป็นหญิงไทยอายุ 53 ปี เข้ารับการผ่าตัดเปลี่ยนลิ้นไมทรัลของหัวใจ
ที่อักเสบเรื้อรังจากโรครูห์มาติก แล้วตายหลังผ่าตัดได้ 5 วัน ตรวจศพนอกจากจะพบโรครูห์มาติกแล้วยังพบ
ฮีแมงจิโอมาขนาดยักษ์ วัดได้ 10.0 x 10.5 x 11.0 ขม. ยื่นจากขอบบนของกลีบข้ายของตับ เชื่อว่าเป็น
ฮีแมงจิโอมาที่ไม่มีอาการ เพราะผู้ป่วยไม่เคยมีอาการและอาการแสดงใด ๆ ที่เกี่ยวกับเนื้องอกก้อนนี้เลย
จากการทบทวนวารสารพบว่าฮีแมงจิโอมาเป็นเนื้องอกไม่ร้ายที่พบบ่อยที่สุดของตับ พบราวร้อยละ 1 ของ
ผู้ป่วยที่ถูกปิดหน้าท้องและจากการตรวจศพ พวกที่เส้นผ่าศูนย์กลางน้อยกว่า 4 ขม. มักไม่มีอาการ พวกที่ขนาดเกิน
4 ขม.มักก่ออาการ ส่วนใหญ่จะเป็นเรื่องของการพบมีก้อนในท้องหรือก้อนกดทับอวัยวะข้างเคียง ฮีแมงจิโอมา
ส่วนมากของตับมักเป็นก้อนเดี่ยว แต่อาจพบหลายก้อนก็ได้ และอาจพบร่วมไปกับฮีแมงจิโอมา ณ ที่อื่น ๆ บางราย
อาจมีเลือดออกง่ายร่วมไปกับเกร็ดเลือดต่ำ เข้าใจว่าเป็นเพราะเนื้องอกทำลายเกร็ดเลือด ถ้าทำลายแองจิโอมา
ให้หมดไปอาการเลือดออกง่ายจะหายไป และระคับเกร็ดเลือดจะกลับเป็นปกติได้ ผู้ป่วยที่นำมารายงานนี้ก็มี
เลือดออกง่ายเหมือนกัน แต่อาการในรายนี้น่าจะเกี่ยวข้องกับการเน่าตายของเขลล์ตับจำนวนมาก ทำให้เกิด
การผิดปกติในการแข็งตัวเป็นลิ่มของเลือด

Metastatic tumors are common in the liver. Primary malignant neoplasms of the liver are less frequent than secondary neoplasms. Benign hepatic tumors are rarer than primary malignant neoplasms. Hemangioma, a developmentally malformed vascular mass, is the most common benign tumor of the liver. The prevalence has been estimated to range from 0.24% to 20% or with an average figure of 1% in routine autopsies. (1-5) Most hepatic angiomas are accidental at laparotomy or postmortem examination at all ages and in both sexes although they are most frequent in adults, especially in women. A female to male ratio is 4:1.⁽²⁾ They are generally less than 2 cm in diameter, symptomless, commonly situated beneath the hepatic capsule, sometimes multiple, and may combine in occurrence with extrahepatic hemangioma.(2,6-11)The purpose of this communication is to report a solitary giant sessile polypoid hemangioma of the liver which is regarded as symptomless.

Case Report

A 53-year-old woman had had a closed mitral valvulotomy because of an old rheumatic endocarditis with mitral stenosis and dyspnea when she was 46-year-old. After that procedure, she was daily on a 0.25 mg tablet of digoxin and was well. Progressive dyspnea, however, returned when she was 52-year-old. The patient, then, was hospitalized for further surgical treatment.

Body temperature was 37°C, pulse rate 75 beats/min, respiratory rate 29/min, and blood pressure 90/70 mm Hg. She had good consciousness. The cardiac apex beat was in the left sixth intercostal space and outside the midclavicular line, and pansystolic murmur was heard in this area. The lungs were clear. The liver and spleen were not palpable. There were cardiomegaly and mild pulmonary congestion in a chest x-ray. Hypertrophy of the left cardiac ventricle was detected in an electrocardiography.

Hemoglobin was 12 gm/100 ml. A leucocyte count disclosed 11,300 cells/mm³ with 66% neutrophils, 2% eosinophils, 30% lymphocytes, and 2% monocytes. Blood glucose was 91 mg/100 ml, BUN 13 mg/100 ml, creatinine 0.9 mg/100 ml, alkaline

phosphatase 26.5 IU, SGOT 52 IU, SGPT 34 IU, albumin 3.75 gm/100 ml, globulin 3.35 gm/100 ml, prothrombin time 9.5 sec (control 12.0 sec), cabon dioxide 15 to 19 mEq/L, sodium 136 mEq/L, potassium 4 mEq/L, and chloride 107 mEq/L. Urinalysis revealed normal findings.

Eight days after hospitalization, the patient underwent an opened cardiac surgery for replacement of the mitral leaflets with Sorin disc valve. The postoperative course was stormy. On the first post operation day, she was reoperated because of cardiac tamponade. Moreover, bleeding tendency, low cardiac output, and acute renal failure were present. The prothrombin time ranged from 16 to 19 sec (control 10 to 11 sec), BUN from 40 to 54 mg/100 ml, and creatinine from 3.2 to 5.4 mg/100 ml. In spite of peritoneal dialysis, the patient died 5 days after surgical intervention, or 2 weeks after hospitalization. There was no record on the level of blood platelets.

