Pediatric Surgery International (2021) 37:555–559
https://doi.org/10.1007/s00383-020-04812-y

ORIGINAL ARTICLE

Therapeutic effectiveness of costal cartilage grafting into both anterior and posterior walls for laryngotracheal reconstruction in acquired subglottic stenosis

Shigeyuki Furuta[1] · Hideki Nagae[1] · Kei Ohyama[1] · Kunihide Tanaka[1] · Hiroaki Kitagawa[1]

Accepted: 16 December 2020 / Published online: 24 January 2021

Abstract

Background We have reviewed the surgical procedures performed and outcomes for low-birth-weight infants with acquired subglottic stenosis.

Methods The gestational age at birth, birth weight, age at Laryngotracheal reconstruction, and therapeutic outcome over the past 26 years were reviewed. Laryngotracheal reconstruction was initially performed by costochondral grafting involving only the anterior wall for the first operation, but since 2010 a costal cartilage was also inserted into the posterior wall of the cricoid cartilage using the BENSON pylorus spreader to split the cartilage.

Results There were 21 patients. The mean gestational age was 29.6 weeks, the mean birth weight was 1127 g, and the first surgery was performed at a mean age of 37.0 ± 21.8 months. Extubation was possible in 11 of 21 patients (52.3%) after the first surgery and in 7 of 7 patients after re-operation. The total extubation rate reached 100% (18/18) excluding three patients (one who suffered sudden death, and two who were lost to follow-up).

Conclusion Sufficient dilatation of the subglottic space could not be achieved by costochondral grafting involving the anterior wall alone. The extubation rate was improved by dilatation of the posterior wall and the insertion of costal cartilage into both the anterior and posterior walls.

Keywords Acquired subglottic stenosis · Low-birth-weight infants · Laryngotracheoplasty

Introduction

Extubation failure occurs in about 2% of low-birth-weight infants [1] and tracheotomy is performed in many cases. Acquired subglottic stenosis (ASGS) is a cause of extubation failure, in which stenosis of the cricoid cartilage is induced by long-term intubation in low-birth-weight infants.

ASGS is treated with laryngotracheoplasty (LTP), such as anterior cricoid split (ACS), after tracheotomy or endoscopic treatment with balloon dilatation and/or laser. Currently, many institutions perform Laryngotracheal reconstruction (LTR), which is a surgical procedure adding a cartilage graft to the ACS. Although the surgical procedure and management are slightly different among institutions, the extubation rate has been reported to be 56–100% [2–6].

We have previously treated ASGS by applying LTR (+ costal cartilage graft) only to the anterior wall of the trachea in infancy, but the extubation rate after the first surgery was only 57%. We, therefore, started adding dilatation of the posterior wall of the trachea in the cricoid cartilage region in 2010 attempting to improve the extubation rate. We report the surgical procedure and treatment outcome.

Materials and methods

Patients

We received approval from the institutional ethics committee at St. Marianna University, Kawasaki, Japan, for this study (No.3831). The gestational age, birth weight, timing of tracheotomy, and timing of LTR, and its treatment outcome

✉ Hiroaki Kitagawa
h2kita@marianna-u.ac.jp

[1] Division of Pediatric Surgery, St. Marianna University School of Medicine, 2-16-1 Sugao, Miyamae-ku, Kawasaki 241-8511, Japan

were retrospectively investigated in the patients with ASGS we treated at our hospital over the last 26 years.

