Delayed presentation of traumatic aortocaval fistula: A report of two cases and a review of the associated compensatory hemodynamic and structural changes

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Chronic aortocaval fistula (ACF) is a rare complication of gunshot wounds to the abdomen. Herein we report two cases of traumatic ACF: one asymptomatic and the other presenting with congestive heart failure (CHF) 20 and 30 years, respectively, after their initial injury. The recent onset of CHF, the presence of a continuous abdominal bruit, and, in the second patient, a history of penetrating trauma suggested the diagnosis of ACF. The diagnosis was confirmed by computed tomography scanning in both patients. Surgical repair of the ACF in the symptomatic patient resulted in resolution of the CHF and reversed the dilatation of the aorta and inferior vena cava. The asymptomatic patient was lost to follow-up. CHF in a young male patient with a history of penetrating abdominal trauma should alert the surgeon to this rare complication. (J Vasc Surg 2006;43:836-40.)

Although infrequent, penetrating injuries of the aorta and vena cava are among the most lethal injuries to the intra-abdominal vessels, with mortality rates ranging from 40% to 45%.[1] Gunshot or fragment wounds, stab wounds, and iatrogenic injuries account for 90% and blunt trauma accounts for the remaining 10% of all such vascular injuries.[1-3]

An unknown number of patients with penetrating injuries to the aorta or vena cava succumb from exsanguination at the scene of the injury. Those who can be resuscitated are transported, often in profound hypovolemic shock, to trauma centers, where emergent laparotomy to control hemorrhage and treatment of concomitant injuries to the adjacent viscera can be undertaken.[3,4]

Delayed presentation of injuries to the abdominal aorta or inferior vena cava is uncommon and may manifest as pseudoaneurysms, aortic or visceral branch occlusion, or arteriovenous (AV) fistula. Herein we describe two patients with aortocaval fistula (ACF) presenting 20 and 30 years after their initial injuries. The clinical presentation, compensatory hemodynamic and structural changes, and management are discussed.

CASE REPORTS

Patient 1. A 50-year-old man presented to his primary care physician complaining of chronic backache localized to the exit site of a previous gunshot wound to the abdomen. Twenty years previously, he had undergone exploratory laparotomy, partial colectomy, and colostomy for a shotgun wound to the abdomen. He was told that there had been an injury to the aorta but was unable to recall any details. His recovery from the initial laparotomy and subsequent colostomy closure was uneventful.

On physical examination, the patient appeared healthy, with a pulse rate of 85 beats per minute and a blood pressure of 130/50 mm Hg. The cardiac examination revealed a regular rate and rhythm without gallops or murmurs. The lung fields were clear. There was a midline abdominal incision from the previous laparotomy and left lower quadrant incision from the colostomy closure. A continuous bruit was heard over the lower abdomen. There was no evidence of pedal edema, and all the peripheral pulses were palpable.

Plain abdominal radiographs demonstrated the presence of gunshot fragments. A contrast-enhanced spiral computed tomography (CT) scan revealed mild dilation of the infrarenal aorta with calcification consistent with a small aortic aneurysm. Just below the aortic dilatation, there was circumferential enlargement of the adjacent inferior vena cava (Figs 1 and 2). The patient refused repair of his ACF and was lost to follow-up.

Patient 2. A 45-year-old man presented to the emergency room with complaints of shortness of breath and a feeling of fullness in his upper abdomen. Over the past 6 months, he was unable to maintain employment as a welder and could walk only short distances because of exertional dyspnea and lower extremity edema. Thirty years previously, he had sustained a gunshot wound to the abdomen for which he underwent exploratory laparotomy, with an uneventful recovery.

On physical examination, the patient was in mild respiratory distress. His pulse rate was 93 beats per minute, and his blood pressure was 136/76 mm Hg. Jugular venous distention to 12 cm above the clavicle was present. Lung examination revealed bilateral basilar rales. Auscultation of the heart revealed atrial fibrillation and a grade 3/6 systolic murmur with an S3 gallop. A thrill and a

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Competition of interest: none.

