BILIARY TRACT HEMORRHAGE: A SOURCE OF MASSIVE GASTRO-INTESTINAL BLEEDING*

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THE UNDETERMINED SITE of massive gastro-intestinal hemorrhage is a constant challenge to the clinician and consultant. In reviewing the origin of hematemesis and melena, the conclusions of several authors[1-3, 5, 8, 27] indicate that 2 to 5 per cent of the cases are tabulated "gastro-intestinal hemorrhage —source undetermined." The biliary tract is rarely considered in such a differential diagnosis of massive hemorrhage. The purpose of this report is to stimulate interest in the liver and biliary tract as a source of bleeding in patients having gastro-intestinal tract hemorrhage which eludes diagnosis by the usual studies.

CASE REPORT

R. P., a 35-year-old Jewish male, was first seen May 22, 1949, with the complaint of epigastric pain and faintness. His past history revealed that in 1931 the right testicle was removed for an embryonal teratoma. This was followed by deep roentgen ray therapy which was given for palpable metastasis. Sufficient roentgen ray was given to produce irradiation dermatitis. In 1934 the deep roentgen ray treatment was repeated for the complaint of recurrent abdominal pain. An appendectomy was performed in 1938 and a Torek procedure on the left testicle in 1939. In 1945, while in Portugal, the patient developed a syndrome of diarrhea, during which ulcerations of the rectum were found. Repeated examinations of the stools failed to demonstrate any pathogens. Shortly after his return to the United States he developed abdominal pain with a definite food relationship which suggested peptic ulcer to his physician. In October, 1947, he was admitted to the hospital for a recurrence of the pain. Marked anemia was present, but roentgenograms showed no lesion in the gastro-intestinal tract. Transfusions failed to produce a satisfactory response and he continued to pass tarry stools. A sub-total gastrectomy was performed as an emergency procedure, although a definite ulcer could not be demonstrated in either the stomach or the duodenum at the time. The pathologist was unable to demonstrate any ulcer in the resected surgical specimen. Six months after the operation he developed an attack of epigastric pain in the left upper quadrant associated with mild shock and was first seen by one of us (M. M.). He was hospitalized, and study demonstrated the mild transient acute pancreatitis evidenced by elevation of the serum amylase, hyperglycemia and glycosuria. He responded well to symptomatic measures and following his recovery a complete gastro-intestinal roentgen ray study and cholecystogram were done. No evidence of pathologic changes in the stomach, small or large bowel, or gallbladder was obtained. At the present admission he again complained of pain in the epigastrium and faintness, and passed a black tarry stool. This pain was not related to the ingestion of food in any way, and was colicky in nature. There had been no vomiting of blood at any time and no associated nausea. Roentgenograms again failed to show any evidence of gastric, marginal, or jejunal ulcer and the barium enema study was again reported negative.

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Gastroscopy showed no intrinsic lesion within the stomach or at the site of the gastro-jejunal anastomosis, but it was noted that there was brownish material welling up in the proximal end of the gastro-jejunostomy loop.

On physical examination the patient was markedly emaciated, his weight being 99 pounds as compared to 130 pounds before his partial gastrectomy. The pertinent findings were limited to malnutrition, anemia and to the abdomen. There was evidence of irradiation, and telangectasia was noted in the skin of the anterior abdominal wall. Moderate epigastric tenderness was noted. The laboratory studies reported a hemoglobin of 74 per cent, R.B.C. 3.4 million. All of the blood chemistry was within normal limits with the van den Bergh, 0.2; the B.S.P. test was negative, cephalin flocculation test was negative. All stools were positive for occult blood. He was given 5 transfusions and was discharged from the hospital with the recommendations that he should submit to laparotomy. A month later he began to bleed again, passing large black tarry stools, and developed weakness and dyspnea with mild exertion. He was again admitted to the hospital. Laboratory and roentgen ray studies were repeated without revealing further information.

Laparotomy was performed on June 22, 1949. On opening the abdominal cavity the viscera were thoroughly explored. It was noted that there was a small tumor in the duodenal stump and after opening the duodenum to remove this polyp it was noted that it was perfectly clean and healthy. The gallbladder was then compressed and, to our amazement, bloody bile exuded through the papilla of Vater. The gallbladder and then the common duct were aspirated and bloody material was found in both. The common duct was opened and a perfect blood clot cast of the common duct and the radicals of the common hepatic duct was found and removed. No stones, ulcer or tumor were found either in the biliary tract or in the gallbladder. Palpation and probing of the hepatic radicals failed to disclose the presence of tumor or stone. Bloody bile came down both the hepatic radicals into the common hepatic duct and it is therefore assumed that the hemorrhage was hepatic in origin. The gallbladder was removed and a T-tube implanted in the common duct. The convalescence was entirely uneventful. During his recovery a specimen of bile was collected and a cytologic study done. The pathologist was unable to find any cells suggestive of tumor. Two cholangiograms done before removal of the T-tube showed no abnormality of the biliary tract. The patient was discharged from the hospital on his sixteenth postoperative day in good condition. Since that time he has had lobar pneumonia, following which he passed tarry stools on 2 days. He has gained 8 pounds of the weight lost. Skin tests and complement fixation tests for schistosomiasis are negative. Repeated studies of his blood, coagulation time, prothrombin time, marrow, etc., have all been normal. There has been no recurrence of bleeding since August, 1949.

