Successful surgical treatment of congenital tracheal stenosis combined with tracheal bronchus and left pulmonary artery sling: a 10-year single-institution experience

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Abstract

Purpose  Radical surgery for congenital tracheal stenosis (CTS) is technically demanding. CTS combined with tracheal bronchus (TB) and pulmonary artery (PA) sling is a particularly challenging condition. We herein report our successfully modified surgical techniques for CTS combined with TB and PA sling.

Methods  Nine patients treated at our institution from July 2010 to December 2020 for CTS with TB and PA sling were enrolled. The patients’ characteristics, operative results, and clinical outcomes were reviewed and analyzed retrospectively.

Results  The mean age at the operation and body weight were 8.0 ± 4.4 months old and 6.5 ± 0.8 kg, respectively. The mean tracheal diameter and length of the stenotic lesion were 3.2 ± 1.0 mm (mean stenosis rate 46.2%) and 25.4 ± 4.9 mm, respectively. All cases were complicated with PA sling at bifurcation stenosis with tracheobronchomalacia. All patients underwent modified posterior-anterior slide tracheoplasty with an inverted Y-shaped incision at the bifurcation and repositioning of the PA. The mean postoperative intubation period was 25.0 ± 32.1 days. There were no major intraoperative or postoperative complications, including hypoxic-ischemic encephalopathy. The mean hospital stay was 92.2 ± 73.4 days. All patients were discharged home without tracheostomy or oxygen support.

Conclusion  Our slide tracheoplasty technique for CTS with TB and PA sling achieved excellent outcomes.

Level of evidence  Level IV.

Keywords  Congenital tracheal stenosis · Tracheal bronchus · Pulmonary artery sling · Slide tracheoplasty · Inverted Y-shaped incision

Introduction

Congenital tracheal stenosis (CTS) is a rare and life-threatening disease in children. Slide tracheoplasty for CTS has become widely accepted, now representing a standard surgical procedure [1–3]. However, CTS combined with tracheal bronchus (TB) and left pulmonary artery (PA) sling remains a particularly challenging condition to treat, as TB of the right upper lobe bronchus (RULB) and stenosis of the bifurcation can cause respiratory failure after ordinary slide tracheoplasty [4–6]. PA sling also sometimes causes tracheobronchomalacia at the bifurcation, leading to a vascular ring surrounding and compressing the trachea. These conditions are thought to be the leading causes of extubation failure [4, 7].

The present study analyzed the clinical features and treatments of CTS combined with TB and PA sling as well as the outcomes of our surgical techniques based on the experience in our single institution.
Materials and methods

Study population and data collection

Fifty-nine patients with CTS were treated at our institution from July 2010 to December 2020; the 9 patients (15.3% of the CTS cases) treated for CTS combined with TB and PA sling were selected for this study (Fig. 1).

A retrospective chart review and data collection were performed after receiving institutional review board approval. The patients’ characteristics, operative results, and clinical outcomes were reviewed based on their medical records. The data that were collected included the age and body weight at the operation, sex, tracheal diameter and length of the tracheal stenosis, associated cardiac anomalies, operative time, surgical complications, postoperative extracorporeal membrane oxygenation (ECMO) and intubation period, follow-up period, and patient condition after the operation, including the extubation rate.

Operative preparation and surgical technique

Before starting the operation, patients were nasally intubated with an endotracheal tube using a flexible bronchoscope. Under general anesthesia, thoracotomy was performed by median sternotomy, and ECMO was introduced. In addition to the repositioning of the PA, cardiac surgery was performed first in cases with other cardiovascular anomalies, after which tracheoplasty was performed.

The position of the upper and lower ends of the tracheal stenosis was confirmed using an intraoperative bronchoscope. We performed a modified slide tracheoplasty technique, as shown in Fig. 2. All patients received posterior-anterior slide tracheoplasty with an inverted Y-shaped incision at the bifurcation and repositioning of the PA. However, we altered our techniques depending on the length of the stenotic lesion (below or above TB).

