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Aortocaval Fistula Resulting From Rupture of Abdominal Aortic Dissecting Aneurysm Treated by Delayed Endovascular Repair

A Case Report

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Abstract: Aortocaval fistula (ACF) after rupture of an abdominal aortic dissecting aneurysm is a rare emergency situation, which has a high mortality. However, the diagnosis is usually delayed, which increases the difficulties of treatment. We describe a case that successfully delayed use of endovascular aneurysm repair (EVAR) for ACF resulting from rupture of abdominal aortic dissecting aneurysm.

We describe a special case of a 70-year-old male with an abdominal aortic dissecting aneurysm rupturing into inferior vena cava (IVC). On account of his atypical presentation, the diagnosis had been delayed for half a year. Due to severe metabolic sequelae of the ACF and pre-existing conditions, the traditional open repair was too risky. Minimally invasive EVAR was performed with a successful result. There were no endoleak or fistula at the follow-up of 9th month.

EVAR is the most suitable method in patients with ACF from rupture of abdominal aortic dissecting aneurysm. Further educational programs should be developed, which may give rise to earlier diagnosis and treatment with better outcomes.

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Abbreviations: AAA = abdominal aortic aneurysm, ACF = aortocaval fistula, Ao = Aorta, CHF = chronic heart failure, COPD = chronic obstructive pulmonary disease, CTA = computed tomography angiography, EVAR = endovascular aneurysm repair, IVC = inferior vena cava, OR = open repair, RAAA = ruptured abdominal aortic aneurysm, VTE = venous thrombus embolism.

INTRODUCTION

Aortocaval fistula (ACF) mostly results from ruptured abdominal aortic aneurysm (RAAA), with a reported

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This study was approved by the Ethics Committee of West China Hospital of Sichuan University.

Informed consent was given by the patient and his family.

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overall prevalence of 2% to 6% in patients with RAAA.^{1,2} These patients are always under emergency situations, which have a high mortality, but their clinical presentations may be quite variable.³ Because this disease has variable clinical manifestations, the diagnosis is usually delayed, which increases the difficulties in treatment.^{1,4} Both conventional open repair (OR) and endovascular aneurysm repair (EVAR) have been used to treat this condition.⁴ However, no patients with abdominal aortic dissecting aneurysm rupturing into inferior vena cava (IVC) have been reported, and there is also no standard therapy for this condition. Because morphologic and hemodynamic characteristics of dissection are different from abdominal aortic aneurysm (AAA), EVAR may be a more valid alternative over OR in these patients. We present a patient with ACF from rupture of abdominal aortic dissecting aneurysm, which had a chronic course of disease. EVAR was successfully performed for this patient, without endoleak and fistula at the follow-up.

CASE REPORT

Prior to referral to our institution, the 70-year-old man had recurrent cough, anhelation, and chest distress. About half a year before he came to our hospital, he had temporary abdominal pain, which disappeared spontaneously and suddenly. Then the patient gradually developed recurrent cough, anhelation, and chest distress. He was treated as pneumonia at the local hospital. However, these symptoms aggravated gradually, so he was transferred to our hospital for further treatment. He had a history of tobacco abuse and chronic obstructive pulmonary disease, but no history of hypertension or heart disease. Physical examination revealed blood pressure of 126/63 mm Hg, heart rate of 85 beats/min, and slight pitting edema of both lower extremities below the knee. Moist crackles could be documented on both lungs, and respiratory sounds were weakened at bilateral lower lobe of lung. He was unable to lay flat. No abdominal tenderness or rebound tenderness was found, but a pulsating mass with continuous vascular bruit was recognized at the lower abdomen, without abdominal thrill. Laboratory results showed no hepatic or kidney dysfunction, HGB of 124 g/L, WBC of $12.29 \times 10^9/L$, BNP of 2625 pg/mL, Troponin-T of 40.6 ng/L.

Chest computed tomography scans before admission showed severe pulmonary edema and inflammation (Figure 1A). A later computed tomography angiography (CTA) revealed an abdominal aortic dissecting aneurysm with an associated aortocaval fistula, no bleeding or hematoma in retroperitoneal space (Figure 1B). Echocardiography demonstrated that left ventricular ejection fraction was 45%, without abnormality of heart structure.