Before surgery, ASGS was diagnosed by CT and rigid and flexible bronchoscopy, and the stenosis was classified (Myer–Cotton classification). In LTR, one-stage Laryngotracheal reconstruction (LTR-A) was performed before 2009, in which costal cartilage was grafted only to the anterior wall of the trachea and the tracheotomy was closed at the same time. When extubation was not possible following LTR-A, the tracheotomy was re-opened and LTR was repeated several years later. From 2010, we placed cartilage grafts in both the anterior and posterior walls of the trachea which we have called Laryngotracheal reconstruction, Anterior and Posterior (LTR-A + P). To do this, an incision was made in the posterior wall of the cricoid cartilage in addition to that in the anterior wall of the trachea, followed by dilatation using a Benson pyloric spreader and placement of a costal cartilage graft in both laryngeal incisions. The posterior cartilage graft was not our original innovation. Our major modification is the use of Benson forceps to spread the cricoid cartilage similar to pyloric stenosis. We then inserted large grafts in both the anterior and posterior subglottic incisions. For 26 years, the same surgeon has performed all the procedures for all the patients.

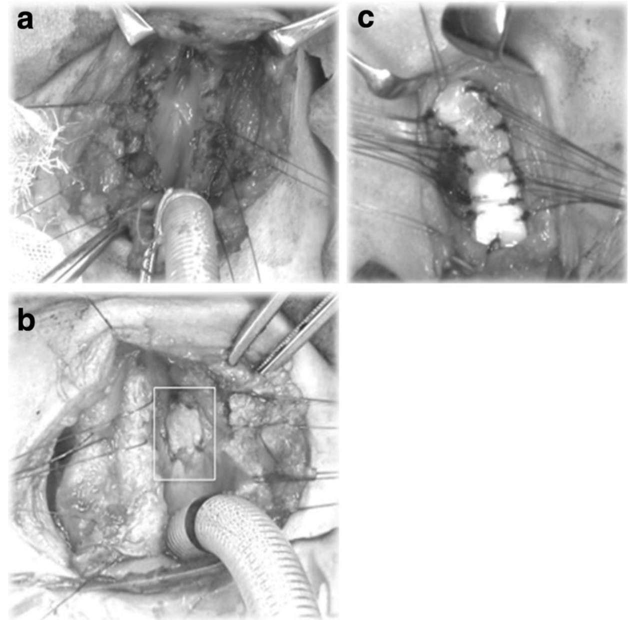

Fig. 1 **a** Anterior cricoid split. An intubation tube was inserted through the tracheotomy aperture for respiratory management. The incision was then advanced through the tracheotomy aperture to the trachea, cricoid cartilage, and upper end of the thyroid cartilage. **b** Posterior cricoid split. The cricoid cartilage posterior wall was incised in the midline with a scalpel. After dilatation with a Benson pyloric spreader, the costal cartilage was sutured to the cricoid cartilage (the mucosa was not sutured). **c** Dilatation of the anterior trachea using costal cartilage. A boat-shaped piece of costal cartilage was sutured to the anterior trachea dilating the tracheal lumen and the cricoid cartilage

Surgical procedure: LTR-A + P

The procedure was performed in a neck-extended position placing a shoulder pillow. A spiral cuffed endotracheal tube (ETT) was inserted into the tracheotomy aperture for respiratory management.

The costal cartilage graft was obtained from the left fifth and sixth costal cartilages. The tracheostomy stoma was circumcised and the incision was then extended transversely either side of the stoma. The anterior cervical muscles and thyroid gland were incised in the midline to reach the anterior trachea. The larynx and trachea above the stoma were exposed up to the upper margin of the thyroid cartilage and inferiorly the incision extended exposed down to the lower end of the tracheotomy aperture. The anterior wall of the trachea, the cricoid cartilage and the thyroid cartilage were incised leaving the upper margin of the thyroid cartilage intact (Fig. 1a). For the posterior wall of the cricoid cartilage, the tracheal mucosa and cartilage were partially incised using a sharp pointed scalpel, and the cricoid cartilage was carefully dilated using the Benson pyloric spreader. A section of costal cartilage, about 7 × 17 mm, was placed in the posterior incision, and the costal and cricoid cartilages were sutured together with 5-0 absorbable monofilament thread, without providing mucosal coverage to the graft (Fig. 1b). The ETT placed in the tracheotomy aperture was replaced with a nasal spiral cuffed ETT, using an introducer guided into the nose from the surgical field. This was inserted and placed as a stent, replacing the cuffed tube originally placed through the tracheostomy. An elongated oval of costal cartilage was then sutured to the anterior incision in the trachea and anterior cricoid ring with 4-0 absorbable monofilament thread (Fig. 1c). After irrigation with saline, a Penrose drain was placed at the anterior trachea, and the wound was closed. The patient was ventilated under sedation for 2 weeks after surgery, followed by extubation in an operating room.

