



Slide thyrocricotracheoplasty for the treatment of high-grade subglottic stenosis in children

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Abstract

Background/Purpose: The aim of this study was to describe our early experiences with a novel surgical procedure, “slide thyrocricotracheoplasty,” for the treatment of high-grade subglottic stenosis in children.

Patients and Methods: A retrospective analysis was performed in 7 children who underwent slide thyrocricotracheoplasty for high-grade subglottic stenosis from 1996 to 2009.

Results: Three patients were male and four were female. The etiology of subglottic stenosis was congenital (n = 4) or acquired (n = 3). All patients had undergone a tracheostomy before slide thyrocricotracheoplasty. Median age at operation was 16 months (range, 1-25 months). The median follow-up period after definitive operation was 58 months (range, 13-156 months). There was one case requiring debridement and re-anastomosis of slide thyrocricotracheoplasty site because of anastomotic disruption caused by a methicillin-resistant *Staphylococcus aureus* infection of the cartilage and one case requiring a minor operation to remove granulation tissue. At final follow-up, all patients did not have any airway cannulation with satisfactory functional outcome in terms of breathing and swallowing. All except one were noted to have acceptable vocal function. The patient with unsatisfactory vocal function continued to receive voice rehabilitation treatment.

Conclusions: Slide thyrocricotracheoplasty offers an efficient surgical treatment option with minimal morbidity for high-grade subglottic stenosis in children.

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Subglottic stenosis (SGS) is defined as a narrowing of the subglottic larynx, the region extending from the inferior margin of the true vocal cords to the inferior margin of the cricoid cartilage [1]. Subglottic stenosis can be either an acquired condition that develops after a considerable period of neonatal intubation or a congenital

condition that manifests as respiratory failure during the neonatal period. Over the last 2 decades, several surgical techniques for treatment of SGS have been devised and developed. These open surgical procedures have usually only been considered for treatment of severe cases of SGS in neonates, including splitting of the anterior cricoids with temporary stenting of the airway [2-3], anterior cricoid resection [4], partial cricotracheal resection and thyrotracheal anastomosis [4-6], and laryngotracheal reconstruction

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with cricoid division [6-7] with or without autografts. The morbidity and mortality rates of patients with SGS have dropped in recent years, although no definitively advantageous surgical procedure has been established because of insufficient experience and a lack of large-scale studies. According to the Myer-Cotton grading system, high-grade SGS is defined as grade III (luminal narrowing above 70%) or grade IV (virtually no lumen) stenosis [8], which includes SGS with severe hypertrophic scar changes after repeated bouginations or prolonged intubation. For grade IV SGS, especially in cases in which the stenotic segment reaches the true vocal cords and part of the thyroid cartilage, reconstructive surgery becomes even more complex, and other reconstructive procedure such as cricotracheal resection has not been considered as a good candidate because of concern for tension and safety margin in relation to true vocal cord. [9] In an animal experiment, Abdelkafy et al [10] applied slide-type laryngotracheal reconstruction to the treatment of SGS. We have also previously reported the feasibility and effectiveness of slide thyrocricotracheoplasty for the treatment of Myer-Cotton stage IV SGS in a child.[11] We have found that this procedure is a viable surgical option in this subgroup of patients for whom other surgical reconstructions are often difficult or inapplicable.

1. Materials and methods

A clinical diagnosis of high-grade SGS was based on the diagnosis of Myer-Cotton grade III or IV SGS obtained from bronchoscopy and 3-dimensional chest computed tomography. Clinical features and the results of investigations were obtained from medical records. During the study period of January 1996 to March 2009, a total of 46 patients younger than 18 years were diagnosed to have an SGS. Slide thyrocricotracheoplasty was performed only for extensive grade IV SGS, in which stenosis extended into the thyroid cartilage and nearly inferior margin of the true vocal cord. Less severe cases with variable degree of SGS were initially managed with conservative therapy, serial dilatations, anterior cricoids split, and steroid use. The other patients underwent other open surgical procedures such as cricoid resection and anastomosis, cricoids splitting, and permanent tracheostomy. Slide thyrocricotracheoplasty was performed as we have described previously [11]. In brief, after the inhalation of anesthesia through a tracheostomy tube, the neck was extended and a transverse collar neck incision was made with a circular skin incision surrounding the tracheostomy site. After transection of the platysma and strap muscles, the upper airway was exposed from the thyroid cartilage to the thoracic inlet. The extent of the stenotic segment was determined by intraoperative bronchoscopic observation of a 24-gauge fine needle tip inserted into the airway. After

