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Application of fibrobronchoscopy-guided aortic suspension in pediatric patients with bronchial compression resulting from repair of coarctation of the aorta: a case report

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Abstract

Background Coarctation of the aorta (CoA) is one of the more prevalent congenital heart diseases (CHD). The conventional treatment approach involves surgical correction of the coarctation. This procedure, however, can be associated with several complications, such as re-stenosis, aneurysm formation and pseudoaneurysm. One rare but significant complication is left bronchial compression, which may result from anterior displacement of the thoracic aorta or compression of the trachea by adherent tissue.

Case presentation We present a case of a 7-month-old postoperative CoA patient who developed stenosis due to compression of the left main bronchus by the thoracic aorta. After comprehensive evaluation, we utilized a fiberoptic bronchoscope to accurately expose the site of aortic compression and subsequently performed bronchial release and aortic suspension procedures to alleviate the symptoms. The child demonstrated a satisfactory recovery following the surgical intervention. This successful case provides valuable insights for managing similar cases in the future.

Conclusions Computing tomography (CT) airway reconstruction enables precise diagnosis of tracheal stenosis secondary to aortic arch pathology or coarctation repair. Intraoperative fiberoptic bronchoscopy provides real-time anatomical localization of stenotic segments, warranting standardization in high-risk aortic surgeries to ensure airway patency. Three-dimensional airway modeling should be incorporated into perioperative reassessment and longitudinal follow-up protocols.

Keywords Coarctation of the aorta, Complications, Bronchial compression, Fibrobronchoscopy-guided surgery

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Background

Coarctation of the aorta (CoA) is one of the more common congenital heart disease (CHD), accounting for approximately 5% of all congenital heart malformations [1]. It is characterized by a localized narrowing of the thoracic aorta at the isthmus. The clinical presentation of CoA varies depending on the severity and extent of the stenosis, as well as the timing of ductus arteriosus closure [2]. CoA was initially described by Johann Friedrich Meckel in 1750, and the first successful surgical repair was accomplished by Clarence Crafoord in 1944 [3]. Advances in surgical techniques have led to significant improvements in treatment outcomes, CoA now has been viewed as a disease that can be entirely cured by adequate surgery. Nevertheless, it is crucial not to overlook potential postoperative complications such as re-stenosis, aneurysm formation or pseudoaneurysm [4]. Bronchial compression emerging as a sequel to surgical reconstruction of the aortic arch for aortic stenosis represents a rare but clinically significant complication, with a documented incidence of 0.7% in the neonatal and infant patient population undergoing this procedure [5]. This report describes a case of left main bronchus stenosis caused by compression from the aortic arch after CoA treatment. Through surgical intervention, we successfully alleviated the bronchial compression, and the patient was discharged in stable condition.

Case presentation

A 7-month-old female infant was admitted to our hospital with a 5-month history of recurrent cough and shortness of breath. The patient was born at 38 weeks of gestation without any significant medical or family history. Two weeks after birth, the patient underwent corrective surgery at another hospital for CoA, atrial defect, and patent ductus arteriosus (Fig. 1a). Postoperatively, the patient experienced recurrent episodes of pneumonia and shortness of breath, leading to her transfer to our facility for further management. A computed tomography (CT) scan performed at our hospital revealed left main bronchial compression secondary to external compression, resulting in hyperinflation of the left lung (emphysema) (Fig. 1b-c). This condition was likely attributable to tracheal narrowing caused by anterior displacement of the thoracic aorta following the previous CoA repair surgery (Fig. 1d). Subsequently, fiberoptic bronchoscopy confirmed significant compression of the left main bronchus, which appeared as a slit-like narrowing, allowing passage of only a 3.1 mm outer diameter (OD) bronchoscopic probe.

