REPRINTED FROM *ANNALS OF OTOLOGY, RHINOLOGY & LARYNGOLOGY*
December 2001 Volume 110 Number 12

SURGERY FOR PEDIATRIC SUBGLOTTIC STENOSIS: DISEASE-SPECIFIC OUTCOMES

CHRISTOPHER J. HARTNICK, MD
BOSTON, MASSACHUSETTS

BENJAMIN E. J. HARTLEY, MD
LONDON, ENGLAND

PETER D. LACY, MD
CINCINNATI, OHIO

JAMES LIU, MD
CINCINNATI, OHIO

J. PAUL WILLGING, MD
CINCINNATI, OHIO

CHARLES M. MYER III, MD
CINCINNATI, OHIO

ROBIN T. COTTON, MD
CINCINNATI, OHIO

To set the foundation to develop a disease-based, operation-specific model to predict the outcome of pediatric airway reconstruction surgery, we performed a retrospective database review of children operated on at a single, tertiary-care children's hospital. Over the 12-year period 1988 to 2000, a total of 1,296 airway reconstruction procedures were performed. Out of these, charts were identified for 199 children who underwent laryngotracheal reconstruction for a sole diagnosis of subglottic stenosis. Children were excluded from the study if their disorder included supraglottic, glottic, or upper tracheal disease. The main outcome measures were Myer-Cotton grade-specific decannulation and extubation rates, including both operation-specific and overall results. There were 101 children who underwent double-stage laryngotracheal reconstruction. The operation-specific decannulation rates for Myer-Cotton grades 2, 3, and 4 were 85% (18/21), 37% (23/61), and 50% (7/14) (χ^2 analysis, p = .0007). The overall decannulation rates were 95% (20/21), 74% (45/61), and 86% (12/14) (χ^2 analysis, p = .04). There were 98 children who underwent single-stage laryngotracheal reconstruction. The operation-specific extubation rates for Myer-Cotton grades 2, 3, and 4 were 82% (37/45), 79% (34/43), and 67% (2/3) (χ^2 analysis, p = .63). The overall extubation rates were 100% (45/45), 86% (37/43), and 100% (3/3) (χ^2 analysis, p = .03). Logistic regression analysis showed no effect of age (less than or greater than 2 years of age) on operation-specific or overall outcome parameters. We conclude that laryngotracheal reconstruction for pediatric subglottic stenosis remains a challenging set of procedures in which multiple operations may be required to achieve eventual extubation or decannulation. Children with Myer-Cotton grade 3 or 4 disease continue to represent a significant challenge, and refinements of techniques are being examined to address this subset of children. Disease-based, operation-specific outcome statistics are the first step in the development of a meaningful predictive model.

KEY WORDS — laryngotracheal reconstruction, outcome, subglottic stenosis.

INTRODUCTION

Surgical management of pediatric subglottic stenosis has undergone an evolution in the methods of diagnosis, workup, and treatment since Grahne[1] applied the Rethi procedure to children in 1971 and Fearon and Cotton[2] introduced the concept of cartilage augmentation and laryngotracheal reconstruction (LTR) in 1972. Since that point, there have been multiple series reporting surgical outcomes for the various operative procedures. Early work focused on the results of double-stage LTR; among these reports was the landmark work of Cotton et al[3] in which they noted an overall decannulation rate of 92% for 203 children who underwent airway reconstruction. More recent work documented the introduction and success of single-stage LTR[4-6] and of cricotracheal resection.[7,8]

Several different methods have been implemented to report the surgical outcomes for procedures designed to treat subglottic stenosis. Initially, articles reported the severity of stenosis according to the Cotton classification system[9] and reported outcomes on the basis of this scale.[3,10] Since 1994, the most common grading system used to assess subglottic stenosis has been that proposed by Myer et al,[11] and surgical outcomes studies have used this system to describe and define the different surgical populations.[8,12,13]

