Rupture of Aortic Arch : A Report of 3 Cases

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Rupture of thoracic aortic aneurysm is a lethal condition that requires urgent surgery¹⁻⁶⁾. When rupture occurred in the aortic arch, surgery on it requires adequate brain protection during cardiopulmonary bypass (CPB) and open procedure for reconstruction of the arch. Mortality rate had been almost 100% in the early experience of cardiovascular surgery and is still high even with current challenge in surgery. We present the case reports of 3 patients operated on within a week for rupture of the aortic arch with different etiology and postoperative course each other.

Case Report

patient 1

A 75-year-old man was admitted to Tohoku Central Hospital with increasing dyspnea and palpitation that suddenly occurred 5 days before. Two years previously, he has been pointed out to have a large aneurysm (60mm) in the aortic arch with no cardiac murmur but had refused operation. On admission, he had a continuous murmur on the left superior anterior chest and edema on both legs. The cervical veins were slightly dilated. Arterial pressure was 100/40 mmHg. Chest roentgenogram showed a large mass shadow lateral to the aortic arch and an increased pulmonary vasculature in the right lung and decreased one in the left (**Fig. 1a**). Three-dimen-

sional (3D) computed tomographic (CT) scan demonstrated a large aortic arch aneurysm of the ductus diverticulum that communicated to the pulmonary artery (**Fig. 1b**). An angiography and oximetry revealed a left-to-right shunt through the aneurysm with oxygen saturation of 48% in the right ventricle and 80% in the pulmonary artery. He was transferred to Yamagata University Hospital and underwent an urgent operation because of severe congestive heart failure that had continued for 7 days.

At operation, the central venous pressure was 15 mmHg and arterial pressure was 50 mmHg. After a median sternotomy, a large aneurysm (60mm) was exposed on the left side of the aortic arch that oppressed the left pulmonary artery. There was no hematoma in the mediastinum. CPB and core cooling were established with an ascending aortic cannulation. Right pulmonary artery was cross-clamped to reduce excessive blood inflow to the pulmonary circulation through the left-to-right shunt. Selective cerebral perfusion was started through three arch arteries with systemic perfusion through the aortic cannula discontinued when the esophageal temperature reached to 20 °C. The aneurysm was excised, in which many intra-luminal thrombi existed. There were small tears at the bottom of the aneurysm and the top of the left pulmonary artery where left-to-right shunt blood flow communicated. The lower body perfusion was re-instituted via a Foley catheter placed in the descending thoracic aorta through the orifice of the aneurysm. The orifice of the aneurysm (3×4cm) with firm fibrous edges was closed with a Dacron patch. The Foley catheter was removed. The perforation of the left pulmonary artery was closed with a direct suture. The patient recovered uneventfully and is doing well for 31 months after the operation.

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Fig. 1. Chest roentgenogram of patient 1 on admission shows a large mass shadow lateral to the aortic arch, increased pulmonary vasculature in the right, and decreased one in the left (a). Three-dimensional CT scan shows direct communication between aortic arch aneurysm and pulmonary artery (b).

Patient 2

A 71-year-old man was admitted to Yamagata Saiseikan Hospital due to loss of consciousness and hemodynamic collapse after sudden left thorax pain. One and half years previously, he had brain infarction and incomplete right hemiplegia. Low arterial pressure (70/35 mmHg) and loss of consciousness recovered with inotropic treatment. Chest roentgenogram with tracheal shift to the right suggested a massive hematoma in the left upper part of the chest (Fig. 2a). Chest CT scan (Fig. 2b) showed a small aneurysm of the aortic arch and hematoma in the posterior mediastinum to parietal extrapleural space. The patient was transferred to Yamagata University Hospital for an emergent operation with receiving fluid supplementation. He was still drowsy before operation, although arterial pressure was stabilized at 110/72 mmHg and the right hemiplegia did not increase. Breathing sound decreased in the left upper chest.

After median sternotomy, CPB and core cooling were instituted with an ascending aortic cannulation. Aortic arch aneurysm was exposed through medi-

astinal hematoma. While using deep hypothermia (lowest bladder temperature = 17.8 °C) and selective cerebral perfusion via arch arteries, systemic perfusion through the aortic cannula was discontinued and the aortic arch just prior to the aneurysm was incised. Proximal ascending aorta was cross-clamped for inducing cardioplegia. There were two small tears in the aneurysm, right and just distal to the left subclavian artery where the aneurysm was most dilated. Distal ascending aorta and total arch were excised and replaced with a branched Dacron graft, because atherosclerotic change was severe in the arch. The patient was weaned from CPB uneventfully but did not awake and died of brain infarction at postoperative day 7. Postoperative CT scan of the brain (Fig. 2c) showed multiple low-density area. At autopsy, the descending thoracic aorta was markedly atherosclerotic including sealed another rupture in the abdominal aorta, where the hematoma was narrowly localized. There were multiple atherosclerotic plaques at the sino-tubular junction of the ascending aorta, arch branches and intracranial arteries. They all could cause new brain