DISCUSSION

The above record illustrates the problems of accurate diagnosis and rational treatment for the patient having repeated massive gastro-intestinal hemorrhage. In this case intrahepatic biliary tract bleeding is the origin of the repeated hemorrhages.

Biliary tract hemorrhage was first reported by Nauyn in 1892 and by Schmidt the following year.[19] We were able to find less than 100 subsequent case reports on the subject. Only reports of an acholemic patient with fatal or near fatal hemorrhage arising from the biliary tract have been considered.

Nauyn (1892) Budinger (1925) and Lichtman (1936),[19] have classified hemorrhage due to diseases of the liver and biliary tract. Lichtman's classification is anatomical, simple and clinically sound. It is as follows:

TABLE I.—*Classification of Hemorrhage Related to Diseases of the Liver and Biliary System (Lichtman 1936).*

- A. Acholemic
  - 1. Portal in origin
    - a. Varices
    - b. Thrombosis of portal vein
  - 2. Hepatic in origin
    - a. Trauma
    - b. Yellow atrophy
  - 3. Biliary tract in origin
    - a. Vascular
      - 1. Ruptured aneurysm
      - 2. Erosion of blood vessel
      - 3. Perforation of gallbladder
    - b. Hemorrhagic cholecystitis
    - c. Neoplasm with ulceration
- B. Cholemic dyscrasia

We have further subdivided biliary tract hemorrhage into intra- and extrahepatic sources because of the difference in clinical manifestations.

TABLE II.—*Sources of Biliary Tract Hemorrhage: Reported.*

- A. Intrahepatic
  - 1. Post traumatic
  - 2. Subacute yellow atrophy
  - 3. Hemangioma
  - 4. Central apoplexy
    - a. Aneurysm hepatic artery
- B. Extrahepatic
  - 1. Bile ducts
    - a. Calculus
    - b. Ulcer—benign with erosion hepatic artery, portal vein, etc.
    - c. Aneurysm
      - 1. Cystic artery
      - 2. Hepatic artery
    - d. Cavernomatous transformation of the portal vein
    - e. Carcinoma ampulla of Vater
  - 2. Gallbladder
    - a. Calculus—erosion cystic artery, etc.
    - b. Ulcer
    - c. Hemorrhagic cholecystitis (hemocholecyst)
    - d. Cancer of gallbladder
    - e. Cholecystitis glandularis proliferans

Intrahepatic hemorrhage into the bile ducts may be due to trauma,[13, 25] subacute yellow atrophy,[19] hemangioma,[5] central apoplexy or rupture of an hepatic artery aneurysm.[28] It should be emphasized that in the reports of post-traumatic hemorrhage, the gastro-intestinal bleeding occurred five to seven weeks after the injury. In one instance, studies for peptic ulcer were made before laparotomy proved the diagnosis of hepato-biliary tract hemorrhage.[13] Therefore, history of injury to the abdomen or lower right chest may lead one to suspect the liver if subsequent hemorrhage occurs. The intrahepatic sources are quite rare as compared to those of the extrahepatic biliary tract. This is a fortunate circumstance since the extrahepatic group is surgically accessible for correction or removal.

The most common cause of extrahepatic biliary tract hemorrhage is the gallstone.[14, 15, 21, 22, 24, 18] The most frequent site of the hemorrhage is in the gallbladder. Other causes of cholecystic bleeding are hemorrhagic cholecystitis, hemocholecyst, benign ulcer, trauma, cancer and finally the rare polypoid condition known as cholecystitis glandularis proliferans.[10, 13, 15-17, 19, 21, 22, 29]

Hemorrhage is an important consideration in cases of cholelithiasis. White[29] reported six cases of massive hemorrhage, one of which resulted from a stone left in the cystic duct stump after cholecystectomy. A considerable number of patients with so-called quiet stones have anemia associated with the presence of occult blood in the stool.[15] In the absence of other sources these findings may suggest previously unsuspected gallbladder disease. We have noted the presence of old blood clots in gallbladders removed for calculus. Lichtman[19] believes that there is evidence of gross hemorrhage in 1 to 5 per cent of all patients with gallbladder pathology. There are several reports in the literature of massive hemorrhage associated with cholecystitis and hemocholecyst. There are only two reports[13, 17] of hemorrhage from the gallbladder following external trauma. These were due to laceration of the cystic artery which produced shock, hematemesis and melena. Although rare, Hudson and Johnson,[15] Fiessinger, *et al.*,[10] Sainburg and Garlock[26] and Lichtman[19] report occasional massive gastro-intestinal bleeding in patients with primary cancer of the gallbladder; *i.e.*, carcinoma and hemangiosarcoma.