Surgical outcomes have also been described by reporting results of different surgical procedures without fully defining the disease population in which the surgery was done. The difficulty with interpreting "surgery-based" outcomes analyses such as these lies in the inherent differences between the different disorders. A posterior costal cartilage graft may well have a different outcome when used for isolated posterior glottic stenosis than when used for either posterior subglottic stenosis or for combined glottic and

From the Department of Pediatric Otolaryngology, Children's Hospital Medical Center, Cincinnati, Ohio.
Presented at the meeting of the American Broncho-Esophagological Association, Palm Desert, California, May 14-15, 2001.
CORRESPONDENCE — Christopher J. Hartnick, MD, Dept of Otolaryngology, Massachusetts Eye and Ear Infirmary, 243 Charles St, Boston, MA 02114.

TABLE 1. OVERALL DEMOGRAPHIC DATA

	No. of Children	*Age (y; mean ± SD)*
Total	199	4.45 ± 5.09
Single-stage procedures	101	4.28 ± 4.46
Double-stage procedures	98	4.62 ± 5.67

Data on gender were missing for 4 children. There were 120 male patients and 75 female patients.

subglottic stenosis.

Other aspects of reporting on the outcomes of surgery for pediatric subglottic stenosis have included overall decannulation or extubation rates and the number of surgical procedures required to achieve this final goal.[3,10] While the number of procedures gives extremely important information to supplement the overall success rates for surgery, the reporting of "operation-specific" success rates (the rates at which an open surgical reconstruction is successful at achieving decannulation or extubation without subsequent open surgical repairs) is not currently part of the classic reporting paradigm. Such data would provide invaluable information to families who are contemplating surgery.

In this report we aim to use the most common classification system for pediatric subglottic stenosis[11] and to give disease-specific outcomes on a grade-by-grade basis. This method supports the use of "operation-specific" as well as "overall" outcomes. This report may provide a framework upon which future studies will flesh out more fully the model of "disease-based" analysis. It is clear that the presence or absence of tracheomalacia or gastroesophageal reflux disease may affect the surgical outcome; exactly how these conditions are defined needs clarification, and documentation to this point has been inexact. Future studies using this form of reporting may more fully characterize the disease process and the role of potential cofactors. We do not purport in this article to specifically compare particular surgical procedures (ie, cricotracheal resection versus anterior-posterior costal cartilage LTR). With these limitations in mind, we hope to establish a common means of presenting outcomes for pediatric airway reconstructive surgery by which results from different centers can be compared and analyzed.

MATERIALS AND METHODS

A retrospective database review was performed from a Microsoft Access database containing 1,296 pediatric airway reconstructions performed at the Children's Hospital Medical Center in Cincinnati (CHMCC) between the years 1988 and 2000. A pediatric airway reconstruction was defined as that performed in a child less than 18 years of age. The database included demographic information on each patient, as well as information regarding the type of surgery performed and the follow-up. Information regarding the date of decannulation for some patients was not available, and supplementary information was obtained from the patients' medical records and added to the database.

Patients were identified from this 12-year period who had isolated subglottic stenosis as their sole disorder. Patients with either concomitant supraglottic or glottic disease or with concomitant suprastomal collapse or tracheomalacia were excluded from the study. Patients whose surgical procedure consisted of an anterior cricoid split also were excluded from the study.

For purposes of uniformity, only those patients whose preoperative subglottic stenosis could be graded according to the Myer-Cotton grading system were included in the study; patients for whom this information was not available were excluded.

Patients who had been previously operated on elsewhere than at CHMCC were included in the study, as long as they met the other requirements listed above. Their initial Myer-Cotton classification was defined as that made when they were first seen at CHMCC.

