Recognition and Surgical Repair of Superior Mesenteric Arteriovenous Fistula

By Robert G. Sumner, M.D., Philip C. Kistler, M.D., William F. Barry, Jr., M.D., and Henry D. McIntosh, M.D.

Systemic-Portal arteriovenous fistulas are uncommon vascular anomalies. A review of the literature revealed only 16 cases with clinical descriptions. Although the site of origin varied, drainage in each instance was ultimately into the portal vein. In one patient the fistula arose from the main superior mesenteric artery, in three the secondary branches of the superior mesenteric artery, in five the hepatic artery, and in seven the splenic artery. Five of the 16 patients died of causes related to this lesion. It would appear that patients with such shunts have a poor prognosis.

The diagnosis of an arteriovenous fistula between the superior mesenteric artery and vein was recently established in our laboratory by radiographic technics. As this patient is the only such case known to have had a successful surgical repair, a detailed report is indicated.

Case Report

E. R., a 27-year-old white single man, was admitted to the Duke Medical Center on March 31, 1962, for evaluation of a painful monoplegia of the right lower extremity, which followed a gunshot wound to the upper abdomen 3 months previously. A .22-caliber bullet had entered the epigastrium and lodged in the posterior thorax at the level of the second lumbar vertebra. Immediate exploratory laparotomy at another hospital was said to have revealed lacerations of the liver and right kidney. A right nephrectomy was performed and 14 units of blood were administered during the procedure. Postoperatively the patient improved slowly. The right leg, however, remained paralyzed and painful. At no time did he com-

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Supported in part by grants HTS-5569, H-6960, and HE-07563-01 from the National Heart Institute, U. S. Public Health Service.

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plain of symptoms referable to the gastrointestinal tract.

Physical examination at the time of admission to the Duke Medical Center revealed a blood pressure of 110/70 mm. Hg and a pulse of 80. The right lower extremity was erythematous. A sensory level was present over the hip with associated loss of motor function and muscle atrophy. The scar from the previous laparotomy extended from the umbilicus to the epigastrium. Of considerable interest was the presence of a systolic thrill and continuous murmur over the entire upper abdomen.

Initial laboratory work included a hematocrit value of 46 per cent; white blood-cell count of 12,500, and an unremarkable urinalysis. Total phenolsulphonphthalein excretion in 2 hours was 55 per cent. Blood chemistries were normal.

The electrocardiogram and chest x-rays were within normal limits. An esophagram and upper gastrointestinal and ileal series were all normal. Films of the lumbosacral spine showed metallic fragments at the level of the second lumbar vertebra. An intravenous pyelogram demonstrated prompt function on the left side but none on the right side; the right renal shadow could not be seen. Liver biopsy (obtained at laparotomy) was interpreted as "normal liver."

Because of the presence of the abdominal bruit, maximal in the supraumbilical region, catheters were inserted percutaneously into the right femoral artery and the right antecubital vein. Hepatic vein catheterization revealed the wedge pressure to be 10 mm. Hg and a free hepatic vein pressure of 5 mm. Hg. Except for the slightly elevated wedge pressure, all pressures and contours recorded in hepatic veins and the inferior vena cavae were within normal limits. There was no step-up in inferior vena cava oxygen saturation beyond that ordinarily seen as a result of renal blood flow (table 1). The hepatic venous oxygen saturation of 86 per cent, however, was remarkably elevated. The cardiac output (indicator-dilution technic) was at the upper limits of normal (5.3 and 5.6 liters per minute). Bromsulfalein clearance was demonstrated to be unusually rapid (fig. 1).

