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Spontaneous aortocaval fistula presenting with acute liver and renal failure: A case report

Akut karaciğer ve böbrek yetmezliğine yol açan spontan aortakaval fistüllü olgu sunumu

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Spontaneous aortocaval fistula is rare, occurring only in 3%-6% of all ruptured abdominal aortic aneurysms. A definitive diagnosis of aortocaval fistula is sometimes difficult, as the classic diagnostic signs (pulsatile abdominal mass with bruit, high-output heart failure and acute dyspnea) are present only in 20% - 50% of all such cases. Pre-operative diagnosis is crucial, as adequate preparation has to be made for the massive bleeding expected at operation. Surgical repair of aortocaval fistula is now standardized repair of the fistula. We report herein a case of spontaneous aortocaval fistula, which presented with liver and renal failure.

Key words: Aortocaval fistula, acute liver and renal failure

Spontan aortakaval fistül nadirdir ve rüptüre abadominal aort anevrizuvalarm %3-6'sını oluşturur. Tanı bazen güçtür ve klasik belirtiler yalnızca %20-50 hastada görülür. Preopenatif tanı, uygun hazırlık için kritiktir. Cerrahi onarım ve standart tedavi yoktur. Burada akut karaciğer ve böbrek yetmezliğine yol açan bir spontan fistül olgusu sunulmuştur.

Anahtar Kelimeler: Aortakaval fistül, akut karaciğer ve böbrek yetmezliği

INTRODUCTION

Aortocaval fistula is reported to be associated with 3%-6% of all ruptured abdominal aortic aneurysms (1, 2). In the physical examination, the presence of pulsatile abdominal mass with bruit, high-output heart failure, acute dyspnea and low back pain should raise the suspicion. Some factors may influence the clinical manifestation of aortocaval fistulas, including the origin, size, and location of the fistula, age of the patient and previous history of liver-renal diseases. A definitive diagnosis offers advantages in surgical management. Computed tomography (CT) is often the initial imaging method, but angiography, duplex scanning, and magnetic resonance imaging (MRI) can also be used for diagnosis. Surgical repair of aortocaval fistula is now standardized repair of the fistula followed by graft replacement of the aneurysm. Operative mortality is related to the degree of acute blood loss, myocardial infarction, coagulopathy and liver-renal failure.

In this report, we describe a case with an atypical presentation characterized by acute liver and renal failure, and a rapidly fatal course.

CASEREPORT

A 65-year-old man was admitted to our hospital with the complaints of nausea, vomiting, and abdominal pain. The abdominal pain had begun four days prior to admission. His medical history was unremarkable. He denied alcohol consumption and smoking. On examination, he was in discomfort, with a heart rate of 108/min, blood pressure 100/60 mmHg, respiratory rate 24/min, oxygen saturation 94% on room air and body temperature of 36.9°C. Auscultation findings of heart and lung

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were normal. On palpation, epigastric tenderness was present. The laboratory analysis yielded hemoglobin (Hb): 12.2 g/dl (12-16), white blood cells: 18400/mm^3 (4500-11000) with 86% neutrophils and no atypical cells in the blood smear, C-reactive protein (CRP): 69 mg/dl (0-6), aspartate amino transferase (AST): 5962 U/L (0-40), alanine amino transferase (ALT): 3600 U/L (0-40), GGT: 92 U/L (8-61), creatinine (Cre): 6.89 mg/dl (0.5-1.4), blood urea nitrogen (BUN): 75 mg/dl, creatinine phosphokinase (CPK): 358 IU/L (22 - 240), creatinine phosphokinase-myoglobin (CPK-MB): 142 IU/L (0-40), lactate dehydrogenase (LDH): 5680 U/L (250-450), antithrombin 3: 57% (80-120), fibrinogen: 178 mg/dl (200-400), D-dimer: 13.1 ug/ml (0-0.5), fibrin derived products: 20 ug/ml (0-5), and in arterial blood gas analysis, pH: 7.1 (mixed acidosis) and O_2 saturation 83% (Table 1). His electrocardiogram and chest radiography were normal. An echocardiogram showed normal left ventricular function and normal cardiac valves. He was admitted to the intensive care unit with the possible diagnosis of acute abdomen including abdominal aortic aneurysm rupture. An urgent CT showed an infra-renal abdominal aortic aneurysm -starting 2 cm distal to renal arteries and ending at common iliac arteries- 4 cm in transverse diameter, with the early detection of contrast material in the inferior vena cava suggestive of an aortocaval fistula (ACF). The ACF was concealed by a huge mural thrombus (Figure 1). Because of acidosis and impaired renal function, hemodialysis was commenced after CT. Due to a simultaneous fever of 38.5°C, and elevated CRP and white blood cells, we decided to start a wide spectrum empirical regimen of piperacillin-tazobactam, with the suspicion of sepsis/multiple organ failure, until all cultures including blood-urine-sputum had been obta-