With these inclusion and exclusion criteria, the charts of a total of 199 children were identified for data review. A Microsoft Access database was created to track demographic data, data on the surgical procedures performed, and outcomes data consisting of operation-specific extubation or decannulation rates and overall extubation or decannulation rates. "Operation-specific" decannulation or extubation rates are defined as the rates at which an *open* surgical reconstruction was successful at achieving decannulation or extubation without subsequent *open* surgical repairs. Endoscopic procedures such as dilation or use of the CO_2 laser do *not* affect the operation-specific calculations.

RESULTS

For the 199 children identified, the average age at the time of operation was 4.42 years (SD, 5.09 years; Table 1). This figure includes the age at the time of the initial operation, as well as the age at subsequent operations. There was a male predominance of 1.6 to 1. The overall rate of decannulation or extubation was 84%.

There were a total of 297 procedures performed, for an average of 1.49 procedures per child (Table 2). Each child's airway was assessed before each reconstruction, and the relative grades of stenosis ob-

TABLE 2. OVERALL RESULTS

Number of procedures	297
Mean number of procedures per child	1.49
Overall decannulation or extubation rate	84% (165/199)
Degree of subglottic stenosis (Myer-Cotton grade) at time of surgery	
Grade 2	91 (31%)
Grade 3	157 (53%)
Grade 4	31 (10%)

served are listed in Table 2.

For purposes of data analysis, children were divided both according to their subglottic grade and according to whether they had a single- or a double-stage procedure. The type of procedure (LTR or cricotracheal resection) and the type of graft used were recorded for each of the populations (Table 3). The operation-specific and overall extubation rates for children with single-stage procedures and for children with double-stage procedures are shown in Table 4. The results for those children who underwent cricotracheal resections are reported in Table 5. Results for children with grade 1 stenosis are not included, because the numbers were too few.

A χ^2 analysis of each data set was performed to look for statistically significant differences. Logistic regression modeling was used to look for statistical differences as a result of age. Examination of the results of the χ^2 analysis for operation-specific and overall outcomes based on grade alone without accounting for age revealed that there was no statistically significant difference among Myer-Cotton grade 2, 3, and 4 stenoses in terms of operation-specific extubation rates for single-stage LTR ($p = .63$). There was, however, a statistically significant difference among the grades in terms of overall extubation rates for this population ($p = .03$). Analysis of the double-stage procedures revealed statistically significant differences among grades in both operation-specific decannulation rates ($p = .0007$) and overall decannulation rates ($p = .04$).

TABLE 3. ANALYSIS OF LARYNGOTRACHEAL RECONSTRUCTIONS

	Single-Stage	*Double-Stage*
Number of children	98	101
Number of procedures	132	162
Mean number of procedures per child	1.3	1.6
Number of children with		
Myer-Cotton grade 2 subglottic stenosis	45	21
Myer-Cotton grade 3 subglottic stenosis	43	61
Myer-Cotton grade 4 subglottic stenosis	3	14
Type of graft		
Costal cartilage	112	132
Thyroid cartilage	7	9
Septal cartilage		6
Auricular cartilage	4	4
Other (cricotracheal resection)	9	11

TABLE 4. RESULTS OF LARYNGOTRACHEAL RECONSTRUCTIONS

Myer-Cotton Grade	*Extubation Rates for Single-Stage Procedures*		*Decannulation Rates for Double-Stage Procedures*	
	Operation-Specific	*Overall*	*Operation-Specific*	*Overall*
2	82% (37/45)	100% (45/45)	85% (18/21)	95% (20/21)
3	79% (34/43)	86% (37/43)	37% (23/61)	74% (45/61)
4	67% (2/3)	100% (3/3)	50% (7/14)	86% (12/14)
χ^2 Analysis	$p = .63$	$p = .03$	$p = .0007$	$p = .04$

To evaluate the effect of age on LTR, we divided the population in 2 ways. The first grouping divided the children according to whether they were less than or more than 2 years of age. The second grouping divided the children according to whether they were less than 2 years of age, between 2 and 5 years of age, or more than 5 years of age. For both groupings, logistic regression analysis revealed no statistically significant differences in overall extubation rates for either the single-stage ($p = .47$) or the double-stage procedures ($p = .96$).