The tip of the aortic catheter was placed at the level of the bullet fragments for the purpose of obtaining cineradiograms and angiograms. Fol-
lowing injection of the contrast medium, a the angiogram (fig. 2) showed immediate visualization of the portal vein in addition to the aorta, superior mesenteric artery, and celiac axis. The left renal artery was demonstrated; the right was not. The portal vein was markedly dilated. Protruding from the right side of the aorta at the level of the second lumbar vertebra was a saccular structure that appeared to be continuous with the portal system. This communication was interpreted as an arteriovenous fistula between aorta and portal vein. The cinefluorograms, on the other hand, showed that the dye injected through the aortic catheter entered the superior mesenteric artery before filling the portal venous system. This study therefore suggested that the shunt arose from the superior mesenteric artery.

Because of uncertainty as to the exact site of the fistula, the patient was recatheterized 4 days later, and again a cinefluorogram and angiogram were obtained.† For these films the patient was rotated 20 degrees in the right posterior oblique position in order to visualize the superior mesenteric artery separately from the aorta.

† For this study the contrast medium used was Anglo-Conray, generously supplied by Mr. James A. Peterson, of Mallinckrodt Chemical Works, St. Louis, Missouri.

Again the angiogram (fig. 3) showed that the contrast medium injected through the aortic catheter immediately filled the portal venous system. The sacular structure, which was presumably the site of the arteriovenous fistula, seemed to arise from the right side of the aorta. Again, however, the cinefluorograms suggested the localization to be between the superior mesenteric artery and portal venous system.

An exploratory laparotomy with total body hypothermia was performed on April 19, 1962. There were numerous adhesions within the peritoneal cavity but no free fluid. The liver and spleen appeared essentially normal; the veins in the mesentery of the small bowel were large and dilated. A pronounced thrill was felt in the root of the mesentery of the small bowel. Palpation of this area revealed a pulsatile mass 20 by 16 by 9 mm. Compression of the mass resulted in obliteration of the thrill. A fistulous communication between the superior mesenteric artery and vein was demonstrated. The fistula had a thick wall and the external appearance of a large artery. The diameter of the superior mesenteric vein was 23 mm., and the intraluminal pressure was 500 mm. of saline. After occlusion of the superior mesenteric artery and vein proximally and distally, a transverse incision was made into the anterior aspect of the fistula. The orifice into the artery was 3 by 7 mm. in diameter; that into the vein 4 by 9 mm. After closure of the venous and arterial openings and release of the clamps, the veins in the mesentery of the small bowel decreased in size and venous pressure fell to 90 mm. of saline. In addition to the fistula, false aneurysms about 1 cm. in diameter projected posteriorly from the superior mesenteric artery and from the right side of the aorta just above the origin of the inferior mesenteric artery. It was decided not to repair these.

Postoperatively, the patient had a satisfactory course; the bruit could no longer be heard. Figure 4 is a representative film from the postoperative angiogram showing successful repair of the fistula. The false aneurysms are seen on the posterior aspect of the superior mesenteric artery and the right side of the aorta. It was this latter aneurysm that made interpretation of the preoperative angiogram difficult. One month after surgery the patient was ambulatory, using a long leg brace, and was pain-free as a result of spinal nerve blocks to the second, third, and fourth lumbar nerve roots.

**Discussion**

This case illustrates how arteriovenous fistulas of the portal system may be asymptomatic and occult. Unlike arteriovenous fistu-
las between other major blood vessels, they do not lead to an increase in cardiac output, probably because of the dampening effect of the hepatic vascular bed. Indeed, prior to the development of signs and symptoms of portal hypertension, the only diagnostic clue to the presence of the lesion may be a bruit over the abdomen.

Superior Mesenteric Arteriovenous Fistulas

Rabhan and his co-workers have described the only other reported case of superior mesenteric arteriovenous fistula. Their patient was similar to ours in that he was a young man who received a gunshot wound to the abdomen. After an immediate exploratory laparotomy, the patient was well until 7