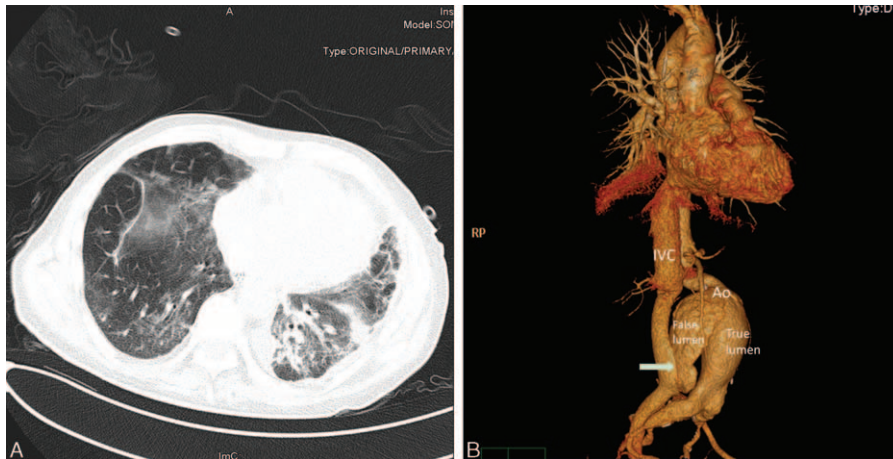


FIGURE 1. A, Chest CT scans before admission revealed bilateral diffuse interstitial infiltrations and patchy shadow in both lungs, partial pulmonary atelectasis of both inferior lobes, together with a little pleural effusion. B, Preoperative reconstructed CT scan showed dissecting aneurysm and early opacification of inferior vena cava (IVC), which indicated dissecting aneurysm had ruptured to form an aortocaval fistula (arrow). CT = computed tomography.

This elderly patient had a severe pulmonary disease and chronic heart failure (CHF), thus traditional OR was extremely risky for him. However, because of the ruptured abdominal aortic dissecting aneurysm and the persistent venous hypertension, this patient had a critical need for operation. Minimally invasive endovascular intervention was performed immediately after the diagnosis was confirmed. Under general anesthesia, bilateral common femoral arteries were punctured for arterial access. This patient's anatomy was suitable for EVAR according to CTA and intraoperative angiography (Figures 2 and 3A). Because the angle of neck was about 65° and this patient's rarely complex condition, we implanted Medtronic Endurant suprarenal aortic devices. A bifurcated stent graft (ENBF2516C170EE), 2 contralateral iliac limbs (ENLW1613C120EE, ENLW1620C120EE), and an ipsilateral extension (ENLW1616C95EE) were placed. After these stent grafts were placed, proximal type I endoleak and fistula could be seen from angiography. To close the endoleak and fistula, balloon dilatation was performed immediately. Finally, the endoleak and aortocaval fistula disappeared, which suggested EVAR was successful (Figure 3).

After operation, the patient was transferred to intensive care unit. He experienced a lengthy postoperative course including 20 days in intensive care unit due to pulmonary edema and pneumonia. This patient was given anti-infective and supportive treatment during postoperative period. He recovered and discharged about 45 days after operation. He was initially maintained on Venous Thrombus Embolism (VTE) prophylaxis with heparin during operation and then switched to low molecular weight heparin after operation. When he discharged, edema of both lower limbs had resolved, and not only the pulmonary edema but also pneumonia had significantly relieved (Figure 4A). On the follow-up of 9th month, CTA showed successful endovascular exclusion without endoleak or recurrent fistula. (Figure 4B and C).

DISCUSSION

It is reported that the overall prevalence of ACF is quite low, about 2% to 6% in patients with RAAA.¹ The most

common cause of ACF is aneurysm erosion, especially in patients with RAAA.^{1,5,6} On account of increased venous hypertension, the ACF may result in some metabolic abnormalities. However, ACF usually results from RAAA, so its most frequent symptoms should be abdominal pain and hemorrhagic shock. In our case, without bleeding in the retroperitoneal space, abdominal aortic dissecting aneurysm ruptured directly into IVC to form a fistula, which led to venous hypertension. Therefore, the hemodynamics and clinical presentations of this patient were different from cases before. In addition, fistula between false lumen of dissecting aneurysm and IVC is small, and the blood flow of false lumen in this case is much lower than that of true lumen. Therefore, the low blood flow of this fistula leads to CHF and chronic pulmonary congestion, and the chronic pulmonary congestion may be the reason of repeated pneumonia for this patient. The dramatic clinical presentation in this case is rare, which leads to delay of diagnosis. Another potential reason of delayed diagnosis may lie in the local doctor's lack of associated knowledge. According to some reports, early diagnosis and intervention can double survival rates from 25% to 50%.⁴ The diagnosis was not made until enhanced computed tomography was performed half a year after initial presentation, which was unfortunate for the patient.