defining the length of the stenotic segment, the airway was divided at the level of the midpoint of the stenotic segment. The cephalad portion of the stenotic trachea, the cricoids, and the lower one third of the thyroid cartilage just below the vocal cords were split at the midline on their anterior surface. The caudal portion of the stenotic trachea was similarly split on its posterior wall and extended until the normal segment of the trachea was encountered caudally. Posterior dissection of the tracheal wall allowed sliding of both segments. Excessive hypertrophied or scarred portions of the anterior cricoid arch and the lower part of the thyroid cartilage were resected and the right-angled corners of the 2 segments were trimmed. After flexing the neck, the superior segment was anastomosed with interrupted 4-0 or 5-0 Prolene sutures to the inferior segment, beginning posteriorly. The tracheostomy tube in the operative field was exchanged for an endotracheal tube. The anterior anastomosis was then completed with interrupted 4-0 or 5-0 Prolene sutures. The previous tracheostomy site was closed after resection

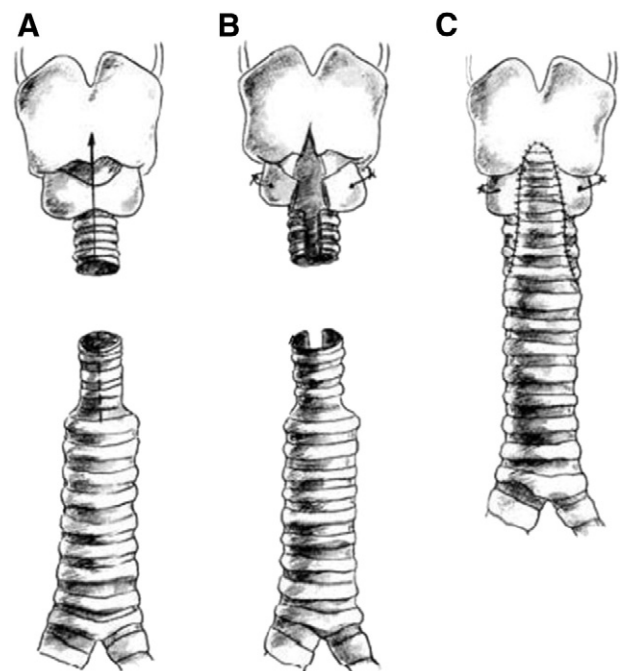


Fig. 1 A, The trachea was divided at the level of the midpoint of the obstructive segment. The cephalad portion of the stenotic trachea, the cricoids, and the lower one third of the thyroid cartilage just below the vocal cords were all split on their anterior surface. The caudal portion of the stenotic trachea was similarly split on its posterior wall. B, Two lateral traction sutures placed from the cricoid cartilage to the prevertebral fascia were necessary to maintain enough space to accommodate the caudal (sliding) portion of the trachea. C, The superior segment was anastomosed with interrupted 4-0 or 5-0 Prolene sutures to the inferior segment, beginning posteriorly. The endotracheal tube was inserted into the trachea and past the posterior anastomosis; the anterior anastomosis was then completed.

of scar tissue (Fig. 1). After slide thyrocricotracheoplasty, all patients were transferred to the pediatric intensive care unit for ventilator care during the immediate postoperative period. Under adequate sedation the neck was maintained in a flexed position with a prefabricated neck brace. We investigated the demographics, etiology of SGS, extent of stenosis, and associated anomalies in each patient. We also analyzed postoperative complications, decannulation, and functional status at final follow-up in terms of breathing, swallowing, and phonation. Study protocols were reviewed and approved by the Yonsei University College of Medicine Research Ethics Committee.

