Case Report

Iliac Arteriovenous Fistula With Renal Insufficiency, Ascites, Hepatomegaly, and Abnormal Liver Test Results

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Arteriovenous fistula presents rarely with liver involvement. A 73-year-old man had new-onset ascites, hepatomegaly, and abnormal liver and renal function test results. An abdominal computed tomogram revealed a 7.6-cm internal iliac aneurysm but no other abnormality to account for his ascites. An aortogram demonstrated a 1.5-cm internal iliac arteriovenous fistula that subsequently was repaired, leading to resolution of his symptoms and laboratory abnormalities. High-output cardiac failure should be considered in the differential diagnosis of patients with new-onset massive ascites, hepatomegaly, and liver test abnormalities.

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ALT = alanine aminotransferase; AST = aspartate aminotransferase; AV = arteriovenous

A common cause for major extrasplanchnic abdominal arteriovenous (AV) fistulas is the rupture of an aortic or iliac aneurysm into a major adjacent vein, occurring in 3% to 4% of all ruptured aneurysms.[1,2] Other causes such as trauma or retroperitoneal surgery (most frequently spine surgery) account for less than 20% of AV fistulas. Isolated iliac aneurysms account for 15% of fistulas[1] and tend to have a more insidious onset. The most common complaints are abdominal, low back, or hip pain, occurring in 78% of cases.[1] The most common physical findings are abdominal bruit (60%), pulsatile abdominal mass (55%), and lower extremity edema, with or without associated skin mottling (40%). Up to 35% of patients may present with symptoms and signs of high-output congestive heart failure. Hematuria and oliguric renal failure are seen in up to 20% of cases.[1] Kazmier and Harrison[3] identified 3 presentations in patients with spontaneous fistulas: high-output cardiac failure, hypotension-oliguria, and back or abdominal pain, all in the presence of a pulsatile abdominal mass and bruit. This triad of pain, pulsatile mass, and bruit occurs in less than 15% of patients.[4] In most cases, the diagnosis is made preoperatively by aortic angiography and, uncommonly, by computed tomography with contrast or Doppler ultrasonography.[5,6] However, in up to 25%, the diagnosis is not made before surgery.[1]

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Surgery is the primary treatment for this condition, consisting of fistula closure and aneurysm repair, usually with an aortic or aortoiliac graft.[4] Operative mortality is 10% to 35% and higher in cases not recognized preoperatively.[1,4]

We report a case of spontaneous iliac AV fistula presenting with renal insufficiency, ascites, and liver test abnormalities.

REPORT OF A CASE

A 73-year-old man presented with a 2-week history of increased abdominal girth, nausea, vomiting, and constipation without abdominal or chest pain. Dyspnea on exertion and decreased urinary output had begun prior to his abdominal symptoms, following a mild upper respiratory infection. He had never smoked and denied alcohol abuse. A rectosigmoid cancer was resected 12 years earlier, without adjuvant therapy. Follow-up colonoscopy 1 year before this current illness showed no recurrence. He had no history of or risk factors for liver disease.

At presentation elsewhere, his aspartate aminotransferase (AST) level was 868 U/L, alanine aminotransferase (ALT) was 814 U/L, and serum creatinine was 2.5 mg/dL. An abdominal computed tomographic scan showed ascites and a 12.3×7.6-cm left internal iliac artery aneurysm with no evidence of rupture. The patient was transferred to our institution for further evaluation of possible progressive liver failure.

Examination revealed a moderately obese man breathing comfortably at rest. Blood pressure was 150/70 mm Hg; heart rate, 90 beats/min; respiratory rate, 24 breaths/min; and temperature, 36°C. Skin and sclerae were anicteric. Jugular venous pressure was increased to 12 cm H_2O. The lungs had bibasilar inspiratory rales without evidence of pleural effusion. Grade 2/6 holosystolic murmur was