Table 1

<table>
<thead>
<tr>
<th>Determinations</th>
<th>Preoperative data</th>
<th>Postoperative data</th>
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</thead>
<tbody>
<tr>
<td>Blood samples (% Saturation)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Arterial</td>
<td>96.4, 97.0</td>
<td>94.8</td>
</tr>
<tr>
<td>SVC</td>
<td>58.8, 59.6, 62.3</td>
<td>70.4</td>
</tr>
<tr>
<td>RA</td>
<td>82.4, 81.5, 75.3</td>
<td>74.6, 72.0</td>
</tr>
<tr>
<td>Hepatic vein</td>
<td>86.0, 86.0</td>
<td>65.2, 63.1</td>
</tr>
<tr>
<td>IVC High</td>
<td>85.6</td>
<td>. . . . .</td>
</tr>
<tr>
<td>Mid</td>
<td>69.2, 66.3</td>
<td>64.6, 66.8</td>
</tr>
<tr>
<td>Low</td>
<td>68.1</td>
<td>70.4</td>
</tr>
<tr>
<td>Cardiac output (L/min.)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>(Dye-dilution method)</td>
<td>5.3, 5.6</td>
<td>4.9</td>
</tr>
<tr>
<td>Pressures (mm. Hg)</td>
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<td></td>
</tr>
<tr>
<td>Hepatic &quot;wedge&quot;</td>
<td>10</td>
<td>8</td>
</tr>
<tr>
<td>Hepatic vein</td>
<td>5</td>
<td>6</td>
</tr>
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months later, when he presented with watery diarrhea and crampy periumbilical pain. The diarrhea became bloody and ascites developed; laparotomy for suspected intestinal obstruction and infarction was accompanied by excessive bleeding, which limited exploration. A second operation was necessary several hours later for continued intraabdominal bleeding. The patient gradually deteriorated and died on his thirteenth hospital day. It was only at postmortem examination that a 2 cm. saccular aneurysm was found 3 cm. from the origin of the superior mesenteric artery, with a 0.5 cm. fistula into the superior mesenteric vein. Intimal proliferation and sclerosis were present in the intrahepatic portal veins. The liver contained areas of regenerating parenchyma, acute necrosis, and fatty infiltration.

Three additional arteriovenous fistulas have been described involving branches of the superior mesenteric artery; all occurred as complications of previous surgery. These cases of arteriovenous fistulas involving smaller arteries appear to have a benign course. The patient described by Movitz and Finne\textsuperscript{2} presented with vague abdominal pain and mild recurrent diarrhea 4 months after abdominal surgery for intestinal obstruction secondary to adhesions and complicated by gangrenous small bowel and mesenteric thrombosis. A bruit was heard over the right lower quadrant of the abdomen and an arteriovenous fistula was suspected. Laparotomy revealed the fistula to be present in the mesentery of the small intestine near its base, in the suture line scar from the previous intestinal resection. This area was surgically excised, and the patient's symptoms abated postoperatively.

Munnell and co-workers\textsuperscript{3} described a patient with regional ileitis who had had an episode of intestinal obstruction in 1952 requiring resection of 45 cm. of terminal small bowel. Over subsequent years she complained of recurrent abdominal cramps, diarrhea, and weight loss. Some 6 years later, a loud continuous murmur and associated thrill were noted in the right lower quadrant of the abdomen. Translumbar aortography showed

**Figure 3**

Representative film and line tracing from preoperative seriogram (20 degrees right posterior oblique projection) in patient with superior mesenteric arteriovenous fistula. The contrast medium was injected through the catheter into the aorta at the level of the bullet fragments. These films suggested that the portal vein filled directly from the aorta, but cinefluorograms suggested that the fistula involved the superior mesenteric vessels. The small insert in the line tracing is from a later film in the seriogram; it shows retention of contrast medium in the false aneurysm arising from the aorta, which led to misinterpretation of the angiogram.

*Circulation, Volume XXVII, May 1963*
filling of the portal vein as well as the aorta. At surgery an arteriovenous fistula arose from a branch of the superior mesenteric artery 2.5 cm. distal to the origin of the middle colic artery. The terminal ileum appeared diseased and was resected with its mesentery, which included the arteriovenous fistula. The patient had a satisfactory postoperative course with gain in weight and development of normal bowel habits. It is difficult to be certain in this case how much of the patient's symptomaticity was attributable to the arteriovenous fistula and how much was the result of the almost completely occluded area of small bowel.

