Temporary bronchial stenting for airway compression in the interstage palliation of functional single ventricle

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ABSTRACT

The Norwood procedure is the first of three palliative surgical procedures offered for hypoplastic left heart syndrome (HLHS). Due to the small size of the thorax and proximity of airway and vascular structures, compression of the airway is possible following the Norwood procedure. We describe the management of an infant with HLHS following Stage I surgical palliation who developed refractory respiratory failure secondary to severe left bronchial compression.

Keywords: Bronchial stent, Glenn procedure, hypoplastic left heart

INTRODUCTION

The Norwood procedure is the first of three palliative surgical procedures offered for hypoplastic left heart syndrome (HLHS). Despite advancements in surgical technique and intensive care unit management, it has the highest postoperative morbidity and mortality of all three palliative procedures. Due to the small size of the thorax and proximity of airway and vascular structures, compression of the airway is possible following the Norwood procedure.[1-3] Potential approaches to alleviate airway compression include anterior sternal aortopexy, bronchopexy, external airway splinting, and, rarely, internal airway stenting.[4-6] We describe the management of an infant with HLHS following Stage I surgical palliation who developed refractory respiratory failure secondary to severe left bronchial compression.

CASE REPORT

A male infant with prenatal diagnosis of HLHS, with near-mitral atresia, aortic atresia, small aortic to left ventricular tunnel, and concern for restrictive atrial septum, was born at 35 weeks' gestation after premature onset of labor and subsequent delivery at an outside facility. Following transfer, he was urgently taken to the cardiac catheterization laboratory for atrial stent placement, which was displaced and required emergent surgical retrieval via median sternotomy. This was performed via bicaval inflow occlusion and was combined with a complete atrial septectomy. He was allowed to recover for a few days due to associated acute kidney and lung injury. On day 4 of birth, he underwent modified Norwood/Sano procedure with a valved Sano conduit using an 8-mm aortic homograft valve and 6-mm Gore-Tex graft.

His postoperative Stage I course was complicated by persistent acute kidney injury requiring short-term peritoneal dialysis, as well as respiratory insufficiency requiring reintubation on three separate occasions. Following a dynamic chest computed tomography (CT) scan demonstrating external vascular compression of the left mainstem bronchus between the left atrium and

Access this article online

Quick Response Code:

Website: www.annalspc.com

DOI: 10.4103/apc.APC_94_18

How to cite this article: Barnes JH, Boesch RP, Balakrishnan K, Said SM, Van Dorn CS. Temporary bronchial stenting for airway compression in the interstage palliation of functional single ventricle. Ann Pediatr Card 2019;12:308-11.

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descending aorta, he underwent posterior descending aortopexy via left lateral thoracotomy at 2 months of age [Figure 1]. Postoperatively, he failed extubation a fourth time and ultimately underwent tracheostomy 1 week later. Despite tracheostomy and positive-pressure ventilation, he continued to have frequent episodes of severe desaturations with left lung collapse seen by chest X-ray [Figure 2]. Serial bronchoscopies continued to demonstrate ongoing severe dynamic left bronchial collapse and compression with intermittent mucous plugging despite aggressive pulmonary toilet and mean airway pressures as high as 25 mmHg. Due to inability to wean from high positive airway pressure support, concerns were raised regarding his ability to proceed to Stage II surgical palliation, which relies on lower mean airway pressures to maintain adequate Glenn flow. After multidisciplinary discussion involving pediatric otolaryngology, pulmonology, cardiology, cardiothoracic surgery, and critical care, he underwent endoluminal left mainstem bronchial stent placement initially with a 4 mm/5 mm × 18 mm Dumon stent [Figure 3]. Chest CT 1-week poststent placement showed improved left bronchial patency as well as left lung aeration and he tolerated significant weaning of his respiratory support to mean airway pressures of 14–16 mmHg. However, due to subsequent spontaneous displacement of the 4 mm/5 mm Dumon stent, a 5 mm/6 mm Dumon was attempted resulting in perforation of the inferior aspect of the left main bronchus just distal to the carina. This was managed with a 6 mm × 22 mm iCAST covered stent [Figure 4] which was later downsized to 5 mm × 22 mm iCAST stent with complete resolution of the bronchial defect. Before undergoing bidirectional Glenn palliation 8 weeks later, the 5 × 22 mm iCAST covered stent was removed by grasping the stent and removing, which was easily performed as the stent covering prevents mucosal ingrowth. This resulted in complete resolution of his bronchial disruption and significant stabilization of his dynamic left mainstem bronchomalacia as well as improvement in the overall bronchial lumen [Figures 5 and 6]. The patient subsequently underwent off-pump Stage II surgical palliation with a Super-Glenn and maintained his Sano conduit [Figure 7]. He was ultimately discharged home, tolerating trials of CPAP and ventilator weaning.