After extensive preoperative preparation, we performed surgery on the patient using the 4th and 5th intercostal incisions. During the operation, significant adhesions were observed between the aortic root, the descending aorta and surrounding tissues. The left main bronchus was found to pass beneath the aortic arch and exhibited substantial adhesion and compression. Using a fiberoptic bronchoscope, we identified the location of airway compression. The severely compressed area was illuminated by the bronchoscope's light source, thereby clearly exposing it within the surgical field and facilitating precise subsequent surgical positioning (Fig. 2a). We exposed the aortic arch and a segment of the descending aorta and performed three 4 – 0 Prolene sutures reinforced with felt pads to secure the aortic arch and descending aorta to the left posterior thoracic wall (Fig. 2b-d). Intraoperative fiberoptic bronchoscopy confirmed that the compression of the left main bronchus was relieved and the trachea was patent, allowing for smooth passage of the bronchoscope probe. Postoperative CT data demonstrated a significant improvement in emphysema symptoms in the patient's left lung, with a marked alleviation of previous symptoms (Fig. 3a-e). However, 3D reconstruction of the CT scans revealed minimal residual constriction of the left main bronchus. Despite this finding, the patient was discharged one week after surgery due to the substantial improvement in respiratory function.

Discussion and conclusions

CoA is a prevalent CHD, characterized by a structural anomaly in the aorta present at birth or developing shortly thereafter [4]. This condition can be diagnosed in infants, children, or adults, and its management and treatment are feasible at any age. Treatment options for addressing aortic obstruction include surgical interventions and interventional management [6]. Although balloon angioplasty and stenting have advanced, most cases of aortic coarctation still require surgical therapy due to anatomical mismatch or vessel size discrepancies. Surgery remains the standard treatment for isolated CoA in neonates and infants [7]. The choice of surgical approach for aortic arch reconstruction is primarily determined by two factors: the patient's age and the location of the narrowing. A comprehensive evaluation of the patient's condition guides the selection of the most appropriate method. For the majority of patients with CoA, resection with extended end-to-end anastomosis (REEEA) has been the preferred procedure [8]. The refinement of surgical techniques has substantially improved survival rates; however, postoperative complications remain a significant concern. The primary reason for reoperation following aortic coarctation surgery include re-stenosis, aneurysm formation or pseudoaneurysm [9]. Of these cases, approximately 90% necessitated repeat surgery owing to restenosis at the site of the initial stenosis [10]. The incidence of postoperative aneurysm or pseudoaneurysm range from 5 to 9%, if left undiagnosed and untreated, the associated mortality rate can be as high as

