Simulated slide tracheoplasty for congenital tracheal stenosis using three-dimensional printed models

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Abstract
Purpose The slide tracheoplasty (STP) is the standard treatment for severe congenital tracheal stenosis (CTS). Understanding the features of the tracheal stenosis in each case and choosing an appropriate incision design are very important for successfully executing the procedure. The present study aimed to evaluate the advantages of three-dimensional (3D) printed models of the trachea for improving CTS.

Methods Three-D tracheal models were created using computed tomography (CT) data from ten patients undergoing STP for CTS. Simulated surgery was performed using the hollow models after reinforcing with them with a coating of gum spray. Clinical outcomes, including patient survival, postoperative surgical interventions, and time required for STP, were compared with the corresponding values in the last ten patients before the introduction of 3D model simulations.

Results All ten patients for whom simulated surgery using a 3D tracheal model were conducted achieved good airway patency after their STP. The surgeons reported feeling that the 3D model simulations were highly effective although there was no significant difference in the clinical outcomes of the groups with or without simulated STP. The models were useful not only for surgical planning but also for sharing important information among the multidisciplinary team and the patients’ family.

Conclusion Our experience using 3D tracheal models demonstrated several features enabling improvement in the surgical treatment of CTS.

Keywords Congenital tracheal stenosis · Slide tracheoplasty · Simulation surgery · Three-dimensional printing model

Introduction
Congenital tracheal stenosis (CTS) is a rare and often life-threatening, anatomical anomaly characterized by complete tracheal rings. The slide tracheoplasty (STP) is currently the gold standard surgical treatment for severe CTS. In STP, the stenotic segment is divided at the midpoint, and the opposite anterior and posterior surfaces of the proximal and distal stenotic segments are incised vertically so that they slide together [1]. Designing the exact dividing point, route, and extent of the incisions is crucial for a successful outcome.

We herein described the utility of a three-dimensional (3D) trachea model for STP which demonstrated utility in improving surgery for severe CTS. Although several, previous reports have described the application of a 3D tracheal model to CTS [2–5], to the best of our knowledge the present study enrolled the largest number of patients with a STP using 3D simulation.

Methods
Patients
Twenty consecutive patients undergoing STP for CTS between August 2018 and February 2022 at Tokyo Metropolitan Children’s Medical Center were included. The patients were divided evenly into two groups with or without 3D simulation. The former group underwent STP
without a 3D simulation, and the latter group underwent a pre-operative STP simulation. Clinical outcomes, including patient survival, postoperative surgical interventions, and time required for STP, were compared between the groups.

The ethics committee of Tokyo Metropolitan Children’s Medical Center approved this study (2020b-70).

**Diagnosis, surgical indications, and management of slide tracheoplasty**

CTS was diagnosed using enhanced computed tomography (CT) and bronchoscopy. Echocardiography was also employed to assess cardiovascular abnormalities. The degree of stenosis was calculated using the formula, narrowest diameter/normal diameter (%), and was graded severe (<40%), moderate (40% ~ 60%) or mild (>60%). Surgery was indicated for moderate and severe stenosis with clinical symptoms.

STP was performed via a median sternotomy with ECMO or cardiopulmonary bypass (CPB). After a cardiovascular surgeon established ECMO or CPB, any cardiovascular anomaly was corrected if necessary. Then, a pediatric surgeon performed the tracheoplasty as previously reported [6]. Briefly, after exposing the anterior wall of the entire trachea, the stenotic segment was circumferentially transected precisely at a ratio of 3:2, with the larger and smaller number representing the proximal and distal segment, respectively. The proximal segment was incised longitudinally and posteriorly, then the distal segment was incised anteriorly. If the stenosis extended to an area near the carina, the distal incision was extended into the right and left main bronchus using an inverted Y-shaped incision as previously reported [7]. The two, tracheal components were then placed together and anastomosed.

Perioperative patient management was carried out using a multidisciplinary, teamwork approach. Patients were sedated for the first three to four days and given muscle relaxants to keep their neck in a flexed position. They were then given sedatives alone for an additional five to seven days.