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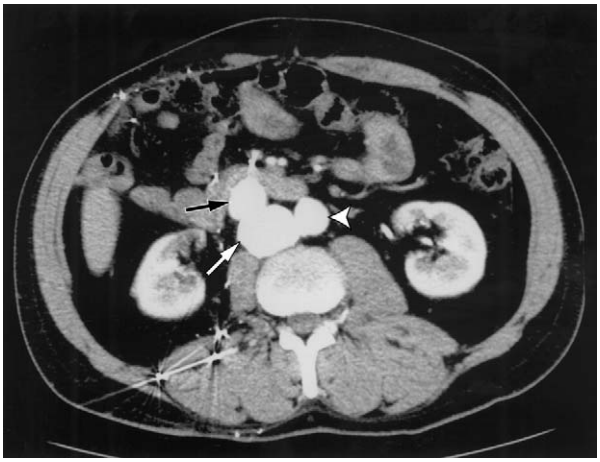

Fig 1. Axial contrast-enhanced computed tomographic scan at the level of L2 shows a traumatic pseudoaneurysm *(white arrow)* to the right of the aorta *(arrowhead)* and posterior to the inferior vena cava *(black arrow)*. Note the multiple gunshot pellets in the region of the right quadratus lumborum muscle.

bruit were detected over the mid abdomen. There was moderate lower extremity edema; the peripheral pulses were all palpable.

An abdominal radiograph, as in the first patient, revealed the presence of gunshot fragments in the abdomen. A chest roentgenogram showed marked cardiomegaly. Cardiac catheterization revealed a mean right atrial pressure of 14 mm Hg, a right ventricular pressure of 55/14 mm Hg, and a pulmonary capillary wedge pressure of 21 mm Hg. The patient's cardiac output was 13.3 L/min, and his systemic vascular resistance 476 dynes/cm^2. The inferior vena caval oxygen saturation was 92%, compared with 60% in the superior vena cava.

An axial contrast-enhanced spiral CT scan showed a markedly dilated inferior vena cava, which was 8 cm in diameter at its widest point. A curvilinear calcification of the aorta immediately adjacent to the ACF was present at the level of the renal arteries (Figs 3 and 4). The patient was taken to the operating room for repair of his ACF. At exploration, the ACF was identified on the right side of the aorta at the level of the right renal artery and left renal vein. The left renal vein was divided to improve exposure of the fistula. After proximal and distal control of the aorta and inferior vena cava was obtained, the pseudoaneurysm was excised, and the defect in the aorta was repaired with interrupted pledgeted polypropylene sutures. The defect in the vena cava was closed with running polypropylene suture, and the left renal vein was reapproximated. The patient had an uneventful recovery. When seen 2 years after surgery, he was asymptomatic but was still taking warfarin for persistent atrial fibrillation. His chest roentgenogram showed a significant improvement in the cardiac silhouette.

DISCUSSION

ACF, first described by Syme[5] in 1831, is most commonly (80%-90%) secondary to rupture or erosion of an abdominal aortic aneurysm into the inferior vena cava. The remaining 10% to 20% are usually the result of penetrating or iatrogenic trauma.[6]

Penetrating trauma due to gunshot and fragment or stab wounds is the most common cause of traumatic ACF. Less commonly, iatrogenic injury during arterial and cardiac catheterization, lumbar disk surgery, and, rarely, blunt trauma have been implicated as etiologic factors.[4,7,8]

There are two modes of presentation of patients with traumatic ACF. Most patients present acutely and either die from exsanguination at the scene of the injury or are transported to major trauma centers, profoundly hypotensive and with extensive associated injuries. The mortality rate in this subgroup of patients ranges from 40% to 45% and is usually the result of the combination of associated injuries, difficulties obtaining exposure, intraoperative exsanguination, hypothermia, and coagulopathy from prolonged surgery.[1,2]

The second, less common, mode of presentation of patients with ACF is usually weeks, months, or years after the initial injury, as was the case in the two patients reported here.[8] The first patient had no symptoms directly attributable to his ACF and was lost to follow-up. The second patient presented in florid high-output congestive heart failure (CHF), as manifested by a cardiac output of more than 13 L/min and systemic vascular resistance of less than 500 dynes/cm^2. Among the factors that influence the severity of the compensatory changes in patients with ACF are the size of the fistula, the diameter of the component artery and vein, the proximity to the heart, and the age of the patient. Young, healthy males are the most likely to sustain traumatic ACF and, because they have very little intrinsic cardiac disease, are more able to adapt to the hemodynamic changes that result from the ACF. They are therefore more likely to have a delayed presentation.