The Fisher exact test was used to test for significance and $p < 0.05$ was judged to be a significant difference.

Results

Twenty-one patients underwent LTR. The mean gestational age at birth was 29.6 weeks. The mean birth weight was 1127.1 ± 560.3 g. Tracheostomy was performed at a mean age of 6.1 ± 3.9 months and the first radical surgery at a mean age of 37.0 ± 21.8 months. Using the Myer–Cotton classification, all patients undergoing ASGS were Grade 3 (71~99% obstruction). Decannulation was possible in 11 (52.3%) of 21 patients after the first radical surgery (LTR-A:

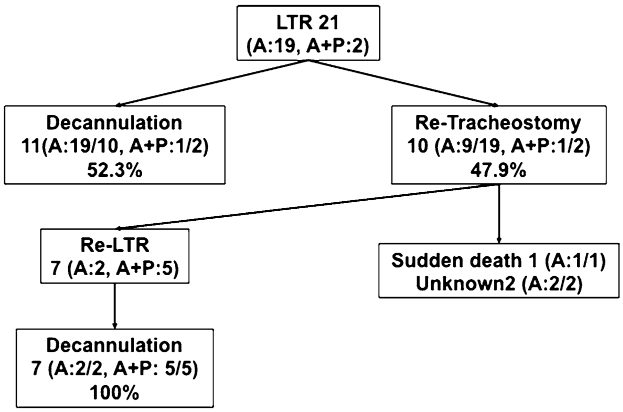

Fig. 2 LTR (Laryngotracheal reconstruction) in 21 patients and outcomes. LTR was performed in 21 patients. In the first surgery, LTR-A (LTP-Anterior) and LTR-A + P (LTR-Anterior and Posterior) were performed in 19 and 2 patients, respectively, and extubation was achieved in 52.3% (11/21). Tracheotomy was repeated in the other 10 patients. Re-operation has been performed in seven and all of them achieved extubation (LTR-A: 2, LTR-A + P: 5)

Table 1 Difference in the extubation rate between the surgical procedures

	Anterior only	Anterior and posterior
Laryngotracheal reconstruction	19	2
Re-laryngotracheal reconstruction	2	5
Decannulation	57.1% (12/21)	85.7% (6/7)

LTR-A (Laryngotracheal reconstruction-Anterior) was performed 21 times in 19 patients and the extubation rate was 57.1%. LTR-A + P (Laryngotracheal reconstruction-Anterior and Posterior) was performed seven times in six patients and the extubation rate was 85.7% (19, LTR-A + P: 2) and in 7 (100%) of 7 patients after re-operation (LTR-A: 2 or LTR-A + P: 5) (Fig. 1).

The total decannulation rate reached 100% (18/18) excluding three patients (one who suffered sudden death, and two who moved to another place and did not return to our hospital for follow-up) (Fig. 2). The operation-specific decannulation rate was 57.1% (12/21) for LTR-A only and 85.7% (6/7) for LTR-A + P (Table 1). Although the extubation rate for LTR-A + P was greater than LTR-A, this did not reach statistical significance ($p = 0.3642$, Fisher exact test).