2. Results

2.1. Demographics and clinical features of the study patients

Demographics and clinical features of the 7 patients who underwent slide thyrocricotracheoplasty are shown in Table 1. There were 3 male and 4 female patients. The median age at corrective surgery was 16 months (range, 1-25.1 months). Subglottic stenosis was congenital in 4 patients and acquired in 3 patients. Among the 4 congenital cases, 2 patients were initially diagnosed with esophageal atresia (EA) (Gross type C). In one of those 2 patients (patient 1), after corrective surgery for EA, an esophageal stricture requiring bougination was found. However, an orotracheal tube (2.5 Fr.) could not be advanced below the level of the vocal cords. The patient eventually received a tracheostomy. The other patient initially diagnosed with EA (Patient 2) was referred from another hospital because of difficulty intubating. The other 2 congenital cases without concurrent EA were referred from other hospitals with tracheostomies because of respiratory insufficiency and difficult orotracheal intubation. All 3 patients diagnosed with acquired SGS were referred from other hospitals with tracheostomies after prolonged orotracheal intubation and

failed extubation attempts. The first patient (patient 5) had viral tracheobronchitis for 1 month and required intubation. Extubation was unable to be performed after a 3-week intubation period. The second patient (patient 6) who received corrective surgery for EA had an eventful postoperative course (long gap EA, Gross type C, and accidental ligation of the right bronchus). After an 8-week intubation period, extubation attempts failed. The final patient (patient 7), who had undergone an anorectal reconstruction for ARM, received a tracheostomy after extubation failure after a 4-week intubation period. All 7 patients showed grade IV SGS. The stenotic segment was purely subglottic in 6 patients and subglottic to the upper fifth tracheal cartilage in one patient.

2.2. Short-term postoperative outcomes of slide thyrocricotracheoplasty

In 4 patients, extubation was possible at a median of 5.5 postoperative days (range, 3-9 postoperative days). In 3 patients, decannulation was delayed; these patients received temporary tracheostomies. The first patient (patient 1) failed extubation twice although bronchoscopy showed no residual stenosis and eventually received a tracheostomy on the 46th postoperative day. The second patient (patient 4) had anastomotic disruption that led to acute respiratory difficulty and marked cervical emphysema. The following day the patient underwent end-to-end re-anastomosis surgery and received a temporary tracheostomy after extensive debridement of necrotic tissue around the anastomotic site. Tissue cultures revealed that a methicillin-resistant *Staphylococcus aureus* (MRSA) infection was associated with the anastomotic disruption. In the last case (patient 5), extubation was attempted several times to no avail. Bronchoscopy revealed bulging granulation tissue at the closed site of the previous tracheostomy with a patent anastomosis of slide thyrocricotracheoplasty. After 1 month of expectant management, the patient underwent bronchoscopic removal of granulation tissue and received a temporary tracheostomy.

Table 1 Summary of the demographics and clinical presentation of the patients who underwent slide thyrocricotracheoplasty

Patient no.	1	2	3	4	5	6	7
Sex	M	F	F	M	F	F	M
Age (mo) at operation	4.4	1.0	7.9	25.1	22	23	16
Etiology	Congenital	Congenital	Congenital	Congenital	Acquired	Acquired	Acquired
Previous tracheostomy	+	+	+	+	+	+	+
Grade of SGS ^a	IV	IV	IV	IV	IV	IV	IV
Involved site	Subglottic	Subglottic + 5th tracheal	subglottic	subglottic	subglottic	subglottic	subglottic
Associated anomaly	EA(C)	EA(C)	PE	BWS	–	EA(C)	ARM
	ASD, PDA		ASD, PDA	ASD, VSD			

M indicates male, F, female; EA(C), esophageal atresia (Gross type C); PE, pectus excavatum; BWS, Beckwith-Wiedemann syndrome; ARM, anorectal malformation; ASD, arterial septal disease; PDA, patent ductus arteriosus; VSD, ventricular septal disease.

