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Management of congenital tracheal stenosis☆

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Abstract

Objectives: Congenital tracheal stenosis is a rare disease. Various methods for treatment exist but there is still much debate as to the appropriate surgical procedure. We present our surgical experiences of patch tracheoplasty and slide tracheoplasty as viable methods for the treatment of congenital tracheal stenosis. **Methods**: From 1994 to 2002, 13 patients were diagnosed with congenital tracheal stenosis. Eight patients (7 symptomatic and 1 asymptomatic) had their stenosis corrected, three by means of pericardial patch tracheoplasty, four by slide tracheoplasty, and one by resection and anastomosis. Concomitant operations were performed on six patients to treat congenital cardiovascular disease. Five patients showing no significant symptoms did not undergo tracheal surgery and received only cardiac procedures. A retrospective review of the hospital course, complications, and long-term results was conducted. **Results**: Among the patch tracheoplasty group, every patient suffered from granulation tissue formation. One patient died of respiratory acidosis and one was hospitalized due to recurrent granulation tissue, which required frequent bronchoscopy. The third patient from this group is free of all symptoms. Among the slide tracheoplasty group, one patient died of anastomosis disruption. The three remaining patients are alive and well. The one patient who received resection and anastomosis is alive without symptoms. **Conclusions**: Surgical repair of long-segment congenital tracheal stenosis exhibited high mortality and morbidity rates. Every patient that underwent pericardial patch tracheoplasty suffered from troublesome granulation tissue. As slide tracheoplasty provided relatively good results in the short and mid-term follow-up periods, it seems to be a preferred method for the treatment of long-segment congenital tracheal stenosis.

Keywords: Trachea; Congenital anomaly; Stenosis

1. Introduction

Congenital tracheal stenosis is a rare disease that has no established surgical procedure, due to insufficient experience and the lack of a large-scale study [1]. Since patients may be symptomatic immediately after birth or develop an accompanying complex cardiac anomaly, surgical repair can be technically demanding. In the case of short-segment stenosis, successful repair has been reported with segmental resection and end-to-end anastomosis [2–4]. However, resection of more than 30% of the tracheal length may result in excessive anastomotic tension that can cause life-threatening complications [5,6]. Although a variety of surgical methods, including rib cartilage augmentation [7,8], pericardial patch augmentation [9–12], and slide tracheoplasty [5,13,14], have been described for the treatment of long-segment stenosis, there seems to be no established surgical technique. We present our surgical experience of patch tracheoplasty and slide tracheoplasty for the treatment of long-segment congenital tracheal stenosis.

2. Materials and methods

Between April 1992 and August 2002, 13 consecutive patients were referred to Seoul National University Hospital

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diagnosed with congenital tracheal stenosis. Five patients with no significant symptoms did not undergo tracheal surgery. They were either treated with conservative management, such as temporary tracheostomy, or were left untreated, as the patients could be weaned from the ventilator after successful correction of complex cardiac anomalies.

Of the remaining eight patients, six were boys and two were girls. Their age at the time of repair ranged from 12 days to 6 months (median 3.2 months) and body weight ranged from 2.6 to 6.6 kg (median 5.1 kg). Seven patients (87.5%) suffered from respiratory distress and six of them (75%) required mechanical ventilation from 3 to 52 days preoperatively. Among them, two patients failed to wean off from the ventilator due to undetected tracheal stenosis after open-heart surgery. The tracheal stenosis was clinically suspected due to respiratory distress or stridor and was diagnosed by a 3D reconstructed computed tomography. Bronchoscopic examination was performed in only one patient. In the remaining patients, it was not feasible to examine the airway preoperatively because of severe respiratory distress. Six patients (75%) were associated with combined cardiac or vascular anomalies, including pulmonary artery sling, aortopulmonary window, coarctation of the aorta, ventricular septal defect, atrial septal defect and patent ductus (Table 1). Six patients (75%) were also associated with bronchopulmonary abnormalities, including tracheal origin of the right upper lobe bronchus (3), stenosis of both main bronchus (2), and hypoplasia of the right lung (1).