We mainly used CT images for pre- and postoperative tracheal evaluations (Fig. 3). After extubation, if the patient was asymptomatic, bronchoscopy was not performed because of the edema at the suture sites. If the respiratory condition deteriorated, then we performed a bronchoscopy, followed by intubation. After re-tracheostomy and achieving respiratory stability, tracheal evaluation was performed by CT and bronchoscopy before re-LTR, and laryngeal fiberscopy was performed to evaluate the larynx. If hoarseness remained after the LTR, laryngeal fiberscopy was performed a few years later. During the stable period following LTR, laryngeal fiberscopy was performed in eight cases, and abnormal findings were observed in seven cases (dislocation of arytenoid cartilage in two cases, vocal cord paralysis in four cases, laryngomalacia in one case). Of these, five were re-LTR cases, so abnormal findings in the vocal cords and larynx may influence the extubation or decannulation rate.

After the initial LTR, if the patient failed several trials at extubation, then re-tracheostomy was performed. The timing of re-operation in the seven patients was all different. The earliest was 10 months after the initial procedure but the longest duration was 10 years after the initial operation. Patients requested re-operation when they felt significant

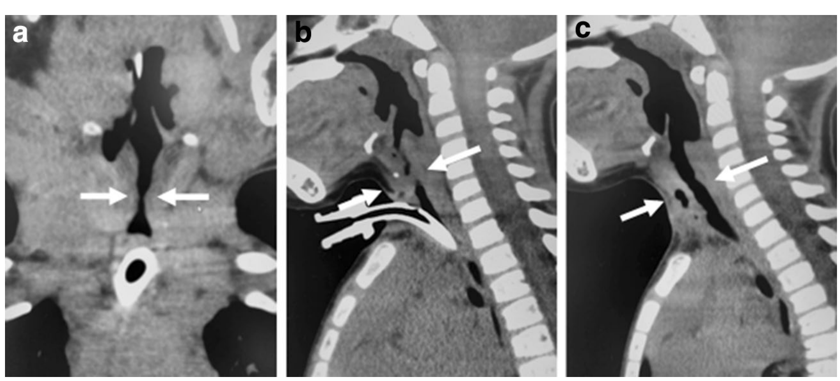

Fig. 3 Evaluation of subglottic stenosis (Tracheal CT image). A boy born at 28 weeks, weighing 1370 g. Due to RDS after birth and difficulty in extubation, artificial respiration was prolonged for 6 months and tracheostomy was performed. At 2 years old, a preoperative coronal CT image showed a ring-shaped subglottic stenosis (Grade 3: Myer–Cotton classification) (**a**). He underwent a LTR-A (Laryngotracheal reconstruction-Anterior). Two months after LTR-A, re-tracheostomy was performed due to difficulty in extubation. Re-stenosis was confirmed on sagittal CT images (**b**), and he underwent re-operation (LTR-A + P: Laryngotracheal reconstruction-Anterior and Posterior) at 3 years and 9 months old. He was extubated 2 weeks after the re-operation. On the sagittal CT image after the re-operation, the subglottic stenosis was released and the respiratory condition was stable (**c**)

inconvenience in their daily life. We did not guarantee to close the tracheostomy after re-operation. In addition, after closure of the tracheostomy, we followed these patients at least once a year until puberty, even if they were are asymptomatic.

Fortunately, there were no cases of re-stenosis after extubation. However, two patients had respiratory problems after extubation. These patients had laryngeal abnormalities, rather than re-stenosis in the subglottic area. One patient had laryngomalacia and the other patient had vocal cord movement abnormality which may relate to the long-term intubation after birth. We are planning to use CO_2 laser for the laryngomalacia patient and night-time BiPAP treatment for the patient with the vocal cord problem.

Discussion

We performed LTR grafting costal cartilage only to the anterior wall of the trachea (LTR-A) for ASGS until 2009 but resolution of the tracheal stenosis was inadequate, and re-stenosis developed after surgery in just over half of the cases, requiring a repeat tracheotomy. From 2010, in an attempt to reduce the rate of repeat tracheostomy, an incision was made in the posterior wall of the cricoid cartilage, in addition to the anterior wall, and the region was dilated with a Benson pyloric spreader followed by costal cartilage grafts in both cartilaginous incisions (LTR-A + P). Extubation was achieved in all but three patients (one who suffered sudden death, and two cases who moved to another place and did not return to our hospital).

