Objective: To determine the utility of performing balloon dilation laryngoplasty of subglottic stenosis (SGS) in children with underlying congenital heart disease (CHD).

Design: Retrospective study.

Setting: Tertiary care academic health center.

Patients: Children with an underlying diagnosis of CHD who subsequently underwent balloon dilation laryngoplasty for SGS from January 1, 2006, through December 31, 2011.

Main Outcome Measures: Clinical improvement and avoidance of tracheotomy.

Results: We identified 16 children who had a diagnosis of CHD and underwent direct laryngoscopy and bronchoscopy. Five patients (3 girls and 2 boys) underwent a total of 11 balloon dilations for SGS. Their ages at initial dilation ranged from 1 to 4 months. All 5 patients had grade III SGS. Only 1 patient required a salvage tracheotomy for a thick glottic web and associated SGS after her first balloon dilation failed to improve airway patency. The remaining 4 patients have had long-term success in avoiding tracheotomy with symptomatic improvement.

Conclusions: Balloon dilation represents a valuable treatment option in patients with CHD and SGS in whom a tracheostomy should be avoided.

for treating SGS, allowing patients to avoid open airway surgery and its potentially increased risk for sternal wound infection or mediastinitis. We sought to review our initial experiences with BDL in patients with SGS and CHD, in anticipation of avoiding tracheotomies in these patients in the future.

**METHODS**

We obtained approval from our institutional review board to perform a retrospective review of patients with SGS and CHD. Patients were initially identified by a review of intradepartmental records. Patients considered candidates for inclusion then underwent more extensive review of their electronic medical records. Those patients 18 years or younger who had diagnoses of SGS and some type of CHD and who had undergone endoscopic BDL during the study period (January 1, 2006, through December 31, 2011) were included for further review. We reviewed their records in detail, and data collection was performed to capture other pertinent data. General demographic information, CHD diagnoses, airway diagnoses (including SGS Myer-Cotton grade), the date and type of cardiac surgery, the date and outcome of BDL, and the length of otolaryngology follow-up were recorded.

**SURGICAL TECHNIQUE**

Patients underwent induction of general anesthesia via mask ventilation in conjunction with pediatric anesthesiologists. Intravenous access was obtained, then direct laryngoscopy was performed using an age- and size-appropriate laryngoscope. Topical lidocaine hydrochloride was applied to the bilateral true vocal folds at a concentration of less than 3 mg/kg to decrease intraoperative coughing and laryngospasm.

After several minutes of repeated oxygenation via mask ventilation, direct laryngoscopy was again performed. Endoscopic examination of the airway was achieved with the use of rod telescopes (Hopkins; Karl Storz). Once the SGS was visualized, the laryngoscope was placed into operative suspension to allow bimanual instrumentation of the airway (Figure 1). Balloon dilation catheters from several different manufacturers have been used at our institution, with the most common being an Admiral XTreme balloon catheter (Invatec Innovative Technologies AG) (Figure 2). While in operative suspension, the distal tip of the balloon catheter was guided into a position traversing the stenotic region of the subglottis using telescopic visual assistance. While the SGS was under direct visualization, an experienced surgical assistant (most often an otolaryngology resident physician) performed balloon inflation to a maximum pressure of approximately 4 atm using a manufacturer-approved insufflation device (Encore 26 inflation device; Boston Scientific). Balloon inflation was performed and maintained until the patient demonstrated oxygen desaturation levels of less than 90%, at which time the balloon was deflated and the patient was intubated with a smaller-than-predicted endotracheal tube. In many cases, this sequence was repeated 2 or 3 times while the patient was under the same anesthesia (Figure 3 and Figure 4). Postoperative management included aggressive medical management of gastroesophageal reflux disease. Postoperative surveillance was performed via bedside transnasal fiberoptic nasopharyngoscopy, and repeated direct laryngoscopy and rigid bronchoscopy were performed for any deterioration of respiratory status or for findings on fiberoptic endoscopy that suggested recurrent stenosis of the subglottic airway.

**STATISTICAL ANALYSIS**

Data were collected and stored in a commercially available spreadsheet program (Excel; Microsoft Corp) on a password-encrypted computer. Basic descriptive statistics were obtained.
RESULTS

We initially identified 16 patients with CHD who had undergone direct laryngoscopy and bronchoscopy. Further review of their medical records identified 5 patients (3 girls and 2 boys) who underwent a total of 11 BDL procedures for SGS. Their mean age at the time of the initial dilation was 2 (range, 1-4) months. Initial SGS severity was assigned Myer-Cotton grade III in all 5 patients (3 girls and 2 boys) who underwent a total of 11 BDL procedures for SGS. Their mean age at the time of the initial dilation was 2 (range, 1-4) months. Initial SGS severity was assigned Myer-Cotton grade III in all 5 patients. Associated CHD diagnoses are included in the Table.