Table 1. **Laboratory Values on Presentation to Mayo Clinic***

Test	Value	Reference range
Hemoglobin (g/dL)	13.5	13.5-17.5
WBC count ($\times 10^9$/L)	11.9	3.5-10.5
Platelet count ($\times 10^9$/L)	207	150-450
INR	1.77	0.8-1.2
PTT (s)	26	21-33
Sodium (mEq/L)	135	135-145
Potassium (mEq/L)	4.1	3.6-4.8
Bicarbonate (mEq/L)	21	22-29
BUN (mg/dL)	106	8-24
Creatinine (mg/dL)	2.7	0.8-1.2
Albumin (g/dL)	3.7	3.5-5.0
AST (U/L)	926	12-31
ALT (U/L)	1163	10-45
Bilirubin (mg/dL)		
Total	1.7	0.1-1.1
Direct	0.8	0.0-0.3
Alkaline phosphatase (U/L)	216	98-251
LDH (U/L)	766	112-257

*ALT = alanine aminotransferase; AST = aspartate aminotransferase; BUN = blood urea nitrogen; INR = international normalized ratio; LDH = lactic dehydrogenase; PTT = partial thromboplastin time; WBC = white blood cell.

best heard at the left midsternal border. The abdomen was moderately distended with a fluid wave and shifting dullness consistent with ascites. There was hepatomegaly (17-cm midclavicular line) without splenomegaly. Distant bruit was heard over the left lower quadrant. Bilateral asymmetrical lower extremity edema to the knee was noted.

Initial laboratory data (Table 1) showed markedly increased transaminase and creatinine levels. Serologic studies for hepatitis were negative, and iron studies yielded normal results. Arterial blood gas analysis on room air showed a pH of 7.44, P_{O_2} of 87 mm Hg, P_{CO_2} of 26 mm Hg with an oxygen saturation of 96%, bicarbonate of 21 mEq/L, and an A–a gradient of 36 mm Hg, consistent with respiratory alkalosis and metabolic compensation. A disseminated intravascular coagulation panel had abnormal D-dimers (>500 μg/L), positive fibrin split products and soluble fibrin monomers, and borderline low fibrinogen level of 153 mg/dL. Peripheral blood smear had no schistocytes.

Chest radiography showed cardiomegaly with signs of pulmonary vascular hypertension and small bilateral pleural effusions. Electrocardiography revealed normal sinus rhythm with first-degree atrioventricular block. Abdominal ultrasonography showed increased liver echogenicity (consistent with fatty infiltration) and moderate ascites. Decreased flow velocity was noted in the portal system, although the veins were patent with forward flow. There was no evidence of obstruction of the hepatic veins. Ascitic fluid had a total protein of 3.0 g/dL, albumin of 1.5 g/dL, and unremarkable cytologic, differential, and culture findings. The serum ascites albumin gradient of 2.2 g/dL and total protein greater than 2.5 g/dL were consistent with a suprahepatic cause of ascites, and further testing was obtained.

Echocardiography showed normal left ventricular function, with an ejection fraction of 65% to 70%, moderate tricuspid regurgitation, moderate right ventricular enlargement with increased pulmonary artery systolic pressure of 44 mm Hg, and no atrial or ventricular septal defects. Lower extremity Doppler ultrasonography was negative for venous thrombosis. A V/Q scan was interpreted as showing low probability for pulmonary embolism. Arteriography showed a 1.5-cm fistula between the left common iliac artery aneurysm and the left common iliac vein (Figure 1).

The patient underwent operative repair of the AV fistula and recovered uneventfully. Postoperative echocardiography showed resolution of right ventricular overload and elevated pulmonary pressure. At follow-up 2 months later, he was asymptomatic without heart failure or ascites. His liver test findings had normalized, and the serum creatinine level had improved to 1.4 mg/dL.

DISCUSSION

Few reported cases of AV fistulas have described marked liver involvement, such as hepatomegaly, ascites, jaundice, or coagulopathy.[7-17] More commonly, patients present with nonspecific complaints such as abdominal pain, anorexia, nausea, vomiting, or rectal bleeding.[8,9] We were unable to find another case of extrasplanchnic AV fistula presenting with marked subacute liver injury as demonstrated by severe liver test abnormalities. This atypical presentation and the subacute course explain the rather extensive work-up that preceded the final diagnosis.