The natural history of an ACF is for the fistula to enlarge over time and only rarely close spontaneously. The adaptive response to a traumatic ACF is manifest locally at the site of injury by a continuous bruit or thrill and systemically as volume overload. Unlike AV fistulas located distally in the extremities, arterial insufficiency is unusual with ACF. In contrast, venous obstruction below the fistula manifests as lower extremity edema and is often a presenting symptom of ACF.

The dilatation of the aorta (3 cm) proximal to the fistula as a result of the increase in blood-flow velocity and shear stress in both these patients resolved in the patient treated surgically. The dilatation and calcification of the proximal aorta evident on both CT scans are of considerable interest in view of the potential contribution of hemodynamic flow disturbances to the development of atherosclerotic aneurysms. Whether the calcification in the aorta observed in these two patients was the end result of prolonged hemodynamic injury or merely evidence of heterotopic or dystrophic calcification of the intramural or periadventitial hematoma as a result of the initial injury is unclear.

Both the artery and vein proximal to the AV fistula undergo structural changes in response to the altered hemodynamic stress. A characteristic finding is an increase in collagen and elastin elements in the media of the artery and

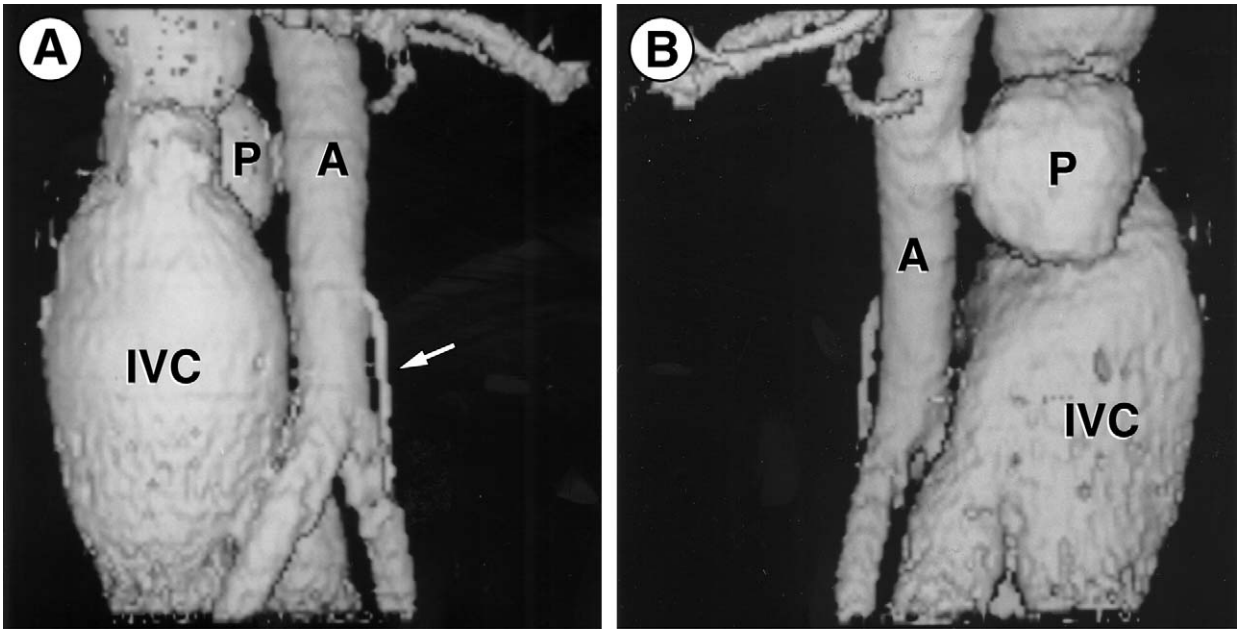

Fig 2. A, Three-dimensional reconstruction and shaded surface rendering of the retroperitoneal vessels after contrast-enhanced helical computed tomographic scanning; the posterior view demonstrates the aortic *(A)* neck of the pseudoaneurysm *(P)* and its position to the dilated inferior vena cava *(IVC)*. **B,** Right anterior oblique view of the aorta (A), IVC, pseudoaneurysm (*P*), and inferior mesenteric artery *(arrow)*.