The ruptured abdominal aortic dissecting aneurysm was a life-threatening condition. And the venous hypertension had led to a series of severe complications, such as pneumonia and heart failure. In addition, patients with ruptured aortic aneurysm need emergency operation according to the guideline. Emergency treatment is therefore advised. It is unknown whether endovascular intervention improves survival compared with traditional OR for this disease.⁷ With the risk related to CHF, massive intraoperative blood loss, and pulmonary infection, the surgical mortality of open repair ranges from 16% to 66%.^{1,7} EVAR is valid in certain anatomical configurations and may be the optimal selection in terms of stent graft available. And EVAR has potential benefits of less blood loss and no need for aortic cross-clamping during operation. Several cases of EVAR for RAAA with ACF have been described, with about 30% mortality, which had partially due to delays in diagnosis.^{4,8} Accompanied with CHF and chronic pulmonary congestion, the

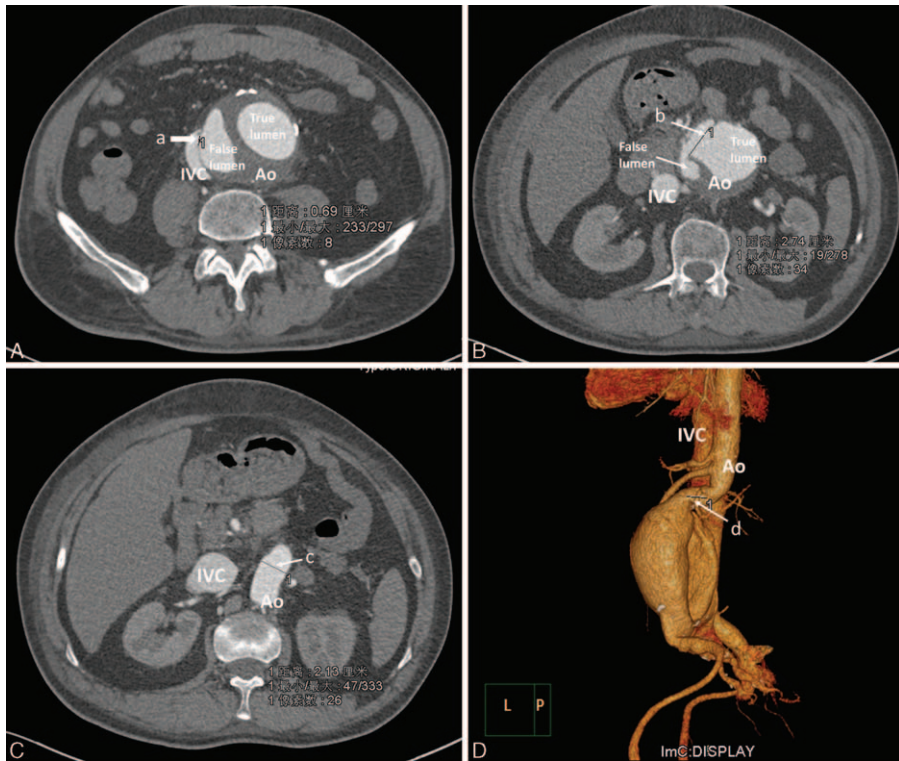


FIGURE 2. Preoperative images of patient with aortocaval fistula and ruptured abdominal aortic dissecting aneurysm. A, Computed tomographic angiography (CTA) revealed false lumen of Aorta (Ao) directly rupturing into inferior vena cava (IVC) through a 6.9 mm fistula (arrow a), and the maximal diameters of this dissecting aneurysm was 89 mm. B, CTA showed the Aorta divides into true and false lumen through a 27.4 mm tear (arrow b), which formed the abdominal aortic dissecting aneurysm. C, CTA showed the diameter of neck was 21.3 mm (arrow c). D, Reconstructed CT scan revealed the length of neck is about 20 mm (arrow d), and the angle of the neck is about 65°. CT = computed tomography.

