

Massive Hemangiomas of the Liver

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Four cases of massive hemangioma of the liver were encountered during the eleven-year period from 1966 to 1976. Two were localized in the right lobe and the third was in the left lobe. The lesion in the fourth case involved both lobes but the celiac angiography failed to demonstrate the vascular nature of the mass. However, splenoportography revealed the large and diffuse hemangiomatosis involving both lobes of the liver with mild portal hypertension. They were successfully removed in the first 3 cases but the last case suffered severe and almost uncontrollable hemorrhage at surgical exploration and was subsequently treated by radiation therapy. The different angiographic finding in the last two cases suggests that hemangioma of the liver may be of two different categories, one is related to hepatic arterial vasculature and the other related to portal venous vasculature. The importance of splenoportography in the diagnosis of hemangioma of the liver when the arteriography fails to demonstrate the vascular lesion is emphasized.

According to Boyd "Small hemangioma on the surface of the liver is a fairly frequent finding in the autopsy room. The tumor forms as a small red or purple mass on the surface from a few millimeters to several centimeters in diameter, and it is apt to be

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mistaken for an infarct. Microscopically it consists of large cavernous blood filled spaces. A massive hemangioma is a rare lesion but it is of importance to the surgeon¹²

In Thailand a large number of patients presented with right upper quadrant masses have eventually proven to be hepatocellular carcinoma. Benign tumors of the liver were rare, however, hemangioma was the most common tumor encountered among these rare conditions.

In 1970 Malt et al. reported 11 cases of liver hemangioma out of 26 cases of all benign hepatic neoplasm collected from Massachusetts General Hospital between 1947-1968.¹² From the Memorial Sloan-Kettering Cancer Center, Adam and associates were able to collect only 22 cases of giant hepatic hemangiomas over a 30 years period.¹ They called the tumor that was larger than 4 centimeters in diameter a giant hemangioma because those that were smaller were usually asymptomatic.

During the eleven years period from 1966 to 1976 we encountered four symptomatic cases of massive type of hemangioma of the liver. They were all larger than 10 centimeters and formed the basis of this communication.

CASE REPORT

CASE 1. Y.L., a 56-year-old man entered the hospital on September 19, 1966 for paraplegia for 3 years and had a mass in the upper abdomen for 2½ years. The patient was a heavy

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Fig. 1. Gross appearance of hemangioma of the right lobe of liver with white fibrinous plaques deposited in the tumor from Case 1 :

drinker since 18 years old. Three years prior to admission, the patient felt weak in both legs and had difficulty of both defaecation and urination. About 6 months later he noted a mass in the right upper quadrant of the abdomen which progressively enlarged. Despite good appetite he began to lose weight because of epigastric fullness after meals. Ten years ago the patient had an episode of jaundice and dark urine. He was told to have viral hepatitis. Physical examination on admission revealed a paraplegic man with a firm, nontender enlarged liver extending one hand breath below the right costal margin without ascites. Rectal examination was unremarkable. There was impairment of pain and tactile sensations from below the umbilicus. Hyporeflexia of all deep tendon reflexes were elicited in both lower extremities.

The hemoglobin was 16.4 gm per 100 ml. The white blood cell was 6,800 per mm^3 with 72 per cent neutrophils and 28 per cent lymphocytes. Urinalysis and stool examinations were unremarkable. The fasting blood sugar was 92 mg, the blood urea nitrogen 9.5 mg, the creatinine 1.2 mg, and the total bilirubin 1.0 mg per 100 ml. The serum albumin and globulin were 4.1 and 2.4 gm per 100 ml respectively.

The chest x-ray was negative. The liver scan by ^{131}I labelled BSP showed an enlarged liver with a space occupying lesion involving inferior portion of the right lobe. Peritoneoscopy revealed a mass in the right lobe of the liver with whitish surface suggestive of a malignancy of the liver.

