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A patient who underwent surgical treatment of an adult-type aneurysm in the nonpatent arterial duct

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Adult-type aneurysms in the arterial duct are rare, and their spontaneous prognosis is poor. We performed surgical treatment of an aneurysm in the arterial duct in a 62-year-old male. The patient had had hoarseness since November 2003, and was referred to our hospital in March 2004. Thoracic CT and aortography demonstrated a sacciform aneurysm in the aorta in the distal arch region on the lesser curvature side. The patient was diagnosed as having an adult-type aneurysm in the nonpatent arterial duct, and underwent surgical treatment in April 2004. Thoracotomy in the fourth left intercostal space was performed up to the thoracic aorta, and a sacciform aneurysm, measuring 35 mm × 32 mm, was detected in the arterial duct. The recurrent laryngeal nerve adhering to the front surface of the aneurysm was overextended. Under partial extracorporeal circulation, the aneurysm was excised, and replaced by an artificial blood vessel. The postoperative course was satisfactory, and the patient was discharged from the hospital 14 days after surgery. The surgical outcome was good, and the hoarseness was improved. Taking possible complications into consideration, surgical treatment can be recommended in the early stage.

Key words: Aneurysm in the arterial duct, CT, aortography, partial extracorporeal circulation, thoracic aorta

INTRODUCTION

Adult-type aneurysms in the arterial duct are rare. We suspected this type of aneurysm in a patient based on physical observation and thoracic radiographic examination, and performed CT and MRI, for the diagnosis of an aneurysm in the arterial duct. We report this patient with a review of the literature.

CASE REPORT

Patient: A 62-year-old male

Chief complaint: Hoarseness

Past-history: Hypertension was noted, but not treated.

Clinical course: Hoarseness occurred in November 2003, and was gradually aggravated. Palsy of the left vocal cord was detected by an otolaryngologist, and an aneurysm in the aortic arch region was suspected by thoracic CT. The patient was referred to our hospital in March 2004.

Conditions at the time of admission: Height, 170 cm; Weight, 77 kg; Blood pressure, 150/80 mmHg without a bilateral difference; pulse, 60/min regular; no cardiac or vascular murmur

Biochemical findings at the time of admission: No abnormalities were detected by hematological and biochemical examination. Inflammatory reaction was normal, and syphilis examination was negative.

Thoracic radiography demonstrated a minor projection of the second left arch alone (Fig. 1). Thoracic CT (Fig. 2) and 3D CT (Fig. 3) revealed an aneurysm of 37 mm projecting from the aortic arch region to the lesser curvature. The vascular lumen was mostly occupied by a thrombus, but blood flow was observed in a region connected to the

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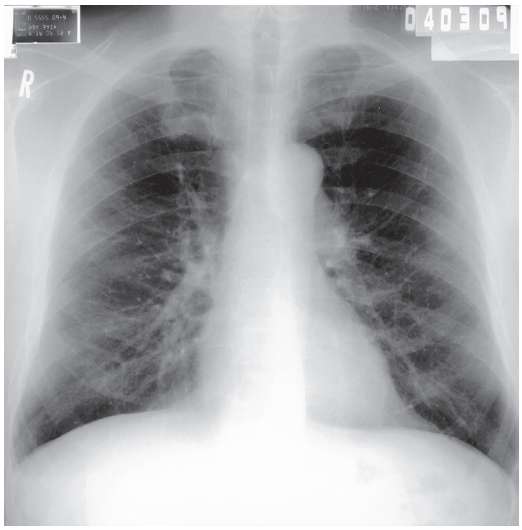

Fig. 1 Thoracic radiography demonstrated a little projection of the second left arch alone.

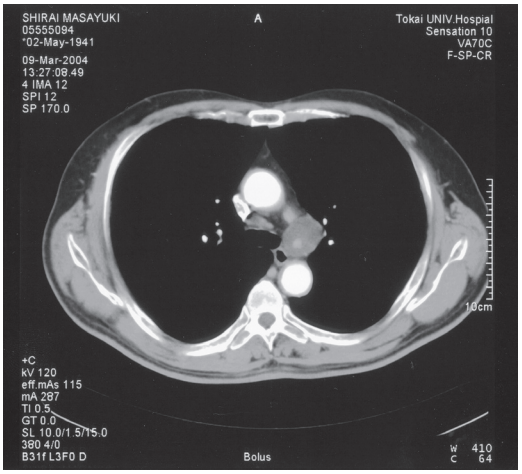

Fig. 2 Thoracic CT
An aneurysm of 37 mm projecting from the aortic arch region to the lesser curvature was observed.