Fig. 1 CTS combined with TB and PA sling. CTS congenital tracheal stenosis; TB tracheal bronchus; PA pulmonary artery; RULB right upper lobe bronchus

Fig. 2 Operative procedure for CTS combined with TB and PA sling using inverted-Y shaped incision. a The trachea was transected above the midpoint of the TB and tracheal bifurcation (black straight line). b The distal segment of the trachea was longitudinally opened along its anterior wall with an inverted-Y-shaped incision at the tracheal bifurcation. c Modified slide tracheoplasty brought the TB and tracheal bifurcation close to each other. d In cases with the stenotic lesion of the trachea extending to the proximal side of the TB, the stenotic trachea was transected just below the TB (black straight line). CTS congenital tracheal stenosis; TB tracheal bronchus; PA pulmonary artery
Tracheal stenosis between TB and tracheal bifurcation

The trachea was transected just above the middle point of the TB and tracheal bifurcation (Fig. 2a). The distal segment of the trachea was longitudinally opened along its anterior wall extending to the tracheal bifurcation with an inverted-Y-shaped incision to enlarge the anastomotic tracheal lumen (Fig. 2b). For the proximal segment of the stenotic trachea, a posterior incision was performed in the 5 o’clock direction to prevent anastomotic stenosis and maintain the blood supply from the lateral surface of the trachea, as shown in Fig. 3a. Slide tracheoplasty was then performed.

Tracheal anastomosis was performed using interrupted 5–0 monofilament absorbable sutures, and the proximal end was just covered over the incised tracheal bifurcation and bronchi (Fig. 2c). The corners of the proximal end should not be trimmed. The tips of the endotracheal tube were positioned just above the TB after tracheoplasty. A drainage tube was placed in front of the trachea below the xiphoid process to close the wound.

Tracheal stenosis from above TB to tracheal bifurcation

In cases in which the stenotic lesion of the trachea extended to the proximal side of the TB (Fig. 4a), the stenotic trachea was transected just below the TB of the RULB, and posterior-anterior slide tracheoplasty was performed (Fig. 2d). This technique brings the TB of RULB and bifurcation closer together, forming a wider triple-bifurcation shape (Figs. 3b, 4b).

Postoperative management

After tracheoplasty, ventilation was resumed, and we confirmed that ventilation was stable. Weaning from ECMO was then attempted in the operating room. In high-risk cases, such as those with severe cardiac anomalies, the cannulation catheter was kept in place for postoperative ECMO management.

To maintain the cervical anteflexion position and tracheoplasty at rest, the patients were fixed and managed using a magic bed with sedation via a muscle relaxant in the pediatric intensive-care unit (PICU) for at least four days. We then evaluated edema, granulation, and tracheomalacia of the anastomotic site of the trachea using a flexible bronchoscope and attempted to wean patients from the ventilator.

Statistical analyses

Data are expressed as the mean ± standard deviation, and ranges, and/or binomial percentages, where appropriate. All statistical analyses were performed with EZR (Saitama Medical Center, Jichi Medical University, Saitama, Japan), which is a graphical user interface for R (The R Foundation for Statistical Computing, Vienna, Austria). More precisely, it is a modified version of R commander designed to add statistical functions frequently used in biostatistics [8].

Ethical approval

This study was performed in accordance with the Ethical Guidelines for Medical and Health Research Involving Human Subjects by the Ministry of Health, Labour, and...
Welfare of Japan in 2014. The study complied with the 1964 Declaration of Helsinki (revised in 2013) and was approved by the Research Ethics Committee of Tokyo Metropolitan Children's Medical Center (registration number: H30b-209). All participants or their parents provided their informed consent for involvement in this study.

Results

Patients’ characteristics

The patients’ characteristics are shown in Table 1. The male/female ratio was 4/5. The mean age at the time of the diagnosis of CTS was 4.9 ± 4.4 (range 0–15) months old. The management for respiratory failure were shown in Table 1. Five cases required intratracheal intubation with a ventilator. One case required CPAP and following O2 administration. One case requires NPPV and following CPAP. Two cases required O2 administration. But of 5 cases that required intratracheal intubation, 4 cases could be extubated after respiratory condition improved. At the timing of radical surgery, 6 cases did not require respiratory management and 3 case required respiratory management (intratracheal intubation: 1, CPAP: 1, O2 administration: 1). The mean age at the time of the operation and body weight was 8.0 ± 4.4 (range 4–18) months old and 6.5 ± 0.8 (range 5.3–7.3) kg, respectively. The mean tracheal diameter of the stenotic lesion and mean stenosis rate were 3.2 ± 1.0 (range 1.7–4.8) mm and 46.2% ± 0.1% (range 26–53%), respectively. The mean tracheal length of the stenotic lesion was 25.4 ± 4.9 (range 20–35) mm. Regarding the site of tracheal stenosis, eight cases had stenosis from the distal side of the TB to the tracheal bifurcation, and one case had stenosis from the proximal side of the TB to the tracheal bifurcation (Table 1, Case 7).