The patient underwent an exploration through a right subcostal incision. There was a tumor mass occupying the right lobe of the liver with reddish-white appearance. There were firm plaques embedded in the reddish white area (Figure 1). The tumor mass felt spongy, compressible, highly vascular, and was confined to the right lobe. A right hepatic lobectomy was performed. The resected specimen weighed 1,340 gm. His postoperative course was uneventful except for low grade fever which subsided spontaneously. He was discharged on 35th post-operative day. He was readmitted on September 28, 1967

for operation for cervical spondylosis, his liver function tests were within normal limits and he had gained 8 kg.

Comment : Peritoneoscopy was erroneously interpreted as hepatocellular carcinoma because of the plaque-like depositions of fibrinous exudate in the tumor mass. Lacking of arteriographic study, the clinically progressive enlarged liver and weight loss together with the unusual peritoneoscopic finding would have favored the diagnosis of malignancy had the exploration not been carried out.

CASE 2. C.T., a 50-year-old Thai housewife was admitted to the hospital on November 20, 1969 for having a painless mass in the right upper quadrant of the abdomen of 5 months duration. The patient had been in good health all her life. While taking a bath, she accidentally felt a fist size mass in her abdomen. Despite the mass became larger, her appetite remained good and there was no change in bowel habits. Physical examination revealed a bulging area in the right side of the abdomen which moved with respiration. On palpation a large nontender 25 x 15 x 15 centimeters mass with rubbery consistency was noted. It was dull on percussion and no bruit or hum was heard over the mass. There was no abnormal pigmentation elsewhere in the body. Rectal and vaginal examinations were unremarkable.

The hematocrit was 33.5 volume per cent. The white blood cell count was 5,100 per mm^3 with 60 per cent neutrophils, 32 per cent lymphocytes, 3 per cent monocytes and 5 per cent eosinophils. Urinalysis and stool examinations were unremarkable. The total bilirubin was 1.36 mg per 100 ml. The alkaline phosphatase was 0.98 Sigma units (Normal 1-2.3 Sigma units). The SGOT was 20 units. The fasting blood sugar was 108 mg and the blood urea nitrogen was 13 mg per 100 ml. The electrocardiogram was normal.

The chest x-ray was negative. Plain films of the abdomen showed a large homogeneous mass in the right side of the abdomen displacing the transverse and hepatic flexure of the

colon inferiorly with no calcification. The GI series showed a large, well circumscribed homogeneous extragastric mass. The stomach, duodenum and small intestine were displaced but not involved by the mass. Hepatic scan by ^{198}Au showed a large space occupying lesion in the anteroinferior portion of the right lobe of the liver.

On November 20, 1969, the patient was explored under general anesthesia through a right subcostal incision. Exploration of the abdominal cavity revealed all viscerae to be normal except for a solitary deep red compressible mass arising from the anteroinferior part of the right lobe of the liver. The tumor was completely removed by segmental resection of the right lobe of the liver. Her post-operative course was benign and she was discharged 9 days after the operation. The patient was seen 2 months later and was doing well.

One year later the patient had undergone an exploration for carcinoma of the sigmoid colon. The liver appeared grossly normal. Careful inspection of the right lobe of the liver showed a 2 x 2 cm reddish and soft plaque on the resected edge of the liver. It was resected and the histological study revealed hemangioma.

Comment: Apart from a rather rapid growing hepatic mass, the patient exhibited no other abnormal gastrointestinal symptoms.

CASE 3. L.P., a 52-year-old Thai woman from the northeast came to the hospital with a chief complaint of having a mass in the epigastric region for 3 months duration. There had been no other abnormal gastrointestinal symptoms. In spite of pressure of the mass the patient had gained 1.8 kg in 3 months.

Physical examination revealed a non-tender and firm mass palpable at the epigastrium approximately 10 centimeters in diameter which moved with respiration. Soft bruit was heard over the mass in a quiet room. Rectal and vaginal examinations were unremarkable.