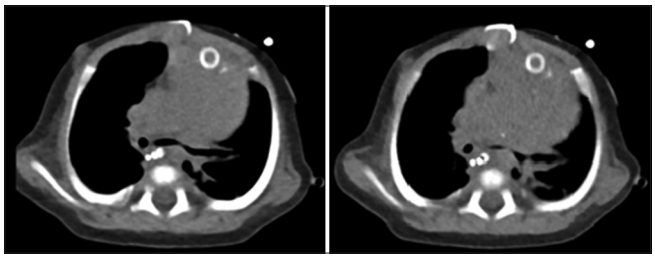

Figure 1: Dynamic computed tomography demonstrating left bronchus collapse upon inspiration (left) and expiration (right) before aortopexy

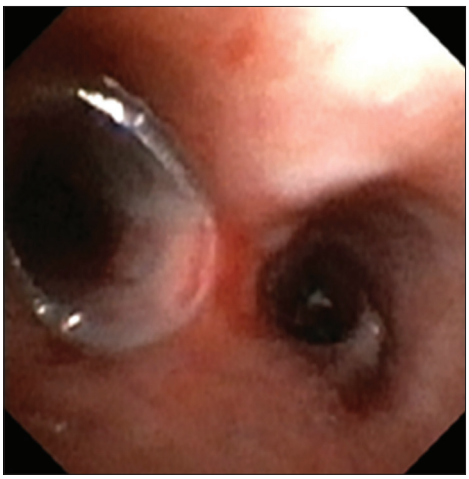

Figure 3: Placement of 4/5, 18 mm Dumon stent in the left main bronchus

DISCUSSION

Significant airway compression is possible following surgical intervention for congenital heart disease, especially for those patients requiring arch reconstruction such as the Norwood procedure.[1] In an attempt to limit compression, reconstruction was performed through adequate mobilization of the proximal descending aorta, complete coarctectomy with the removal of all ductal tissues, and using the interdigitating technique with cutback incision in the posterior proximal descending

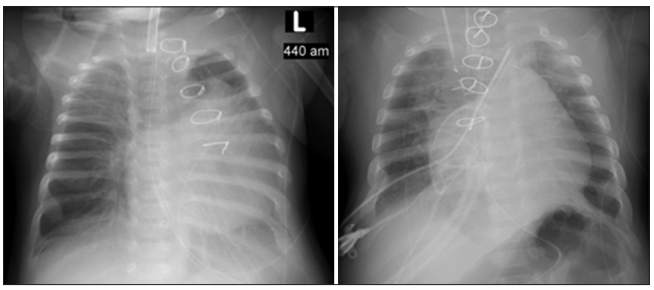

Figure 2: Chest X-ray demonstrating severe left lung collapse before stent placement with consequent resolution following stent placement and Glenn procedure

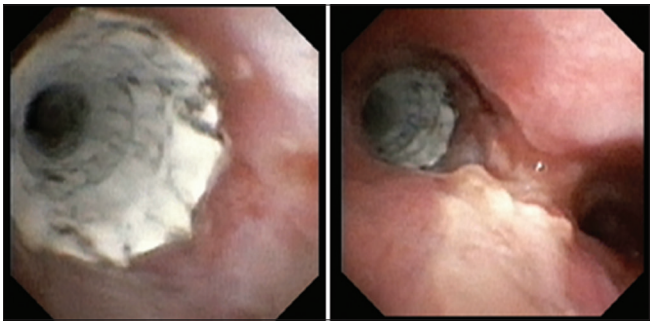

Figure 4: Placement of the 6 mm × 22 mm iCast stent

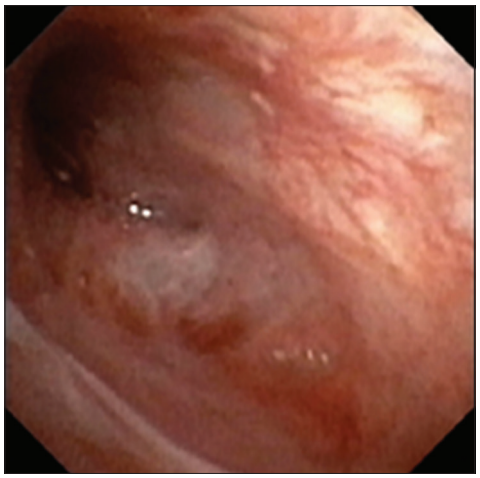

Figure 5: Resolution of bronchial perforation after the removal of 5 × 22 iCast stent

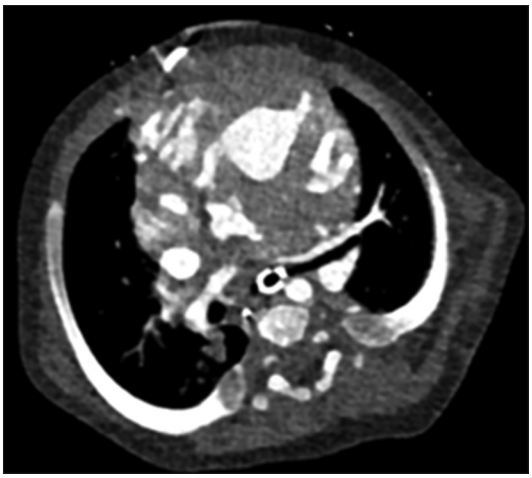

Figure 6: Computed tomography chest taken roughly two weeks after the final stent placement demonstrating continued left bronchus patency