The degree of tracheal stenosis was measured preoperatively by a 3D reconstructed computed tomography and intra-operatively by direct vision (Fig. 1). The length of stenosis ranged from 7 to 40 mm (median 32 mm). The luminal diameter at the narrowest site ranged from 2 to 7 mm (median 2 mm).

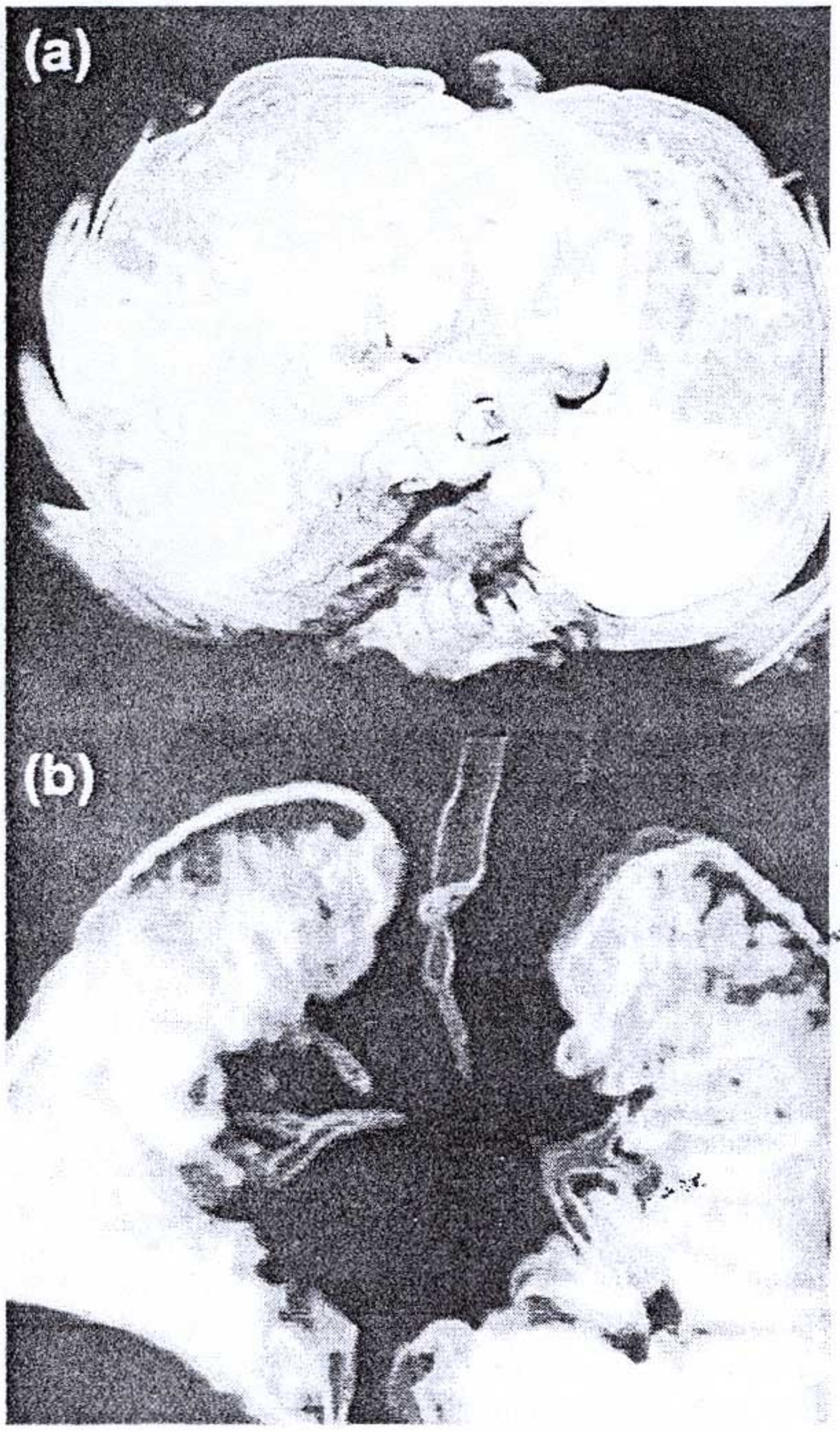

Fig. 1. Preoperative 3D computed tomography of a 6-month-old baby whose trachea showed long-segment stenosis as well as narrowing of both main bronchi. The left pulmonary artery arises from right pulmonary artery and passes behind the trachea to the left lung.

Table 1
Summary of the associated cardiovascular anomalies

Patient No.	Tracheal procedure	Cardiovascular anomalies	Tracheal stenosis
1	Slide	AP window, PDA	Lower 1/3
2	Patch	CoA, VSD, PDA	Whole trachea
3	Patch	Pulmonary artery sling	Lower 1/3
4	Patch	ASD, absent RPA	Lower 1/3
5	R/A	None	Mid trachea/two rings
6	Slide	None	Lower 2/3
7	Slide	Pulmonary artery sling	Lower 2/3
8	Slide	VSD, ASD, PDA	Lower 2/3

R/A, resection and end-to-end anastomosis; AP window, aortopulmonary window; CoA, coarctation of Aorta; VSD, ventricular septal defect; PDA, patent ductus arteriosus; ASD, atrial septal defect.

2.1. Surgical technique

All operations were performed via median sternotomy with the use of cardiopulmonary bypass (CPB) except in the case of one patient, who was approached through a low collar incision and partial upper sternotomy without using CPB. Seven patients with long-segment stenosis underwent repair, three by means of pericardial patch tracheoplasty and four by slide tracheoplasty. The patient with short-segment stenosis was repaired by resection and end-to-end anastomosis. In the six patients who had additional cardiac anomalies, four patients were treated concomitantly. For two patients in whom tracheal stenosis had not been detected initially, tracheal surgery was performed in a staged manner.

