GHM Open. 2024; 4(2):91-94.

CORRESPONDENCE

DOI: 10.35772/ghmo.2023.01029

Direct aortic suture technique for anomalous systemic arterial supply to the basal lung: A retrospective cohort study

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Abstract: Anomalous systemic arterial blood supply to the basal lung (ABLL) is a rare congenital malformation. Although surgical resection is the standard treatment, the surgical techniques for aberrant arteries remain poorly discussed. Herein, we evaluated the efficacy of our direct suture closure technique in preventing aneurysmal changes in aberrant artery stumps through a retrospective review of the medical records of patients who underwent surgical resection of ABLL at our institution between January 2013 and January 2023. The diagnosis of ABLL was based on enhanced computed tomography (CT) findings. To treat ABLL, we performed lateral thoracotomy through the 5th intercostal space *via* a ~10 cm skin incision. After anatomical pulmonary resection, the aortic stump of the aberrant artery was sutured directly with a felted non-absorbable thread. In one patient, we further examined the postoperative blood flow using 4D-flow magnetic resonance imaging. Overall, 5 consecutive patients, including four (80%) females with a median age at operation of 59-year-old, were assessed. The median operative time was 166 min, and the median blood loss was 34 ml. There were no cases of perioperative mortality or morbidity, and the median hospital stay was 8 days. No vortex flow was observed in 4D-flow evaluation of blood flow. Histological changes were observed in the aberrant artery, including fibrous intimal thickening, atherosclerosis, intramural thrombus, and collection of foam cells and lymphocytes. Thus, we present this technique as a safe treatment for ABLL that allows for the preservation of blood flow and complete resection of abnormal vessels.

***Keywords*:** direct aortic suture technique, anomalous systemic arterial supply to the basal lung, pulmonary sequestration, 4D-flow MRI, aberrant artery

Introduction

Pulmonary sequestration (PS) is a rare congenital malformation first described by Pryce in 1946. PS is subdivided into two types: intrapulmonary and extrapulmonary fractionation (*1*). The condition initially classified as Conventional Pryce type III, defined as an anomalous systemic arterial supply to the basal lung (ABLL), is now considered an independent disease (*2*). The presentation of this disease varies widely, with patients ranging from asymptomatic hemoptytic, requiring emergency surgery. However, surgical resection and/or endovascular treatment is generally recommended after diagnosis in all cases (*3*). PS is rare, with an incidence of only 0.15-1.7% among all congenital malformations (*4*). Therefore, surgical techniques and postoperative complications remain poorly discussed.

Damage to the lungs can cause secondary infection and hemoptysis (*5*), therefore, pulmonary resection is essential. Distinct from usual pulmonary resection, in PS surgical methods to treat aberrant arteries must be performed in addition to lung resection. Division of the aneurysmal aberrant artery has been described as having a high risk of rupture during surgical dissection or re-aneurysmal changes of the arterial stump following surgery (*6*). The effects of endovascular treatment, such as embolization or stent grafting, on aberrant arteries from the descending aorta have previously been demonstrated (*7,8*).

Recently, thoracoscopic surgery has been proven to be a minimally invasive alternative to conventional thoracotomy. In this technique, the aberrant artery can be resected using a stapler during thoracoscopic surgery (*9,10*). Previous reports have also described endovascular treatment of aberrant arteries performed prior to lung resection (*7-9,11*). Various methods for this technique have been reported by many institutions. The bifurcations and roots of aberrant arteries can be observed as slightly saccular protrusions from the descending aorta. This sac-shaped protruding stump can lead to turbulent blood flow in the descending aorta.

However, no consensus has yet been reached regarding the treatment of aberrant artery stumps.

In our department, which includes both cardiovascular and thoracic surgeons, during surgical management of such patients, we apply a side clamp to the descending aorta after anatomical lung resection, and use felt for direct suture closure of the resected stump. In the present study, we evaluated the results of surgical cases managed using this novel technique. Throughout the study period, we assessed the perioperative clinical course, pathology of the aberrant artery, and blood flow in the descending aorta after resection in 5 patients. Further, we examined postoperative blood flow using 4D-flow magnetic resonance imaging (MRI) in one patient.