Patient 3 required a salvage tracheotomy for a thick congenital glottic web with a significant SGS component that did not respond to initial dilation attempts. This patient also had DiGeorge syndrome. Her glottic web and associated SGS were discovered incidentally during intubation for her cardiac procedure. Therefore, the risk of immediately performing a tracheotomy or laryngotracheal reconstruction (LTR) at the time of initial diagnosis was deemed unacceptable by the cardiothoracic surgery service. Every attempt was made to improve the airway using BDL until her sternotomy had healed and her risk of mediastinitis was judged to be sufficiently low. She subsequently underwent an LTR with anterior and posterior costal cartilage grafts at the age of 19 months. She underwent a total of 5 BDL procedures and eventually developed recurrent stenosis, skewing our analysis of the overall number of procedures. Although her airway patency is improved, it is not adequate enough to allow decannulation, and she is currently awaiting revision LTR.

The remaining 4 patients underwent a total of 6 procedures, and none required tracheotomy. One patient underwent a single-stage LTR with an anterior thyroid alar cartilage graft at the age of 3 months, with the hope of improving his airway patency in anticipation of eventual surgical repair of his ventricular septal defect. A tracheotomy was never required, and BDL successfully provided controlled postoperative dilation of his subglottis, as described by Bent et al. The remaining 3 patients required only 1 BDL procedure each and have remained symptom free, with follow-up times with the Pediatric Otolaryngology service ranging from 2 to 3 months. All patients have continued to be observed by the Pediatric Cardiology clinic at our institution to the present and have not required additional consultation with the Pediatric Otolaryngology service.

COMMENT

Subglottic stenosis is estimated to have an incidence of only 0.24% in the neonatal population. Although this number may seem low, the condition is common enough that pediatric otolaryngologists must be familiar with its definitive management. Children with underlying CHD are hypothesized to be at an even higher risk for SGS for a number of different reasons. Small series of children with CHD have estimated the coexisting incidence of SGS in this patient population to range from 1.08% to 2.3%, with 1 group estimating slightly higher incidences in children younger than 1 year.

Several reasons for the increased risk for SGS development in patients with CHD have been reported. In utero, the embryologic development of the respiratory and cardiac systems is temporally and spatially related, thus resulting in a higher incidence of coexisting anatomic abnormalities if normal development is perturbed in any way. The associated medical complexities in these patients can in many cases require periods of endotracheal intubation, a well-described risk factor for the development of acquired SGS. Because of the need for higher airway pressures after cardiac surgery, larger and/or cuffed endotracheal tubes are sometimes required. Finally, poor tissue oxygenation and hypoperfusion likely affect laryngeal mucosal wound healing, with a correlate drawn to gastrointestinal tract hypoperfusion after cardiopulmonary bypass.

Treatment options for SGS include open and endoscopic approaches, with the traditional management paradigm favoring open approaches in more severe cases. Tracheotomy has been recognized for a long time as a criterion standard for managing the airway obstruction caused by SGS, although it does not treat the underlying cause. However, a tracheotomy in any patient—and certainly in a small child—carries a high risk for morbidity in addition to the risk for mortality. Cardiothoracic surgeons traditionally avoid the early placement of tracheotomies in their patients who have undergone recent median sternotomy because of a higher risk for sternal wound infection and mediastinitis.

Balloon dilation laryngoplasty appears to offer a less invasive option to treat severe SGS definitively in children and thereby avoid complications associated with open surgical procedures such as tracheotomy-associated sternal wound infections. Laryngeal dilation with fixed dilators or bougies was performed for many years before falling out of favor owing to an increased risk of causing more tissue damage and for lack of success in treating the underlying stenosis. With the advent of balloon dilation catheters used in interventional cardiology and radiology, the technique has been revived.

Figure 4. Photograph of the subglottis in patient 2 immediately after balloon dilation.
The first reports of balloon dilation in the pediatric airway were published in 1991 and included several reports of subglottic dilation. Durden and Sobol published their experience of using BDL in 10 patients with SGS and demonstrated a 70% success rate in adequately treating the underlying SGS so that no further procedures were required. The authors advocated the use of BDL as a viable treatment option in evolving SGS. Over time, more clinicians have adopted the technique. The largest series to date involved 52 balloon dilation procedures in 44 children, with an overall success rate of 71%. The authors further classified their success rates in primary and secondary procedures (ie, after a prior LTR) but did not find a statistically significant difference in success rates between the groups. Bent et al also described their use of balloon dilation as an adjunct procedure for recurrent stenosis after LTR in 10 patients.