After the induction of general anesthesia, standard median sternotomy was performed. The thymus was either excised or divided between lobes and retracted laterally, and the innominate vessels were mobilized. After the pericardium was opened, the trachea was further exposed between the aorta and the superior vena cava. During the dissection, meticulous care was taken so as not to interfere with the lateral blood supply of the trachea and to preserve the recurrent laryngeal nerves. Patients were given heparin and they were cannulated for cardiopulmonary bypass via the right atrium and ascending aorta. After the institution of the CPB, the endotracheal tube was removed, and then either rigid or fiberoptic bronchoscopy was performed to confirm the extent and degree of stenosis.

In the case of pericardial patch tracheoplasty, a rectangular patch of autologous pericardium was harvested. A longitudinal incision on the anterior surface of the trachea was placed through the entire stenotic segment. Then, the pericardium was sewn over using 6-0 PDS®, enlarging the trachea. The pericardial patch was suspended to the surrounding mediastinal tissues by tacking several sutures to prevent airway collapse. In cases of slide tracheoplasty, the trachea was divided transversely through the middle of the stenosis, and the procedure then proceeded in a manner described by Grillo et al. [5,13] using 6-0 Vicryl® sutures. The thymic tissue was generally used to interpose between the tracheal suture line and innominate artery to prevent potential erosion of the artery. After the procedure was completed, an anesthesiologist reinserted the endotracheal tube under direct visual guidance. Mechanical ventilation was then initiated and the cardiopulmonary bypass was withdrawn. After heparin reversal, the sternotomy was closed over a mediastinal drain in a standard fashion.

After patch tracheoplasty, we put patients on the ventilator for at least a week to maintain airway stenting. On the other hand, after slide tracheoplasty or resection and anastomosis, we tried to remove the ventilator as soon as possible.