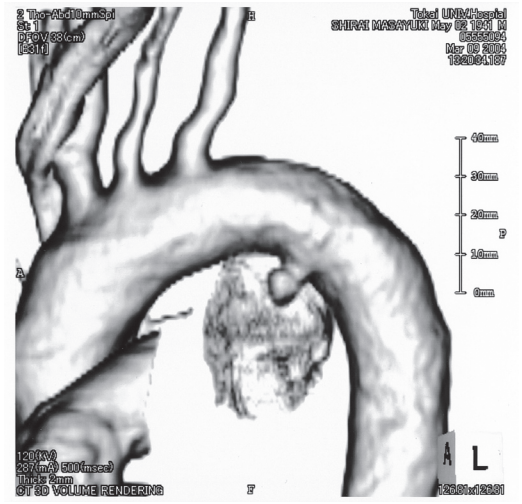

Fig. 3 3D CT VOLUME RENDERING
An aneurysm of 37 mm projecting from the aortic arch region to the lesser curvature was observed. The vascular lumen was mostly occupied by a thrombus, but blood flow was observed in a region connected to the aorta.

aorta. No stenosis was observed by coronary angiography.

Observation during surgery: Posterolateral excision of the fourth left intercostal space was performed up to the thoracic aorta, and the mediastinal pleura was incised up to the aneurysm. The aneurysm, measuring 30 mm × 28 mm, was sacciform in the arterial duct region of the peripheral left subclavian artery in the distal arch region. Neither inflammation nor adhesion was detected around the aneurysm, but the aneurysm wall was partially thinned, and adhesion of the ligament of the arterial duct to the bottom of the aneurysm was observed. The recurrent laryngeal nerve was overextended due to adhesion to the front surface of the aneurysm, and partially removed (Fig. 4). Partial extracorporeal circulation was performed from the right femoral artery to the right cardiac atrium via the right femoral vein. Blocking of arteries around the aneurysm was performed immediately distal to the left subclavian artery on the central side and at the descending part of the aorta on the distal side. Mild arteriosclerotic changes and thrombosis on the wall were observed in the aneurysm. After the aneurysm was

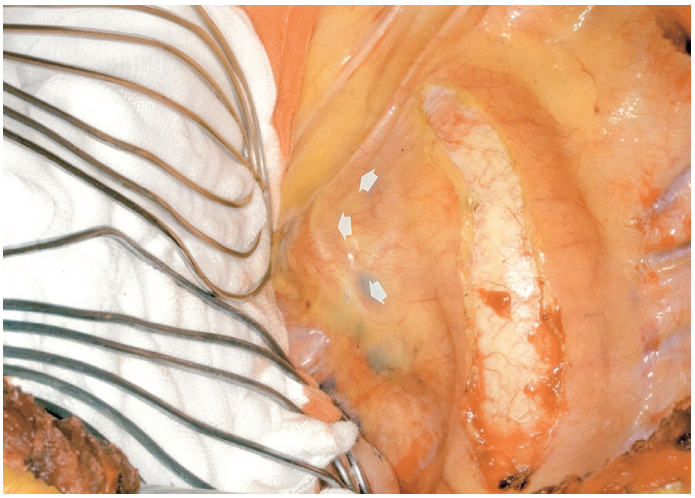

Fig. 4 Photograph taken during surgery
As shown by an arrow, adhesion of the ligament of the arterial duct to the bottom of the aneurysm was observed, and the recurrent laryngeal nerve was extended.

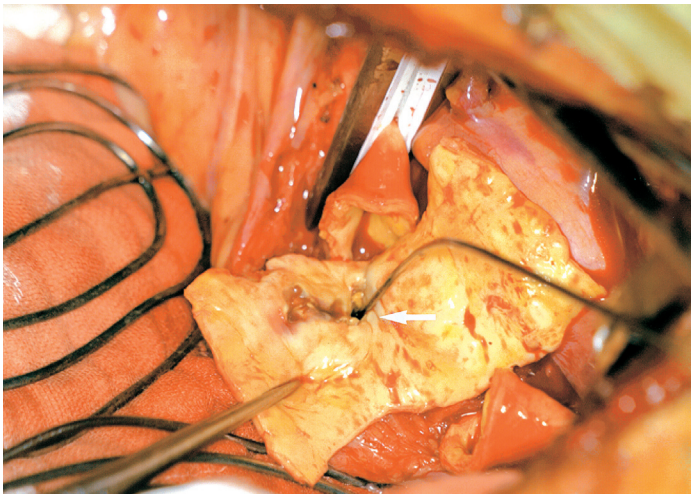

Fig. 5 Photograph taken during surgery
Arteriosclerotic changes and thrombi on the wall were observed by incision of the aneurysm. The aneurysm was connected to the pulmonary artery, shown by a probe (arrow), by ligament-like tissues, but there was no patency between the aneurysm and pulmonary artery.

removed, an aneurysm opening of about 10 mm was observed on the wall, but there was no patency between the aneurysm and pulmonary artery although connection of these arteries by ligament-like tissues was observed (Fig. 5). The affected region was replacement with a 20 mm Hemashield Graft was performed.