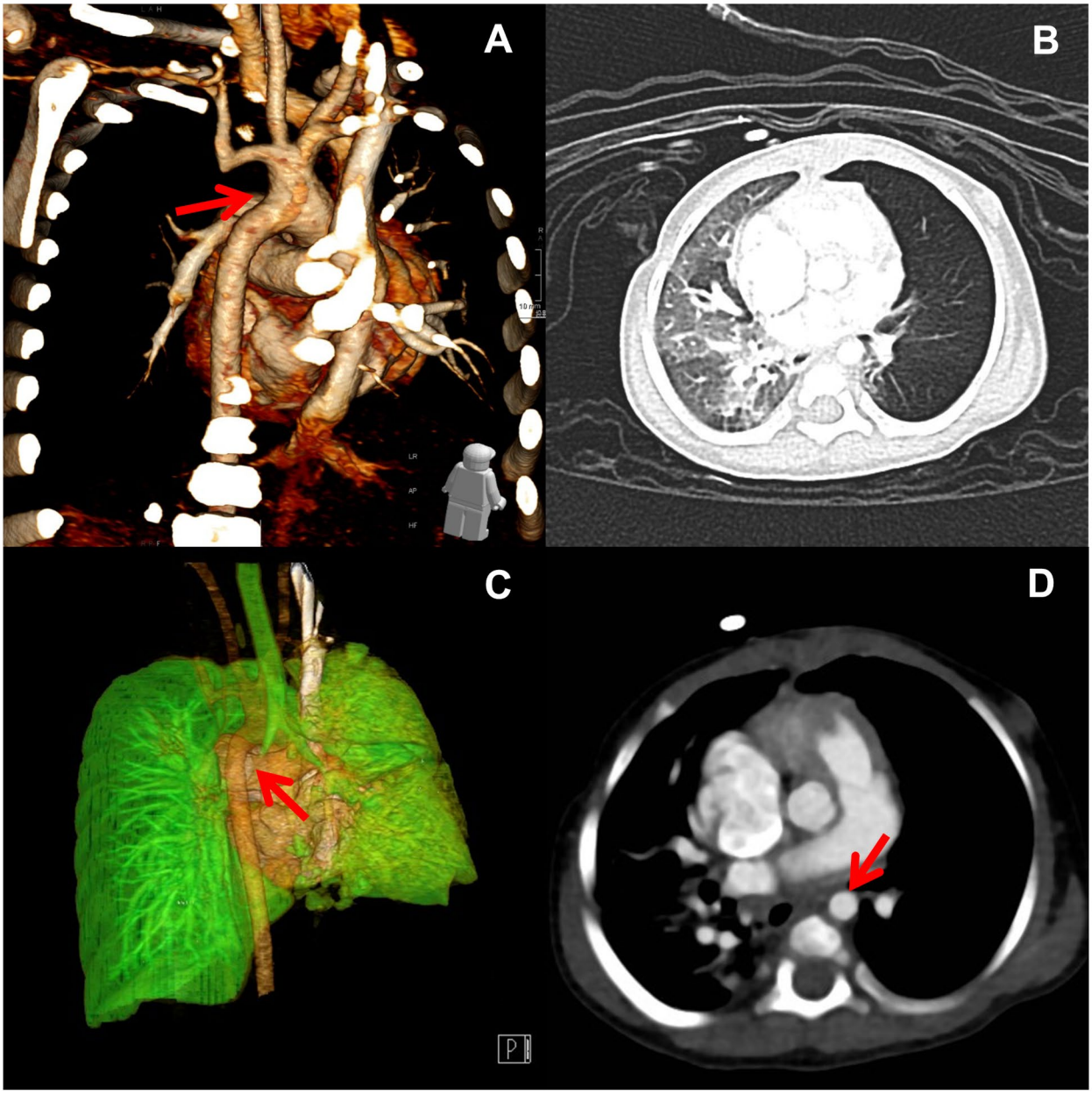

Fig. 1 Preoperative imaging data. (**a**) The 3D reconstruction image of the heart and major blood vessels in the pediatric patient following CoA surgery (the arrow indicates the site of aortic reconstruction); (**b**) The chest CT images of the pediatric patient obtained upon hospital admission; (**c**) The 3D reconstruction image of the child patient's airway (the arrow indicates the region of airway stenosis); (**d**) Enhanced CT image of the child's heart (the arrow indicates the anteriorly displaced aorta)

36–100% [11, 12]. Consequently, a comprehensive understanding and prompt management of such postoperative complications are essential for improving patient outcomes and survival rates.

However, airway stenosis is a rare but significant complication associated with this surgical approach. Postoperative left bronchial compression can result from compression of the tracheobronchial by the newly reconstructed aortic. Therefore, it is crucial for surgeons to consider the position of the left bronchus when selecting and performing the surgical treatment for CoA. In aortic arch reconstruction, stenosis of the left bronchus is rarely encountered during resection with extended end-to-end anastomosis. However, in complex aortic arch reconstructions such as those performed in Norwood procedures, addressing the spatial relationship between the aorta and the left bronchus presents a significant challenge for surgeons. To alleviate the compression of

