Subglottic Stenosis Complicating Cardiac Surgery in Children*

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Objective: To highlight the incidence of subglottic stenosis (SGS) as a complication of surgery for congenital heart disease and the role of single-stage laryngotracheoplasty in treating this complication.
Design: Retrospective case series.
Setting: University-based referral center specializing in surgery for congenital heart disease and complex airway problem management.
Intervention: Laryngotracheal reconstruction (LTR).
Main outcome measure: Successful airway expansion.
Results: At last follow-up, 87.5% (7 of 8) of patients remain free of obstructive airway symptoms.
Conclusion: SGS can complicate surgery for congenital heart disease in children. Single-stage LTR is an effective treatment modality for this problem.
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C hildren in whom cardiac surgery is performed frequently need mechanical ventilation in the immediate postoperative period. Although this life-supporting modality is critically important in determining the outcome of the procedure, it can lead to debilitating laryngotracheal injury. The most common cause of acquired laryngeal stenosis is endotracheal tube injury, which accounts for approximately 95% of all cases of laryngeal stenosis.[1] Intraluminal tissue edema in the subglottis readily causes ischemia of the mucosa due to compression between the tube and nondistensible cricoid ring. The associated inflammation and infection lead to perichondritis and chondritis, which heal with circumferential fibrotic scarring and the formation of a stricture or complete stenosis.

Koh et al[2] observed laryngeal edema in 19 of 181 (10%) children after cardiac surgery for congenital heart disease and associated it with tracheal intubation. Mucosal hypoperfusion due to perioperative hypotension and the use of extracorporeal circulation, which is known to impair gas exchange, may predispose the larynx to hypoxic endotracheal tube trauma. Although most cases of laryngeal trauma secondary to intubation resolve spontaneously, a few may progress to subglottic stenosis (SGS) which can

aggravate the existing cardiovascular condition or result in serious morbidity even after successful cardiac surgery. We present a series of eight pediatric patients who developed severe SGS following cardiac surgery. To the best of our knowledge, this complication has not been previously reported in the medical literature. The management of these patients and their outcomes are discussed.

PATIENTS AND METHODS

A retrospective study was carried out on all pediatric patients who had undergone surgery for congenital heart disease from January 1994 to December 1995 to identify those who had developed airway complications following the procedure. The patients who were referred to our service for the management of severe SGS from January 1994 to December 1995 were studied in detail. Eight patients were entered into the study (five female and three male patients). Their ages at the first cardiac procedure ranged from 3 weeks to 2 years. Details of the duration of mechanical ventilation, the number of intubations, difficulty in intubation, and postextubation stridor were recorded for each of the eight patients.

SGS was confirmed by direct laryngotracheoscopy in all patients and was graded according to Cotton's classification.[3] Grade 1 included less than 70% obstruction of the lumen. Grade 2 included approximately a 70 to 90% obstruction of the lumen. Grade 3 included obstruction greater than 90%, no matter how narrow, in which there was an identifiable lumen. Grade 4 included a complete obstruction. The management of subglottic stenosis and the complications encountered thereof were noted. The associated airway lesions encountered and the subsequent clinical course of the patients were recorded (Table 1). Laryngotracheal reconstruction (LTR) was planned depending on the patients' cardiopulmonary statuses and on the results of a discussion with the treating cardiovascular surgeon. In this procedure, a preliminary direct laryngotracheoscopy was performed to assess the severity and length of the stenotic segment. The laryngotracheal complex was then exposed via an external incision and opened in the midline from the lower one third of the thyroid cartilage down to include the lower four to five tracheal rings. An autologous rib graft was then shaped to fit into this elliptical defect and sutured in place, thus expanding the airway and closing the tracheotomy. This was followed by extubation in the operating room or a brief period of nasotracheal intubation in the ICU. No other stenting was required. All patients had a direct laryngotracheoscopy in the follow-up period after surgery to confirm airway adequacy.

RESULTS

The results are summarized in Tables 1 and 2. In the 2 years of the study, 300 open cardiac procedures were performed. In this series, the incidence of SGS following open cardiac surgery was 2.3%. The mean duration of mechanical ventilation following cardiac surgery was 7 days (maximum, 17 days; minimum, 3 days). The average number of intubations per patient was 1.5 (maximum, 3; minimum, 1). There was no documentation of any traumatic intubation or accidental extubation in any of these patients. Patchy atelectasis was noted on chest x-ray films of all patients postoperatively. However, there was no record of tracheitis or bacterial pneumonitis. Stridor was documented in three patients after extubation. Two responded satisfactorily to medical management with racemic epinephrine, systemic steroids, and cold humidification. One patient required a tracheotomy for severe respiratory distress secondary to laryngeal edema which was confirmed by direct endoscopy. The absence of SGS prior to cardiac surgery was inferred from anesthesia records which did not mention any difficult intubation or inability to pass a normal-sized tube for the child's age. There was no relationship between the number of cardiac procedures and the severity of subsequent SGS in this study.