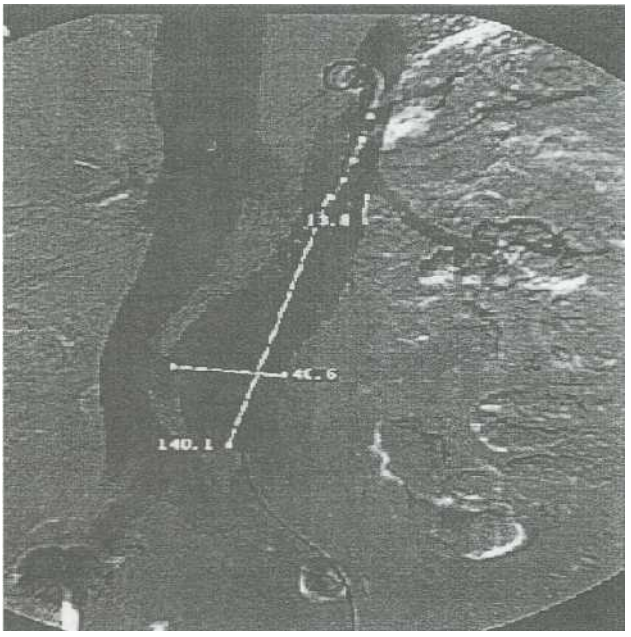

Figure 1. Preoperative abdominal computed tomography showing a large abdominal aortic aneurysm and aortocaval fistula

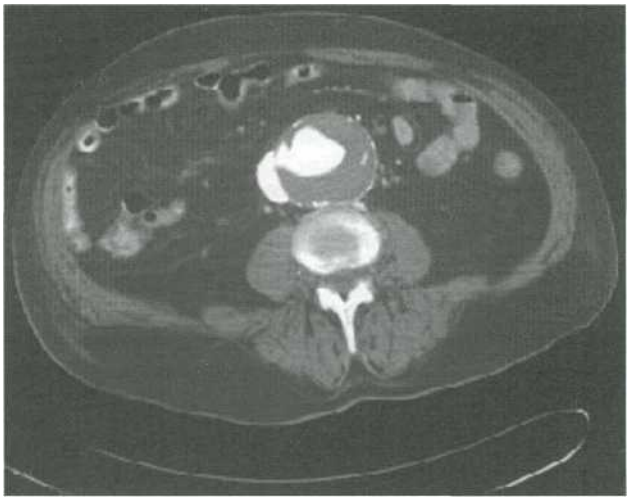

Figure 2. Aortography showing the aortocaval fistula

Table 1. The consecutive laboratory analysis of the patient

Parameter	OrL admission	second day	third day	after operation (5^{th} day)
Hb (g/dl)	14.7	13.5	12.1	11.4
WBC (/mm^3)	18400	12100	11700	13500
PLT (/mm^3)	66400	51800	45000	108000
ALT (U/L)	3600	2700	1800	146
AST (U/L)	5962	3674	2563	196
AP (U/L)	190	194	187	155
GGT (U/L)	92	119	121	46
BUN (mg/dl)	75	86	93	97
Cre (mg/dl)	6.89	7.86	7.6	6.9
CPK (U/L)	358	400	1999	455
CPK-MB (IU/L)	142	90	86	62
CRP (mg/L)	69	93	26	24
Urine output (cc/h)	15			

