Case Report

Spontaneous Rupture of the Ascending Thoracic Aorta Resulting in a Mimicking Pseudoaneurysm

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We report on the rare and successful surgical treatment of a case of spontaneous rupture of the ascending thoracic aorta resulting in a mimicking pseudoaneurysm. A 72-year-old male who had complained of sudden onset of severe chest pain was admitted to our hospital. Initially, acute type A closing aortic dissection was suspected because computed tomography (CT) showed a small ulcer-like projection (ULP) in the posterior aspect of the ascending aortic wall, but it also revealed no intimal flap, false lumen or aortic aneurysm. CT and magnetic resonance imaging (MRI) indicated a change in the radiographic aspect of the ULP and revealed a mimicking saccular-type pseudoaneurysm and gradual increasing size of the pseudoaneurysm. Surgery was performed after considering the risk of pseudoaneurysmal rupture. We replaced the ascending aorta and diagnosed it as a spontaneous aortic rupture by histological examination of the rupture site after failing to observe an aneurysm or dissection. We discuss these results with reference to the literature, including our pathological and radiographical findings. (Ann Thorac Cardiovasc Surg 2006; 12: 223–7)

Key words: spontaneous aortic rupture, pseudoaneurysm, graft replacement

Introduction

Spontaneous rupture of the thoracic aorta in the absence of trauma and without aneurysm or dissection is a rare but fatal event. To our knowledge, the present case is the 16th of spontaneous rupture to have occured in the ascending aorta. We report on the rare and successful surgical treatment of a case of spontaneous rupture of the ascending thoracic aorta resulting in a mimicking pseudoaneurysm, and discuss it with reference to the literature.

Case Report

A 72-year-old male who had complained of sudden onset of severe chest pain was admitted to our hospital. He had a past history of hypertension (HT), partial liver resection for hepatic cell carcinoma, and chronic liver dysfunction with splenomegaly caused by hepatitis C. On admission, he was conscious, but he was in a state of shock with a systolic blood pressure of 70 mmHg. His blood pressure improved after treatment by intravenous drip combined with dopamine and he was stabilized with a systolic pressure between 100 mmg and 130 mmHg. Electrocardiogram (ECG) showed no evidence of myocardial ischemia and chest radiography revealed pleural fluid on the left side without an enlarged mediastinal and cardiac silhouette. Initially, acute type A closing aortic dissection was suspected because computed tomography (CT) showed a small ulcer-like projection (ULP) in the posterior aspect of the ascending aortic wall, but he was followed up with medication because his vital signs remained stable. In addition, CT did not reveal an intimal flap, false lumen or aortic aneurysm (Fig. 1), and echocardiography did not show any aortic valvular insufficiency. The presence of a mobile intimal flap or aortic dissection, and slight pericardial effusion with no signs of tamponade