Associated cardiac anomalies were shown in Table 1. All cases involved PA sling with bifurcation stenosis and tracheobronchomalacia, and the other associated cardiac anomalies were tetralogy of Fallot (TOF) in one, atrial septal defect (ASD) in four, and persistent left superior vena cava (PLSVC) in three.

The operative results and clinical course

The operative results and clinical outcomes were shown in Table 2. All patients received slide tracheoplasty with an inverted Y-shaped incision at the bifurcation and repositioning of the PA. Among the cases with cardiac anomalies, the one case of TOF and four cases of ASD underwent one-stage
surgery at the same time as tracheoplasty. The three patients with PLSVC underwent conservative follow-up.

The mean operative time was 690.1 ± 251.0 (range 464–1302) min. There were no intraoperative complications. Postoperative ECMO was required in 5 cases (55.6%, mean duration: 16.4 ± 18.9 days, range 7–50 days). In three cases, ECMO was continuously performed from the operating room; the other two cases were able to be weaned from ECMO temporarily in the operating room after tracheoplasty, but after entering the PICU, ventilation failure occurred, and ECMO management was required again within 12 h after surgery in both cases. The mean postoperative intubation period was 25.0 ± 32.1 (range 8–108) days.

In case 2, continuous hemorrhaging from the drain was recognized due to mismanagement of anticoagulation. However, the hemorrhaging was resolved by appropriate anticoagulation. In case 4, minor leakage from the tracheal anastomosis and right heart failure was observed after surgery but improved with conservative management. There were no postoperative hypoxic episodes or subsequent ischemic encephalopathy. The mean hospital stay was 92.2 ± 73.4 (range 47–283) days. All patients were discharged home without tracheostomy or oxygen support. The mean follow-up period was 81.8 ± 3.9 (range 32–136) months. No patients showed tracheal stenosis, intratracheal granulation, or tracheomalacia during the follow-up period.

### Discussion

CTS combined with TB and PA sling is a life-threatening disease that is difficult to manage not only during surgery but also after tracheoplasty [4–6]. We modified a slide tracheoplasty technique for CTS combined with TB and PA sling.

The major findings of this study were as follows: (1) We modified the slide tracheoplasty technique to bring the TB and bifurcation closer and formed a wider triple-bifurcation shape; (2) All cases were complicated with PA sling at the

#### Table 1 Patients’ characteristics of CTS with TB and left PA sling

| Case | Sex | Age at diagnosis (Month) | Management for respiratory failure (age: d; day; m; month) | Respiratory management at radical surgery | Age at operation (Month) | Body weight at operation (kg) | Tracheal diameter (mm) (stenosis rate: %) | Length of the stenosis (mm) | Associated cardiac anomalies without PA sling |
|------|-----|--------------------------|----------------------------------------------------------|------------------------------------------|--------------------------|---------------------------|-------------------------------------|--------------------------|---------------------------------------------|
| 1    | M   | 0                        | CPAP (0-3d), O₂ administration (4 m)                     | No                                       | 4                        | 7.3                       | 2.4 (50%)                           | 23                       | –                                           |
| 2    | F   | 2                        | Intratracheal intubation (2 m)                           | No                                       | 5                        | 6.4                       | 2.5 (50%)                           | 20                       | TOF<sup>a</sup>, PLSVC                     |
| 3    | F   | 8                        | Intratracheal intubation (5 m/8 m)                       | No                                       | 11                       | 5.8                       | 3.5 (50%)                           | 22                       | –                                           |
| 4    | F   | 6                        | Intratracheal intubation (6 m)                           | No                                       | 9                        | 7.3                       | 4.3 (50%)                           | 24                       | ASD<sup>a</sup>                           |
| 5    | M   | 15                       | Intratracheal intubation (16 m)                          | No                                       | 18                       | 7                         | 3.6 (49%)                           | 32                       | –                                           |
| 6    | M   | 5                        | O₂ administration (5 m)                                  | No                                       | 9                        | 7.1                       | 3.5 (49%)                           | 23                       | ASD<sup>a</sup>, PLSVC                     |
| 7    | F   | 3                        | NPPV (5 m) → CPAP (5 m)                                  | CPAP                                     | 5                        | 5.6                       | 4.8 (53%)                           | 35                       | ASD<sup>a</sup>, PLSVC                     |
| 8    | M   | 5                        | Intratracheal intubation (5 m)                           | Intratracheal intubation                  | 5                        | 5.3                       | 2.5 (39%)                           | 25                       | –                                           |
| 9    | F   | 0                        | O₂ administration (1-6 m)                                | O₂ administration                        | 6                        | 6.7                       | 1.7 (26%)                           | 25                       | ASD<sup>a</sup>                           |