A retrospective review was conducted for evaluating the hospital course, complications, and long-term results.

3. Results

There were two operative deaths, resulting in an early mortality rate of 25% (2/8). A 6-month-old girl with a history of atrial septal defect and absence of right pulmonary artery underwent repair by means of pericardial patch tracheoplasty. The patient died at 29 days postoperatively of respiratory acidosis, resulting from granulation tissue. A 1-month-old boy diagnosed with aortopulmonary window and patent ductus underwent total repair. He failed to wean off from the ventilator due to unexplained respiratory acidosis. A bronchography was performed on the 22nd postoperative day and revealed an undetected tracheal stenosis. He underwent slide tracheoplasty through a low collar incision and partial sternotomy without CPB. Unfortunately, however, the patient died at 4 days postoperatively of anastomosis disruption.

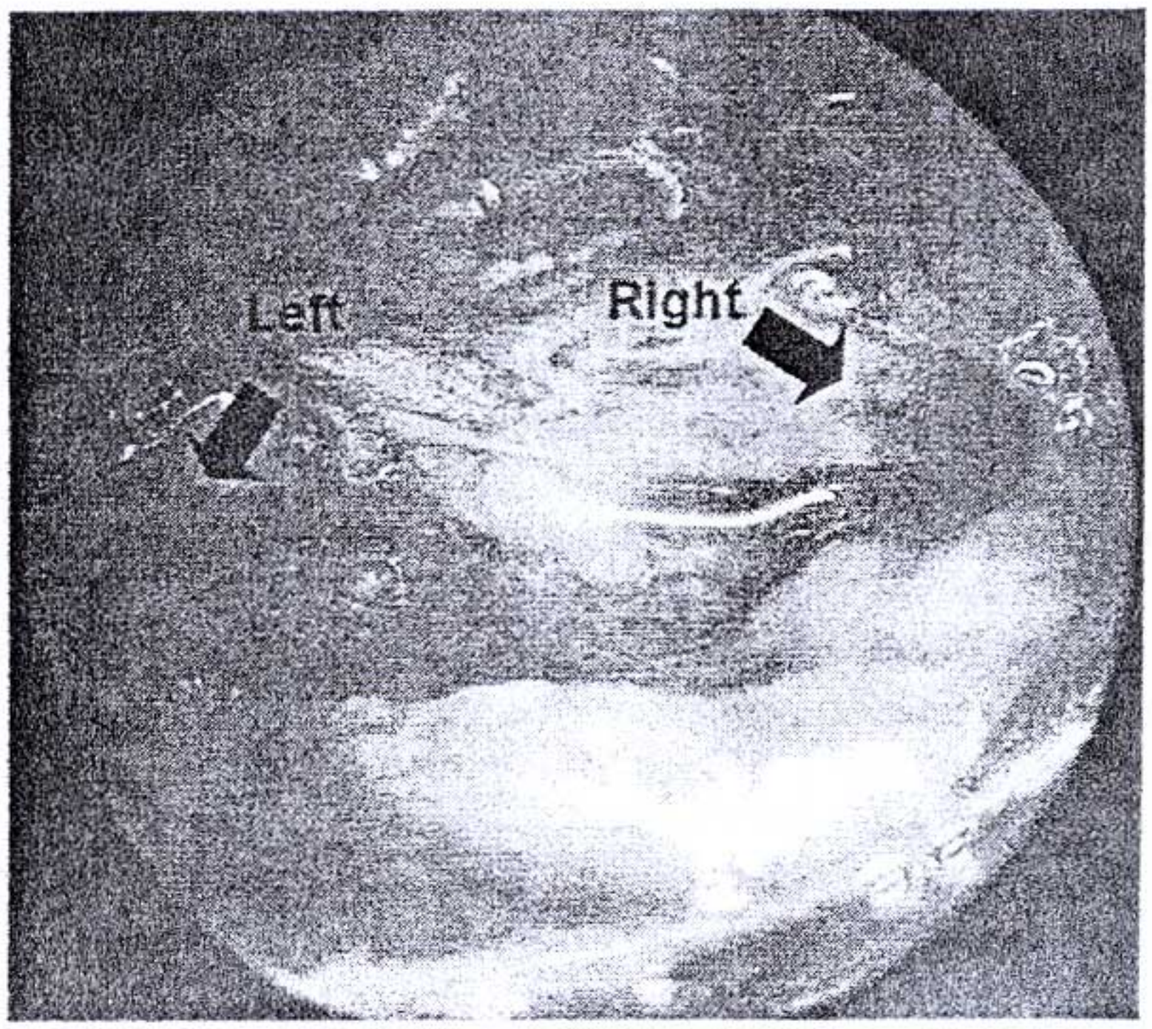

Fig. 2. Bronchoscopic finding of a 4-month-old baby complicated with recurrent overgrowth of granulation tissue after pericardial patch tracheoplasty. Note that both main bronchi are severely narrowed (arrows). Despite repeated rigid bronchoscopy, the patient died of respiratory failure 47 months after tracheal surgery.

There was one case of late death in a 7-month-old boy who underwent pericardial patch tracheoplasty and suffered from granulation tissue formation and bronchomalacia, which required re-operation, stent insertion and repeated bronchoscopy (Fig. 2). He eventually died at 45 months postoperatively of chronic respiratory failure. All the remaining five patients were long-term survivors with a follow-up period ranging from 13 months to 9 years (median 24.4 months). These long-term survivors are now free of symptoms and report no airway problems. All patients with patch tracheoplasty suffered from granulation tissue. Two patients died of respiratory acidosis due to recurrent granulation tissue. The remaining patient survived despite the formation of granulation tissue and he remains alive without complications (Table 2). After patch tracheoplasty,

Table 2