Seven out of the 8 (87.5%) patients had tracheotomies prior to LTR due to airway insufficiency or failed extubation. The tracheostomy tubes were in place from 3 months to 4 years (mean, 26 months) prior to LTR. The diagnosis of SGS was confirmed after tracheostomy with direct laryngotracheoscopy in seven patients. Patient 1 was referred to our service from the operating room subsequent to failure of intubation by an anesthesiologist prior to a third cardiac procedure. Direct examination revealed approximately 70% narrowing of the subglottis with a posterior glottic web. After consultation with the cardiovascular service, it was decided to proceed directly with LTR. The other seven patients had at least two direct laryngoscopies and bronchoscopies performed by an attending pediatric otolaryngologist 2 to 6 months apart to assess the severity of stenoses prior to surgery.

At present, 7 out of the 8 (87.5%) patients remain free of obstructive airway symptoms. Following LTR, 5

Table 1—*Airway Lesions and Their Management**

Patient	Airway Lesions	Grade of Stenosis[†]	Management[†]	Age at LTR	Present Airway Status
1	Glottic web, SGS	2	LTR, AG	7 yr	Asymptomatic
2	SGS	2	Tracheotomy, LTR, AG	12 mo	Asymptomatic
3	SGS, tracheomalacia	3	Tracheotomy, LTR, AG	31 mo	Intermittent aspiration
4	SGS, tracheomalacia	3	Tracheotomy, LTR, AG	5 yr	Tracheotomy
5	SGS, tracheomalacia	2	Tracheotomy, LTR, AG	29 mo	Asymptomatic
6	Bilateral vocal cord granulomas, SGS	3	Tracheotomy, LTR, AG	25 mo	Asymptomatic
7	SGS	3	Tracheotomy, LTR, AG	14 mo	Asymptomatic
8	Vocal cord paresis, SGS	3	Tracheotomy, LTR, AG&PG	7 yr	Asymptomatic

*AG=anterior rib graft; PG=posterior rib graft.
[†]Cotton's classification (1984).[3]

Table 2—*Summary of the Patient's Cardiac Profiles**

Patient	Cardiac Defects	No. of Procedures[†]	Age	Present Status
1	Fallot's tetralogy	2	24 mo	Asymptomatic
2	COA, PDA	1	3 wk	Congestive heart failure
3	AVC defect	1	8 mo	Asymptomatic
4	AVC defect, subaortic stenosis	2	6 wk	Residual subaortic stenosis
5	COA, PDA, VSD	2	2 wk	Residual VSD
6	ASD, AS	1	1 mo	Recurrent AS
7	PDA, ASD	1	3 mo	Asymptomatic
8	ASD	1	4 mo	Asymptomatic

*Abbreviations are as follows: AS=aortic stenosis; ASD=atrial septal defect; AVC=atrioventricular canal; COA=coarctation of aorta; PDA=patent ductus arteriosus; VSD=ventricular septal defect.

[†]The number of open cardiac procedures undergone prior to the identification of SGS.

(62.5%) patients were extubated in the operating room. Patient 3 required emergency intubation 2 days later due to collapse of the graft but was successfully extubated after 10 days. Patient 4 failed extubation 3 times. A tracheotomy was performed after endoscopy revealed dynamic collapse of the trachea with poor vocal cord movement. Patient 6 developed pulmonary edema after the procedure and needed mechanical ventilation for 3 days; this was followed by successful extubation. The present airway status of the patients is shown in Table 1. Direct laryngotracheoscopy after surgery showed a satisfactory airway in 7 of the 8 (87.5%) patients.

DISCUSSION

Major advances in cardiovascular surgery over the last 30 years permit the total correction of previously inoperable congenital cardiac defects in children. Surgery is now being performed at progressively younger ages with better outcomes. Recently, concern has been raised over complications such as embolization, hypoxia, cerebral and gastrointestinal hypoperfusion, and biochemical imbalances that occur following major cardiac surgery with cardiopulmonary bypass.[4-6] However, the issue of laryngeal complications arising after cardiac surgery remains inadequately addressed in the medical literature.