**3D printing model**

Three-dimensional data on the patients’ trachea were extracted from chest CT data and converted to the Standard Triangulated Language (STL) format for 3D printing. The models were printed by stereolithography (Form 2, Form-labs). Three different materials were used: clear resin (hard), elastic resin (soft), and flexible resin (medium). To create hollow, tubular structures, the STL data were imported into Mimics® inPrint (Materialise). Ten to 30 min were needed to convert the CT data for printing, and about 3–7 h were required for printing.

**Simulated surgery**

The hollow, 3D tracheal models in elastic and flexible resin were created life-sized for use in simulated surgery. The models were fixed by adhesive tape to the stage, and a slide tracheoplasty was performed as in actual surgery (Fig. 1). Sawing and tying surgical thread lacerated the wall of the models. However, applying a coating of gum spray was able to prevent this problem.
| Group                          | Case | Age at STP, month | Weight at STP, kg | Sex | Trachea, minimum diameter, mm | Trachea, stenosis length, mm | PAS Cardiovascular anomaly | Trachea, abnormal arborization | Other complications | Time required for tracheal anastomosis, min | Time required for tracheal anastomosis per stenotic length, min/mm | Postoperative surgical intervention |
|-------------------------------|------|-------------------|-------------------|-----|-------------------------------|-------------------------------|-----------------------------|-------------------------------|--------------------------|--------------------------------|----------------------------------------|----------------------------------|
| With simulation              | 1    | 1                 | 7.1               | F   | 2.5                          | 20                           | -                          | +                            | Branchial arch syndrome, ear deformity, micrognathia | 135                          | 5.2                          | No                                  |
|                              | 2    | 3                 | 6.1               | M   | 2.5                          | 44                           | + VSD, PAS                  | -                            | Duodenal atresia, radial deformity, vertebral deformity | 127                          | 3.5                          | No                                  |
|                              | 3    | 2                 | 4.0               | M   | 3.0                          | 26                           | + VSD, PAS                  | -                            | HIE                      | 113                          | 4.3                          | No                                  |
|                              | 4    | 20                | 11                | F   | 3.5                          | 36                           | -                          | -                            | Trisomy 21, TAM             | 143                          | 3.4                          | No                                  |
|                              | 5    | 15                | 10                | M   | 3.8                          | 50                           | -                          | -                            | Right lung hypoplasia,                  | 60                           | 6.7                          | No                                  |
|                              | 6    | 8                 | 5.5               | M   | 4.0                          | 26                           | - cAVSD                     | -                            | Left lung agenesis                      | 195                          | 3.8                          | No                                  |
|                              | 7    | 51                | 12                | F   | 3.8                          | 42                           | + ASD, PAS                  | -                            | Trisomy 21                  | 155                          | 3.5                          | No                                  |
|                              | 8    | 16                | 7.4               | M   | 3.3                          | 9                            | - ASD                       | -                            |                          | 163                          | 4.8                          | No                                  |
|                              | 9    | 5                 | 5.6               | F   | 3.0                          | 34                           | + ASD, PAS                  | -                            |                          | 195                          | 3.8                          | No                                  |
|                              | 10   | 17                | 9.3               | M   | 4                            | 51                           | -                          | -                            | Trisomy 21                  | 135                          | Mean 4.3                      | No                                  |
| Median                       | 11   | 5                 | 3.4               | F   | 3.0                          | 20                           | - VSD                      | -                            | Trisomy 21                  | 63                           | 3.2                          | No                                  |
| Without simulation           | 12   | 1                 | 2.1               | F   | 2.0                          | 44                           | + PAS                      | -                            | Tracheal bronchus, pharyngeal stenosis | 140                          | 3.2                          | No                                  |
|                              | 13   | 23                | 8.7               | M   | 2.0                          | 18                           | - CoA, ASD, VSD, PDA        | +                            |                          | 108                          | 6.0                          | No                                  |
|                              | 14   | 2                 | 2.7               | M   | 3.0                          | 26                           | - TOF                      | -                            | Left lung agenesis                      | 89                           | 3.4                          | No                                  |
| Group | Case | Age at STP, month | Weight at STP, kg | Sex | Trachea, minimum diameter, mm | Trachea, stenosis length, mm | PAS Cardiovascular anomaly | Trachea, abnormal arborization | Other complications | Time required for tracheal anastomosis, min | Time required for tracheal anastomosis per stenotic length, min/mm | Postoperative surgical intervention |
|-------|------|------------------|------------------|-----|-----------------------------|-----------------------------|---------------------------|-----------------------------|---------------------------|---------------------------------|---------------------------------|--------------------------------------|
| 15    | 6    | 6.7              | F                | 1.7 | 26                          | +                           | ASD, PAS                  | +                           | Right lung hypoplasia, tracheal bronchus, anal atresia, horseshoe kidney, left renal hypoplasia, vertebral deformity | 130                             | 5.0                             | No                                  |
| 16    | 5    | 2.7              | M                | 4.0 | 24                          | –                           | ASD, PAS                  | –                           | Trisomy 21, laryngomalacia                                | 120                             | 5.0                             | Brachiocephalic artery ligation, tracheostomy |
| 17    | 15   | 8.6              | F                | 2.0 | 30                          | +                           | PAS                       | –                           | Thoracic deformity, multiple arthrogryposis               | 114                             | 3.8                             | No                                  |
| 18    | 5    | 4.3              | M                | 2.0 | 38                          | –                           | –                         | –                           | Thoracic deformity, multiple arthrogryposis               | 154                             | 4.1                             | No                                  |
| 19    | 6    | 8.2              | M                | 3.5 | 35                          | +                           | PAS                       | –                           | Thoracic deformity, multiple arthrogryposis               | 121                             | 3.5                             | No                                  |
| 20    | 16   | 11               | M                | 2.3 | 25                          | –                           | +                         | Tracheal bronchus                              | 103                             | 4.1                             | No                                  |
| Median|      | 5.5              | 5.5              | 2.1 | 26                          |                             |                           |                             |                                            | Mean 120                         | Mean 4.1                       |                                     |