The Myer–Cotton classification is widely used for grading stenosis [7], but the Modified Myer–Cotton classification including the condition of the vocal cords (such as supraglottic stenosis and vocal cord paralysis) and underlying diseases (heart, respiratory, neurological, digestive, and hereditary diseases) has been proposed, emphasizing the need to pay attention to impact of other lesions (not just the SGS) in assessing the treatment outcome [8]. Due to these varying classification regimes, it is very difficult to compare the treatment outcome of SGS between institutions. Recently, we performed flexible laryngoscopy without anesthesia in four children several years after LTR (two patients awaiting re-operation and two patients with night-time dyspnea) and evaluated the larynx and vocal cords. Hoarseness was noted in all four children. There was unilateral vocal cord dislocation in one child (7 years old), bilateral vocal cord dilatation failure in another child (6 years old), laryngomalacia in a 14-year-old child and unilateral vocal cord paralysis in the 4th child (13 years old). It may be difficult to diagnose these in infancy because of the difficulty in communicating with the patient. Morita et al. reported that stenosis reached the glottis or supraglottis in 72.7% (24/33) of SGS patients and this pathology was frequently noted in children in whom intubation was prolonged, with these patients being classified as "Extended to glottis or supraglottis" [9]. The decannulation rate was only 14.3% even after laryngotracheal resection in cases with this pathology, showing that these are cases are very difficult to treat [9]. In our patients, the mean duration of intubation from the neonatal period was long (6.1 months) and hoarseness is still persisting after surgery in 10 of 13 patients, suggesting that many cases were "Extended to glottis or supraglottis", or that they had vocal cord pathology.

Despite these conditions, the extubation rate was improved by the addition of a cartilage graft in the posterior wall of the cricoid cartilage. According to seven papers that performed a relatively large number of LTR-A + P, excluding case reports, the extubation rate of LTR-A + P was 93–100%, which is an excellent result [10–16]. However, as in our report, the target pathological conditions and surgical methods (single-stage LTR, double-stage LTR, etc.) are not uniform in any of the papers, so a clear answer as to the best approach could not always be derived. In many cases, it was stated that LTR-A + P was selected for more severe SGS cases compared to LTR-A alone. Younis et al. [11] reported that the LTR-A + P procedure was targeted to patients with SGS Grade 3–4, or problems with their interarytenoid region or their vocal cord fixation, suggesting that these conditions were indications to perform a re-operation. In our surgical procedure, the thyroid cartilage was incised to the upper limit of the thyroid cartilage together with the anterior wall of the trachea. The posterior wall of the cricoid cartilage was also dilated, extending a part of the infraglottic cavity up to the glottic region. Slightly opening the vocal cords may have secured maintenance of the airway but this may have been the cause of the high frequency of postoperative hoarseness. If decannulation is aimed for in surgery for ASGS, the quality of voice may be impaired.

There were several limitations of this study. First, the number of patients was small. Second, the evaluation of the surgical procedure was compromised because LTR-A and LTR-A + P were both included in the assessment of both the first surgery and any re-operation. The extubation rate was superior in the LTR-A + P cases, although this was not no statistically significant. It is not yet clear whether LTR-A + P should be employed as the first surgery or used only for re-operations. Given the frequent need for re-operation in LTR-A patients, we are now using LTR-A + P as our primary procedure in severe SGS patients. The SGS and the larynx and vocal cords should be carefully evaluated before and after LTR, but making an accurate diagnosis in infancy is difficult.

Conclusion

All our ASGS patients had high-grade stenosis. Up to 2009, ASGS was treated with LTR grafting costal cartilage only to the anterior wall of the trachea. However, the subglottic stenosis and the tracheal stenosis was not always sufficiently corrected and re-stenosis occurred, requiring a repeat tracheotomy in just over half of the cases. From 2010, the posterior wall of the cricoid cartilage was dilated with Benson forceps to sufficiently expand the stenotic region followed by a costal cartilage graft in both the anterior and posterior incisions in both the first surgery and in re-operations, and extubation was achieved in all patients.