ined. All cultures yielded no bacterial growth and all viral serological markers for hepatitis B-C, human immunodeficiency virus, cytomegalovirus and Epstein-Barr virus were negative. On his follow-up, we found edema, cyanosis, and coldness, and arterial pulses were absent in lower extremities. Further investigations for his ACF and of the lower extremity were then commenced. An urgent abdominal-aortic angiography showed the same findings (Figure 2) as the CT. The patient underwent an emergency operation with retroperitoneal approach. The aorta was clamped proximally between the origins of the celiac and superior mesenteric arteries and distally at the common iliac bifurcations. An ulceration of the abdominal intima, measuring 5x4 cm, 3 cm above the abdominal aortic bifurcation was detected. After removing the small fresh thrombus on it, massive venous bleeding was encountered. The fistula was oversewn from within the aneurysm sac with 4/0 prolene, and the aneurysm replaced with prosthetic aortic Y-graft. During the operation, the cardiovascular surgeon decided to take a biopsy from the liver, due to its color and shape. Pathological findings were consistent with ischemic liver injury. After operation, hypothermia and hypotension ensued, and his liver and renal functions progressively worsened. His condition did not improve and he arrested. He did not respond to resuscitation and unfortunately died four days after operation.

DISCUSSION

Although the principal hemodynamic effects of arteriovenous (AV) fistulas have been well characterized, the specific clinical features of large vessel central AV fistulas may be quite variable (1). An ACF is found in 1% of operations performed for an abdominal aortic aneurysm and in 4% of operations for ruptured aneurysms (2, 3). Most abdominal aortic aneurysms rupture either into the retroperitoneum or into the peritoneal cavity. Rupture into the inferior vena cava, duodenum, iliac vein or left renal vein is very rare. The most common cause of an AV fistula involving the abdominal aorta or iliac arteries is spontaneous rupture of the aorta into the adjacent venous system (3, 4). However, there are reported cases resulting from rupture of syphilitic or mycotic aneurysms (5, 6), as well as aneurysmal lesion seen in Marfan's syndrome (1, 3), Ehlers-Danlos syndrome, Takayasu's arteritis, and iatrogenic causes (abdominal surgery, cardiac catherization). Very rarely, neoplasms may also cause a major AV fistula by erosion of adjacent arterial and venous structures (3,5).

A definitive diagnosis of ACF is sometimes difficult because the classic diagnostic signs (pulsatile abdominal mass with bruit, high-output heart failure, acute dyspnea and low back pain) are present only in 20%-50% of all such cases (7,8,9). We did not note abdominal bruit in our case, due to mural thrombus within the aneurysm sac, which caused partial obstruction of the fistula and obliterated the typical continuous bruit. Decompensated congestive heart failure due to increased venous return occurs in only 35% of such patients. In such patients, hematuria and acute renal failure may occur as a result of a renal infarction due to renal arterial problems, or of renal congestion due to a perforation of an abdominal aortic aneurysm into the renal vein. Furthermore, pre-renal failure due to heart failure and the lowered arterial blood pressure lead to lowered renal pressure. Our patient was anuric and renal function was impaired, due to the above-mentioned mechanisms. Thus he was treated with hemodialysis. Because of ACF and sepsis, his liver functions were impaired. To our knowledge, this is the first case reported in the literature in whom both liver and kidney functions were impaired due to ACF fistulas (1-6). After operation and antiobiotic treatment, his serum liver enzymes levels progressively decreased but his renal functions did not improve.

A definitive diagnosis offers advantages in surgical management. CT is often the initial imaging method when evaluating abdominal aortic aneurysm because it is a non-invasive technique. An early enhancement of the inferior vena cava is a direct clue for the diagnosis of ACF. Angiography, duplex scanning, and MRI can also be used for diagnosis (1, 2, 3, 10).

Most AV fistulas involving great abdominal vessels require prompt surgical repair. The most important problem is the control of venous bleeding from the fistula. Application of the proximal aortic cross clamp decreased the AV shunt. Venous bleeding from the fistula can be controlled by direct finger tamponade, but in our case that alone was not sufficient to control the bleeding. Aortic repair is then achieved by insertion of a prosthetic graft. Our patient died after operation due to sepsis and kidney dysfunction.

Operative mortality of the ACF is about 30% and appears to be no greater than that seen with rup-

tured abdominal aortic aneurysm in general (1, 2, 11). The mortality is mainly related to the degree of acute blood loss, myocardial infarction, coagulopathy and liver-renal failure.

We wish to alert physicians to the importance of early recognition and treatment of ACF, which is essential in reducing the mortality and complication of this disorder. In conclusion, ACF must be included in the differential diagnosis in patients with abdominal pain and acute renal-liver dysfunction.

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