Systemic AV fistulas result in considerable hemodynamic alterations due to diversion of blood flow from the high-resistance arterial circuit into the venous circuit. The physiologic effects of this shunt[18,19] depend on the size of the fistula and the caliber of the involved vessels. Arterial flow into the venous system increases venous volume and pressure and consequently venous return.[20] A simultaneous decrease in peripheral vascular resistance due to shunting results in a hyperdynamic circulation characterized by increased stroke volume, heart rate, and cardiac output.[21,22] One study reported the mean preoperative cardiac output of 12.2 L/min decreased to 5.4 L/min after surgery.[1] The increased workload may often lead to cardiac failure.[23] Abnormalities in liver test results are noted frequently in patients with congestive heart failure.[24,25] They are far more common in patients with acute (49%) vs chronic (5%) heart failure, usually 2 to 3 times above normal levels for AST and ALT.[24,25] Patients with severe rheumatic heart disease and with acute heart failure with hypotension or shock may

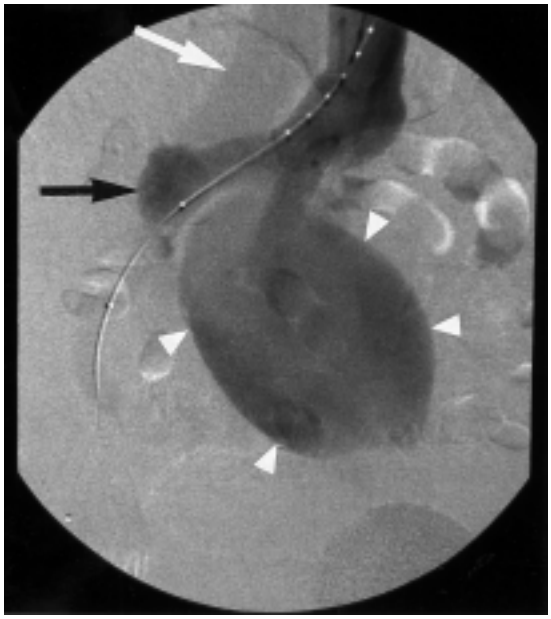

Figure 1. Distal aortic arteriogram shows a large common iliac artery aneurysm (arrowheads) and early filling of the inferior vena cava (white arrow) simultaneously with the contralateral common iliac artery (black arrow), indicating the presence of arteriovenous fistula.

have more extreme elevations.[25] Increased serum bilirubin level has been reported in 27% to 70% of patients with congestive heart failure, especially in those with acute and predominantly right-sided failure.[25] A prolonged prothrombin time was reported in 80% to 90% of patients with liver congestion, and it usually ranges between 50% and 80% of normal, again, more common and more severe in patients with acute heart failure.[25]

The explanation for the unusually high transaminase levels in our patient is unclear. Ischemic hepatitis or "shock liver" may be associated with marked increases in transaminase levels and rapid improvement after resuscitation.[26] However, our patient had no evidence of hemodynamic deterioration at any point during his illness, and preoperatively his liver test results remained elevated without marked fluctuations. Moreover, his liver test abnormalities as well as ascites and renal failure resolved quickly after surgery. It is difficult to determine if the compensated disseminated intravascular coagulation contributed to the liver injury, in addition to the passive congestion from heart failure, since both resolved after surgery.

We describe an interesting and unusual presentation of an AV fistula with hepatic and renal dysfunction and an excellent outcome after surgery. To our knowledge, no similar cases have been reported previously. Although high-output cardiac failure is an unusual cause of new-onset massive ascites, hepatomegaly, and marked liver test abnormalities, it should be considered in the differential diagnosis in these patients, particularly those with known aneurysms or unexplained bruits.