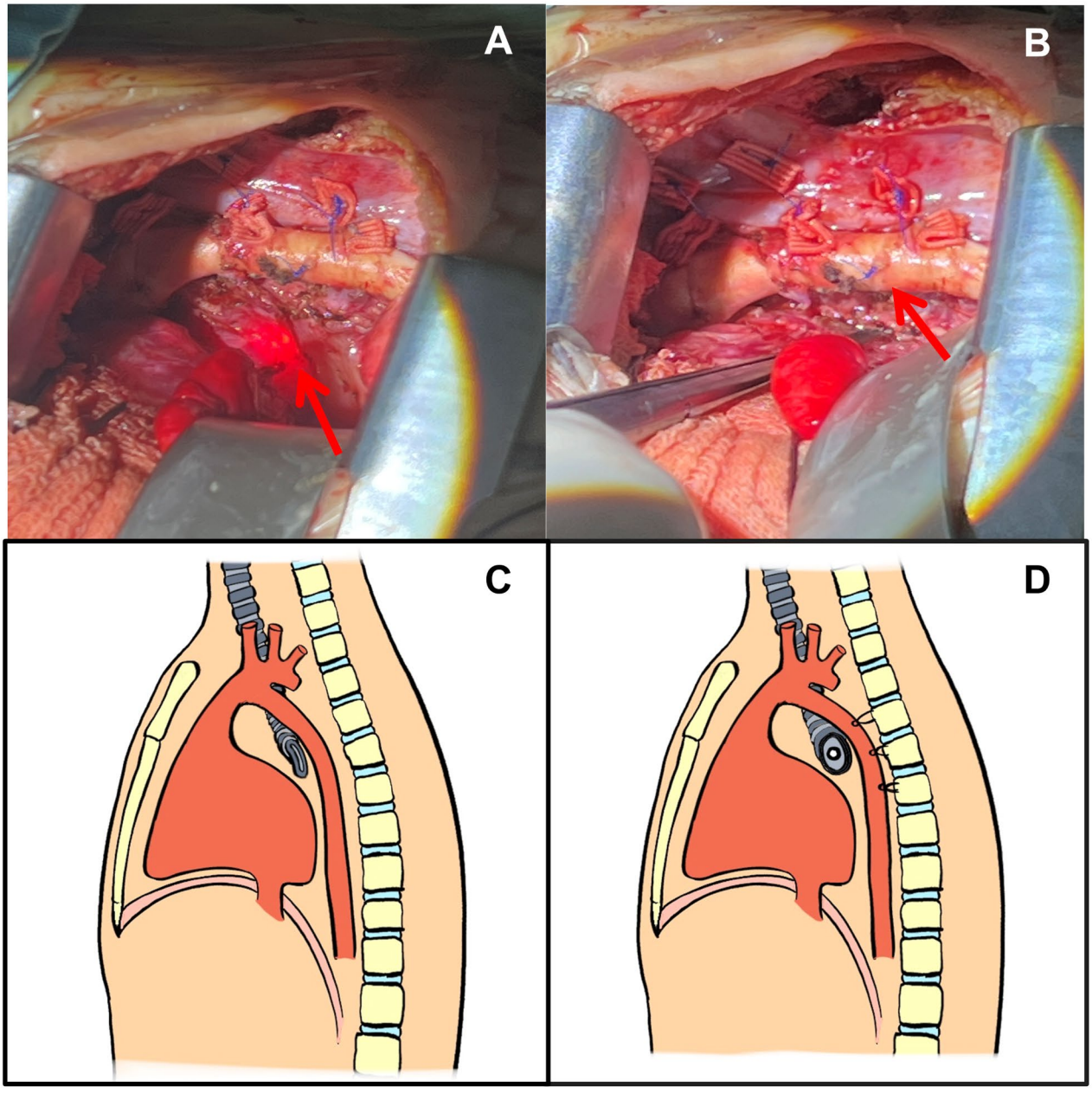

Fig. 2 Intraoperative images. (**a**) The fiberoptic bronchoscope was utilized to assess the extent and location of airway compression during the procedure (The red spot indicated by the arrow represents the light source of the bronchoscope); (**b**) The thoracic aorta was carefully dissected and suspended posteriorly towards the paravertebral region of the spine during the procedure (The arrow indicates the suspended aorta); (**c**) Schematic diagram of the aorta following the repair surgery for coarctation of the aorta in the pediatric patient; (**d**) Schematic diagram of the aortopexy procedure

the left bronchus caused by the reconstructed aortic arch during surgery, it has been proposed to utilize glutaraldehyde-treated autologous pericardium to elevate the aortic arch or to reposition the right pulmonary artery anteriorly to reduce tracheal pressure [13, 14]. Nonetheless, these interventions may have long-term implications for the child's hemodynamics.