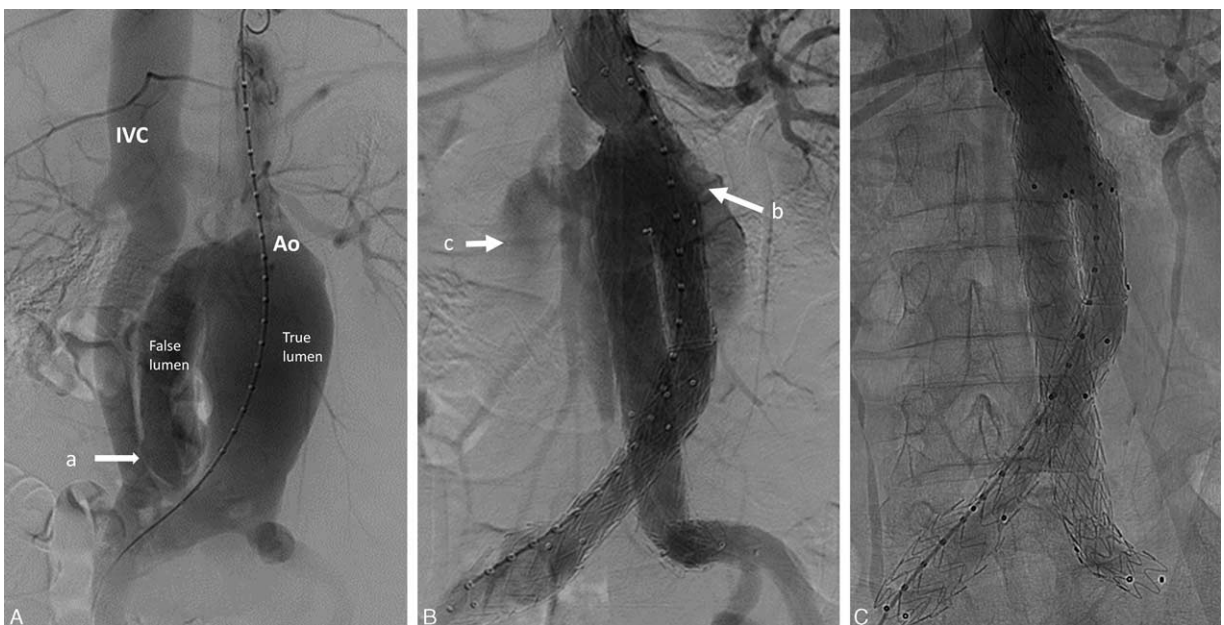


FIGURE 3. Intraoperative view during endovascular repair. A, Angiography showed fistula (arrow a) between false lumen of dissecting aneurysm and early opacification of inferior vena cava (IVC). B, After endografts were placed, angiography revealed proximal type I endoleak (arrow b) and early opacification of IVC (arrow c). C, After balloon dilatation was performed, angiography showed the endoleak and aortocaval fistula had disappeared.

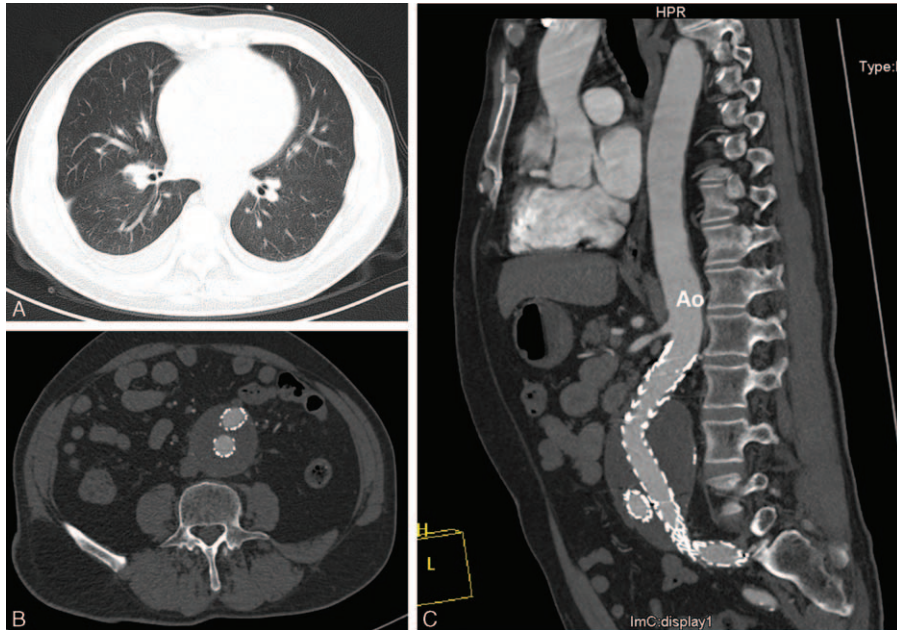


FIGURE 4. A, Chest CT scan before discharge showed pulmonary edema and inflammation had significantly relieved. From (B) and (C), CTA at the follow-up of 9th month after operation revealed no endoleak or fistula. CT=computed tomography, CTA=computed tomographic angiography.

general condition of the elderly patient in our case was poor, he was not suitable for OR. Meanwhile, the anatomical configurations of our patient were suitable for EVAR. Furthermore, the morphologic and hemodynamic characteristics of dissecting aneurysm in our case are different from AAA or dissection before. Therefore, EVAR was the best choice for this patient.