REFERENCES

1. Brewster DC, Cambria RP, Moncure AC, et al. Aortocaval and iliac arteriovenous fistulas: recognition and treatment. *J Vasc Surg*. 1991;13:253-264.
2. Pagni S, Halene S, Kwass W, Khachane V. Ruptured aortic pseudoaneurysm: a rare presentation as aortocaval fistula. *J Cardiovasc Surg (Torino)*. 1997;38:165-168.
3. Kazmier FJ, Harrison CE Jr. Acquired aortocaval fistulas. *Am J Med*. 1973;55:175-183.
4. Davis PM, Gloviczki P, Cherry KJ Jr, et al. Aorto-caval and ilio-iliac arteriovenous fistulae. *Am J Surg*. 1998;176:115-118.
5. Mansour MA, Russ PD, Subber SW, Pearce WH. Aorto-left renal vein fistula: diagnosis by duplex sonography. *AJR Am J Roentgenol*. 1989;152:1107-1108.
6. Sheward SE, Spencer RR, Hinton TR, Hightower CW, Patel PV, Suckarieh JG. Computed tomography of primary aortocaval fistula. *Comput Med Imaging Graph*. 1992;16:121-124.
7. Alexander JJ, Imbembo AL. Aorta-vena cava fistula. *Surgery*. 1989;105:1-12.
8. Nennhaus HP, Javid H. The distinct syndrome of spontaneous abdominal aorto-caval fistula. *Am J Med*. 1968;44:464-473.
9. Reckless JPD, McColl I, Taylor GW. Aorto-caval fistulae: uncommon complications of abdominal aortic aneurysms. *Br J Surg*. 1972;59:461-462.
10. Morris GC Jr, Arnold HF, McMurrey JD. Acute spontaneous aorto-caval fistula. *JAMA*. 1962;182:72-73.
11. Du Toit DF, van der Merwe DM, Groenewald JH, DeRoubaix JA. High-output cardiac failure associated with an iliac artery aneurysm and arterio-venous fistula. *S Afr Med J*. 1983;63:293-294.
12. Beall AC Jr, Cooley DA, Morris GC, Debakey ME. Perforation of arteriosclerotic aneurysm into inferior vena cava. *Arch Surg*. 1963; 86:137-146.
13. Eiseman B, Hughes RH. Repair of an abdominal aortic-vena caval fistula caused by rupture of an atherosclerotic aneurysm. *Surgery*. 1956;39:498-504.
14. Ross HB. Aneurysm of abdominal aorta with aorto-caval fistula. *Proc R Soc Med*. 1962;286-288.
15. Shumacker HB Jr, Jontz J. Rupture of abdominal aortic aneurysm into inferior vena cava: successful surgical repair. *Am Surg*. 1956;22:1182-1185.
16. Guillem P, Mohieddine T, Hassan H, Triboulet JP. Acute liver failure revealing aorto-caval fistula. *Gastroenterol Clin Biol*. 2000; 24:461-463.
17. Schneider M, Schellong S, Hanig V, Voigt JU, Daniel WG. Endovascular stent in spontaneous arteriovenous fistula of the common iliac artery [in German]. *Vasa*. 1997;26:228-231.
18. Holman E. The physiology of an arteriovenous fistula. *Arch Surg*. 1923;7:64-82.
19. Holman E. Clinical and experimental observations on arteriovenous fistulae. *Ann Surg*. 1940;112:840-878.
20. Nickerson JL, Cooper FW Jr, Robertson R, Warren JV. Arterial, atrial and venous pressure changes in the presence of an arteriovenous fistula. *Am J Physiol*. 1951;167:426-434.
21. Frank CW, Wang HH, Lammerant J, Miller R, Wegria R. An experimental study of the immediate hemodynamic adjustments to acute arteriovenous fistulae of various sizes. *J Clin Invest*. 1955; 34:434-438.
22. Elkin DC, Warren JV. Arteriovenous fistulas: their effect on the circulation. *JAMA*. 1947;134:1524-1528.
23. Shumaker HB Jr, Stah NM. A study of the cardiac frontal area in patients with arteriovenous fistulas. *Surgery*. 1949;26:928-944.
24. Kubo SH, Walter BA, John DHA, Clark M, Cody RJ. Liver function abnormalities in chronic heart failure. *Arch Intern Med*. 1987; 147:1227-1230.
25. Dunn GD, Hayes P, Breen KJ, Schenker S. The liver in congestive heart failure: a review. *Am J Med Sci*. 1973;265:174-189.
26. Kamath PS. Clinical approach to the patient with abnormal liver test results. *Mayo Clin Proc*. 1996;71:1089-1095.