The hematocrit was 30 volume percent. The white blood count was 3,600 per mm^3 with 53 per cent neutrophils, 30 per cent lymphocytes and 9 per cent eosinophils. The platelet was 210,000 per mm^3 . Urinalysis and stool examinations were unremarkable. The fasting blood sugar was 90 mg, blood urea nitrogen 13 mg, total bilirubin 1.0 mg per 100 ml. Serum albumin and globulin were 4.1 and 3.3 mg per 100 ml respectively. The SGOT was 13 units, SGPT 4 units and the alkaline phosphatase 1.3 Sigma units. The chest x-ray was normal. The abdominal films showed a round mass in the epigastrium. The oral cholecystogram revealed a normal functioning gall bladder. The GI series showed the stomach being slightly displaced anteriorly and to the left by a mass suggestive of pancreatic in origin. A small diverticulum was noted on the second portion of the duodenum. $^{99\text{m}}\text{Tc}$ sulfur colloid liver scan showed an enlarged extra hepatic mass in the portahepatis. The uptake in the liver was normal. Selective angiography revealed a tumor mass in the left lobe of the liver with abnormal vascularity consistent with hemangioma of the liver.

The patient was then explored and a 10 x 10 x 3 centimeters deep red colored mass was found in the inferior surface of the left lobe of the liver. It was soft and characteris-



Fig. 2. A hemangioma of the left lobe of the liver from Case 3 with marked compressibility of the tumor mass.

tically compressible. (Figure 2). The rest of the liver appeared normal. Exploration of the other abdominal viscerae were within normal limits. Left hepatic lobectomy was done. The patient made an uneventful recovery and was discharged on 7th postoperative day. The tumor was diagnosed histologically as hemangioma of the liver.

Comment: The huge tumor mass confined to the left lobe of the liver produced a pressure displacement of the stomach mimicking a pancreatic lesion. This patient also exhibited no abnormal gastrointestinal symptoms except for the presence of an abdominal mass.

CASE 4. B.S., a 45-year-old man came to the hospital on November 13, 1973 complaining of abdominal discomfort and bulging of the abdomen for 3 years. Shortly before entry he experienced a pressure and tightness in the upper abdomen especially at inspiration and after meals. He was unable to sit up straight but found bending over the most comfortable position. He also noted that he had rather become constipated and had lost some weight. There had been no fever or nausea and vomiting. He had been drinking one glass of whisky every day for years but this was stopped about one year previously be-

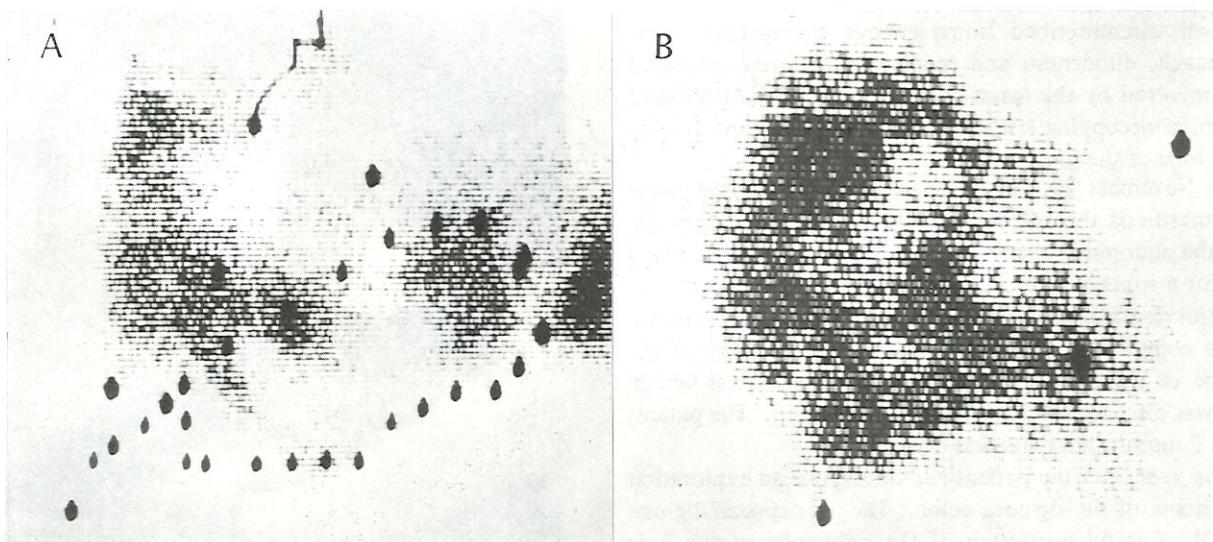


Fig. 3. An antero-posterior (A) and lateral (B) views of the liver scan from Case 4 demonstrating multiple hemangiomatosis of the liver.

