

Crossing a Turbulent Bridge: A Rare Case of a Bridging Bronchus

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ABSTRACT

The bridging bronchus is a rare congenital bronchial anomaly in which an aberrant right bronchus arises from the left main bronchus^[2], crosses the mediastinum and continues its trajectory towards the right lung. This condition often co-exists with other congenital anomalies, namely congenital cardiac malformations and vascular malformations. Patients with bridging bronchus may experience wheezing, stridor and respiratory distress attributed to the presence of tracheal or bronchial stenosis^[1,2]. Owing to its infrequent occurrence, there is currently no standard resection and reconstruction technique for stenosis involving a bridging bronchus.

Keywords: Bridging bronchus, congenital anomaly

INTRODUCTION

An Anomalous bronchus from the left main bronchus to the right lung is a form of tracheobronchial arborization known as a bridging bronchus^[1]. This condition is associated with other congenital anomalies such as a right upper lobe bronchus, congenital cardiac malformations and vascular malformations^[1,2]. The diagnosis is suspected in the neonates present with respiratory distress, wheeze, chronic coughing or stridor. We report an unusual case of a newborn with stridor at birth who was diagnosed to have a bridging bronchus.

CASE REPORT

A 35-year-old gravid lady of Chinese ethnicity with an unremarkable antenatal history, delivered a full-term baby girl with a birth weight of 3.2kg via spontaneous vaginal delivery at a private center. Upon delivery, although the APGAR score at 1 min was 9, the newborn had

persistent biphasic stridor with subcostal recessions with an inability to adequately maintain saturation and was therefore placed on supplemental oxygenation and transferred to a tertiary center for further management. At 6 hours of life, with increasing oxygen requirements and worsening subcostal recessions, the newborn baby intubated with an endotracheal tube (ET) size 4mm ID. A flexible bronchoscopy performed via the endotracheal tube revealed complete tracheal rings. With the airway secured, a contrasted computer tomography of the thorax was performed which reported a right bridging bronchus arising from the left main bronchus with an associated proximal stenotic segment and an aberrant left pulmonary artery (Figure 1-3). Following a multidisciplinary discussion involving the respiratory, cardiothoracic, ENT, anaesthetic, paediatric surgery, and intensive care units, the patient was transferred to a tertiary teaching faculty with

the appropriate supporting expertise and facilities for further management. Following a joint decision involving the various specialities and obtaining the appropriate informed high-risk consent from her parents, the patient underwent a rigid bronchoscopy, and repair of the left pulmonary artery which was approached through a median sternotomy with concurrent cardiopulmonary bypass so as to ensure intraoperative oxygenation. A staged slide tracheobronchoplasty was then undertaken via a lateral thoracotomy incision to address the bridging bronchus. Despite the best attempts, the patient succumbed to multiorgan failure during the early postoperative period.

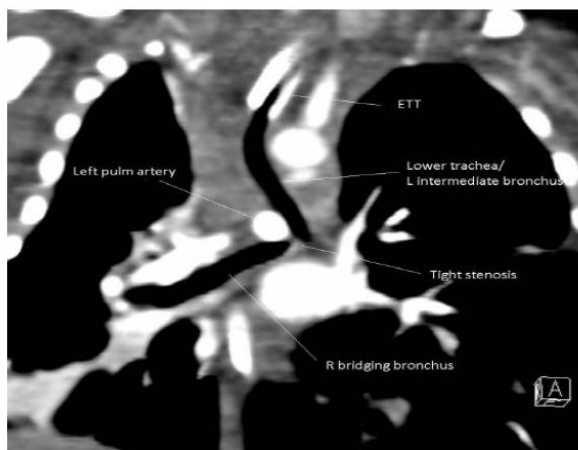


Figure 1: contrast enhanced Computer tomography of the thorax, coronal view showing a right bridging bronchus arising from the left main bronchus with a proximal stenotic segment



Figure 2: 3-dimensional reconstruction of CT Thorax demonstrating the bridging bronchus.

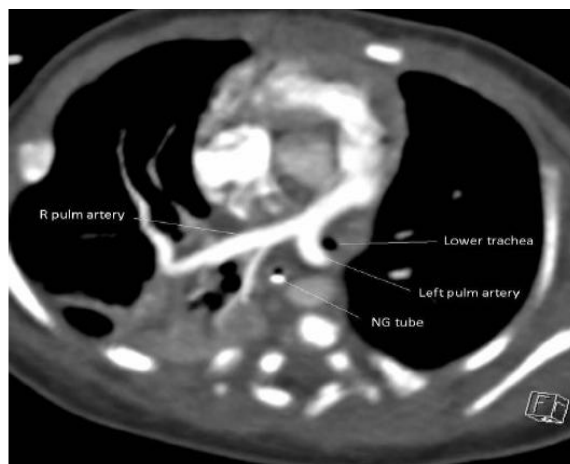


Figure 3: Axial view CT Thorax demonstrating the left pulmonary artery sling with airway narrowing.

DISCUSSION

Anomalies of tracheobronchial arborization have been reported in literature to occur in 0.1-1.9% of children [1]. A variant of these malformations is a Bridging Bronchus. The bridging bronchus is an extremely rare bronchial malformation that was first reported by Gonzalez-Crussi et al [2] in 1976 who coined the term. These account for only 4.7% of patients with congenital tracheal stenosis [3]. It is often associated with other congenital anomalies, the most common being a left pulmonary artery sling. Other reported associations include VACTERL syndrome (Vertebral anomalies, Anal atresia, Cardiac anomalies, Tracho-Esophageal-fistula, renal anomalies, limb anomalies [4]. The bridging bronchus is often but not exclusively discovered in newborns and infants. They may be present with less distressing symptoms late childhood. Stridor, wheezing and respiratory distress attributed to stenotic segments found in the trachea and/or left main bronchus are often the presenting symptoms that may mimic other conditions related to the lower airways. Pathologically, these stenoses occur as a consequence to the presence of circumferentially complete, lumen-narrowing cartilage rings found in the posterior wall of the narrowed airways and/or the absence of the pars membranacea [5]. This results in a partial to near complete airway obstruction. Literature reports mortality rates amongst patients with

congenital trachea-bronchial airway stenosis to be as high as 50% which is proportionate to the extent and site of narrowing observed^[6]. Challenges in diagnosis present itself whether patients present with stridor and respiratory distress or with less distressing symptoms. This is because diagnosis often requires extensive endoscopic examinations complemented with radiological evidence of the condition. Therapeutic challenges compound the clinical problem owing to its infrequent occurrence, and the fact that currently no standard resection and reconstruction technique for stenosis involving a bridging bronchus exists. Previous authors have addressed short and long segment stenotic segments differently. They propose an end to end anastomosis for a short segment bridging bronchus, and a slide tracheoplasty using the bridging segment for long segment stenosis as used in this case^[7,8]. Often associated anomalies involving the heart and vasculature need also to be addressed surgically therefore posing yet another challenge. Surgical planning is usually complex in these cases and must therefore involve a multidisciplinary team approach. Even with the best intentions, facilities and surgical skills, mortality rates remain high in these cases.

CONCLUSION

Bridging bronchus is rare, potentially life-threatening congenital anomaly that is well documented. The diagnosis often requires high clinical suspicion and is usually made after comprehensive endoscopic examinations complemented by appropriate radiological investigations. These complex conditions require clinical vigilance and are best to be addressed in a multidisciplinary fashion as each case requires tailored surgical

management as no one approach is fit for all cases.

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