Acknowledgements The authors thank to Professor Kevin Pringle in Department of Obstetrics and Gynecology, School of Medicine & Health Sciences, University of Otago, Wellington, New Zealand for his help with English editing for this manuscript.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval All the procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and the 1964 Declaration of Helsinki and its later amendments or comparable ethical standards.

Informed consent Informed consent was obtained from all individual participants included in this study.

References

1. Santos D, Mitchell R (2010) The history of pediatric airway reconstruction. Laryngoscope 120(4):815–820
2. Holinger LD, Stankiewicz JA, Livingston GL (1987) Anterior cricoid split: the Chicago experience with an alternative to tracheotomy. Laryngoscope 97(1):19–24
3. Eze NN, Wyatt ME, Hartley BE (2005) The role of the anterior cricoid split in facilitating extubation in infants. Int J Pediatr Otorhinolaryngol 69(6):843–846
4. Rotenberg BW, Berkowitz RG (2006) Changing trends in the success rate of anterior cricoid split. Ann Otol Rhinol Laryngol 115(11):833–836
5. Agrawal N, Black M, Morrison G (2007) Ten-year review of laryngotracheal reconstruction for paediatric airway stenosis. Int J Pediatr Otorhinolaryngol 71(5):699–703
6. Marom T, Joseph RA, Grindle CR, Shah UK (2014) Tracheostomy after laryngotracheoplasty: risk factors over 10 years. J Pediatr Surg 49(8):1206–1209
7. Myer CM, O'Connor DM, Cotton RT (1994) Proposed grading system for subglottic stenosis based on endotracheal tube sizes. Ann Otol Rhinol Laryngol 103(4 Pt 1):319–323
8. Monnier P, Ikonomidis C, Jaquet Y, George M (2009) Proposal of a new classification for optimising outcome assessment following partial cricotracheal resections in severe pediatric subglottic stenosis. Int J Pediatr Otorhinolaryngol 73(9):1217–1221
9. Morita K, Yokoi A, Bitoh Y, Fukuzawa H, Okata Y, Iwade T, Endo K, Takemoto J, Tamaki A, Maeda K (2015) Severe acquired subglottic stenosis in children: analysis of clinical features and surgical outcomes based on the range of stenosis. Pediatr Surg Int 31:943–947
10. Smith LP, Zur KB, Jacobs JN (2010) Single- vs double-stage laryngotracheal reconstruction. Arch Otolaryngol Head Neck Surg 136(1):60–65
11. Younis RT, Lazar RH, Astor F (2003) Posterior cartilage graft in single stage laryngotracheal reconstruction. Otolaryngol Head Neck Surg 129(3):168–175
12. Koltai PJ, Ellis B, Chan J, Calabro A (2006) Anterior and posterior cartilage graft dimensions in successful laryngotracheal reconstruction. Arch Otolaryngol Head Neck Surg 132(6):631–634
13. Gustafson LM, Hartley BE, Liu JH, Chadwell J, Koebbe C, Myer CM 3rd, Cotton RT (2000) Single-stage laryngotracheal reconstruction in children: a review of 200 cases. Otolaryngol Head Neck Surg 123(4):430–434
14. Zalzal GH (1993) Treatment of laryngotracheal stenosis with anterior and posterior cartilage grafts. A report of 41 children. Arch Otolaryngol Head Neck Surg 119(1):82–86
15. Schmidt RJ, Shan G, Sobin L, Reilly JS (2011) Laryngotracheal reconstruction in infants and children: are single-stage anterior and posterior grafts a reliable intervention at all pediatric hospitals? Int J Pediatr Otorhinolaryngol 75(12):1585–1588
16. Younis RT, Lazer RH, Astor F (2003) Posterior cartilage graft in single-stage laryngotracheal reconstruction. Otolaryngol Head Neck Surg 129(3):168–175