CASE REPORT

Two stage surgical treatments for a chronic type B dissecting aortic aneurysm with aberrant right subclavian artery and Kommerell Diverticulum ; a case report

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Kommerell憩室と異所性右鎖骨下動脈を伴ったB型慢性解離性大動脈瘤に対し二期的手術を行った1例

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Abstract

We report a case of two stage operations for a chronic type B dissecting aortic aneurysm with aberrant right subclavian artery(ARSA) and Kommerell's diverticulum (KD). A 62-year-old man with a type B aortic dissection from 9 years before underwent follow-up Computed Tomography (CT) revealing aortic arch with KD expanded to 53mm and aberrant right subclavian artery expanded to 23mm in diameter. We performed two stage operations. In the first, we performed ARSA division at the proximal part of vertebral artery and transposed the distal end to right common carotid artery transposition. Subsequently, we performed replacement of descending aortia and over sewing of the aneurysmal of the origin ARSA under circulatory arrest. KD with ARSA is a rare anomaly of the aortic arch, and it may develop complications such as distal embolization, subclavian-esophageal fistula, dissection and rupture. We performing in two stages, subclavian reconstruction before the intrathoracic repair would be expected to reduce the subsequent risk of distal ischemia or subclavian steal.

和文要旨

我々は，異所性右鎖骨下動脈（ARSA）とKommerell憩室（KD）を伴った慢性B型解離性大動脈瘤に二期的手術を行った１例を報告する。症例は，９年前にB型大動脈解離を来した62歳の男性でCTによる定期検査を受け，KDを伴う53mmに拡大した大動脈弓と直径23mmに拡大した異所性右鎖骨下動脈を認めた。治療は，２期的手術の方針とし，初回は椎骨動脈の近位部でARSAを離断し，末端部を右総頸動脈に転位吻合した。二期目は，循環停止下で下行大動脈置換と瘤化したARSAの起始部を縫合した。ARSAを伴ったKDは稀な大動脈弓異常であり，末梢部の塞栓形成，鎖骨下動脈食道瘻，解離や破裂などの合併症を呈する可能性がある。我々が二期的に行った，大動脈置換前の鎖骨下動脈の再建は，末梢の虚血または鎖骨下動脈盗血のリスクを低下させると思われた。

Key words: diverticulum; aberrant subclavian artery; dissecting aortic aneurysm

Introduction

Kommerell's diverticulum (KD) with aberrant right subclavian artery (ARSA) is a rare anomaly of the aortic arch. However, an ARSA is the most common of the intrathoracic major arterial anomalies, with an incidence of 0.4-2%.[1)] Most patients with KD, further complicated by ARSA, are asymptomatic. Aneurysms originating at ARSA from the descending thoracic aorta with KD include complications of distal embolization, compression of adjacent structures, dissection, and rupture.[1)~2)] A number of surgical procedures are performed for aneurysmal treatment of these KD with ARSA. The patient consented to use of case information and images for publication.

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Case report

A 62 year-old man with hypertension and 40 years history of smoking had previously undergone a partial gastrectomy by gastric ulcer perforation at the age of 26. The patient developed type B aortic dissection 9 years before and had been receiving antihypertensive medication. Systolic blood pressure was controlled to <120mmHg. During the follow-up period in outpatient department, abdominal aortic replacement was performed for dissected abdominal aortic aneurysm one year before. Computed Tomography (CT) revealed aortic arch with KD was expanded to 53mm and ARSA with retroesophageal segment was expanded to 23mm in diameter. ARSA and KD were complicated dissection, and the lesion was continuous from the aorta. (Figure1)

first operation

Surgical approach performed as a right supraclavicular incision in the supine position. We made an incision of sternocleidomastoid and anterior scalene muscle. The recurrent laryngeal nerve could not seen through this incision. The right phrenic nerve was identified and preserved and the vagal nerve was identified in the posteromedial aspect of the jugular vein. The right common carotid artery (CCA) and subclavian artery were divided. We performed ARSA division at the proximal part of right vertebral artery (VA) and transposed the distal end to right CCA transposition of distal portion with preservation of the right VA. Brain oxygenation monitoring during operation did not show significant changes. Postoperative complications did not develop.

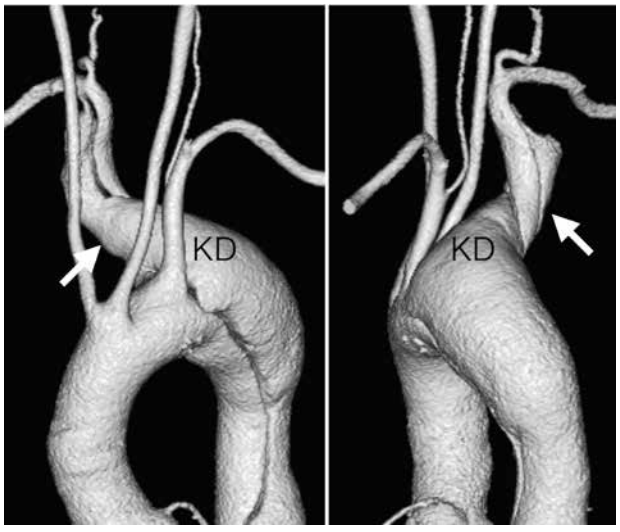

Figure 1 : Preoperative three-dimensional CT image of a dilated aortic arch with KD and ARSA (arrow) with dissection.

second operation

Surgical approach was performed in a left posterolateral thoracotomy in fourth intercostal space using cardiopulmonary bypass in the right lateral decubitus position. We performed resection and replacement of the descending aorta and over sewing the aneurysmal of the origin aberrant subclavian artery in deep hypothermia (20℃) under circulatory arrest.

outcome

Postoperative three-dimensional CT image demonstrated that ARSA was anastomosed to right CCA and showed the descending aorta replacement. (Figure 2) The postoperative blood pressure showed no difference between the right and left arms and no ischemic symptoms. The patient had delayed post hypoxic leukoencephalopathy, without any other circulatory disorders. He underwent rehabilitation and returned to work 7 months later. At the 2-year follow-up, CT angiography revealed patent anastomosis of the ARSA to right CCA transposition and dissected descending aorta had no dilatation.