^a Based on the Myer-Cotton classification system [8]: grade I, up to 50% obstruction; grade II, from 51% to 70%; and grade III, greater than 70% with any detectable lumen. An airway with no lumen was designated as grade IV.

Table 2 Short-term and long-term postoperative outcomes of slide thyrocricotracheoplasty

Patient no.	1	2	3	4	5	6	7
Postoperative complication	–	–	–	+ ^a	–	–	–
Postoperative decannulation	28 mo	3 d	5 d	53 mo	14 mo	9 d	6 d
Excision of granulation	+	–	–	–	+	–	–
F/U period (mo)	156	111	74	58	44	27	13
Dyspnea	–	–	–	–	–	–	+ ^b
Noisy breathing	–	–	–	+ ^c	–	–	+ ^c
Coughing/choking	–	–	–	–	–	–	–
Voice impairment	–	–	–	–	–	– ^d	+ ^e
De-glutition	nl	nl	nl	nl	nl	nl ^f	nl

nl: Normal.

^a Anastomotic disruption because of MRSA infection.^b With mild exertion.^c With moderate exertion.^d Minimally asthenic voice that is socially communicable.^e Aphonia, received voice rehabilitation at 13 months postoperatively.^f Intermittent GER symptoms.

There was no mortality during the immediate postoperative period (Table 2).

2.3. Long-term postoperative outcomes of slide thyrocricotracheoplasty

All 4 patients extubated within the first 10 days postoperatively had an uneventful clinical course thereafter. The other 3 patients, whose extubation was delayed during the immediate postoperative period, required tracheostomies. After several trials of corking and sealing off the tracheostomies, these patients were eventually extubated postoperatively at 28 months (patient 1), 53 months (patient 4), and 14 months (patient 5), respectively. The median follow-up period after airway reconstruction was 58 months (range, 13–156 months). None of the patients, with the exception of one, demonstrated signs of dyspnea. One patient (patient 7) exhibited dyspnea with mild exertion. Five patients demonstrated normal patterns of breathing. Two patients (patient 4, 7) exhibited noisy breathing after moderate exertion. Five of the patients had no voice impairment at all; however, 2 patients (patients 6 and 7) demonstrated voice impairment (Table 2).

3. Discussion

The survival rate of neonates continues to increase as the outcomes of prolonged respiratory care in the NICU improve. Some survivors may develop airway stenosis, with most of the cases involving the subglottic region. These patients usually require a tracheostomy with no voice with a