Postmortem examination. The following findings were found: old rheumatic endocarditis involving mural endocardium of the left ventricle, hypertrophy of the heart (400 gm), old infarct of the spleen and right occipital lobe of the brain, chronic passive congestion of the lungs, liver, and spleen; severe centrilobular necrosis of the liver cells, fatty metamorphosis and cholestasis of the liver, acute tubular necrosis of the kidneys, hemopericardium (50 ml), right hemothorax (500 ml), hemoperitoneum (200 ml), and congestion of the lungs (750 gm). The surgical specimen of the mitral leaflets was not received for pathologic study.

Protruding from the superior border of the left lobe of a 1,600-gm liver was a sessile polypoid mass, $10.0 \times 10.5 \times 11.0$ cm (Fig. 1). Its thick capsule was partly adherent by the diaphragm and omentum. Its bulging, rubbery, and purplish-red cut surface was partly honey-combed (Fig.2).

Microscopically, numerous vascular channels lined by endothelium and filled by blood comprised the lesion. The vascular walls were variable in thickness. The fibrous septa separated the vascular channels (Fig. 3).

The pathologic diagnosis was a solitary giant sessile polypoid hemangioma of the liver.



Figure 1. Interior (visceral) surface of the liver showing a giant sessile polypoid hemangioma attaching to the superior border of the left lobe. Note fatty (yellow) liver.

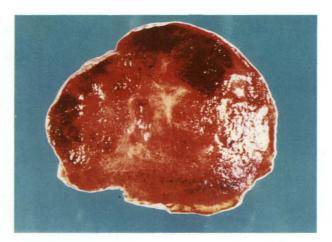


Figure 2. The purplish-red bulging cut surface of the hemangioma is interspersed party by dark honey-combed foci. The white flecks on the surface are related to light reflection in photography.

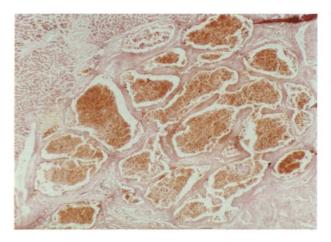


Figure 3. Hemangioma displaying microscopic appearance. Note many vascular spaces comprising the lesion which is surrounded partly by liver cells. H and E, \times 100.

Discussion

The definition of giant hemangioma of the liver differs among authors. Adam et al, $^{(6)}$ for example, regarded an angioma of more than 4 cm in diameter as giant. They based the definition on the presence of symptom. No hemangiomas of the liver of less than 4 cm across were symptomatic. Edmondson and Craig, $^{(7)}$ on the other hand, considered the size of 10 to 12 cm in diameter of an angioma of the liver to be more suitable for designating it as giant. The size of $10.0 \times 10.5 \times 11.0$ cm of the current hemangioma of the liver, thus, can be regarded as a giant lesion by these criteria. $^{(7,9)}$ The size of a giant hepatic hemangioma may be up to 45 cm in greatest dimension or 10 times of normal weight of the liver. $^{(8,15)}$

Hemangioma of the liver of less than 4 cm across is usually symptomless. (5) Only 13.5% of hepatic angiomas were symptomatic. (2) The common complaints are the presence of intra-abdominal mass or pressure upon adjacent abdominal viscera. (2,3,13) A murmur (bruit) may be heard on auscultation, especially in relation to arteriovenous shunting within the lesion which may lead to cardiac failure. (3,5) Rupture and intra-abdominal hemorrhage have been reported which resulted in abdominal pain and shock which necessitated laparotomy and which accompanied by high motality rate. (13) The hemangioma, moreover, may produce vague abdominal pain, nonspecific digestive disturbances, jaundice, distension of gallbladder, and ascites. (3,14) The current angioma is regarded as symptomless although it is gigantic. The patient did not complain of any aforementioned symptom throughout her course of ailment on two hospitalizations of 6-year interval.

Hemangiomatosis has been described. this condition, many angiomas are embedded within the liver or in association with hemangiomas of extrahepatic sites. (5,8-10) It should not be confused with the term "multiple hemangiomas" in which only a few hemangiomas are present in the liver. When angioma protrudes above the hepatic surface the term "polypoid hemangioma" can be used. The polypoid hemangioma may be sessile as in the current However, a fifth to a sixth of polypoid hemangiomas of the liver are pedunculated. (2) Some of the latter are indeed enormous to reach the pelvic cavity. Rubin, (15) for example, described a 33-yearold woman in whom a pedunculated hemangioma of the liver had grown into the pelvic cavity and caused obstetric difficulty during her ninth pregnancy. The resected hemangioma weighed 10,450 gm.

Kasabach and Merritt⁽¹⁶⁾ first described the relation between hemangioma and bleeding tendency. A 2-month-old boy with capillary hemangioma of the skin in the left thigh and lower part of the obdomen developed extensive purpura in the skin overlying the lesion and scrotum as well as thrombocytopenia. the hemangioma regressed after radiotherapy and radium implants. The platelet count returned to normal level. This phenomenon is now regarded as a well defined clinical syndrome (hemangioma-thrombocytopenia syndrome, or Kasabach-Merritt syndrome).(11,17,18) The syndrome usually occurs in the first few months of life. Bleeding is thought to be related to trapping and destruction of thrombocytes within the hemangioma and depletion of circurating clotting factors. (18) The present patient also had bleeding tendency as well as giant hemangioma of the liver. However, it is unfortunate that blood platelet count was not known. Hence, it is not possible to consider her bleeding tendency to be associated with hemangioma-thrombocytopenia syndrome. author, on the other hand, suggests extensive centrilobular necrosis of the liver cells to be responsible in the occurrence of prolonged prothrombin time and bleeding tendency in this adult patient.

In symptomatic cases, surgical resection is the treatment of choice. (3,9) Radiation therapy is of value. (16) Biopsy can be harmful and fatal because of internal bleeding. (3) Hence, it should be avoided when a hemangioma hepatis is suspected.

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