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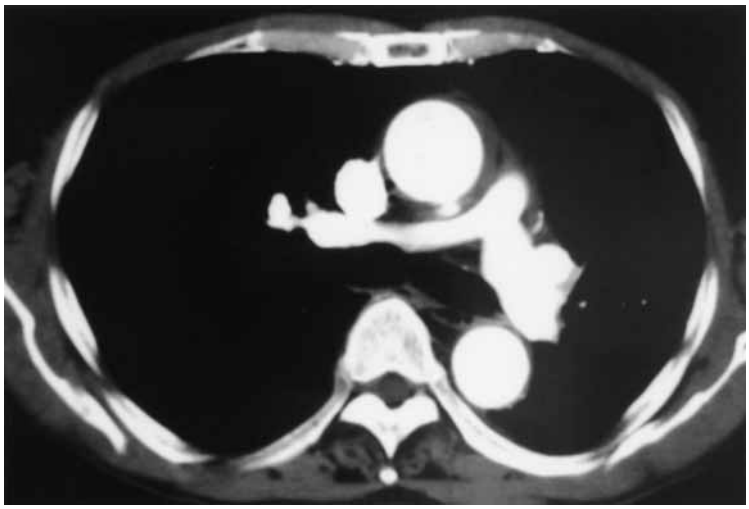
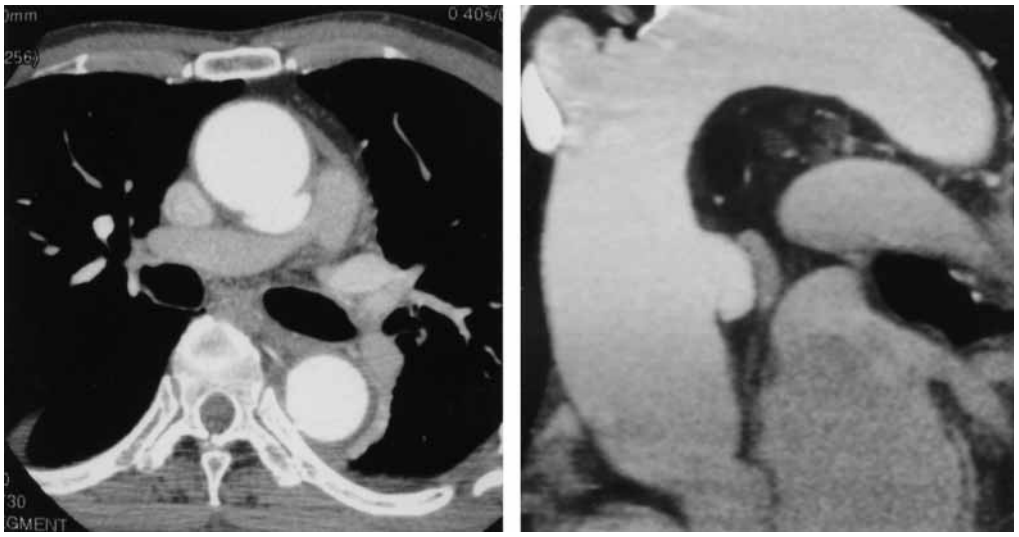


Fig. 1. Computed tomography (CT) showed an intramural or medial hematoma as a small ulcer-like projection (ULP) in the posterior aspect of the ascending aortic wall, but revealed no intimal flap, false lumen or aortic aneurysm.



a | **b**

Fig. 2. CT (**a**) and magnetic resonance imaging (MRI) (**b**) on the 8th day of hospitalization indicated a change in the radiographic aspect of the ULP and revealed a mimicking saccular-type pseudoaneurysm measuring 20×10 mm with pleural fluid on the left side.

were observed. Laboratory findings did not indicate remarkable changes, excluding anemia with a hemoglobin level of 11.7 g/dl and thrombocytopenia with a count of 67,000/mm^3. However, CT, magnetic resonance imaging (MRI), and aortography on the 8th day of hospitalization indicated a change in the radiographic aspect of the ULP and revealed a mimicking saccular-type pseudoaneurysm measuring 20×10 mm with pleural fluid on the left side (Fig. 2). Furthermore, CT on the 12th day of hospitalization revealed that the pseudoaneurysm was gradually increasing in size and surgery was performed after considering the risk of pseudoaneurysmal rupture. Macroscopically, a longitudinal intimal tear without a mobile intimal flap and dissection 3 cm in length was found in the posterolateral wall of the ascending aorta 1 cm from the aortic valve. There also existed a mimicking oozing ruptured pseudoaneurysm 2 cm in diameter in the center of the intimal tear and a hematoma in the periadvential region of this site (Fig. 3). The ascending aorta was thus subsequently replaced. Histological examination of the aortic wall neighboring the tear of the rupture site showed no evidence of dissection, cystic medial necrosis, and atherosclerosis in the intima, but the tear extended to the adventitia of the pseudoaneurysm and had produced a hematoma around the aortic wall without medial dissection. Furthermore, the adventitia of the pseudoaneurysm at the site of the rupture was walled off by organizing granulation tissue with hemorrhaging (Fig. 4). The pa-