Fig. 2. Chest roentgenogram of patient 2 on admission (a) shows dark matter in the left lung field and mediastinal shift to the right. Chest CT scan on admission (b) shows a small aneurysm of aortic arch with hematoma in the posterior mediastinum to parietal extrapleural space. Brain CT scan (c) shows multiple low-density area with brain edema.

infarction, but it was not identified which lesion caused this embolic event.

Patient 3

A 72-year-old man was admitted to Tohoku Central Hospital due to hemodynamic collapse after left-sided thorax pain. Low arterial pressure of 60/34 mmHg on admission was recovered to 110/72 mmHg after inotropic support and fluid supplementation. Consciousness was clear. Although massive hematoma was apparent in the posterior mediastinum to parietal extrapleural space in chest roentgenogram (**Fig. 3a**) and CT scan, 3D-CT scan did not demonstrate any aneurysms (**Fig. 3b**). However, repeated CT scan at 1.5 hours after the initial scan demonstrated the contrast medium around the distal arch in the non-enhanced hematoma (**Fig. 3c**). Diagnosed as a spontaneous rupture of the distal aortic arch, he was transferred to Yamagata University Hospital with receiving fluid supplementation.

The emergent operation was performed through median sternotomy while using deep hypothermic

CPB with an right axillary arterial cannulation. Systemic perfusion was discontinued while the brain was selectively perfused at 19.4 °C. The arch exposed through the hematoma was not dilated. There was a 5mm × 15mm tear just distal to the left subclavian artery. The distal aortic arch was excised for 3 cm in length, and replaced with a Dacron graft. He had infectious mediastinitis but was treated successfully with debridement and omentopexy with pectoral muscle flap at postoperative day 15. He was discharged at postoperative day 124, but died of pneumonia 19 month after the operation and 2 months of re-hospitalization for gradual loss of general activity.

Discussion

Surgery for aneurysm of the thoracic aorta still carries high mortality rate, especially when rupture has occurred. In 1980, Pressler and McNamara reported that 62 patients died of thoracic aortic rupture

Fig. 3. Chest roentgenogram of patient 3 on admission (a) shows dark matter in the left. Three-dimensional CT scan (b) shows no aneurysm. Repeated CT scan (c) at 1.5 hours after the initial scan demonstrates that contrast medium surrounds distal aortic arch.

among 176 patients of thoracic aortic aneurysm¹⁾. Johansson and colleagues assumed the incidence of aneurysm rupture in the thoracic aorta as 5/100,000 per year, and reported 2 operative survival cases in 1989²⁾. In 1991, Crawford and colleagues reported that 30-days survival was 76% among 117 patients with ruptured thoracic and thoracoabdominal aneurysm in their large operative series³⁾. Von Segesser and colleagues, in 1996, reported that 30-days mortality increased from 6% to 15% when rupture occurred with thoracic and thoracoabdominal aneurysm⁴⁾. In Japanese experience, Hayashi and colleagues reported in 1997 that 30 days mortality was 30 or 40% when rupture occurred in the thoracic descending or arch aneurysm, whereas it was 4 or 14% without rupture in the same regions^{5, 6)}. Our experience with three patients with one in-hospital death carries the same mortality as summarized above.

Aneurysm of the ductus diverticulum is thought to occur after failure of closure of the aortic side of ductus⁷⁻¹¹⁾. Abnormal ductal tissue left in the aortic wall may contribute to aneurysmal formation and

early rupture. More than one-half of the patients with aneurysm of the ductus diverticulum died of erosion or rupture when the aneurysm was not repaired surgically. Rupture into the pulmonary artery was rarely reported, where hemodynamic collapse may be mild because left-to-right shunt may occur as in patient 1 instead of catastrophe by rapid exsanguination. This type of aneurysm may develop promptly due to tissue abnormality, and rupture into the other region, bronchus, esophagus, pericardium, or pleural cavity, may cause earlier death through rapid exsanguination. Therefore, early operation should be planned and follow-up continued even when operation is refused. In operation, over inflow of the perfused blood into the pulmonary artery should be reduced during CPB. In patient 1, deep hypothermic selective cerebral perfusion with lower body perfusion maintained systemic circulation during the open procedure with clamping the right pulmonary artery.