Although our series is small, we believe that it constitutes the first description of BDL use in the specific patient population with comitant CHD and SGS. Others have identified this population as being at increased risk for SGS compared with the general pediatric patient population. In addition, although the existing literature contains some contradictory findings, enough data suggest concern for an increased risk for sternal wound infection in the presence of an indwelling tracheotomy tube. Thus, we believe that BDL provides an efficacious and safe alternative for treating SGS in children with underlying CHD, in whom pediatric cardiothoracic surgeons want to avoid a tracheotomy or any other type of open airway procedure. Some of these patients may also be too sick to undergo definitive airway reconstruction, such as LTR, immediately. Balloon dilation laryngoplasty may be used in primary cases as the first choice of airway reconstruction, such as LTR, immediately. Balloon dilation laryngoplasty may be used in primary cases as the first choice of airway reconstruction, such as LTR, immediately.

Submitted for Publication: June 26, 2012; final revision received August 14, 2012; accepted September 7, 2012.

Correspondence: William O. Collins, MD, Department of Otolaryngology, University of Florida College of Medicine, PO Box 100264, Gainesville, FL 32610 (William .collins@ent.ufl.edu).

Author Contributions: Drs Collins, Kalantar, and Silva had full access to all the data in the study and take responsibility for the integrity of the data and the accuracy of the data analysis. Study concept and design: Collins and Kalantar. Acquisition of data: Kalantar and Rohrs. Analysis and interpretation of data: Kalantar and Silva. Drafting of the manuscript: Collins. Critical revision of the manuscript for important intellectual content: Kalantar, Rohrs, and Silva. Statistical analysis: Collins. Administrative, technical, and material support: Rohrs.

Conflict of Interest Disclosures: None reported.

Previous Presentation: This study was presented as a poster at the Spring Meeting of the American Society of Pediatric Otolaryngology; April 28, 2012; San Diego, California.

References

4. Zestos MM, Hoppen CN, Bellenky WM, Viruppanavar V, Stricker LJ. Subglottic who do not have CHD, thus possibly allowing a future comparison of success rates in CHD and non-CHD patient populations. As with any retrospective studies, some limitations are inherent. The only data available for review are contained within the medical record. An unknown population of patients in whom SGS developed may exist but, because clinical symptoms did not develop, the need for bronchoscopy never materialized. However, we believe that BDL represents a safe and efficacious option when using minimally invasive techniques to treat SGS definitively in children with CHD. Thus, we can avoid open airway procedures and their associated risks.

<table>
<thead>
<tr>
<th>Patient No./Age, mo</th>
<th>Other Diagnosis</th>
<th>Cardiac Diagnosis</th>
<th>SGS Description</th>
<th>No. of BDL Procedures</th>
<th>Adjuvant Therapy</th>
<th>Follow-up Length</th>
</tr>
</thead>
<tbody>
<tr>
<td>1/4</td>
<td>Trisomy 18</td>
<td>Atrioventricular canal defect</td>
<td>Thin circumferential SGS</td>
<td>1</td>
<td>Mitomycin</td>
<td>2 mo with clinic FFL</td>
</tr>
<tr>
<td>2/1</td>
<td>None</td>
<td>Truncus arteriosus</td>
<td>Thin circumferential SGS</td>
<td>1</td>
<td>Mitomycin</td>
<td>23 mo with clinic FFL</td>
</tr>
<tr>
<td>3/1</td>
<td>DiGeorge syndrome</td>
<td>Aortic arch repair</td>
<td>Thick congenital glottic web with subglottic component</td>
<td>5</td>
<td>None</td>
<td>25 mo with rigid bronchoscopy</td>
</tr>
<tr>
<td>4/1</td>
<td>None</td>
<td>Hypoplastic aortic arch</td>
<td>Thin circumferential SGS</td>
<td>1</td>
<td>Topical triamcinolone acetonide (Kenalog)</td>
<td>1 mo with clinic FFL</td>
</tr>
<tr>
<td>5/3</td>
<td>None</td>
<td>Ventricular septal defect</td>
<td>Thick anterior shelf of SGS</td>
<td>3</td>
<td>Mitomycin</td>
<td>21 mo with rigid bronchoscopy</td>
</tr>
</tbody>
</table>

Abbreviations: BDL, balloon dilation laryngoplasty; CHD, congenital heart disease; FFL, flexible fiberoptic laryngoscopy; SGS, subglottic stenosis.

a The SGS Myer-Cotton grade was III in all 5 patients.