Study design for direct aortic suture technique

Ethical considerations

This study was approved by the Institutional Review Board of the Hamamatsu University School of Medicine (approval number 23-126). The need for informed consent was waived due to the retrospective study design.

Data analysis

Herein, we retrospectively reviewed the medical records of patients who underwent surgical resection of ABLL at our institution between January 2013 and January 2023, and further extracted the relevant clinical data. The diagnosis of ABLL was made based on the findings of enhanced computed tomography (CT), according to the conventional Pryce classification type III, which require that abnormal arteries return only to the PS and intrapulmonary sequestration shared visceral pleura with the normal lung. The resection range was defined as the area of the normal pulmonary artery bifurcation, inflow of the aberrant artery, destructive changes, and secondary infection. Patients who received oxygen therapy or were unable to tolerate general anesthesia due to organ failure were excluded.

Surgical technique

Lateral thoracotomy was performed through the 5th intercostal space. A skin incision of approximately 10 cm was made for segmentectomy or lobectomy. The aberrant arteries were dissected 2 cm apart, and resected using a staple. After anatomical lung resection, a side-biting clamp was placed on the descending aorta around the aberrant artery in the same surgical view, and the staple was removed by clamping the descending aortic staple transection. Felt-reinforced mattress sutures were then placed to close the stump of the aberrant artery, passing through the aortic wall just beside the aberrant artery take-off (Figure 1).

4D-flow magnetic resonance imaging

A 3-Tesla magnetic resonance scanner (Discovery MR750 or MR750w; GE Healthcare, Waukesha, Wis, USA) was used for 4D-MRI. Contrast-enhanced 3D magnetic resonance angiography was performed first to define the shape of the aortic wall. Subsequently, a bolus injection of 0.1 mmol/kg gadolinium chelate (Omniscan; Daiichi Pharma Co., Japan) was administered at an injection rate of 2.0 mL/s, followed by saline (20 mL) at the same injection rate. Respiration compensated for retrospective cardiac gating was used for 4D-flow imaging. Raw data were transferred to a personal computer for postprocessing and flow visualization in the Digital Imaging and Communications in Medicine (DICOM) format. The flow analysis software Flova (R'Tech Co, Hamamatsu, Japan) was used to visualize intraaortic flow information at a spatial resolution of 2 × 2 × 2 mm.

Clinical Characteristics of 5 ABLL patients

Five consecutive patients were assessed in this study. The median age at operation was 59-year-old, and 4 of the 5 patients (80%) were female. The only male patient was a current smoker. Four patients presented with symptoms, including repeated pneumonia from childhood in two patients, and epigastric pain and hemoptysis each in one patient. The respiratory function measured in elective operation cases was within the normal limits. Respiratory function could not be evaluated in one patient who required emergency surgery due to hemoptysis. The number of aberrant arteries was usually one, and only one case showed two aberrant arteries.

Perioperative outcome included postoperative blood flow

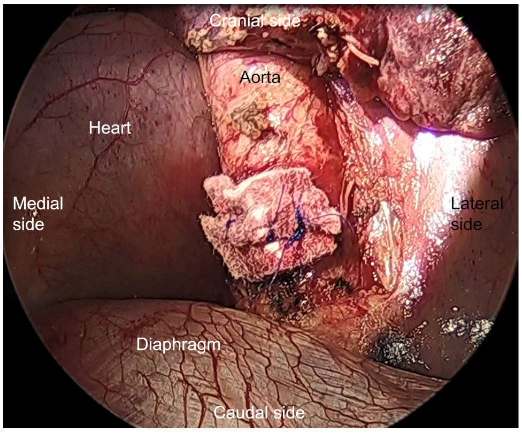

Figure 1. Intraoperative findings after lung resection and direct suture of the aorta. The stump of abnormal vessel was enclosed using felt and non-absorption thread.

We performed segmentectomy in three patients and lobectomy in two patients. The median operative time was 166 min, and the median blood loss was only 34 ml. There were no cases of perioperative mortality or morbidity. The median hospital stay was 8 days (range 5-13 days). One month after surgery, the descending aorta was smooth, and no internal vortex flow was observed on 4D-flow evaluation of blood flow (Figure 2).