Study of these reports of patients presenting massive hemorrhage which originated in the gallbladder leads to the following conclusions. Nearly all patients with stones gave a history of previous episodes suggesting colic before the hemorrhage. Those having tumor or hemorrhagic cholecystitis presented a right upper quadrant mass in the region of the gallbladder associated with tenderness, fever and hemorrhage. In the cases of traumatic hemorrhage the history of injury was always given even though the gallbladder was not suspected as the source until operation. We believe, therefore, that we should be able to predict the origin of hemorrhage in this group of patients by past history, physical findings and exclusion of the more common causes of massive hemorrhage.

The diagnosis of primary extrahepatic bile duct hemorrhage is more difficult. Massive bleeding from the ducts has been reported due to stone, benign ulcer,[19] rupture of hepatic artery aneurysm,[11, 19] erosion of the cystic,[29] hepatic or portal vessels, and to cavernomatous transformation of the portal vein with rupture into the common bile duct.[19] It is of considerable interest that several authors make no mention of hemorrhage being associated with benign papilloma[4, 6, 9, 20] of the common duct, papilla of Vater or the gallbladder. One may conclude that it must rarely occur with these benign tumors. However, malignant ulcerations[5, 7-9, 20, 23] of the ampulla of Vater are frequently associated with gross and massive hemorrhage. Cooper[7] states that occult or frank hemorrhage occurs in all patients having ampullary car-

cinoma. Eusterman[8] reported that in a group of patients undergoing surgery for undiagnosed gastro-intestinal bleeding, carcinoma of the ampulla of Vater was a frequent unexpected finding. This finding emphasizes the value of duodenal drainage for the study of its contents in patients having negative gastro-intestinal roentgen ray studies. Cytological study of these contents should be a further aid in establishing a preoperative diagnosis of ampullary carcinoma at an earlier date.

SUMMARY

1. A case report of intrahepatic biliary tract hemorrhage is presented.

2. The problem of biliary tract hemorrhage as a source of massive gastro-intestinal bleeding is discussed.

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DISCUSSION.—DR. JOSEPH E. J. KING, New York: I have enjoyed Doctor Blakemore's paper very much and am pleased to see his most intriguing apparatus for control of this terrible condition. You need see only one of these patients die before your eyes to realize your helplessness in such a situation. It's like watching a man drown without being able to do anything about it.

About ten years ago I operated upon a man who was chief officer on a merchant marine ship for an encapsulated brain abscess of the left temporal lobe. It was one of the easiest to deal with and operate upon that I have ever seen, and we expected a good result in a fairly short time. He was about 42 years old and a known syphilitic. About the fifth postoperative day a massive hemorrhage of bright blood took place and his condition became very poor. We felt sure the hemorrhage was from varices of the esophagus but we did not know how to control it. I knew that electrocoagulation had been done on a few occasions successfully, but we did not want to attempt it in this case because of possible rupture of the brain and provoking hemorrhage through struggling. So we did nothing for the time being but observe him and wait. We then gave him a small transfusion of blood and he improved. Just about the time he seemed to be doing fairly well he had another severe hemorrhage. This recurred five times. Each time we gave him a little blood and his condition became somewhat better, he would have another hemorrhage. After the fifth one, he died on about the fifteenth postoperative day.

Autopsy revealed a large opening in a varix, so large that it admitted the blunt end of a mortician's needle. The spleen was greatly enlarged to about three times its normal size. The liver, instead of having the appearance of a normal liver, was discoid in shape, about 12 inches in diameter, and was somewhat the shape of a loaf of black Polish bread, narrower at the edge than in the middle. I would say it was not more than 3.5 inches in thickness at its central portion. Of course, the hemorrhage could have been controlled readily had one been able to put his finger on the small hole and hold it, but this could not be done. I thought of all sorts of things to do, like tamponading the esophagus with a gauze packing, or making some sort of apparatus shaped like the old-fashioned cattail that grows in a swamp, and pushing this down into the esophagus and holding it there. However, none of these things were done.

Just a few weeks ago, shortly after I received the program for this meeting, I glanced through it and saw the title of Doctor Blakemore's paper. I had not the slightest idea what it was about so far as the rubber bag was concerned. A few evenings later I was called away from a surgical meeting by one of my colleagues. He told me about a patient with an enormous plum-colored hemangioma occupying about half of his face, who had sustained a hemorrhage of bright blood from his throat. I was told he had had a hemorrhage from a varix. Having remembered the one word "balloon" from the