Clinical outcomes of slide tracheoplasty in congenital tracheal stenosis†

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Abstract

OBJECTIVES: Treatment of long-segment congenital tracheal stenosis (CTS) remains challenging. Recently, slide tracheoplasty has become the standard approach in many centres. The aim of this study was to evaluate the clinical outcomes of slide tracheoplasty.

METHODS: Between 2004 and 2011, 18 patients underwent slide tracheoplasty in our centre. The median patient age was 2.5 months (range, 18 days–4 years) and the median body weight was 4.2 (range, 2.2–17.7) kg at operation. Eleven (61%) patients were on a mechanical ventilator prior to surgery. The median stenotic segment estimated by a computed tomography scan was 52% of the length of total trachea (range, 18–84%). Five (28%) patients had proximal bronchial stenosis, 3 (17%) had tracheal bronchus, 2 (11%) had tracheobronchomalacia and 1 (6%) had agenesis of the right lung. Thirteen (72%) patients had a combined cardiac anomaly, including 8 patients with a pulmonary artery sling. Ten (56%) patients had associated extracardiac anomalies. Slide tracheoplasty was performed on cardiopulmonary bypass in all patients, and cardiac lesions were corrected.

RESULTS: There was no early death. The patient with agenesis of the right lung died of left bronchial stenosis 3 months after the surgery. Two (11%) patients were reoperated on for tracheal restenosis. In the other 15 patients, the median duration of ventilator support was 8 (range, 5–34) days and the median duration of hospitalization was 31 (range, 12–79) days. During the follow-up (median duration of 17 months; range, 2–77 months), 13 (72%) patients were symptom-free and 2 (11%) underwent tracheostomy for tracheomalacia.

CONCLUSIONS: Based on this study, slide tracheoplasty seems to be an effective technique for CTS. However, shortening of the trachea after reconstruction may give rise to recurrent obstruction.

Keywords: Congenital tracheal stenosis • Tracheal surgery • Pulmonary artery/abnormalities

INTRODUCTION

Congenital tracheal stenosis (CTS) is a rare but life-threatening disease in children. The stenotic lesion characterized by complete tracheal rings may occur anywhere and have variable length [1]. The disease is frequently associated with other congenital anomalies, most commonly in the circulatory system, which may disguise the airway problem.

Surgical reconstruction is the only treatment, but still remains challenging mainly because of high morbidity and lack of surgical experience due to low incidence. Several techniques have been introduced to correct long-segment tracheal stenosis [2–6]. Among them, slide tracheoplasty has shown superior clinical results and seems to be effective for complicated lesions such as bronchial stenosis [7], tracheal bronchus [8] and hypoplasia of the unilateral lung [9]. This technique was adopted in our institute in 2004.

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The aim of this study was to evaluate our clinical outcomes after slide tracheoplasty for CTS in infants and children.

PATIENTS AND METHODS

Study population

We retrospectively reviewed patients’ characteristics, operative data and clinical course through the medical record. Patients were identified using the database of our centre. Our Institutional Review Board approved this study and waived the need for individual consent.

During the study period of January 2004–December 2011, 20 patients with CTS underwent surgery at our institute. Tracheal resection and end-to-end anastomosis were performed in 2 patients. Eighteen patients who underwent slide tracheoplasty were enrolled in this study. The patients included 11 boys and 7 girls. All patients showed respiratory symptoms and 11 (61%)...
required preoperative ventilator support. The median age was 2.5 months (range, 18 days–4 years) and the median body weight was 4.2 (range, 2.2–17.7) kg at operation. To evaluate the degree and length of the tracheal stenosis, computed tomography (CT) with three-dimensional reconstruction was performed in all patients and bronchoscopy was performed in 3 patients. All patients showed diffuse tracheal stenosis with a complete tracheal ring involving 18–84% (median, 52%) of the tracheal length.

Five (28%) patients had proximal bronchial stenosis, 3 (17%) had tracheal bronchus, 2 (12%) had tracheobronchomalacia and 1 (6%) had agenesis of the right lung. Preoperative cardiac echocardiography was performed to assess combined cardiovascular anomalies. Thirteen (72%) patients had various cardiovascular anomalies, including 8 (44%) patients with a pulmonary artery (PA) sling. Six patients had undergone prior operations, including 1 patient who underwent tracheoplasty with an anterior pericardial patch at another institute. The patient characteristics are summarized in Table 1.

### Operative techniques

All procedures were performed on cardiopulmonary bypass. Single atrial cannulation was used in patients with isolated tracheal stenosis. Combined cardiovascular defects were corrected during the same operation with bicaval cannulation and moderate hypothermia. Our technique has been described previously [10] and is similar to that of Tsang et al. [11]. After mobilization, the trachea was transected at the mid-portion of the stenotic segment. The superior trachea was incised posteriorly, the inferior trachea was incised anteriorly through the entire stenotic portion and the corners of both ends of the resected trachea were trimmed. The two components were slid together and anastomosed with multiple interrupted 7-0 polydioxanone sutures, avoiding suture materials in the mucosa. The endotracheal tube was positioned in the middle of the newly reconstructed trachea. When combined with the PA sling, we routinely divided the left PA and implanted it to the main PA before slide tracheoplasty.