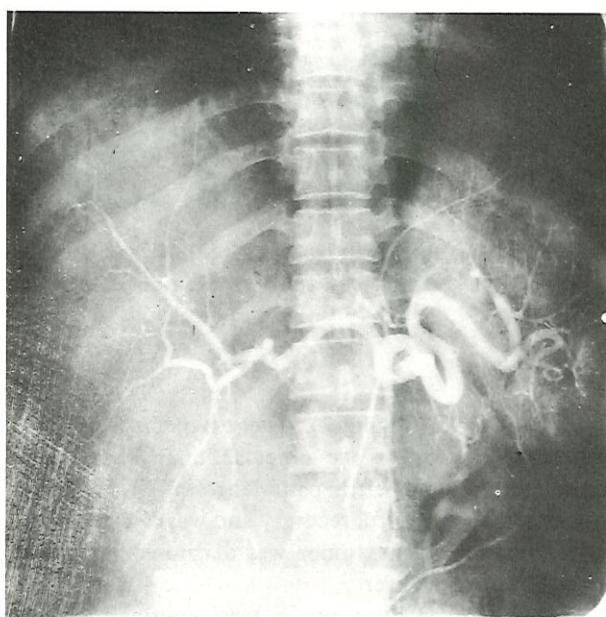


Fig. 4. Celiac angiogram in Case 4 showing a large tortuous splenic artery. Intrahepatic branches of hepatic artery were separated by multiple masses and no contrast material entered into the masses of hemangiomas.

cause it caused more abdominal discomfort. There was no stigmata of cirrhosis. Heart and lungs were unremarkable. Abdominal examination revealed an enlarged liver about one hand breath below the right costal margin. There were several soft and compressible areas interspersed on what appeared to be a hard liver mass. The mass was not tender and no friction rub nor bruit could be detected. The spleen was two finger breaths below the left costal margin. Rectal examination was unremarkable.

The hemoglobin was 13.2 gm per 100 ml. The white blood cell count was 5,900 per mm³ with 49 per cent neutrophils, 35 per cent lymphocytes, 1 per cent basophils and 14 per cent eosinophils. The platelet count was 196,000 per mm³. The fasting blood sugar was 94 mg, the blood urea nitrogen 8 mg, the total bilirubin 1.0 mg, the cholesterol 106 mg per 100 ml. Serum albumin and globulin were 3.8 gm and 4.0 gm per 100 ml respectively. The SGOT was 56 units, the SGPT 23 units and the serum alkaline phosphatase 2.0 King-Armstrong units. The prothrombin time was 34.2 per cent. The partial thromboplastin time was 80 seconds and the thrombin time was 1.9 seconds. Urinalysis was unremarkable. Stool examination showed opisthorchiasis and hook worm ova as well as hook worm larvae.

The chest x-ray was normal. The GI series showed marked hepatomegaly. The stomach and duodenum were displaced laterally by the enlarged liver but their mucosa appeared intact. Liver scan by ^{99m}Tc showed hepatomegaly with multiple space-taking lesions throughout both lobes of the liver (Figure 3). Celiac angiography revealed enlarged liver and spleen with dilated and tortuous splenic artery. Intrahepatic branches of the hepatic artery were spread apart. There was no abnormal tumor stained (Figure 4). A splenoportogram showed splenomegaly with enlarged splenic and portal veins (Figure 5A) and the splenic pulp pressure measured 260 mm H₂O. No collateral veins were seen. In the late films the contrast material concentrated in the big venous spaces in the liver (Figure 5B). Peritoneoscopy showed markedly nodular and enlarged liver with multiple blueish-red cystic lesions.