During the patient's previous therapeutic intervention, the thoracic aorta was displaced anteriorly, leading to significant vascular compression of the left main bronchus due to stenosis caused by its passage under the aortic arch with substantial adhesion. Furthermore, considering the trachea's directly connection to the external environment, there is a potential risk of anastomotic infection. Therefore, our primary objective was to

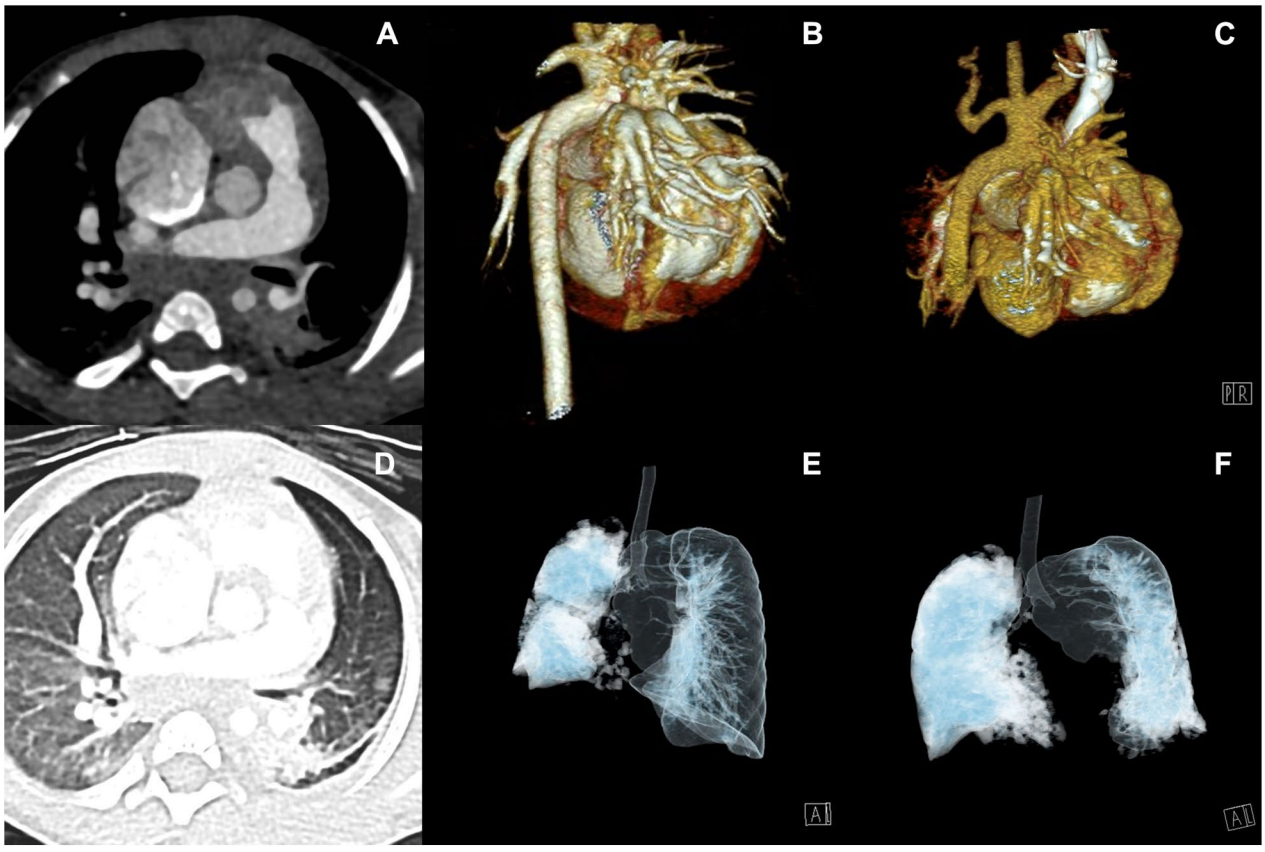

Fig. 3 Postoperative imaging data. **a.** The postoperative enhanced CT demonstrated a posterior displacement of the thoracic aorta; **b-c**. The 3D reconstruction results demonstrate that, in comparison to the preoperative state (**b**), the distance between the aorta and the pulmonary artery has significantly increased postoperatively (**c**); **d.** The postoperative lung CT scan demonstrated a notable improvement in the emphysema of the left lung; e-f. The 3D reconstruction of the postoperative airway (**f**) revealed a marked improvement in pulmonary ventilation conditions compared to the preoperative state (**e**)