STP slide tracheoplasty, PAS pulmonary artery sling, VSD ventricular septal defect, CoA coarctation of the aorta, ASD atrial septal defect, PDA patent ductus arteriosus, TOF tetralogy of Fallot, cAVSD complete atrioventricular septal defect, HIE hypoxic-ischemic encephalopathy, TAM transient abnormal myelopoiesis
Results

Patient characteristics and demographic data

Table 1 shows the patient demographics and general characteristics. There were no deaths in the entire cohort. In terms of the postoperative surgical interventions, one patient in each group had a tracheostomy for hypoxic-ischemic encephalopathy and laryngomalacia with vocal cord palsy, respectively. One patient in the group without simulation required a ligation of the brachiocephalic artery on postoperative day 9 for a fistula between the trachea and the brachiocephalic artery.

Case presentation

We performed a simulated slide tracheoplasty in ten patients. Below are two characteristic instances. The first case was that of a patient whose tracheal bronchus required more complicated dissection at the proximal trachea because the tracheal bronchus arose from the right posterior wall of the trachea. The second case was that of a patient with a left pulmonary artery sling. In such cases, not only the structure of the trachea but also the possibility of friction between the transplanted pulmonary artery and main bronchi need to be considered. Using two, different 3D models of the trachea and blood vessels facilitated the simulation.
Case 1

An 8-month-old, female infant with no cardiovascular anomaly had abnormal tracheal arborization in which the tracheal bronchus communicated with the azygos lobe (Fig. 2). Usually, we incise the stenotic upper segment posteriorly and the lower segment anteriorly, but in the present case the tracheal bronchus crossed the midline of the posterior wall. We decided therefore to make the incision in the upper segment anteriorly and the incision in the lower segment...
posteriorly (Fig. 3), a pattern that previously worked well in a simulation. The STP was performed as in the simulation, and the postoperative course was uneventful. Figure 4 shows the 3D tracheal models before and after the simulation surgery. The shape of the model after the simulation was quite similar to that of a model constructed on the basis of postoperative CT data, suggesting that the method used was both reliable and feasible.

Case 2

A 3-month-old, male infant with long-segment CTS had the unusual anatomical feature of two left pulmonary arteries. The dominant branch showed normal branching, but the other branch originated in the right pulmonary artery, running behind the trachea in the manner of a pulmonary artery sling (Fig. 5A) and requiring transplantation surgery. We made a 3D tracheal model with clear resin and modeled the vessels using flexible resin (Fig. 5B), which allowed us to simulate the effect of a tracheoplasty on the surrounding blood vessels. As the stenotic segment was long (44 mm), there was a possibility of the lower trachea and bilateral main bronchus migrating upwards after the STP and causing friction between the blood vessels and main bronchi. STP with laryngeal release and left pulmonary artery transplantation with an appropriate location for anastomosis was successfully performed. The postoperative course was excellent.