He was operated upon on November 21, 1973 through a right subcostal incision. There were multiple small and large vascular spaces varying the size from one millimeter to six centimeters involving both lobes of the markedly enlarged liver. Those cystic masses were compressible as has been experienced preoperatively. Direct portal pressure measurement

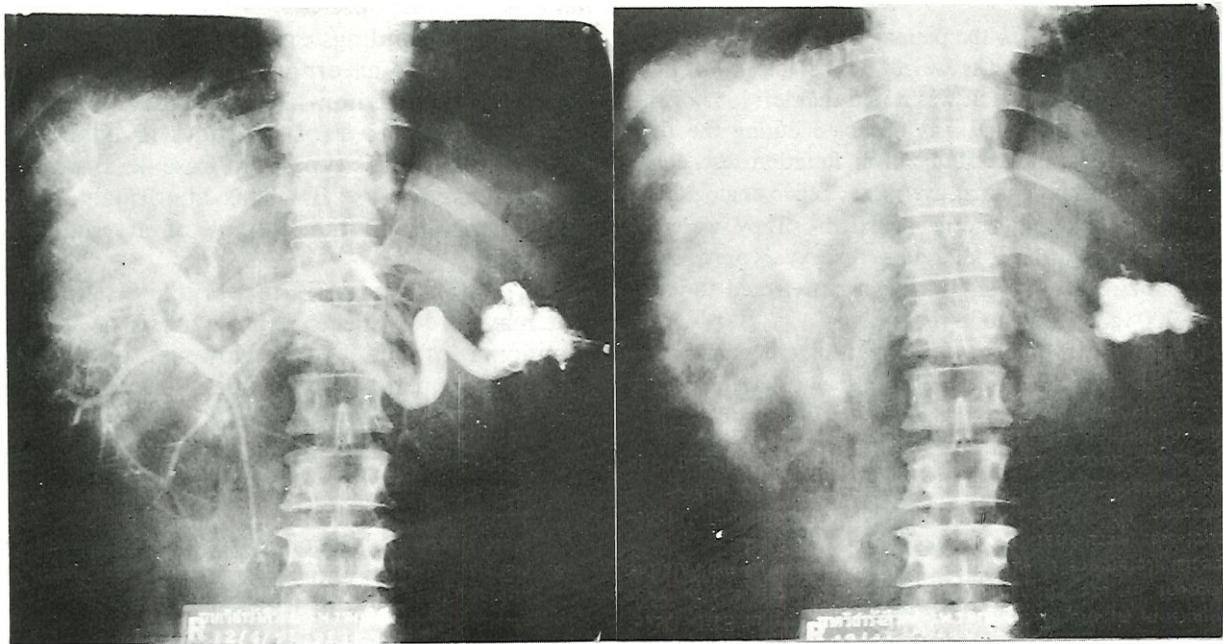


Fig. 5. Splenoportogram in Case 4 showing multiple blood filled spaces in the liver being filled with contrast material via the portal vein (A), more contrast material filled spaces in the liver in the late splenoportogram film (B).

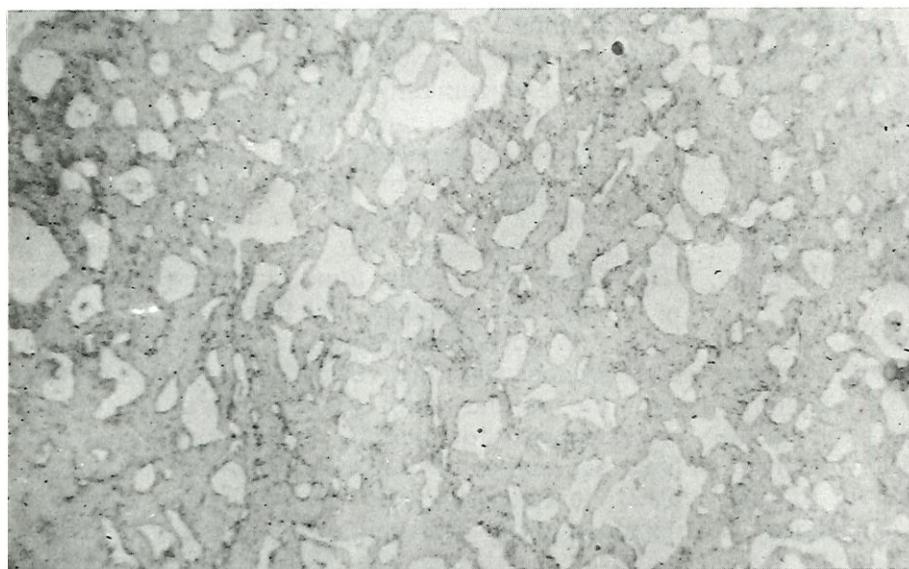


Fig. 6. Photomicrograph of hemangioma of the liver from Case 4 showing markedly dilated venous blood channels with red blood cells in their lumina x 200.