Patient 2 had initially lost consciousness due to hemodynamic collapse. Although it was recovered to a level enough for hospital transfer and emer-

gency operation, this initial event and the past history of brain infarction indicated high risk for perioperative brain infarction in patient 2. We replaced the ascending aorta and transverse aortic arch because atherosclerotic change was severe. However, multiple plaques at the proximal ascending aorta and cervical arteries revealed at autopsy might also have potentially caused brain embolism in patient 2. We selected ascending aorta instead of femoral artery as a site for blood return of CPB in patients 1 and 2 to prevent brain embolism from descending aorta, as Westaby and Katsumata advocated¹²⁾. The marked atherosclerosis in the descending aorta revealed at autopsy in patient 2 may justify this perfusion. However, atherosclerotic ascending aorta itself carries high risk of brain embolism especially during CPB. In 1986, Culliford and colleagues advocated to prevent brain embolism during CPB by reduction of flow velocity in the atherosclerotic ascending aorta with a long cannula put in the arch¹³⁾. Alternatively, Baribeau and colleagues advocated arterial inflow via an axillary artery graft for patients with severely atheromatous aorta in 1998¹⁴⁾. We used direct cannulation at axillary artery in patient 3, and currently moved to use arterial inflow via an axillary artery graft in operations of arch aneurysm, where no brain infarctions occurred until today. Use of this CPB technique and further extension of the arch replacement both to the proximal ascending aorta and arch arteries would reduce the risk of brain embolism even when atherosclerosis is severe as in patient 2. Deep hypothermia and selective cerebral perfusion may provide sufficient safe period for complete inspection of the aorta and extended procedures. Intra-operative ultrasonic imaging, when available, would assist this inspection of the aorta. Fomon, Juvonen and their colleagues assumed the risk for rupture as almost 0% of thoracic aneurysm with diameter of less than 5cm^{15, 16)}. Spontaneous rupture of the thoracic aorta without aneurysm is, therefore, extremely rare¹⁷⁻²¹⁾. Ma and Ang showed in 1996 that only 5 among 13 patients with spontaneous rupture of thoracic aorta survived through operation²⁰⁾. Atherosclerosis as well as cystic medial necrosis would cause this rare catastrophe. Rupture site was various and extremely difficult to diagnose before lethal exsanguination. Several recent reports showed that CT scan would occasionally suggest the rupture site but aortogram did not¹⁷⁻¹⁹⁾. The initial CT scan in Patient 3 did not show any aneurysm and rupture site. However, repeated CT scan demonstrated the contrast medium just around the distal arch,

which suggested the rupture site that should be replaced. Although we took a CT scan 1.5 hours later, interval of 0.5 to 1 hour would be enough for the contrast medium to accumulate just outside the rupture.

Recent reports of ruptured thoracic aorta with^{22, 23)} and without aneurysm¹⁷⁻²¹⁾ showed that there were certain duration between the initial episode and death or operation, where immediate death was rather rare. All three patients with different etiologies reported herein had this duration, during which diagnosis, hospital transfer and emergency repair were accomplished. CT scan including 3-dimensional reconstruction and repeated scan provided useful diagnosis. Deep hypothermia with selective cerebral perfusion was helpful for open procedure in the arch, but one patient died of perioperative brain infarction. Complete inspection of the aorta with branching arteries, subsequent extended replacement of the atherosclerotic lesions during the long safe period provided by selective cerebral perfusion and use of axillary arterial inflow would be recommended in cases of atherosclerotic aneurysmal rupture as a safe strategy for operation.

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ABSTRACT

A 75-year-old man had sudden congestive heart failure with continuous murmur due to ruptured aneurysm of ductus diverticulum into the pulmonary artery. Patch repair was successfully performed with deep hypothermic selective cerebral perfusion, and adjunctive lower body perfusion through the orifice of the aneurysm. A 71-year-old man with deep shock due to ruptured aneurysm of the aortic arch underwent total aortic arch replacement but died of severe brain infarction due to severe atherosclerosis on postoperative day 7. A 72-year-old man with deep shock due to spontaneous rupture of distal aortic arch was successfully operated after diagnosed with repeated computed tomography. Although rupture of the aortic arch is a lethal condition, patients may survive when an urgent operation is performed after appropriate diagnosis and adequate medical support during initial period. Deep hypothermic selective cerebral perfusion is helpful for these patients, but the risk of cerebral embolism due to severely atherosclerotic aorta should be reduced.

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Key Words : aortic arch rupture, ductus diverticulum aneurysm, spontaneous aortic rupture, hypothermic selective cerebral perfusion, repeated CT scan

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