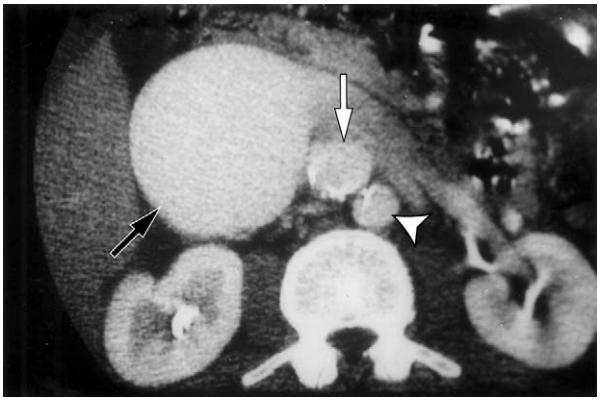

Fig 3. Axial contrast-enhanced computed tomographic scan at the level of the renal hilum demonstrates a partially calcified pseudoaneurysm *(white arrow)* located between the aorta *(arrowhead)* and the massively dilated inferior vena cava *(black arrow)*. The left renal vein is dilated and displaced anteriorly.

the vein and more vaso vasorum in the venous adventitia. Eventually, however, the arterial wall undergoes degenerative changes with atrophy of smooth muscle fibers, fragmentation and a reduction in the number of elastic elements, and, ultimately, the formation of an atherosclerotic plaque. These structural changes may be irreversible if the fistula persists for prolonged periods (>2 years). The distal artery and vein tend to remain unchanged.[9]

The physiological changes associated with ACF have been elucidated by the experimental studies of Holman,[10] who observed an increase in pulse rate, a decrease in blood pressure, and an increase in cardiac size in developing dogs after creation of an ACF. The cardiomegaly was accompanied by a corresponding increase in the diameter of the aorta and vena cava proximal to the fistula. Growing animals were able to tolerate considerable degrees of cardiac enlargement and dilation without decompensation.[10]

Several factors have been implicated in the dilatation of the aorta and vena cava proximal to the fistula. The most obvious physiologic change concomitant with the formation of an AV fistula is the severalfold increase in blood flow, especially during diastole.[10,11] Other etiologic factors implicated in the aortic dilatation associated with an ACF include vibratory forces and impairment of the nutrition of the arterial wall as a result of changes in the number and diameter of the vaso vasorum.[11] D'Silva and Fouche[12] have provided experimental evidence to support the influence of increased blood flow on progressive elongation and dilatation of up to 30% in the femoral artery proximal to an AV fistula. It seems likely that all these etiologic factors acting in concert account for the changes in diameter observed in the aorta and vena cava proximal to the fistula.

The molecular mechanisms that underlie the arterial elongation and dilation associated with ACF are not fully understood. Sho et al[13] and Guzman et al[14] have provided evidence for the role of metalloproteinases 2 and 9, nitric oxide, and other as-yet unrecognized cellular signals in the structural changes that cause the artery to dilate and elongate. The dilatation and elongation of the aorta were reversed after surgical correction of the ACF

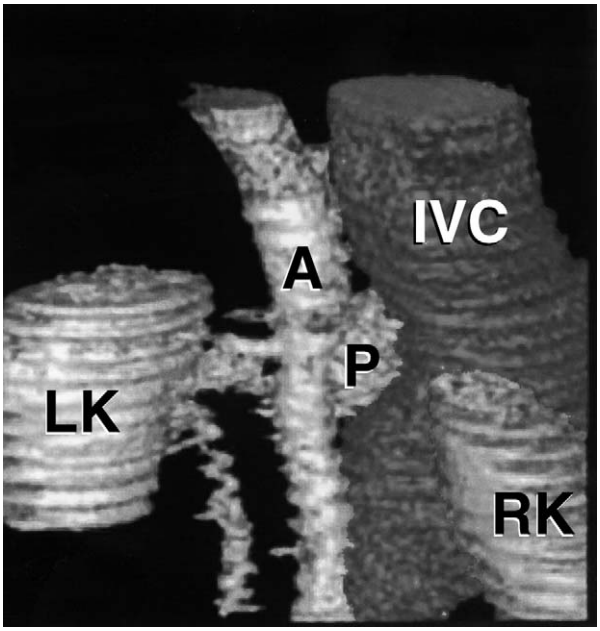

Fig 4. Three-dimensional reconstruction of a helical computed tomographic scan with shaded surface rendering shows the left and right kidney *(LK* and *RK)*, the aorta *(A)*, and the dilated inferior vena cava *(IVC)*. The pseudoaneurysm *(P)* is associated with the aortocaval fistula.