In patients with combined bronchial stenosis, the lower incision was extended onto the stenotic bronchus and the tongue of superior trachea was overlapped on the incised bronchus [7]. In 2 of 3 patients with tracheal bronchus, we used a technique (Fig. 1A) similar to that described by Le Bret et al. [8] Recently, we performed a lateral slide tracheoplasty (Fig. 1B) in the third patient.

### Postoperative care

For the first 3 patients, we used a guardian chin stitch with a neck brace to maintain neck flexion and sedated patients with muscle relaxants. We then performed a tracheotomy on the following day.
relaxants for 7 days postoperatively. However, as the accumulation of our experience convinced us of the safety of shortening the postoperative sedation period, it has now become our routine to sedate patients for only 48 h postoperatively when the ventilation is satisfactory. Furthermore, chin stitch is no longer used and postoperative bronchoscopy is not routinely performed unless patients show relevant symptoms or signs related to the airway.

RESULTS

There was no early death. One patient died 3 months after tracheoplasty due to left bronchial obstruction. This patient had been on a ventilator preoperatively and was a preterm baby with agenesis of the right lung and a large ventricular septal defect (VSD). Closure of VSD and tracheoplasty were performed through a right thoracotomy 1 month after birth, when her body weight was 2.4 kg. She suffered from recurrent obstruction of the left main bronchus, which was suspected to be compressed between the aorta and the vertebrae (Fig. 2).

Reoperation was required in 2 patients. One patient with a PA sling and Down syndrome showed stenosis at the proximal anastomosis site and underwent resection and end-to-end anastomosis. The other patient who had an extensive tracheal stenosis and a sling of the left upper PA underwent reoperation using an autologous rib cartilage and tracheostomy for tracheomalacia.

All other patients were weaned off from a ventilator at a median of 8 (range, 5–34) days and were discharged at a median of 31 (range, 12–79) days after surgery. The median follow-up period was 17 (range, 2–77) months. Sixteen patients were discharged. Among these patients, 13 (13/18, 72%) patients were asymptomatic at the latest follow-up. Two patients had mild dyspnoea during exercise and 1 patient underwent tracheostomy for subglottic stenosis. No patient needed an additional tracheal stent insertion or reoperation for tracheal stenosis after discharge. These data and postoperative respiratory complications are summarized in Table 2.

DISCUSSION

Frequently, CTS is characterized by complete tracheal rings, which presents with various symptoms depending on the grade of stenosis. Long-segment CTS often presents as a life-threatening emergency within a few weeks of birth. CTS may be associated with a PA sling, other cardiac anomalies and gastrointestinal abnormalities. These complicated features and the low incidence of the disease make it difficult to diagnose and manage. Surgery is the only option to properly treat this disease. Except for the results at a few centres with significant experience, overall mortality of CTS after surgical intervention has remained high (28%) until recently. The highest mortality rate is observed in patients younger than 1 month (73%) and in those with intracardiac anomalies (53%) [12].
Many surgical techniques for tracheoplasty have been proposed. However, tracheal augmentation using a patch such as autologous pericardium [3], rib cartilage [4], tracheal autograft [5] and homograft [6] has shown unsatisfactory results mainly because of recurrent granulation formation. Since Tsang et al. [11] reported the first slide tracheoplasty, this technique has shown superior results and is considered as the standard approach in many centres [13, 14]. We adopted this technique in 2004 and have reported promising outcome compared with patch tracheoplasty [10].

