External stenting: A reliable technique to relieve airway obstruction in small children

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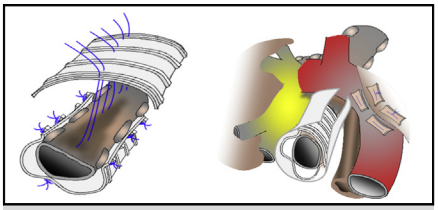
ABSTRACT

Objective: Airway obstruction in children may be caused by conditions such as vascular compression and congenital tracheobronchomalacia. Obstructive pulmonary vascular disease may be a detrimental sequel for patients with congenital heart disease. We evaluate our own original external stenting technique as a treatment option for these patients.

Methods: Ninety-eight patients underwent external stenting (1997-2015). Cardiovascular anomalies were noted in 82 (83.7%). Nine patients had hypoplastic left heart syndrome and 6 had other types of single-ventricular hearts.

Results: The median age at the first operation was 7.2 months (range, 1.0-77.1 months). The mechanisms were tracheobronchomalacia with (n = 46) or without (n = 52) vascular compression. Patients underwent 127 external stentings for 139 obstruction sites (62 trachea, 55 left bronchus, and 22 right bronchus). The stent sizes varied from 12 to 16 mm. There were 14 (8 in the hospital and 6 after discharge) mortality cases. Nine required reoperation for restenosis and 3 required stent removal for infection. The actuarial freedom from mortality and any kind of reoperation was 74.7% ± 4.6% after 2.8 years. The negative pressure threshold to induce airway collapse for congenital malacia (n = 58) improved from −15.9 to −116.0 cmH_2O. A follow-up computed tomography scan (>2.0 years interval from the operation; n = 23) showed the mean diameter of the stented segment at 88.5% ± 13.7% (bronchus) and 94.5% ± 8.2% (trachea) of the reference.

Conclusions: External stenting is a reliable method to relieve airway compression for small children, allowing an age-proportional growth of the airway. (J Thorac Cardiovasc Surg 2017;■:1-11)



The scheme of the external stent technique using a ringed polytetrafluoroethylene graft.

Central Message

External stenting, suspending the airway to the surrounding rigid prosthesis, is a reliable method to relieve airway compression for small children.

Perspective

External stenting, suspending the airway to 2 separate pieces of rigid prosthesis, is a reliable method to relieve airway obstruction in small children. This is equally effective for tracheobronchomalacia with or without vascular compression, and may allow age-proportional growth of the airway. This technique may provide an alternative or adjunct to established practices such as aortopexy.

CONG

Severe airway obstruction in children may result from a congenital cartilage ring, intrinsic tracheobronchomalacia (TBM) or vascular compression syndrome.[1-3] Many of these patients, especially with vascular compression, have coexisting congenital heart diseases. Even if the patient does not exhibit a life-threatening hypoxic spell, secondary pneumonia and atelectasis may lead to obstructive pulmonary vascular disease.[4] Therefore, for the maintenance of cardiopulmonary function, the obstruction should be relieved at the earliest age possible.[5]

An aortopexy offers a reasonable surgical option for TBM and vascular compression, but may not always assure airway expansion. An external stenting (ES)—suspending the airway to the externally placed rigid prosthesis—may be an alternative option,[6] but this prosthesis could interfere with the airway growth in small children. We herein evaluated the outcomes of our own original ES technique, using 2 separate pieces of ringed polytetrafluoroethylene (PTFE) prosthesis with expectation of age-proportional growth of the airway.

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Abbreviations and Acronyms

CT = computed tomography
ES = external stenting
LB = left bronchus
PFTE = polytetrafluoroethylene
RB = right bronchus
T = trachea
TBM = tracheobronchomalacia

