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3D-modelling of transient left bronchial obstruction following bidirectional superior cavo-pulmonary shunt

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ABSTRACT

Extrinsic compression of airways is one the most important causes of respiratory insufficiency in the perioperative period in children with congenital heart disease. This is especially true of pathologies that involve surgery of the aortic arch or conduit replacement of the right ventricular outflow tract. However bronchial obstruction is uncommon in the setting of bidirectional cavopulmonary shunt alone.

We report the case of an infant with a functionally univentricular heart who had a bidirectional superior cavopulmonary shunt and disconnection of the main pulmonary artery from the ventricular mass with oversewing of pulmonary valve. Post-operatively the patient desaturated due to compression of left main bronchus by the left pulmonary artery anteriorly and the descending aorta posteriorly. This was clearly defined by CT based on 3D-modelling of the airways and great vessels. The child was managed conservatively by ventilator support, selective bronchial suctioning and systemic steroids with a successful outcome.

Keywords: bronchial compression, left pulmonary artery, descending aorta, CT angiography, 3D-modelling

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INTRODUCTION

The current approach to the surgical management of patients with univentricular hearts is staged repair, which includes neonatal surgery to establish a source of controlled pulmonary blood flow and eliminate systemic outflow obstruction, followed successively by bidirectional superior cavopulmonary shunt (BSCPS) and a Fontan completion. Respiratory compromise is an important cause of desaturation following a BSCPS and is usually due to consolidation or collapse of the lung parenchyma and/or collections of fluid or air in the pleural space. Respiratory compromise due to bronchial obstruction is uncommon in this setting.

We present a patient with a functionally univentricular heart who had a normal airway. Following a BSCPS, she developed desaturation with inability to wean from ventilator. Brochoscopy and CT angiography revealed compression of left main bronchus by pulmonary artery anteriorly and descending aorta posteriorly. The site and cause of obstruction was clearly defined by CT-based 3D-modelling of the trachea, bronchi and great vessels. The patient improved with conservative management and was extubated and discharged home without any residual airway obstruction.

CLINICAL REPORT

A full-term baby was diagnosed with double inlet left ventricle (DILV), levo-transposition of great arteries (L-TGA), large unrestrictive ventricular spetal defect (VSD), and an atrial septal defect (ASD). Aorta originated from the non-dominant anterior ventricle and pulmonary artery came from the dominant posterior ventricle. A small patent ductus arteriosus (PDA) was also present. The main pulmonary artery was banded and the PDA ligated in the neonatal period. Follow up echocardiography showed pulmonary artery band gradient of 71mm Hg with no sub-aortic obstruction.

At 5 months of age a bidirectional superior cavopulmonary shunt was performed. The main pulmonary artery was disconnected from the ventricular mass and the pulmonary valve was oversewn. The child was extubated soon after surgery, but had respiratory distress, requiring reintubation. Auscultation of the chest showed diminished air entry into the left lung, which was attributed the position of the endotracheal tube. Chest x-rays were normal.

Echocardiography showed unobstructed shunt, unobstructed branch pulmonary arteries, good ventricular function and no atrioventricular valve regurgitation. Echo-imaging injection of agitated saline in the right upper limb vein was not suggestive of pulmonary arteriovenous malformations. Ventilator strategy to maintain relative hypercarbia to improve superior venacaval return did not improve saturations. Inhaled nitric oxide also showed no improvement.

Cardiac catheterization showed patent BSCPS and branch pulmonary arteries and no decompressing veins. Femoral arterial saturation was 56%, and the left and right pulmonary artery saturations 37% superior venacaval, right and left pulmonary artery pressures were 17 mm Hg. Mean left and right atrial pressures were 4 mm Hg and the left ventricular end diastolic pressure was 5 mm Hg.

During cardiac catheterization it was observed from chest screening that left lung expansion was poor. The position of the tube was optimized but there was no improvement in the left lung expansion. The endotracheal tube was maneuvered into the left main bronchus and hand ventilation attempted, but it was too difficult to inflate the left lung, and this was clearly observed on screening. This raised a strong possibility of bronchial obstruction. Bronchoscopy was therefore performed which showed extrinsic pulsatile compression of the left main bronchus. CT angiography confirmed impingement of the left main bronchus between pulmonary artery anteriorly and descending aorta posteriorly (Figure 1).

The site and cause of obstruction was clearly defined by CT-based 3D-modelling of the airways and great vessels. The patient was managed conservatively with ventilator support, selective bronchial suctioning and mucolytic installation under bronchoscopic guidance and systemic steroid were given for one week, the child was successfully extubated to nasal CPAP and was subsequent discharged home with oxygen saturation in 80s.

METHOD FOR 3D MODELLING

CT scans were obtained via a Siemens Sensation 64 with a slice thickness of 1.0 mm and a slice increment of 0.8 mm. DICOM were imported into Mimics (Materialise, Leuven, Belgium) for 3D reconstruction of the blood volumes in the single ventricle, aorta and pulmonary artery. The processed files were exported as STL files into 3-matic (Materialise, Leuven, Belgium) to create the various images of interest.