In our case, EVAR alone appears to have successfully isolated abdominal aortic dissecting aneurysm and closed the ACF without the need of a stent-graft in the IVC. Because of aortocaval fistulas and type II endoleak, persistent communication between the aneurysm and the IVC may exist. In some literatures, early placement of stent graft in the IVC has been advocated to avoid persistent type II endoleak.^{9,10} However, no evidence demonstrated stent graft of IVC should be placed at the time of initial EVAR. Because the chance of persistent aortocaval fistula after EVAR is rather small, endoleak and fistula may disappear spontaneously after EVAR. Some researchers suggested that EVAR can be performed alone without further intervention.⁸ If persistent aortocaval fistula occurs, stent graft in IVC can be performed in the future. In our case, no fistula or endoleak were found at the follow-up of the 9th month. In addition, because aneurysm sac debris may dislodge, prophylaxis of pulmonary embolism should be addressed. In our case, VTE prophylaxis with anticoagulant therapy was maintained during the perioperative periods. It is reported that IVC filter should be placed before operation.¹¹ However, the chance of clinically significant pulmonary embolism is minute, and IVC filter placement may complicate the possible further IVC stent-graft placement. Preoperative IVC filter placement does not appear to be necessary.⁸

CONCLUSION

We introduce a case of ACF resulting from rupture of abdominal aortic dissecting aneurysm. Diagnosis of this

patient was delayed, but he fortunately had a successful operation and recovered finally. Despite his poor general status and the high mortality associated with this condition, his complete recovery presents the potential for recovery with appropriate treatment. EVAR is the most suitable selection in this condition for its minimal invasion. Further educational programs should be developed, which may give rise to earlier diagnosis and treatment with better outcomes.

REFERENCES

1. Cinara IS, Davidovic LB, Kostic DM, et al. Aorto-caval fistulas: a review of eighteen years experience. *Acta Chir Belg.* 2005;105: 616–620.
2. Brightwell RE, Pegna V, Boyne N. Aortocaval fistula: current management strategies. *ANZ J Surg.* 2013;83:31–35.
3. Gerassimidis TS, Papazoglou KO, Kamparoudis AG, et al. Endovascular management of ruptured abdominal aortic aneurysms: 6-year experience from a Greek center. *J Vasc Surg.* 2005;42: 615–623.
4. Akwei S, Altaf N, Tennant W, et al. Emergency endovascular repair of aortocaval fistula—a single center experience. *Vasc Endovascular Surg.* 2011;45:442–446.
5. Kotsikoridis I, Papanas N, Papanas N, et al. Aortocaval fistula formation due to ruptured abdominal aortic aneurysms: a 12-year series. *Vasc Endovascular Surg.* 2012;46:26–29.
6. Psathas ED, Lioudaki S, Doulaftsis M, et al. Clinical manifestations of aortocaval fistulas in ruptured abdominal aortic aneurysm: report of two cases. *Case Rep Surg.* 2012;2012:123081.
7. Davidovic L, Dragas M, Cvetkovic S, et al. Twenty years of experience in the treatment of spontaneous aorto-venous fistulas in a developing country. *World J Surg.* 2011;35: 1829–1834.

8. van de Luijtgaarden KM, Bastos GF, Rouwet EV, et al. Conservative management of persistent aortocaval fistula after endovascular aortic repair. *J Vasc Surg.* 2013;58:1080–1083.
9. Melas N, Saratzis A, Saratzis N, et al. Inferior vena cava stent-graft placement to treat endoleak associated with an aortocaval fistula. *J Endovasc Ther.* 2011;18:250–254.
10. Siepe M, Koeppel S, Euringer W, et al. Aorto-caval fistula from acute rupture of an abdominal aortic aneurysm treated with a hybrid approach. *J Vasc Surg.* 2009;49:1574–1576.
11. De Rango P, Parlani G, Cieri E, et al. Paradoxical pulmonary embolism with spontaneous aortocaval fistula. *Ann Vasc Surg.* 2012;26:739–746.