Laryngeal edema has been documented in children receiving ventilatory support after cardiac surgery; a higher incidence of edema has been observed in children less than 1 year of age.[2] Tanaka et al[7] noted that the risk of respiratory and other complications increased in pediatric patients less than 1 year of age on whom cardiac surgery was performed if they were intubated for more than 4 days. These studies did not attribute the airway problems encountered solely to endotracheal intubation and mechanical ventilation and suggested that cardiac surgery also may have played a role. In the present series, 7 of the 8 patients were less than 1 year of age at the time of their first cardiac operation. At the Le Bonheur Children's Medical Center, 2,058 children were admitted to the pediatric ICU during the period under study. Almost all children required mechanical ventilation for a period of at least 24 h. The incidence of severe SGS requiring LTR in

the noncardiac surgical group was 0.8% as compared with 2.3% in the studied group. The small number of patients precluded establishing a relationship between the duration of intubation and airway stenosis. However, no relationship was seen between the number of cardiac procedures performed per patient and the severity of SGS.

Ischemic neurologic and gastroenterologic events occurring after surgery for congenital heart disease have been well documented.[5,8] An association was identified between the length of cardiopulmonary bypass and a period of low perioperative perfusion pressure. Additionally, intrapulmonary shunts have been found to be a major contributor to impaired gas exchange before, during, and after cardiac surgery.[9] The laryngeal subglottis, which is the narrowest part of an infant's airway, is lined with respiratory epithelium below which is loose areolar tissue surrounded by a rigid complete cartilaginous cricoid ring. The intraluminal laryngeal mucosa has a poorer vascular supply than that of the gut or nervous system, which is likely to make it more prone to ischemia, edema, and ulceration when compressed by an endotracheal tube. When the source of trauma is ultimately removed from the subglottis, the denuded wall attempts healing by collagen production and contracts circumferentially, producing a severe degree of stenosis.

SGS in infants represents one of the most difficult problems facing the otolaryngologist. It is a more severe form of disease than the congenital variety and almost always requires operative intervention. Holinger et al[10] identified endotracheal intubation as the cause of SGS in 37 of 43 (86%) children studied. Several factors contribute to laryngeal trauma from endotracheal tubes. Foremost among them are traumatic and multiple intubations, inappropriately large tubes, and infection. The contributory role of the duration of intubation remains a matter of controversy.[10]

The surgical treatment for children with SGS has progressed from dilatations and long-term tracheotomies to single-stage LTR. Long-term tracheotomy has a considerable morbidity and a high mortality rate ranging from 1.6 to 24%.[11,12] However, it may be essential for the immediate relief of severe respiratory distress. In the present study, 7 children had a tracheotomy in place for a mean period of 26 months. There were no

tracheotomy-related complications documented. Cotton[13] is of the opinion that in children with SGS, a tracheotomy without insertion of a stent results in a more severe stenosis. Contamination of the injured larynx through the tracheotomy wound prolongs healing and predisposes the patient to more prolific scarring. Surgical reconstruction of the larynx should be considered in children with SGS who require a tracheotomy.[3] The safety and efficacy of single-stage LTR has been well established.[14,15] The procedure can be used to facilitate early decannulation and, in some patients, to avoid tracheotomy altogether as in patient 1. Since most of the cardiac procedures are performed in patients at a very young age, the early decannulation following an LTR permits the return of normal laryngeal function and the development of spoken language. In this series, four patients were successfully extubated in the operating room after the LTR and had no further problems. The longest postoperative intubation was for 10 days to stent a collapsed graft. At the last review, seven of the eight patients had satisfactory airways, and in one of them an additional cardiac operation was performed uneventfully.

Many suggestions have been put forward to minimize the risk of complications secondary to hypoperfusion and impaired gas exchange during cardiac surgery. Mythen and Webb[8] recommend using perioperative plasma volume expanders with colloid to improve circulation and prevent hypoxic events. Early extubation after cardiac surgery is now being advocated because evidence suggests that it does not affect morbidity or mortality.[16,17] The anesthesia technique used and the patient's medical condition are the major factors involved in accomplishing this goal. Widespread acceptance of this policy along with the use of properly sized tubes, good endotracheal tube fixation, and adequate sedation when the patient is intubated will probably reduce the incidence of endotracheal tube trauma. Any child who has a difficult postextubation period following cardiac surgery should be examined with a flexible fiberoptic laryngotracheoscope before he or she leaves the ICU. If laryngeal edema or ulceration has occurred, the cardiac anesthesia team should be alerted before any subsequent procedure. Adequate mucosal perfusion during surgery and early extubation following it may prevent the progression to SGS.

CONCLUSION

SGS is a hitherto unreported complication of cardiac surgery in children. The severity of stenosis does not appear to be related to the number of cardiac procedures. Single-stage LTR is a safe and effective method of treating established stenosis.

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