PATIENTS AND METHODS

From April 1997 to July 2015, 98 patients underwent a total of 127 ES procedures. There were 43 female and 55 male patients. This was a 2-center study composed of 47 patients operated at Matsuda Municipal Hospital by the second author (Y.N.) and the remaining 51 patients at Sakakibara Heart Institute by the first author. The bronchoscopic evaluation was performed by the third author (H.H.) for all 98 patients. The mechanisms of airway obstruction were congenital TBM with (n = 46) or without vascular compression (n = 52). The operations were indicated due to severe hypoxic (dying) spells (n = 36), failure to wean from the ventilator (n = 34), respiratory distress requiring continuous or bilevel positive pressure breathing (n = 18) and recurrent respiratory infections (n = 10). The hypoxic spells were typically associated with tracheomalacia. Before undergoing operation, the details of the obstruction were assessed using a video-assisted flexible bronchoscopy and computed tomography (CT) scan. The median birth weight was 2.7 kg (range, 0.7-4.4 kg). Cardiovascular anomalies were noted in 82 patients (83.7%) (Table 1). Surgical corrections for these abnormalities were attempted for all patients. A total of 103 ES procedures were performed for these 82 patients. ES procedures were performed before (n = 39), simultaneously with (n = 25), or after (n = 39) the final repair. Patients with arch obstruction and 2 ventricles underwent direct anastomosis of the arch. Patients with hypoplastic left heart syndrome underwent the Norwood operation except for 1, who died after bilateral pulmonary arterial banding. Operations performed on these patients are listed in Table 2. There were 39 patients with chromosomal abnormality, including Down syndrome (n = 17), 22q11 deletion (n = 5), asplenia (n = 2), polysplenia (n = 2), Jacobsen syndrome (n = 2), Smith-Lemli-Opitz syndrome (n = 1), Smith-Magenis syndrome (n = 1), trisomy 8 (n = 1), Turner syndrome (n = 1), and Weaver syndrome (n = 1) with 6 others having unspecified multiple organ malformations. Eight patients had undergone an unsuccessful aortopexy in the referring hospital. None of the remaining patients had undergone operations related to the airway.

TABLE 1. Cardiovascular anomalies

Cardiovascular anomaly	n
Ventricular septal defect	22
Hypoplastic left heart syndrome or variants	9
Arch obstruction with a heart with two ventricles	9
Atrial septal defect with or without anomalous pulmonary venous connection	6
Functionally single ventricle other than hypoplastic left heart syndrome	6
Complete atrioventricular septal defect	5
Double aortic arch	4
Transposition of the great arteries	4
Absent pulmonary valve syndrome	4
Tetralogy of Fallot or double outlet right ventricle	3
Patent ductus arteriosus	2
Right aortic arch with left subclavian artery and ligamentum	2
Pulmonary artery sling	1
Others	5
Total	82

TABLE 2. Operations performed for cardiovascular anomalies and airway obstructions

Operation	n
Pivotal operation performed prior to the first external stenting	n = 46
Intracardiac biventricular repair	15
Norwood operation	8
Aortic arch repair with intracardiac biventricular repair	8
Pulmonary arterial banding	7
Systemic-to-pulmonary arterial shunt	3
Glenn operation	2
Bilateral pulmonary arterial banding	1
Patent ductus arteriosus ligation	1
Vascular ring repair	1
Operation performed simultaneously with the first external stenting	n = 24
Intra-cardiac biventricular repair	15
Vascular ring repair	5
Aberrant innominate artery repair	1
Patent ductus arteriosus ligation	1
Systemic-to-pulmonary arterial shunt	1
Aortic arch repair with intracardiac biventricular repair.	1
Operation performed after the first external stenting	n = 3
Second-time external stenting (planned)	18
Intracardiac biventricular repair	9
Glenn operation	1
Fontan operation	1
Pulmonary arterial banding	1
Second operation after the first stenting	n = 6
Intracardiac biventricular repair	3
Third-time external stenting (planned)	2
Second-time external stenting (planned) with intracardiac biventricular repair	1

Negative pressure testing—to measure the threshold at which airway collapse is induced—was performed before and after 58 ES procedures for TBM. This testing was performed under general anesthesia. The airway was observed using a bronchoscope, which was connected by a rubber tube to a vacuum system and a watertight *U* tube. The water level of this tube was considered to reflect the amount of negative pressure imposed on the airway. A follow-up CT scan was performed in 23 patients (10 trachea [T] and 13 right bronchus [RB] or left bronchus [LB]; maximum age = 8.7 years) and the mean diameter of the stented area on the coronal and saggital planes were obtained. These were all patients who visited our outpatient clinic from March to July 2014 for follow-up. The inclusion criterion was a minimum of 2 years interval from the ES. Hence, there were no bias (ie, presence of a respiratory symptom) in selecting these patients. As a reference for T (mean age, 6.5 ± 1.9 years), a CT scan was performed for 11 age-matched volunteers (6.2 ± 0.2 years; $P = .26$). For the RB and LB, the maximal diameter of the main bronchus for the same patient was used.

The end point of the study included mortality, reoperation, and removal of the mechanical ventilator or tracheostomy tube. The last date of data