was 250 mm H₂O. Liver biopsy was obtained and bleeding from biopsy site controlled. Unfortunately during exploration one of the masses was inadvertently ruptured and severe hemorrhage resulted. Direct suturing of the lesion and the liver was inadequate for hemostasis. The whole left lobe of the liver was then tied with umbilical tape in order to achieve control of the massive hemorrhage. The episode of intra-operative hypotension lasted for about one hour before it was corrected by massive blood transfusion and Ringer Lactate infusion. The post-operative course was very stormy. Another 3,500 ml of blood transfusion was required in the following 24 hours. The

patient required mechanical respiratory support and external abdominal pressure in order to stabilize the vital signs. Despite pneumonia, pleural effusion, congestive heart failure, *Pseudomonas* urinary tract infection, the patient eventually recovered and was discharged from the hospital 26 days later. The liver biopsy showed hemangioma (Figure 6) with hemosiderosis and early hemochromatosis.

On April 4, 1976 he was readmitted for dysuria. Transurethral resection of prostate gland was done. He tolerated the procedure well and was discharged on May 3, 1976. The patient still had abdominal discomfort as previously expe-

rienced.

On September 7, 1976 the patient was again hospitalized with the same complaint. His weight was stable at 48 kilograms. He was not jaundice. It was noted that left lobe of the liver became harder than that was observed during the first admission. The hematologic study, liver function test, chest x-ray, GI series, barium enema, liver scan, celiac angiography and the splenoportography were repeated. They were all essentially unchanged from studies done three years previously. We decided not to operate on him again but elected to try a course of 2,000 rads of radiation therapy.

Comment: This patient has multiple hemangiomas throughout the liver. The wall of each vascular space was very thin and friable. Alarming hemorrhage occurred when it was inadvertently ruptured during exploration. It was rather unusual that these hemangiomas received blood supply solely from the portal venous system as demonstrated in the splenoportogram but not in celiac angiographic study. Mild degree of portal hypertension observed in this case may be secondary to portal venous congestion from massive hemangiomatosis. The failure of celiac angiography to demonstrate the vascular lesion of the intra-hepatic mass did not completely exclude the possibility of the lesion being a hemangioma.

DISCUSSION

Of all benign hepatic neoplasm, hemangioma is the most common. However, the massive type is rare.^{1,2,6,12} In Adam's report among one hundred and twenty six cases of benign liver tumors, one hundred and six cases were hemangiomas and only twenty two cases were larger than four centimeters.¹ The largest hemangioma of the liver weighing 18 kg was recorded in a patient by Major and Black.¹¹ This patient also had associated hemangioma of the scalp and bilateral cystic adrenal glands.

This tumor occurs among patients in third and fifth decade. The ratio between female to male varies from 1:1 to 10:1.^{1,6} It is known to occur since birth. Clatworthy⁴ and associates regarded cavernous hemangioma in the newborn as a surgical emergency condition since it had a high tendency to rupture. However, Braun et al. reported only 4 instances of rupture in the review of over 100 cases in the literatures and advocated a nonoperative approach with supportive cardiac therapy and systemic use of steroids.³ Twelve cases of spontaneous rupture of hepatic hemangioma had been reported by Sewell and Weiss with a mortality of 75 per cent.¹⁵

The leading cause that brings the patient to the physician was the presence of a mass. Some patient may have the abdominal mass for as long as 30 years.¹⁶ Digestive disturbance such as anorexia, nausea vomiting, tightness in the abdomen, epigastric pain etc. may be experienced on the basis of the pressure from the mass. Other unusual presentations such as obstetrical difficulty,¹⁴ thrombocytopenia,⁵ gastrointesti-

nal obstruction¹⁸ are extremely rare.