alleviate the airway compression. After careful deliberation of our surgical strategy, we opted to address the thoracic aorta in situ rather than intervene with the trachea. This decision was primarily driven by the infant's small size, which precluded stent insertion. Before the operation, we utilized three-dimensional airway reconstruction technology to preliminarily predict the location and extent of airway stenosis in the child. During the procedure, we employed a fiberoptic bronchoscope to precisely identify the site of stenosis in the left main bronchus and accurately determined the thoracotomy site using the bronchoscope's light source, thereby enhancing the precision of the surgery. Finally, we examined the entire left bronchus following aortic suspension using the fiberoptic bronchoscope, and no significant stenosis was detected.

Currently, the utilization of fiberoptic bronchoscopy intraoperatively to assess the outcomes of airway plasty has become a adopted auxiliary technique [15, 16]. Studies have demonstrated that bronchoscopy-assisted aortic fixation can significantly enhance the medium- and long-term prognosis for patients who experience central airway obstruction following congenital heart surgery [17]. In this case, we integrated three-dimensional airway reconstruction with fiberoptic bronchoscopy to perform a comprehensive evaluation of the patient's tracheal stenosis. Additionally, we innovatively utilized fiberoptic bronchoscopy for real-time guidance, directly exposing the stenotic site within the surgical field, thereby assisting the surgeon in achieving more precise aortic fixation. Postoperatively, the child's left lung ventilation improved significantly, and the overall pulmonary condition was notably enhanced. However, it is important to highlight that the three-dimensional (3D) reconstruction images indicated that the left main bronchus remained stenotic. This condition may be attributed to tracheal softening resulting from prolonged aortic compression. Consequently, the presence of airway compression should have been assessed during the initial surgical intervention. However, considering the absence of significant symptoms and the young age of the child, it is reasonable to hypothesize that tracheal function may normalize as the infant matures and the compression is relieved. Alternatively, if complete normalization do not occur, surgical

intervention should be considered as an alternative option at an appropriate age.

The successful management of this case affords critical insights for handling similar scenarios that may arise in the future. For potential postoperative airway stenosis, the intraoperative use of a fiberoptic bronchoscope is essential to assess the airway condition. Aortic suspension may be employed as a conventional surgical approach if required. Additionally, airway reconstruction should be considered a critical postoperative evaluation to promptly identify any potential airway compression. If necessary, timely intervention and treatment via surgical or interventional methods should be implemented to prevent tracheal softening and collapse due to prolonged compression.

Tracheal stenosis following aortic arch reconstruction is an uncommon but clinically significant complication. The successful management of this case offers valuable insights for addressing potential similar occurrences in the future. Moving forward, surgeons should meticulously evaluate the anatomical location and morphology of the trachea when selecting a surgical technique for repairing CoA, particularly in pediatric patients.

Abbreviations

CoA	Coarctation of the aorta
CHD	Congenital heart disease
CT	Computed Tomography
OD	Outside diameter
REEEA	Resection with extended end-to-end anastomosis
3D	Three dimensional

Supplementary Information

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Supplementary Material 1

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Author contributions
Conceptualization, J.J.L; Writing-original draft preparation, H.Y.T and X.K.Z; investigation, Y.F.Y and S.Z.W; Picture drawing, K.G; Writing-review and editing, S.J.W. All authors have read and agreed to the published version of the manuscript.

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Data availability
The data and materials that support the findings of this study are available from the corresponding author upon reasonable request.

Declarations

Ethics approval and consent to participate
Approval was obtained from the ethics committee of the Second Xiangya Hospital of Central South University. The procedures used in this study adhere to the tenets of the Declaration of Helsinki.

Consent for publication
Since the child was a 7-month-old female infant, written informed consent for publication of her clinical details was obtained from her parents. A copy of the consent form is available for review by the Editor of this journal.

Competing interests
The authors declare no competing interests.

Clinical trial number
Not applicable.

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