The third case of an arteriovenous fistula in the mesentery was reported by Reams.4 This fistula developed after a gastrectomy and involved the middle colic artery and vein. The patient had noted the presence of an upper abdominal thrill in the immediate postgastrectomy period. During the next 2½ years he complained of vague epigastric discomfort. A translumbar aortogram revealed the lesion which was subsequently excised from the mesenteric without any necessity for bowel resection. The patient's postoperative course was uneventful.

### Splenic Arteriovenous Fistulas

The seven cases in the literature10-16 of arteriovenous fistulas involving the splenic artery have been reviewed recently by Murray and his co-workers.16 Splenic artery aneurysms were involved in the arteriovenous fistula in at least four of these cases. This association led Murray et al.16 to speculate that congenital splenic artery aneurysms may rupture into the splenic vein to produce the shunt. The investigators cite the well-known observation that splenic artery aneurysms rupture during parturition, producing retroperitoneal hemorrhage. Fortuitously, certain of them may rupture into the splenic vein. Of the cases of splenic arteriovenous fistulas in the literature, no attempt was made to correlate them to childbirth except for the patient described by Cassel et al.,15 who presented 2 months after delivery.

The clinical manifestations are usually the result of years of abnormal blood flow, and
portal hypertension is almost invariably present, including gastrointestinal bleeding or ascites, or both. Only one of these seven patients did not have evidence of portal hypertension. This patient was a 67-year-old woman who complained of anorexia, weight loss, lassitude, and vomiting. Laparotomy showed an enlarged spleen containing two arteriovenous fistulas arising from splenic artery aneurysms.\(^{13}\)

Of the six cases with laboratory or clinical evidence of portal hypertension, four had at least one episode of massive gastrointestinal bleeding,\(^{10-12,14}\) and three of these four had demonstrable esophageal varices. One patient\(^{10}\) died with gastrointestinal hemorrhage thought to be secondary to mesenteric venous thrombosis. Esophageal varices were not present. Ascites was the presenting symptom in this patient as well as in the case reported by Cassel and co-workers. In the latter patient, the ascites disappeared following resection of the fistula. Hypersplenism was present in three patients.\(^{11,14,16}\)

**Hepatic Artery-Portal Vein Fistulas**

Of the five reported cases of hepatic artery-portal vein fistulas, two were the result of gunshot wounds and associated with systemic arteriovenous fistulas as well. The other three were probably congenital; the origin of the right hepatic artery was anomalous in one\(^{5}\) and a hepatic artery aneurysm was associated with another fistula.\(^{7}\) Such an association is unusual, however, as only one of 99 cases of hepatic artery aneurysm reported by Sheridan\(^ {17}\) ruptured into the portal system. Esophageal or duodenal varices were demonstrated at autopsy or laparotomy in all of the three patients with congenital hepatic-portal arteriovenous fistulas. Moreover, all three had profuse upper gastrointestinal bleeding. One patient had ascites as well.

**General Comment**

Portal hypertension does not consistently develop in the experimental animal as a result of systemic-portal fistulas. Anastomosis of hepatic, splenic, or renal arteries to the portal vein or its tributaries does not produce more than transient elevation of portal pressure.\(^ {18}\) Nor does chronic progressive occlusion of the portal vein lead to portal hypertension.\(^ {19}\) However, if these two experimental technics are combined in the dog,\(^ {20}\) then pressures in the portal system remain elevated above normal levels and esophageal varices are consistently found.

Notwithstanding failure of attempts to produce portal hypertension in laboratory animals by portal arteriovenous shunting alone, it is certain that these lesions produce portal hypertension in man. Of the 17 systemic-portal fistulas reported in the literature (including this case), portal system pressure was measured directly or indirectly in seven cases and was found to be elevated in all.\(^ {4,7,8,11,12,16}\) The superior mesenteric venous pressure of 500 mm. of saline observed in our case is especially striking.