Physical findings entirely depend on the type of hemangioma. A cavernous type usually presents with a rubbery to firm non-tender mass in the upper abdomen in contrast to a soft multicystic mass of multiple blood filling spaces type as in Case 4. Bruit is elicited in some as in Case 3. This abnormal auscultatory sound depends on the volume of arterial feeding to the tumor.

There is no definite peripheral blood picture that may give a clue to the diagnosis of a massive hemangioma of the liver. However in rare instance thrombocytopenia may occur.⁵ Liver function test in all cases were unremarkable. Despite the presence of large and multiple space occupying hemangiomas in the liver the serum alkaline phosphatase remained normal in all cases.

Contrast study such as gastrointestinal series and barium enema will show displacement of internal viscerae and may give information as to whether or not hollow viscerae are involved. Liver scan demonstrates only space taking lesion but offers no definite characteristics. Peritoneoscopy may be helpful if a deep red mulberry mass is seen but due to limitation of the visualized field the correct diagnosis at times may be difficult. In some cases peritoneoscopic findings may be erroneously interpreted as in Case 1. A blind liver biopsy is definitely contraindicated because it will lead to an exanguinating hemorrhage.

The most useful investigation for hemangioma of the liver is angiographic study of the celiac axis. It is widely agreed that the findings in celiac or hepatic arteriography are diagnostic in the massive type of hepatic hemangioma. But this was not the case in Case 4 since the vascular tumor was demonstrated only by splenoportography and thus it was exclusively related to portal venous system without appreciable arteriovenous shunting. In the case reported by Kato et al. the vascular tumor was demonstrable by hepatic arteriography but not by splenoportography and from the survey of Japanese literature they found other investigators who likewise were unable to demonstrate the hemangioma by splenoportography.⁸ However, they learned that Moriyasu and Levine both had reported that splenoportography proved valuable in the diagnosis of hepatic hemangioma and this was supported by the findings in our Case 4.

It appears that the majorities of hemangioma of the liver are associated with or having their feeding vessels from the hepatic arterial branches. Our Case 4 showed the tumor to have its feeding vessels exclusively via the portal venous system. The case of hemangioma of the liver diagnosed by splenoportogram and reported by Levine¹⁰ resembled our Case 4 because of the similar splenoportographic findings and the evidence of associated portal hypertension but unfortunately no hepatic arteriography was performed

to confirm that the tumor was exclusively associated with portal venous blood supply. Slovis¹⁷ et al. reported an unusual hemangioma of the liver in a one-day-old infant with hepatic artery-hepatic vein and portal vein shunting. Retrograde filling of the portal vein in large hemangiomas of the liver had been reported but their feeding vessels primarily came from hepatic artery.⁷

As the liver receives dual blood supplies, from both hepatic artery and portal vein, thus it is conceivable that hemangioma of the liver may be of two different types. One is hemangioma of the hepatic arterial system and the other of the portal venous system, i.e. hepatic hemangioma and portal hemangioma. The awareness of the possibility of portal hemangioma is important when hepatic arteriography fails to show the vascularity of the tumor. Splenoportography is indicated in such case before a percutaneous needle biopsy is being attempted.

As to what should be done to this massive vascular tumor depends entirely on the extent of the tumor. When the tumor is localized hepatic lobectomy or segmental resection should be performed. In Case 4 whose extensive bilobar hemangiomatosis made surgical extirpation impossible, we decided to try radiation therapy as advocated by Park & Philip.¹³ It is still too early to make any conclusion regarding the result of the treatment given. We had considered porta systemic decompression as a mean to reduce the tumor vascularity and to prevent the possible spontaneous rupture of the tumor in this case but the operation had not been undertaken. It has been shown that a porta systemic decompression will be well tolerated if the Zimmon-Kessler⁹ test shows a good hepatic arterial response to portal venous diversion. This approach is planned for Case 4 if radiation therapy fails to alleviate the patient's symptoms and the danger from spontaneous rupture of the hemangioma makes porta systemic decompression become necessary.

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