Mean ± SD 4.9 ± 4.4 8.0 ± 4.4 6.5 ± 0.8 3.2 ± 1.0 (46.2 ± 0.1%) 25.4 ± 4.9

<sup>a</sup>One-stage surgery at the same time as slide tracheoplasty

CTS congenital tracheal stenosis; TB tracheal bronchus; PA sling pulmonary artery sling; CPAP continuous positive airway pressure; NPPV non-invasive positive pressure ventilation; TOF tetralogy of fallot; PLSVC persistent left superior vena cava; ASD atrial septal defect
bifurcation stenosis with tracheobronchomalacia and underwent posterior-anterior slide tracheoplasty with an inverted Y-shaped incision at the bifurcation and repositioning of the PA; (3) Postoperative ECMO management was required in 55.6% of cases; (4) There were no major intraoperative or postoperative complications, including hypoxic episodes and subsequent ischemic encephalopathy; and (5) All patients were discharged home without tracheostomy, and no patients showed tracheal stenosis, intratracheal granulation, or tracheomalacia during the long-term follow-up period.

TB is defined as a congenital anomaly in which the RULB arises directly from the trachea and above the tracheal bifurcation in the narrow sense [9]. Cases with CTS combined with TB tend to have a combination of tracheal abnormalities and cardiac anomalies, resulting in a high postoperative mortality rate [10, 11]. Although various surgical procedures have been reported, which is the most appropriate tracheoplasty procedure remains controversial. Tracheal end-to-end anastomosis was performed for short-segment CTS cases with a stenotic trachea below the TB [1, 12]. For cases with long-segment CTS, slide tracheoplasty described by Tsang and Grillo has been widely accepted and become the standard tracheoplasty procedure [1, 2]. Left–right slide tracheoplasty for CTS with TB was reported to avoid the need for an incision and a suture line closing the orifice of RULB [13]. However, Morita et al. did not recommend this procedure, as their cases had severe tracheomalacia after left–right slide tracheoplasty and required tracheostomy. They pointed out two reasons for this complication with left–right slide tracheoplasty [5]. One reason was that a transverse figure-eight deformity occurred at the reconstruction site and easily collapsed [6]. The other reason was that the tracheal walls receive their blood supply from the lateral surface of the trachea [14], and ischemia occurred in the central part of the trachea at the anastomotic site [10]. We, therefore, modified the posterior-anterior slide tracheoplasty technique for long-segment CTS below or above the TB, as reported by Beierlein and Elliott [13].

In our procedure, the stenotic trachea was transected just above the middle point between TB and tracheal bifurcation and slide tracheoplasty was performed to make TB and tracheal bifurcation close forming a tracheal trifurcation. The tip of the endotracheal tube was able to be placed just above the TB after tracheoplasty. This allowed the suction tube to easily reach the anastomotic site without leaving a long distance from the intubation tube and prevented critical postoperative airway obstruction caused by edema of the anastomotic site and bloody secretions. We also performed another modification of the slide tracheoplasty technique for TB: the proximal segment of the trachea was incised slightly to the left lateral side of the dorsal midline (5 o’clock direction) to maintain the blood supply. The TB arises from the posterolateral aspect of the trachea in most patients [5], so this incision approach avoided the need to perform suturing near the orifice of the TB. In our study, airway obstruction was prevented in all cases at the TB and bifurcation using this method. As a result, all cases were able to be extubated safely.