*Hepatic venous oxygen content and arterial-hepatic venous oxygen difference determined before and after operation in patient with superior mesenteric arteriovenous fistula.*

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The arterial-hepatic vein oxygen differences in the present case of 2 volumes per cent pre-operatively and 5.8 volumes per cent post-operatively (fig. 5) indicate that the shunt flow was relatively high. Cardiac output (5.3, 5.6 L./min.) however, was not elevated above normal levels, nor has increased cardiac output been described in other cases unless a systemic arteriovenous shunt was also present.

The clearance of bromsulfalein by our patient before surgery was at least twice as rapid as after surgery (fig. 1). This phenomenon has been described previously in human subjects with portal arteriovenous fistulas and in dogs with experimental systemic-portal fistulas. The rapid extraction is probably the result of increased blood flow to the liver secondary to the arterialization of portal vein flow produced by the fistula.

In order to make the diagnosis of an arteriovenous fistula involving the portal system, the physician must routinely listen for bruits over the abdomen, and, if present, recognize their possible significance and attempt to define their origin. The characteristic feature of the bruit of an arteriovenous fistula is its continuous quality. Following thorough clinical and laboratory evaluation of the patient, the diagnosis is made by radiographic visualization of the fistula itself. Injection of contrast medium can be accomplished by a translumbar needle or catheter inserted in the femoral artery. The latter method is probably safer, in view of the increased vascularity that may be associated with arteriovenous fistulas. In such studies on this patient the contrasting virtues of angiography and cinefluorography were apparent. The angiogram outlined vascular size and detail concisely, whereas the actual flow patterns were delineated by the cinefluorograms.

Summary

A patient is described with a traumatic arteriovenous shunt from the superior mesenteric artery to the superior mesenteric vein. The patient had no symptoms referable to this fistula 3 months after a gunshot wound to the abdomen. The diagnostic feature was a continuous bruit in the supraumbilical area. The radiographic technics used in determining the nature of this patient’s fistula are described. This is the first reported case of a traumatic superior mesenteric arteriovenous fistula suspected clinically, demonstrated radiographically, and successfully repaired surgically. Although experience with such fistulas is limited, the fatal outcome of the only other case in the literature (not recognized ante mortem) indicates the need for careful auscultation over all abdominal surgical incisions and sites of trauma at regular intervals.

References

The Hazards of Progress

The greatest risk that confronts us is not precisely unawareness of new discoveries and the latest findings. No, it is the slow breaking down of the rigour of our scientific method which was devised to ensure certain bases for our knowledge and to demand proofs before admitting the truth of what is new. It is the risk of forgetting Cartesian doubt, that doubt which has been the backbone of our scientific posture. The flood of medical literature not infrequently impedes serene reflection and drowns critical judgement. What we gain in erudition we lose in wisdom.

There are yet other risks. Those prodigies of technique which fill us with enthusiasm and which all too easily deceive us, sometimes cause us to confound the delicacy of the procedure with the rigour of the method. The complicated and wonderful instrument becomes both an aid and a menace. Furthermore there is the heady speed at which we live and the contagion of enthusiasm which finally produces collective illusions and turns us into victims of fashions. Who can doubt that we are suffering these things? It is enough to remember the number of drugs that only yesterday filled the magazines with their flattering results; everybody used them. And today? Who remembers their names?—Dr. Ignacio Chávez. Speech delivered at the Inaugural Ceremony of the IV World Congress of Cardiology. Universidad Nacional Autónoma de México, México, D.F., 1962, p. 7.
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Circulation. 1963;27:943-950
doi: 10.1161/01.CIR.27.5.943
Circulation is published by the American Heart Association, 7272 Greenville Avenue, Dallas, TX 75231
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Print ISSN: 0009-7322. Online ISSN: 1524-4539

The online version of this article, along with updated information and services, is located on the World Wide Web at:
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