Discussion

In 1936, Kommerell reported an aortic diverticulum in a patient who had an ARSA originating from the descending thoracic aorta of a left-sided aortic arch.[3] The KD consisted of an aneurysm of thoracic aorta as well as an aneurysmal originating from aberrant subclavian artery.

An aneurysm of ARSA is an extremely rare condition. 60% of patients have an aberrant subclavian artery

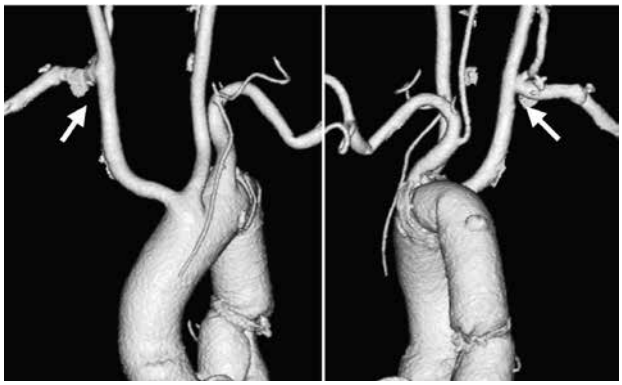

Figure 2 : Postoperative three-dimensional CT image demonstrated that right aberrant subclavian artery was anastomosed right carotid artery (arrow) and descending aorta was replacement.

from KD.[4] ARSA is caused by abnormal development of the right fourth arch and the vestigial remnants of the right dorsal primitive aorta. In most cases, ARSA crosses the retroesophageal space to the right arm.[5]
Most patients with KD are asymptomatic, but dilatation of KD results in compression of the surrounding structures, such as dysphagia, dyspnea, stridor, wheezing, cough, recurrent pneumonia, obstructive emphysema, or chest pain. Patients with KD have been observed having serious complications involving embolization, dissection and rupture.[2),5),6)] Moreover, there may be a risk of subclavian-esophageal fistula with severe gastrointestinal bleeding in the long-term.[7)]

Though our patient was asymptomatic, we observed dilatation to 23mm of ARSA and 53mm of KD. Furthermore, he had a type B dissected aortic aneurysm.

Surgical indications have not been established because of the rareness, and the unknown natural history, of aneurysms associated with KD. Cina and colleagues reported that in a review of aneurysms associated with 32 patients, patients who had a right aortic arch with aberrant left subclavian artery experienced rates of rupture or dissection of 53%. The mortality was 8.3%.[5)] Austin and Wolfe reported a rate of rupture of 19% among 32 patients, all of whom died. Their operative mortality for elective treatment of aneurysm with KD was 16.6%.[6)] Cina and colleagues suggest aggressive treatment for aneurysms with a diameter of 3 cm or greater in good-risk patients. Ota and colleagues recommended surgical indications for symptomatic patients who had KD with a diameter of more than 5cm.[8)] Our patient with complicated dissection at onset 9 years before, had an aorta with KD diameter of 4cm. KD was less than 5 cm in diameter, therefore we recommended early intervention appropriate.

In 1946, Gross described an initial report about the surgical treatment of this anomaly. According to the report, ARSA was divided and ligated by a left thoracotomy. Children have the ability to develop collaterals to the right upper extremity, but adults develope upper extremity claudication and steal syndromes.[9)] Kamiya and colleagues reported that treating ARSA aneurysms using one approach is very difficult and that 2-staged surgical approach results were excellent and without complications. Their method, using the supraclavicular approach and a left thoracotomy, enables secure and complete repair of ARSA aneurysm.[10)] However, some cases that did not show revascularization of the subclavian artery caused a steal syndrome. We considered if transposition of the ARSA or carotid subclavian bypass was necessary. [9),10)]

In the literature, the majority of procedures were 2-staged operations, of which descending aorta and ARSA were approached seperately.[5)-6),8),10),11)] There are reports to recommend in situ reconstruction8, but in certain cases, ARSA can become aneurysmal or dissected, therefor we recommend 2-stage operations in the elective case. This approach also has the additional benefit of preventing the possible occurrence of subclavian-esophageal fistula.

In recent years a report on endovascular repair of KD was founded.[11)] We consider surgical procedure case by case. However, as in our case, endovascular repair is restrictive in cases with aortic dissection.

It is indispensable that careful preoperative imaging and surgical plans for anatomy are considered.[5)]

Conclusion

In conclusion, we completed anatomical repair of the KD with ARSA aneurysm and type B dissected aortic aneurysm performed in two stages, (the supraclavicular approach and left posterolateral thoracotomy). Performing subclavian reconstruction before the intrathoracic repair would be expected to reduce the subsequent risk of distal ischemia or subclavian steal.

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