in the symptomatic patient, thus confirming the observations of Holman.[10] The cardiac silhouette, however, remains mildly enlarged at 2 years. A possible explanation for the persistence of mild cardiomegaly comes from experimental and human studies demonstrating that the sustained volume overload caused by an AV fistula produces cardiac myocyte hypertrophy, biventricular dilation, and, ultimately, ventricular decompensation. It is possible that prolonged hemodynamic stress results in alterations in the remodeling of the ventricular wall, which ultimately results in irreversible cardiomegaly.[11,15,16] These changes in myocardial function in patients with high-output cardiac failure associated with AV fistulas may be mediated via the renin-angiotensin mechanism.[16] Potentially, the administration of an angiotensin-converting enzyme inhibitor may improve myocardial function. Whether the administration of an angiotensin-converting enzyme inhibitor would prevent or improve cardiomegaly in symptomatic patients or whether these agents could be used to prevent decompensation in asymptomatic patients remains unclear because of the small number of patients with this condition.[16]

Cut film or digital subtraction catheter angiography has been the standard preoperative imaging technique used in patients with abnormalities of the abdominal aorta and inferior vena cava. Alternative noninvasive imaging techniques include color Doppler sonography, echocardiography, CT angiography, and magnetic resonance angiography.[17-20] Although color Doppler ultrasonography can confirm the presence of an ACF, contrast-enhanced spiral or helical CT scanning with three-dimensional reconstruction is superior to ultrasonography in demonstrating the surgically relevant vascular anatomy. Multislice helical CT scanning has largely replaced conventional angiography as the gold standard for the imaging of aortic pathology.[19] Magnetic resonance angiography and digital subtraction angiography with carbon dioxide as a contrast agent[20] are excellent alternatives to CT angiography in patients with a history of contrast reaction or renal failure.

The treatment of traumatic or secondary ACF is presently surgical closure of the fistula. Sufficient blood and blood products should be available, because hemorrhage can be catastrophic. The aorta is mobilized above and below the fistula and occluded between clamps after systemic heparinization. As can be seen on the CT scan of these patients, the vena cava is extremely enlarged, and great care should be taken when this vessel is mobilized circumferentially. Occlusion of the vena cava with vascular clamps, occlusion balloons, or sponge stick compression above and below the defect has been used to control venous bleeding and to allow oversewing of the defect. In our patient, the dense fibrous capsule of the intervening pseudoaneurysm (present in approximately 60% of patients with traumatic ACF) required excision before the defects in the vena cava or aorta could be located and repaired.

ACF associated with ruptured abdominal aortic aneurysms and traumatic ACF are both amenable to endovascular repair. This mode of therapy may be the preferred treatment if the diagnosis can be established before surgery.[21-23] Careful assessment of graft size is necessary, especially because the dimensions of the aorta can be expected to decrease to normal once the fistula has closed. An aortic extension cuff of appropriate diameter and length would close the defect in the aorta. In patients with ACF associated with abdominal aortic aneurysms,[22] the potential for endoleaks exists and, if untreated, may result in persistence of the fistula. Persistence of the ACF has not been a problem in the short-term follow-up of patients with aneurysm-associated ACF treated with aortic endografts.[23]

In summary, these two patients evaluated 20 and 30 years after their initial injuries, one asymptomatic and the other with symptoms of CHF, represent the clinical extremes of the presentation of patients with ACF. In both patients, helical CT imaging studies were helpful in delineating the anatomy of the ACF and allowed planning of the treatment in the patient who underwent successful surgical repair. The recent onset of CHF in a young male patient with a history of penetrating abdominal trauma should alert surgeons to this rare but remedial complication.

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