DISCUSSION

One of the central aims of reviewing large retrospective series of surgical outcomes is to be able to better predict the results of future operations. Such predictions are fraught with potential error because of the immense selection bias implicit in the choices of which charts or cases should be reviewed in this retrospective fashion. Selection biases may be involved in which patients are chosen for review or the outcomes parameters themselves that are chosen. The temptation is to choose those patients whose

TABLE 5. RESULTS OF CRICOTRACHEAL RESECTIONS (N = 20)

Myer-Cotton Grade	*Extubation Rates for Single-Stage Procedures*		*Decannulation Rates for Double-Stage Procedures*	
	Operation-Specific	*Overall*	*Operation-Specific*	*Overall*
2	0		100% (1/1)	
3	75% (6/8)	88% (7/8)	75% (6/8)	88% (7/8)
4	100% (1/1)		67% (4/6)	83% (5/6)

success rates are the highest. However, to provide meaningful information, both to compare between differing studies and to educate families whose children are faced with the need for airway reconstructive surgery, a clear and uniform reporting system would be invaluable. For example, outcomes for squamous cell carcinoma of the head and neck are reported according to disease-specific criteria by defining the "T (size), N (nodal status), and M (presence or absence of metastasis)" stage.[14,15] Such a system does not currently exist to define the disorders for which pediatric airway reconstructive surgery is required.

It is well known that results of surgical procedures vary according to the disorders the procedures aim to address. With reference to the field of airway reconstruction, it has been documented that the results of single-stage LTR vary according to whether tracheomalacia exists as a co-morbid factor along with subglottic stenosis.[5] Clearly defined "disease-specific" outcomes parameters are a first step in establishing a classification system as a predictive model for outcome. Combining "disease-specific" with "grade-specific" data according to well-established grading systems further facilitates the development of such a model. As stated above, this "disease-specific" model will require future modification to take potential co-morbid factors such as tracheomalacia and gastroesophageal reflux disease into account. This will be best accomplished by establishing the criteria to be examined and then performing prospective database collection. Our results represent an example of how such a model would allow for meaningful analysis of surgical outcomes.

The results described above fall within the ranges for surgical outcomes that have been previously reported.[3,5-8,10,12,13] It is important to note that the children seen at CHMCC often have undergone attempted repair of their airway disease before being seen at CHMCC; the surgical results therefore may not necessarily mirror the results of operating on a child's airway for the first time. Another important point to note is that as the time frame for charts examined extended until January 2000, several of the children were in the process of moving toward, but were not yet at, the point of decannulation; the success rates necessarily reflect this limitation. Nevertheless, several global comments can be made. It is reasonable that both the operation-specific and overall results for the single-stage procedures would be somewhat higher than those for the double-stage procedures. The very decision to perform a single-stage procedure implies that the surgeon believes that there is a significant chance of success and that a double-stage procedure is not essential. Another aspect worthy of mention is the overall results for grade 3 and 4 subglottic stenoses. This disease severity remains a difficult surgical dilemma; the results serve to reinforce why alternative means such as cricotracheal resection are being employed to treat these difficult cases. Exactly which procedures are best utilized according to the specific disorders remains to be elucidated.

It is of interest that in our series, age was not a statistically significant factor when children were operated on above and below the 2-year age mark. Although success with LTR before the age of 2 years has been reported,[10] the impact of age upon surgical outcome has not been previously evaluated in a large series. Similar to the comparison of single-stage versus double-stage procedures, the statistical comparison of outcomes regarding airway reconstruction according to age criteria may *not* reveal underlying selection bias in terms of those children who are chosen to be operated on at a younger age. Most commonly, only those children who, in the experience of the senior surgeon, have the greatest likelihood of surgical success will be either scheduled for surgery at a young age (less than 2 years of age) or scheduled for a single-stage procedure. Exactly what defines this delineation remains to be studied in a prospective analysis; as has been described above, variables would include the presence or absence of other co-morbid medical problems such as gastroesophageal reflux disease, tracheomalacia, and concomitant glottic disease. However, for select children, early airway reconstruction is clearly a feasible option.