The advantages of slide tracheoplasty include (i) reconstructing the trachea with tracheal tissue, which provides a stable cartilaginous wall with normal epithelial lining; (ii) potential of tracheal growth [15, 16] and (iii) the flexibility of use for complicated lesion such as tracheal bronchus, unilateral hypoplasia or agenesis of the lung [8, 9]. However, our experience has also highlighted a problem that is inherent in this procedure: shortening of the trachea. In spite of doubling the diameter, the length of the involved trachea should be reduced by half. It would be most troublesome in such patients with agenesis of the right lung or extensively long-segment stenosis as shown in our series. In unilateral lung agenesis, mortality and risk of vascular compression are known to be higher with right lung agenesis [17]. The only patient who died in our series had agenesis of the right lung, VSD and tracheal stenosis (Fig. 2A). The 2.4-kg baby was weaned off from a ventilator after VSD closure and slide tracheoplasty through a right thoracotomy, but developed tachypnoea and desaturation a month later. We found a newly developed obstruction at the distal end of the trachea and the proximal part of the left main bronchus. We performed aortopexy to the anterior chest and insertion of a tissue expander to the right thoracic cavity to relieve the rotation of the heart and the tension on the aorta. The obstruction improved after the procedure, and the patient could be extubated and transferred to the general ward (Fig. 2B and C). But afterwards, the obstruction eventually progressed (Fig. 2D). The possible mechanism of the recurrent obstruction in the patient is suggested in Fig. 3. Another patient had tracheal stenosis involving the entire thoracic trachea and a sling of the right upper PA. After the slide tracheoplasty and reimplantation of the right upper PA at the age of 2.5 months, tracheomalacia and granulation tissue developed, which might be because of excessive tension on the anastomosis. We performed another anterior tracheoplasty using an autologous rib cartilage. During follow-up, several bronchoscopic procedures to remove granulation tissues were required, twice under support with veno-venous extracorporeal membrane oxygenation; the patient still had residual obstruction and tracheostomy. Laryngeal release through another collar incision might be helpful in this case.
patient to make a tension-free anastomosis, although many surgeons, ourselves included, believe that adequate mobilization of the trachea could be achieved by a sternotomy in such small infants as in our study (median age of 2.5 months) [18, 19].

Correct diagnosis and postoperative care are important for successful outcomes of tracheal reconstruction surgery, especially in infants and small children. Recent imaging techniques such as chest CT with three-dimensional reconstruction allow the noninvasive assessment of CTS pre- and postoperatively. In the first 3 patients of our series and in our earlier experiences using different tracheoplasty techniques, we routinely performed bronchoscopy for the assessment of CTS. However, it was difficult to evaluate the tracheal lumen beyond the stenotic lesion before surgery, and sometimes the procedure was risky in patients with severe respiratory distress. Recently, we have not performed bronchoscopy as a routine procedure if a patient follows an unusual course before and after surgery. Notwithstanding that, bronchoscopy would be the best option to diagnose and treat patients with complications, such as granulation formation, malacia and restenosis. Successful management of CTS depends on an attentive, multidisciplinary team approach, especially when complicated by these situations. This concept has already been emphasized by other authors [20].

In conclusion, slide tracheoplasty seems to be an effective and safe technique. However, the treatment of CTS is still challenging, especially in patients with complicated lesions. Careful surgical planning is needed because shortening of the trachea after reconstruction may give rise to recurrent obstruction in patients with extensively long-segment CTS or unilateral agenesis of the lung. Attentive postoperative management and multidisciplinary team approach are essential for optimal results.

**Conflict of interest:** none declared.
REFERENCES


APPENDIX. CONFERENCE DISCUSSION

Dr E. Le Bret (Le Plessis Robinson, France): The patient characteristics in your study are quite similar to what we have in Marie Lannelongue. I think it reflects real life. Each patient with tracheal rings is very different. You can have a pulmonary artery sling, one or two lungs, you can have tracheal bronchus, funnel shape with hypoplasia of the main bronchus, severe tracheomalacia, and even cardiac anomalies which most often need to be treated at the same time.

The median age in your series is 2.5 months, which is very young, and 11 out of 18 were preoperatively ventilated. That shows that your population had very severe abnormalities. Nevertheless your results are very good with only one death and two reoperations occurring in two patients with pulmonary artery sling.

I have two questions about the pulmonary artery sling. First of all, which technique did you use? What do you do with the pulmonary artery sling, do you reimplant the pulmonary artery?

Dr Chung: That’s right. We usually correct pulmonary artery sling with the reimplantation technique.

Dr Le Bret: And the second thing about pulmonary artery sling is that in our team we also have worse results with tracheal hypoplasia with pulmonary artery sling. Do you have an explanation for this? Do you think when we have a pulmonary artery sling and we have to dissect the posterior part of the trachea as well as dissecting the anterior part to do the slide tracheoplasty, we impair the vascular blood supply?

Dr Chung: I’m not quite understanding what you’re saying.

Dr Le Bret: Let’s try another question. Regarding the possible tracheobronchial compression due to the bronchial mobilization, we never had any. But to be honest, I don’t understand why. Do you release the larynx when you do the slide tracheoplasty?

Dr Chung: In our institution, we did routinely perform procedures of hilar release, neck flexion, and hyoid release to minimize tracheal tension during surgery. However when slide tracheoplasty is performed in severe long-segment tracheal stenosis, we also agree that division of the suprahyoid muscles should be performed for sufficient laryngeal release as you mentioned.

Dr R. Cesnjever [Erlangen, Germany]: In my experience, granulations are very common in these patients. What is your treatment concept for these granulations? Do you balloon them? Do you remove them via bronchoscopy? Or do you sometimes even stent them?

Dr Chung: If we have a granulation at the anastomotic site, we sometimes perform balloon dilatations and granulation removal. We showed improvement with granulation removal using the bronchoscope under V-V ECMO support if needed. But in our last case the baby was too small to insert a stent, so we just repeated granulation removal.