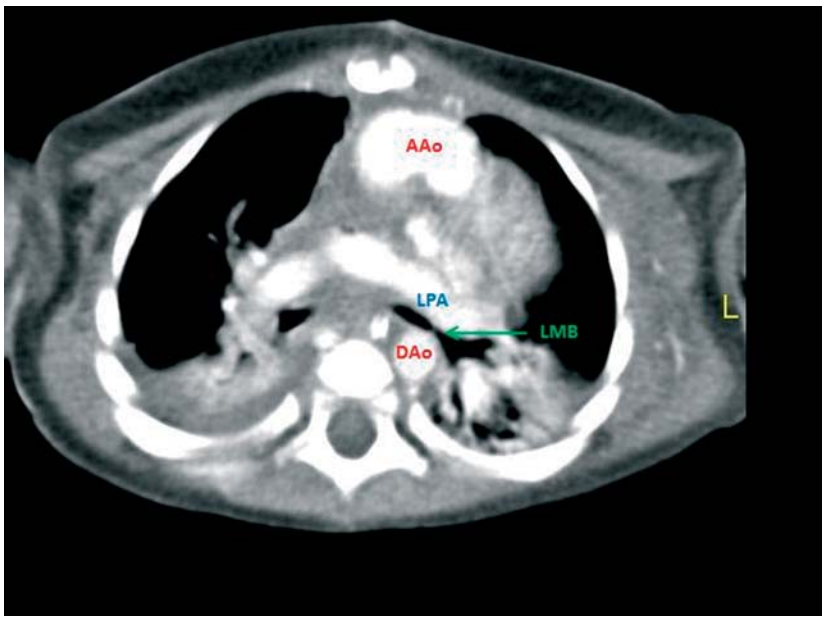

Figure 1. Slice CT scan showing the discrete obstruction in the left main bronchus with the left pulmonary artery directly anterior and the descending aorta directly posterior to the site of obstruction.

DISCUSSION

Causes of desaturation flowing bidirectional superior cavopulmonary shunt include anastamotic obstruction, presence of decompressing vein from the cavopulmonary circuit to the inferior vena cava territory or to the atrium, high pulmonary vascular resistance, ventricular dysfunction, and, in rare cases, acute pulmonary arteriovenous malformations. Bronchial obstruction has been commonly described following aortic arch repair and conduit reconstruction of the pulmonary outflow tract[1,2] but it is uncommon following BSCPS. Compression of the left main bronchus between the left pulmonary artery anteriorly and the descending aorta posteriorly has been described in a 3-month-old child following patch augmentation of aortic arch and closure of VSD.[3] They called it the "pincer effect". In their patient, augmented aortic arch was the main culprit, which was surgically elongated to relieve the obstruction. A 13-year-old patient in their series was the only case of a functionally univentricular heart, who, after an extra cardiac Fontan operation, developed left bronchial compression by pincer effect between the posterior side of the ascending aorta and the anterior side

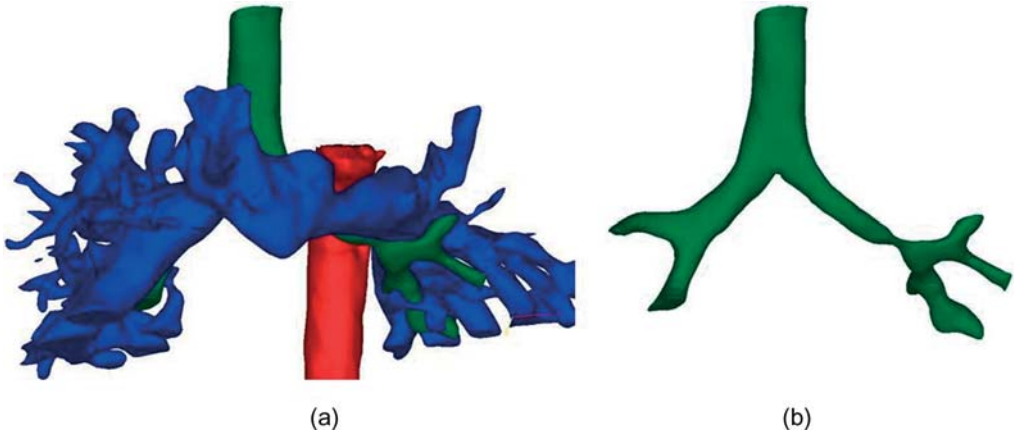

Figure 2. 3D reconstruction of the pulmonary artery, trachea and aorta. (a) The left main bronchus (shown in green) is compressed between the left pulmonary artery (shown in blue) anteriorly, and the descending aorta (shown in red) posteriorly. (b) Reconstruction of the trachea and main bronchi clearly showing the site and extent of the compression.

of the descending aorta. This patient needed aortopexy and placement of a stent in the left main bronchus to relieve the obstructions. We believe that in our patient disconnection of the main pulmonary artery from the ventricular mass caused the branch pulmonary arteries to fall back into the posterior mediastinum where the left pulmonary artery caught the left main bronchus between itself and the posteriorly placed descending aorta (Figure 2). In combination with inflammatory edema that follows surgery, the left bronchus was trapped between two big vessels was obstructed. With time, conservative management, and steroids, the edema subsided and the compression on the bronchus was relieved.

Bronchoscopy is helpful in visualizing luminal obstruction to the left main bronchus. 3D reconstruction based on medical imaging is an effective method of defining the cause of respiratory obstruction. Computed tomography scanning is particularly useful for demonstration changes in airway caliber, in addition to the location, degree and extent of the airway narrowing.[4] CT angiography was successful in confirming the mechanism of airway obstruction and planning therapeutic intervention in 17 patients who developed airway obstruction following operations that involved reconstruction of the aortic arch or the right ventricular outflow tract.[3]

CONCLUSION

Transient left bronchial obstruction following a BSCPS is described as a cause of desaturation. Conservative management was successful, leading to full recovery. The use of 3D modelling described here represents a major refinement for accurately determining the site and cause of the obstruction, and can be repeated using MRI if necessary to determine the response to therapy.

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