CTS were sometimes associated with PA sling and complete vascular ring [15]. But there were no cases that

| Case | Operative time (min) | Intraoperative complications | Postoperative ECMO period (days) | Postoperative intubation period (days) | Postoperative complications | Tracheostomy/O2 support | Postoperative hospital stay (days) | Follow-up period (months) |
|------|---------------------|------------------------------|---------------------------------|--------------------------------------|----------------------------|--------------------------|----------------------------------|---------------------------|
| 1    | 611                 | –                            | 11                              | 33                                   | –/–                        | –/–                      | 70                               | 136                       |
| 2    | 1302                | –                            | 50a                             | 108                                  | Bleeding from drain        | –/–                      | 283                              | 119                       |
| 3    | 808                 | 6a                           | 17                              | –                                    | –/–                        | –/–                      | 87                               | 108                       |
| 4    | 599                 | 8                            | 15                              | –                                    | –/–                        | –/–                      | 53                               | 91                        |
| 5    | 662                 | 7                            | 15                              | Minor leakage of tracheal anastomosis/Right heart failure | –/–                      | –/–                      | 62                               | 69                        |
| 6    | 464                 | 8                            | 12                              | –                                    | –/–                        | –/–                      | 60                               | 67                        |
| 7    | 534                 | 9                            | 12                              | –                                    | –/–                        | –/–                      | 100                              | 61                        |
| 8    | 533                 | –                            | 8                               | 15                                   | –/–                        | –/–                      | 68                               | 53                        |
| 9    | 698                 | –                            | 8                               | –                                    | –/–                        | –/–                      | 47                               | 32                        |
| Mean ± SD | 690.1 ± 251.0     | 0                            | 16.4 ± 18.9                     | 25.0 ± 32.1                          | 2 (22.2%)                  | 0/0                      | 92.2 ± 73.4                  | 81.8 ± 3.9                |

ECMO extracorporeal membrane oxygenation

*a ECMO management was started within 12 h after entering the PICU.
underwent prenatal diagnosis of CTS associated with PA sling and complete vascular ring in our case series. All cases underwent a definitive diagnosis of CTS due to postnatal respiratory failure. PA sling is the most common cardiac anomaly and is complicated with CTS in 50–65% of cases [16, 17], causing airway obstruction in the neonatal and infant periods due to complete cartilage rings surrounded by PA. Tracheobronchomalacia at the bifurcation induced by compression of the vascular ring was found in 27% of cases, and this condition was a risk factor for mortality [7, 18]. Muraji et al. reported the first survivor after concomitant reconstruction of extensive CTS and PA sling in 1998 [19]. However, the lumen of the tracheal bifurcation could not be enlarged sufficiently with conventional posterior-anterior slide tracheoplasty. Extubation was reportedly difficult in about 30–40% of cases due to the flattened bifurcation after conventional slide tracheoplasty [20], requiring additional aortopexy, intratracheal stenting, and tracheostomy postoperatively [4, 6, 20].

Since 2005, we have performed an inverted Y-shaped incision for cases with tracheobronchomalacia at the tracheal bifurcation [7]. In all our cases, no tracheobronchomalacia was recognized at the bifurcation after slide tracheoplasty, and extubation was achieved without additional surgery. This technique is thus worth attempting before considering other approaches, such as prolonged stenting and tracheotomy.

PA sling associated with CTS has a high rate of cardiac anomalies and a high mortality rate (up to 46%) [21]. This high mortality is due to the severity of cardiac anomalies in addition to the presence of CTS. Staged repair is beneficial for CTS with complex cardiac anomalies with keeping intra- and postoperative stable circulatory dynamics potentially affect the outcomes of combined tracheal and cardiac surgery [11]. In our institution, we proactively use postoperative ECMO management for CTS cases with severe cardiac anomalies, depending on the cardiac disease, to improve the survival rate.

Postoperative ECMO management was performed in 5 cases (55.6%). Three of them underwent continuous ECMO management during and after surgery, ultimately achieving ECMO/ventilator weaning and extubation without respiratory complications or hypoxic-ischemic episodes following encephalopathy.

Conclusion

Several limitations associated with the present study warrant mention. Namely, we investigated few patients, and the results were thus obtained from a relatively small population. However, our modified tracheoplasty approach was able to prevent ventilation and respiratory failure after tracheoplasty. As a result, all patients were discharged home without tracheostomy or oxygen support, and none of our cases had any postoperative problems during the follow-up period. Our results suggest that slide tracheoplasty using an inverted Y-shaped incision and forming a wide triple-bifurcation shape is an effective surgical technique for CTS combined with TB and left PA sling.

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Author contributions T.H. and S.I. wrote the main manuscript text. N.S. and A.S. collected the patients information, and K.K. and S.H. prepared the study design. H.T. prepared figures 1-4. All authors reviewed the manuscript.

Declarations

Competing interests The authors declare no competing interests.

Conflict of interest The authors declare no conflicts of interest in association with the present study.

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