Histopathological examination of aberrant artery

Histopathological examination revealed fibrous intimal thickening in three patients, atherosclerosis in three patients, intramural thrombus in two patients, and collection of foam cells and lymphocytes in one patient. Inflammatory cell infiltration was observed in 4 patients, bronchial epithelial metaplasia in 3, hemorrhage in 3, and chronic inflammation in 2.

Clinical outcome of direct aortic suture technique

In the present study, we found that the walls of aberrant arteries invariably show pathological changes that may underlie future aortic problems. Our direct suturing technique enables complete removal of the modified tissue, allowing the creation of a smooth aortic internal surface. In our study, absence of a protruding remnant or vortex flow was observed on 4D flow MRI in the one patient who underwent this analysis. As the exposure of the descending aorta is excellent after anatomical lung resection, this method can be performed safely through the same surgical field without prolonging the operative time or increasing blood loss. All patients, including those who underwent emergency surgery, were discharged within a short period without experiencing postoperative complications.

Histopathological changes in aneurysms of aberrant arteries

Several studies have previously demonstrated the formation of aneurysms in aberrant arteries (*11-14*). However, the mechanisms by which aberrant arteries form aneurysms remain unclear. However, pathological changes in aberrant arteries may be one such mechanism. Aneurysms of aberrant arteries caused by arteritis or arteriosclerosis in childhood have long been reported (*4*), and enlarged aberrant arteries were further observed inside a PS that had lost its normal structure due to chronic inflammation and interstitial fibrosis (*15*). Various histological changes, including those described previously, were observed in this study. Although there have been no prior case reports of clinical problems linked to treatment, Shibano *et al.* reported concerns regarding reaneurysm formation on the stump of an aberrant artery (*5*).

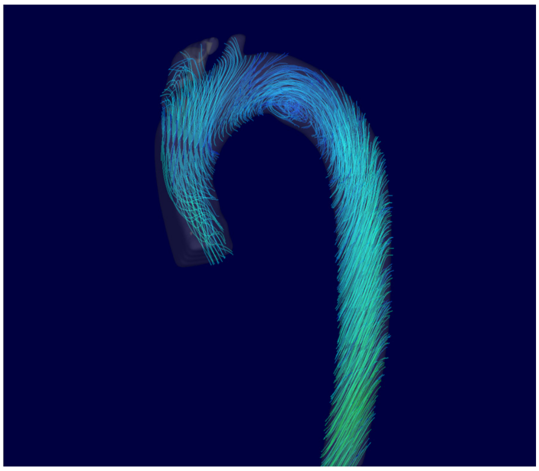

Figure 2. Results of postoperative 4D-flow magnetic resonance imaging (MRI) in the patient.

Postoperative blood flow in the aorta

Another concern which should be considered in cases of remnant protrusion is the turbulent flow within the protrusion. In a previous study, we showed that wall shear stress (WSS) is low in saccular aneurysms with a sac depth/width ratio > 0.8, which is caused by vortex flow within the aneurysms (*16*). The aberrant arterial remnant after stapling had this shape. A low WSS promotes atherosclerosis, and may cause aneurysm formation (*17*). Malek *et al.* previously demonstrated the biological responses to low WSS and the morphological transformation of endothelial cells (*18*). Overall, we believe that our method may help prevent histological changes by maintaining laminar blood flow in the aorta.

Limitation

This study has several limitations which should be mentioned. First, this method can only be performed in cases in which the lesion is located on the left side. Second, only five cases were performed at a single institution; thus, the number of cases was small, and the long-term results for more than ten-years remain unknown. Third, our method is not a procedure that can readily be performed by thoracic surgeons alone, but instead requires joint collaboration with cardiovascular surgeons. Furthermore, previous studies have demonstrated the incidence of intraoperative bleeding from aberrant arteries (*10*).

In conclusion, the present study showed that our direct suture closure technique is safe for ABLL. This procedure allows the complete resection of abnormal vessels and avoids vortex flow in the descending aorta, which may prevent future aortic complications.

Funding: This work was supported in part by the Smoking Research Foundation (grant no. 108).

Conflict of Interest: The authors have no conflicts of interest to disclose.

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Received December 9, 2023; Revised May 29, 2024; Accepted June 10, 2024.

Released online in J-STAGE as advance publication June 14, 2024.

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