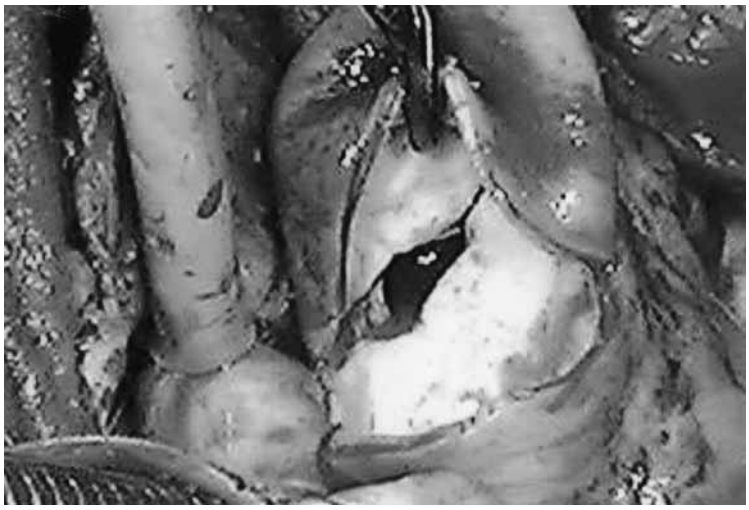


Fig. 3. Intraoperative findings revealed a longitudinal intimal tear without a mobile intimal flap and dissection 3 cm in length in the posterolateral wall of the ascending aorta with a mimicking oozing ruptured pseudoaneurysm 2 cm in diameter in the center of the intimal tear.

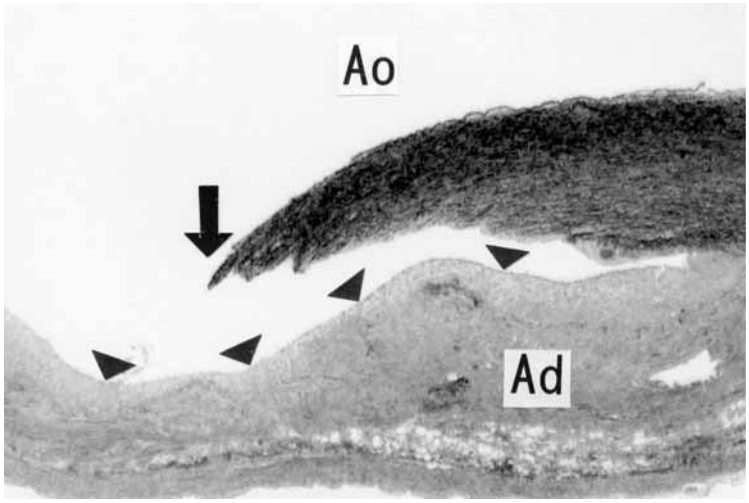


Fig. 4. Histological examination of the aortic pseudoaneurysm neighboring the tear of the rupture site showed no evidence of medial dissection, cystic medial necrosis, and atherosclerosis in the intima. However, the tear extended to the adventitia of the pseudoaneurysm at the site of the rupture, which was walled off by organized granulation tissue and was hemorrhaging (Elastica van Gieson stain).
Arrowheads, organized granulation tissue; arrow, complete disruption of the media; Ao, aortic lumen; Ad, adventitial pseudoaneurysm.

tient was later discharged in good health on the 43rd postoperative day.

Discussion

The occurrence of a spontaneous aortic rupture without aortic aneurysm, dissection, trauma, inflammation of the aortic wall or erosion from a neoplastic mass is rare. After reviewing the literature for cases of spontaneous rupture of the ascending aorta, only 16 were found, including our case (Table 1).[1-13] Among these, the mean patient age was 62.6 years and ranged from 47 to 76 years and nine cases were male and five cases were female, including unknown in two cases. Eleven cases had a history of HT (69%), and it thus appears that HT and aging caused spontaneous ascending aortic rupture. The majority of the patients had severe acute chest pain or back pain with shock and hemopericardium which resulted in severe hemopericardium with an enlarged cardiac silhouette on chest roentgenograms. Furthermore, aortography and CT were useful diagnostic methods for evaluating ascending aortic wall abnormalities to identify the rupture sites. However, it was difficult to differentiate spontaneous aortic ruptures from the clinical or radiographical findings of ruptures of classic aortic dissection or aneurysm preoperatively. As a preoperative misdiagnosis, aortic dissection, acute myocardial infarction or acute hemopericardium of unknown origin and a definitive diagnosis in most cases was made usually only after histological examinaton during surgery or autopsy. Our review also found that eight of the cases died of the rupture and eight survived after surgery and all non-surgically treated cases died. Ten cases underwent aortic repair, which involved suture closure in four cases, and graft replacement in six depending on the