Discussion

In the present study, we performed preoperative simulations of STP in ten patients. Clinical outcomes, including patient survival, postoperative surgical interventions, and operative time for tracheal anastomosis, did not differ significantly between the groups. However, we found that a 3D tracheal model had several, useful features enabling improved CTS treatment.

First, it allowed the trachea to be examined carefully from multiple viewpoints. The 3D model enabled the size, shape, undulation, and direction or angle of the branching to be readily grasped. In an actual STP, surgeons can observe only the anterior surface of the trachea; thus, having a 3D model prior to the actual surgery is extremely helpful in anticipating difficulties caused by peculiarities of the patient’s tracheal anatomy. Several, previous studies reported a similar approach to surgical planning of STP for CTS using solid, 3D tracheal models in a small number of subjects [2–4].

Second, the 3D model enables simulation prior to the actual surgery. In patients with a complicated tracheal structure as in case 1, we used a 3D model to perform several simulations testing various patterns of dividing and incising the trachea. Based on a comparison of the results, we determined the final design for the actual surgery. Thus, the 3D surgical simulations are very helpful for testing different types of surgical procedure. Richardson et al. also recently reported a simulated STP using a 3D model and compared the postoperative anatomic outcomes in three, different, tracheal transection designs [5]. In the multidisciplinary team, repeated simulations by the surgeon, surgical assistants, and scrub nurses may also be helpful for reminding us of details of the procedure so that it can be performed smoothly with minimum difficulty during an actual operation.

Third, the 3D model is useful for sharing important information with physicians besides surgeons, nurses, and the patients’ family. STP for CTS is highly demanding and requires multidisciplinary teamwork. For ICU physicians and anesthesiologists, choosing an appropriate endotracheal tube, then placing it correctly, are important for avoiding unnecessary granulation or unexpected extubation. For the patients’ family, the 3D model is helpful for understanding the nature of the patients’ condition and the complicated surgical technique involved.
Fourth, the 3D model is an excellent educational tool for trainees and medical students. Simulated surgery provides a good opportunity for trainees to exercise their surgical skills before attempting surgery on a patient. The present study has some limitations. First, the 3D tracheal data derived from CT data and were imported into editing software to create the hollow, tubular structure of the trachea with a 1-mm thickness. The thickness and firmness of the tracheal wall may have differed by location, especially in the area near the bifurcation, one of the most difficult but important areas during STP.

Second, the models must be durable enough to last through the simulation. Gum spray was extremely helpful in preventing lacerations of the models, but an alternative method of reinforcing the artificial trachea against sawing and tying damage is desirable. Recently, Hashimoto et al. described a sophisticated, 3D tracheal model for adult tracheobronchial surgery [8] in which the damage caused by sawing and tying the surgical thread during simulations was prevented by using various types of soft material to mimic the different parts of the trachea, such as the cartilage, connecting tissue, and membranous wall, thus also providing some indication as to sophistication possible in 3D models.

Third, creating a 3D model is costly and time-consuming. Three-D printing machine (Form2, Formlab) costs about 500,000 JPY, and a software subscription needed for hollow model production (Mimics® inPrint, Materialise) costs about 800,000 JPY per year. The running costs for materials like resin and gum spray are about 1,000 JPY per model. Previous reports have extensively described the costs involved [2]. Improvements in the current technology to enable the creation of better quality, more cost-effective models are highly desirable.

In conclusion, our experiences of using 3D trachea models for STP demonstrated several features that were useful for improving the treatment of patients with CTS. The findings should prove helpful not only to the surgeon, but also enable the multidisciplinary treatment team and the patients’ family in better understanding the particulars of the patients’ case. The models may also be used to good effect for educational purposes.

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Author contributions N.S. wrote the main manuscript text. N.S., A.S., H.T. and S.H. performed simulation using models as operating surgeons. All authors reviewed the manuscript.

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Declarations
Conflict of interest The authors declare that they have no conflicts of interest.

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