Histological observation: No histological findings characteristic of this patient were obtained. Though the morphology of true aneurysms with a 3-layer structure was maintained, most elastic fibers and muscular components of the tunica media had disappeared.

In the tunica intima, calcification and marked atheromatous Degeneration were observed.

Patch closure is indicated in patients with a relatively small Entry of arterial

duct aneurysms without surrounding atherosclerotic changes. However,since this patient showed marked atherosclerosis in the entry area and surrounding tissue, graft replacement was selected for the prevention of recurrence.

Postoperative observation: The postoperative course was satisfactory without complications, but the hoarseness was not improved. The patient was discharged from the hospital 21 days after surgery, and has consulted the outpatient department. The hoarseness due to palsy of the left recurrent laryngeal was improved about 3 months after surgery.

DISCUSSION

Though about 100 infants with arterial duct aneurysms have been reported, adults with this disorder have been very rare. To our knowledge, only 32 adult cases including autopsy cases [1-10] have been reported since the report by Thoma *et al.* in 1890, and surgical resection with a definite diagnosis has been performed only in 22 cases.

Arterial duct aneurysms are classified into: 1) the spontaneous infantile type that is present at birth and rapidly grows and 2) the adult type that grows during childhood or adulthood [2]. Falcone *et al.* [3] classified this disorder into 3 types: I) aneurysms that are patent on both the aorta side and pulmonary artery side of the arterial duct, II) those that are patent on the pulmonary artery side, and III) those after operation for patent ductus arteriosus, and described that type I is frequently observed in children/infants and type II in adults.

Concerning the pathogenesis of this disorder, the arterial duct is physiologically closed soon after birth in general, but complete anatomical closure due to vascular endothelial proliferation requires about 1 month. This organic closure occurs from the pulmonary artery end of the arterial duct and terminates at the aortic end. Therefore, the cause of arterial duct aneurysms in infants has been suggested to be a delay in medial smooth muscle constriction due to hypoxemia and acidosis and a resulting closure delay at the aortic end of the arterial duct [4]. However, since some infantile aneurysms spontaneously regress, the pathogenesis of arterial duct aneurysms in adults may not always be the same as that in infants. This patient had a history of hypertension. Past history was clearly described in 26 of the 32 previously reported patients, and 14 of them had a history of hypertension. In particular, 3 of 4 patients in whom arterial duct aneurysm was detected in their 20s had a history of hypertension. Unlike elastic arteries such as the pulmonary artery and aorta, the physiological arterial duct has only a few elastic fibers in the media. The aneurismal wall shows medial necrosis and mucus degeneration as is observed in Marfan syndrome, and the arterial duct is readily dilated [5]. Adult type aneurysms of the arterial duct is frequently observed at the age of 60-69 years [6], suggesting an age-related increase in the degree of the involvement of the atherosclerotic factor. Therefore, in addition to congenital factors, atherosclerosis as an acquired factor may be involved in this disorder.

In the 32 reported patients, symptoms most frequently described were hoarseness and dyspnea (about 40%), followed by cough and sputum, hemoptysis, chest pain, back pain, and swallowing difficulty. However, many recently reported patients underwent operation in the absence of symptoms.

The frequent complications are rupture into the surrounding organs and recurrent nerve paralysis, and other symptoms include embolism and infection. The closest attention should be paid to the high incidence of rupture due to the fragility of the arterial duct wall. The cause of death was rupture in 10 of 16 patients. Falcone *et al.* [3] reported that 57 of 61 collected cases were autopsy cases, which supports the importance of this complication. Therefore, Mitchell *et al.* [7] described that an aneurismal diameter of 3 cm or more is an indication for surgery even in the absence of symptoms.

For surgery, either median sternotomy or left thoracotomy can be selected. In our patient, the latter was selected because no pulmonary dysfunction was observed, and interruption was possible immediately below the left subclavian artery. However, medial sternotomy, which facilitates central side interruption, has been reported to be useful in patients with aneurysmal rupture [8]. As assisting measures, we consider that partial extracorporeal circulation using an artificial heart-lung machine is safe because this aneurysm has the risk of accidental hemorrhage during dessection

[9]. Aortic repair is generally performed by patch closure. However, in patients such as ours with marked atherosclerotic changes, pseudoaneurysms at the anastomosis site may develop [8], and no arterial duct tissue is left. Therefore, in this patient, graft replacement was selected.

CONCLUSION

We reported a 62-year-old male with an arterial duct aneurysm showing hoarseness who was treated by graft replacement after left thoracotomy under partial extracorporeal circulation. The adult type of this disorder is very rare, and its pathogenesis and surgical procedures were also discussed.

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