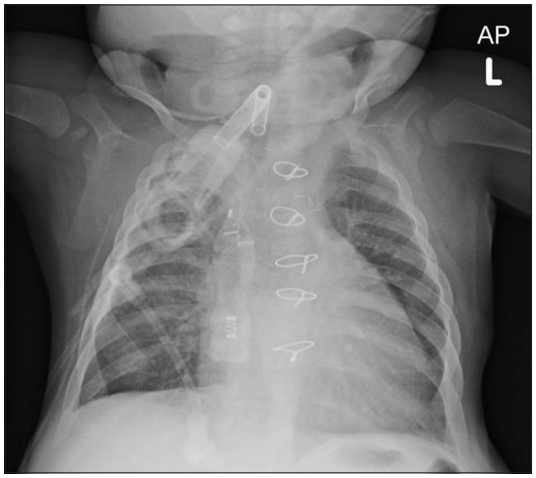

Figure 7: Post operative x-ray following stent removal with resolution of lung collapse

aorta to create a large interdigitating tissue-to-tissue connection between the open distal arch and the proximal descending aorta. Despite this, narrowing resulted. In instances of airway narrowing not due to external structures (e.g., tracheomalacia), anterior aortopexy is accepted as an effective treatment for life-threatening disease.[7] In the setting of severe, localized tracheal narrowing secondary to vascular compression, anterior aortopexy is also a common first treatment approach.[8] However, for isolated left mainstem bronchial narrowing due to more distal vascular compression involving the transverse arch or descending aorta, posterior aortopexy may be indicated.[9] In one series, Arcieri *et al*. described 18 patients undergoing posterior aortopexy; of these, 6 patients required stent placement for residual severe left mainstem bronchomalacia. However, they focused on aortopexy and did not describe the results of stent placement in detail. In our patient, posterior aortopexy was performed for severe, focal, left mainstem bronchial compression without significant improvement in his respiratory symptoms, nor did it allow for weaning of his high ventilator settings. The novel use of a temporary intraluminal bronchial stent to alleviate focal bronchial compression resulted in significant weaning of his ventilator support allowing for successful Stage II Super-Glenn palliation and intact Sano conduit, without complications of bronchial stenosis or erosion. Although the left pulmonary artery was normal in size, Super-Glenn was chosen due to elevated pressures of 30/16 mmHg. If a traditional bidirectional Glenn had been placed, the patient would have experienced bidirectional flow resulting in superior vena cava syndrome, hypoxia, and low cardiac output. To help prevent this, a right-sided bidirectional Glenn was placed to volume off-load his single right ventricle while the Sano conduit was left in place to continue to provide higher arterial blood flow to the higher pressure left pulmonary artery.

This case involved the use of both fixed-diameter (Dumon) and expandable covered (iCast) stents, neither of which has previously been described in this clinical setting. In this instance, however, we experienced two main complications: Dumon stent dislodgment and bronchial perforation. In considering methods to avoid these in the future, stent choice is of utmost importance. Dislodgment was due to the stent being too small, while perforation was due to a stent that was too large. In the future, we would likely measure the airway with a positive pressure breath hold on CT, instead of endoscopic measurement. The iCast stent was particularly beneficial because it offers the possibility of creating a flared end to secure the stent. We would now likely choose a Gore Medical VBX stent which offers increased flexibility to accommodate the curve of the bronchus and still allows creation of a flared end. Finally, an absorbable polydioxanone (PDS) stent would also be a good option, but is not currently available in the United States.

Little published data support endoluminal airway stent placement following unsuccessful posterior aortopexy

for aortic compression causing severe bronchomalacia. In this case, we describe a neonate with HLHS who developed severe bronchomalacia requiring high ventilator settings after undergoing Norwood Stage I palliation. Despite undergoing a posterior aortopexy, he ultimately required temporary endoluminal bronchial stent placement to relieve his persistent bronchial compression. Fortunately, over time, his severe focal left mainstem bronchomalacia improved, allowing for both safe removal of his endoluminal stent and successful Stage II surgical palliation.

CONCLUSION

Temporary endoluminal bronchial stent placement is a novel approach used to alleviate focal bronchial compression causing bronchomalacia in an infant with HLHS following Stage I Norwood/Sano surgical palliation procedure. After attempting to alleviate his external compression with a posterior aortopexy, a multidisciplinary approach led to the placement of an intraluminal left main stem bronchus stent that ultimately resulted in relief of his left bronchomalacia. This subsequently allowed for Stage II surgical palliation with a Super-Glenn, weaning of high ventilator support and discharge from the hospital. To our knowledge, this is the first description of such a case as well as the first detailed description of the use of Dumon and covered balloon-expandable wire-mesh stents in the infant bronchial airway.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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