considerable risk of airway obstruction. The overall mortality in patients receiving a tracheostomy is known to be approximately 5.9% [12]. Most of the patients in our study were referred from other hospitals after some period of intubation or tracheostomy. All patients had grade IV SGS, for which no previous reconstruction surgery has been reported to be universally successful [6]. Previously, we performed a slide-type laryngotracheal reconstruction in a child with grade IV SGS and enjoyed a favorable outcome [11]. We continued to perform this procedure in our department for patients with high-grade SGS whom we considered less appropriate candidates for other types of reconstruction. We are encouraged by the results which show that slide-type laryngotracheal reconstruction (thyrocricotracheoplasty) with native tissue can be applied to even the most severe form of SGS (circumferential narrowing of the subglottis with virtually no lumen) with all the advantages of slide tracheoplasty. There was no mortality in our series of patients and the morbidity was acceptable. At final follow-up, no patients had a tracheostomy. We would like to highlight several important aspects of slide thyrocricotracheoplasty. We found that slide thyrocricotracheoplasty was very effective even in high-grade SGS, whether it was congenital or acquired in origin. Within 10 postoperative days, more than half of the patients had undergone successful decannulation. The 3 patients whose decannulation was delayed during the immediate postoperative period eventually all underwent successful decannulation. Serial bronchoscopy in the first patient (patient 1) revealed no residual stenosis at the anastomotic site. We thought that the cause of the failed decannulation during the immediate postoperative period resulted from spontaneous inward prolapse of the swollen arytenoids and supraglottis into the newly created airway lumen with each inspiratory movement. In the second patient (patient 4), decannulation was delayed because of the anastomosis problem described above (MRSA infection). In the last patient (patient 5), the primary problem was not with the anastomosis but rather resulted from luminal bulging growth of granulation tissue growing from the closed site of the previous tracheostomy. Thus, although the number of patients in our series was small, the anastomosis was found to be intact in all patients with the exception of one (MRSA infection) and the overall decannulation rate was 100% after slide thyrocricotracheoplasty, which is actually higher than other series reporting postoperative decannulation rate in patients with high-grade SGS [9,13–17]. This high success rate of overall decannulation in our study can be explained by several advantages specific to slide-type reconstruction. The first is that slide-type reconstruction has the advantage of using vascularized autograft, thereby facilitating healing of the anastomosis with minimal formation of scar tissue. Postoperative follow-up bronchoscopy in our study revealed with a patent anastomosis in all patients except one patient having granulation tissue at the closed site of the previous tracheostomy. The second is the

widely accepted notion that slide tracheoplasty does not hinder tracheal growth [18]. During the follow-up of our patients, we have found that the anastomosis further stabilizes as the laryngeal box grows. Lastly, as suggested by Acosta et al [19], slide-type reconstruction has only one half the tensions distributed over an oblique anastomosis that is longer than the circumferential length of a resection and anastomosis approach. No revisional surgery was necessary in our series of patients except one patient with infection because of devascularization or partial necrosis of the anastomosis, which has been anecdotally reported in airway reconstruction in patients having grade IV SGS [17]. Thus, better blood supply and less tension in slide-type reconstruction improve the integrity of the anastomosis. It is also noteworthy that the function and quality of the voice gradually improved in some patients in this study. Our greatest concern regarding the application of this technique in high-grade SGS was minimizing postoperative dysphonia because the area of surgical manipulation was very close to the true vocal cords in all patients. However, postoperative voice function was quite satisfactory in most cases, which suggests that damage to the critical nerves and muscles necessary for normal vocal function can be minimized during the midline procedure. In particular, continuation of the midline incision up to the inferior border of the thyroid cartilage in cases of severe SGS produces a beneficial effect without affecting glottis function. Among the 4 patients whose decannulation was successful during the immediate postoperative period, one patient (patient 6) exhibited aphonia for 6 months. Her voice function gradually improved to a socially acceptable level. However, one patient in our study still exhibited aphonia at 13 months postoperatively. In this patient the vocal cords assumed a paramedian position when the infant tried to phonate. We believe that the inadequate position of the vocal cords in this patient resulted from insertion of the lower tracheal segment formed during the slide thyrocricotracheoplasty. It has been suggested that enlargement of the anterior laryngeal commissure dramatically increases the degree of postoperative dysphonia [20]. If the anterior split is too close to the true vocal cords, as was the case in one of our patients, closure of the vocal cords is incomplete and normal vocal function is impaired. However, given that an adequate airway is a prerequisite for safe decannulation, the creation of a large interarytenoid space after reconstruction should not be considered a failure in this patient. Based on the outcomes of this study we cannot yet claim that slide thyrocricotracheoplasty is the most ideal operative method for high-grade SGS because of our lack of a control group, the small number of patients, and incomplete follow-up. However, in this study, slide thyrocricotracheoplasty resulted in successful laryngotracheal reconstruction in most of our patients. Based on the results from our experience, slide thyrocricotracheoplasty is a viable surgical

option for single-stage repair of high-grade (Myer-Cotton stage IV) SGS in children.

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