Table 1. Clinical findings of spontaneous rupture of the ascending aorta

Author (year)/ref. no.	Age/sex	HT	Pathlogy	Operation	Outcome	Autopsy
Castleman (1970)[1]	62/M	+	Atheroma	-	Dead	Hemopericardium Left hemothorax
Murray (1973)[2]	-	–	CMN	-	Dead	-
Murray (1973)[2]	-	–	CMN	-	Dead	-
Widder (1983)[3]	60/M	+	Unknown	Suture closure	Alive	
Le Moigne (1984)[4]	67/F	+	Unknown	Graft replacement	Alive	
Padro (1988)[5]	47/M	+	CMN	Graft replacement	Alive	
Ando (1991)[6]	63/M	+	Unknown	Suture closure	Alive	
Aoyagi (1991)[7]	68/F	+	CMN	-	Dead	Hemopericardium
Shkrum (1992)[8]	49/M	+	Minimal atherosclerosis	-	Dead	Hemopericardium Hematoma stenosing right coronary artery AMI
Handa (1994)[9]	49/M	–	Minimal atherosclerosis	Suture closure	Alive	
Tomita (1996)[10]	74/M	–	Atheroma	-	Dead	Hemopericardium
Moussarih (1998)[11]	76/F	+	Atheroma	Graft replacement	Dead	-
Moussarih (1998)[11]	69/M	+	Atheroma	Graft replacement	Dead	-
Akashi (2003)[12]	66/F	+	CMN	Graft replacement	Alive	
Bito (2004)[13]	55/F	–	Unknown	Suture closure	Alive	
Hirai (2005)	72/M	+	Unknown	Graft replacement	Alive	

HT, hypertension; atheroma, atheromatous change; CMN, cystic medial necrosis; AMI, acute myocardial infarction.

size of the aortic tear. Afterwards, eight cases survived and two died. Graft replacement is the best procedure in terms of patient survival considering the size of the aortic tear and fragility of the aortic wall near the rupture. Regarding pathologic examination of the rupture site, our review of the literature also found atheromatous changes in six cases involving minimal atherosclerotic changes, cystic medial necrosis in five cases, and no significant pathologic changes in four cases. The leading pathologic findings from the spontaneous aortic rupture were arteriosclerosis with longstanding HT. Our case showed no evidence of atherosclerosis, cystic medial necrosis or aortitis and no significant gross or histological changes were noted. Since there were no changes that would clearly explain the rupture, we diagnosed it as a spontaneous aortic rupture through an intimal tear resulting in a mimicking pseudoaneurysm. Recently, Stanson et al. reported as a new clinical entity that penetrating atherosclerotic ulcers (PAUs) can be regarded as a cause of aortic rupture by atherosclerosis.[14] PAU of the aorta is ulceration of an atherosclerotic plaque that penetrates the intima, and it results in aortic intramural hematoma, adventitial pseudoaneurysm formation, or aortic rupture.[15] Also in our case, the tear extended to the adventitia of the pseudoaneurysm at the site of the rupture, which was walled off by organized granulation tissue, such as in a mimicking aortic ulcer. Furthermore, CT images also revealed arteriosclerotic lesions with longstanding HT in another part of the thoracic aorta and those images were similar to the above-mentioned consecutive progression, such as the existence of a relatively narrow orifice compared with a large lesion in differentiating a transmural aortic rupture with a hematoma. These findings might be confirmed by change from an intramural hematoma to a pseudoaneurysm formation, and may lead to aortic rupture due to PAU, although microscopic examination of the rupture site showed thin intima with no severe atherosclerosis. In general, aortic local dissection is regarded as a cystic medial necrosis with an intimal tear or medial hemorrhage leading to localized false lumen separating the intima and adventitia. Intramural hematoma is thought to be spontaneous localized hemorrhage into the aortic wall in the absence of aortic dissection, intimal tear or PAU. The aortic rupture through a small intramural hematoma with ruptured arteriosclerosis plaque could have been interpreted clinically as a spontaneous aortic rupture.[16] We suspected that some spontaneous aortic rupture due to atheromatous plaque as previously reported might have been due to the perforation of PAU. Therefore, we believe that PAU might be recognized as a cause of aortic rupture with increasing frequency in the future by sensitive imaging techniques, such as three dimensional

(3D)-CT, MRI, transesophageal echocardiography (TEE), intravascular ultrasound (IVUS), and precise pathological examination.

Conclusion

We reported on a rare surgical case of spontaneous rupture of the ascending thoracic aorta resulting in a mimicking pseudoaneurysm, and discussed it with reference to the literature, including the pathological and radiographical findings.

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