Finally, the operation-specific extubation and decannulation rates seem at first glance to be remarkably low. However, when coupled with the overall success rates, the figures become more recognizable. This means of representing the data is similar to the more traditional means of reporting overall success rates and the average number of procedures used to achieve these figures. When faced with a family member who wants counsel as to what to expect as a result of an upcoming operation, the ability to predict the likelihood of success for that given procedure is important. It remains understood that each open surgical procedure will require several endoscopic evaluations and minor manipulations after the major procedure. Nevertheless, the ability to predict whether and how often subsequent major procedures will be required is a first step in the development of a disease-based, operation-specific predictive model.

ACKNOWLEDGMENT — The authors thank Judy A. Bean, PhD, Department of Biostatistics, Children's Hospital Medical Center, for her assistance with statistical analysis.

REFERENCES

1. Grahne B. Operative treatment of severe chronic traumatic laryngeal stenosis in infants up to three years old. Acta Otolaryngol (Stockh) 1971;72:134-7.

2. Fearon B, Cotton R. Surgical correction of subglottic stenosis of the larynx. Preliminary report of an experimental surgical technique. Ann Otol Rhinol Laryngol 1972;81:508-13.

3. Cotton RT, Gray SD, Miller RP. Update of the Cincinnati experience in pediatric laryngotracheal reconstruction. Laryngoscope 1989;99:1111-6.

4. Lusk RP, Gray S, Muntz HR. Single-stage laryngotracheal reconstruction. Arch Otolaryngol Head Neck Surg 1991; 117:171-3.

5. Gustafson LM, Hartley BE, Liu JH, et al. Single-stage laryngotracheal reconstruction in children: a review of 200 cases. Otolaryngol Head Neck Surg 2000;123:430-4.

6. Seid AB, Pransky SM, Kearns DB. One-stage laryngotracheoplasty. Arch Otolaryngol Head Neck Surg 1991;117:408-10.

7. Monnier P, Savary M, Chapuis G. Cricotracheal resection for pediatric subglottic stenosis: update of the Lausanne experience. Acta Otorhinolaryngol Belg 1995;49:373-82.

8. Stern Y, Gerber ME, Walner DL, Cotton RT. Partial cricotracheal resection with primary anastomosis in the pediatric age group. Ann Otol Rhinol Laryngol 1997;106:891-6.

9. Cotton RT. Pediatric laryngotracheal stenosis. J Pediatr Surg 1984;19:699-704.

10. Narcy P, Contencin P, Fligny I, Francois M. Surgical treatment for laryngotracheal stenosis in the pediatric patient. Arch Otolaryngol Head Neck Surg 1990;116:1047-50.

11. Myer CM III, O'Connor DM, Cotton RT. Proposed grading system for subglottic stenosis based on endotracheal tube sizes. Ann Otol Rhinol Laryngol 1994;103:319-23.

12. Monnier P, Lang F, Savary M. Cricotracheal resection for pediatric subglottic stenosis. Int J Pediatr Otorhinolaryngol 1999;49(suppl 1):S283-S286.

13. Monnier P, Lang F, Savary M. Partial cricotracheal resection for severe pediatric subglottic stenosis: update of the Lausanne experience. Ann Otol Rhinol Laryngol 1998;107:961-8.

14. Beahrs OH. Staging of cancer. CA Cancer J Clin 1991; 41:121-5.

15. Weymuller EA. Head and neck cancer staging. In: Cummings CW, Fredrickson JM, Harker LA, Krause CJ, Richardson MA, Schuller DE, eds. Otolaryngology–head and neck surgery. Vol 1. St Louis, Mo: Mosby, 1998:136-44.