Summary of the outcomes for the eight patients who underwent tracheal surgery

Patient no	Procedure	Complications	Outcome
1	Slide	Disruption of anastomosis	Dead
2	Patch	Granulation tissue	Alive
3	Patch	Granulation tissue, respiratory acidosis	Dead
4	Patch	Granulation tissue, respiratory acidosis	Dead
5	R/A	None	Alive
6	Slide	Chronic lung disease	Alive
7	Slide	None	Alive
8	Slide	Mediastinitis	Alive

R/A, resection and end-to-end anastomosis.

we planned to put patients on the ventilator for at least a week for the purpose of airway stenting. One patient could be weaned off the ventilator at the 12th postoperative day. Two patients could not be weaned off the ventilator as planned because of some troublesome granulation tissue. One of them died of respiratory acidosis caused by granulation tissue on the 29th postoperative day and the other patient was weaned off the ventilator at 7 postoperative months after repeated removal of granulation tissue through the rigid bronchoscopy as well as re-operation. Unfortunately, the patient was re-intubated in a month and eventually died of respiratory failure after 45 months despite repeated bronchoscopic removal of granulation tissue and tracheal stent placement.

Except for one patient who died of anastomosis dehiscence, all the other members of the slide tracheoplasty group survived without suffering from airway problems. None of the patients who received slide tracheoplasty experienced granulation tissue-related complications (Fig. 3). The one patient who received resection and anastomosis is alive without symptoms (Table 2). After slide tracheoplasty or resection and anastomosis, we tried to remove the ventilator as soon as possible. The patient who underwent resection and anastomosis was extubated on the day of operation. The patient who underwent slide tracheoplasty was weaned off the ventilator on the second postoperative day. The patient who died of anastomosis disruption at postoperative day 4 could not be taken off the ventilator. The remaining two patients needed prolonged ventilator care for 1 month, not because of tracheal surgery, but due to emphysematous lung disease and postoperative mediastinitis, respectively.

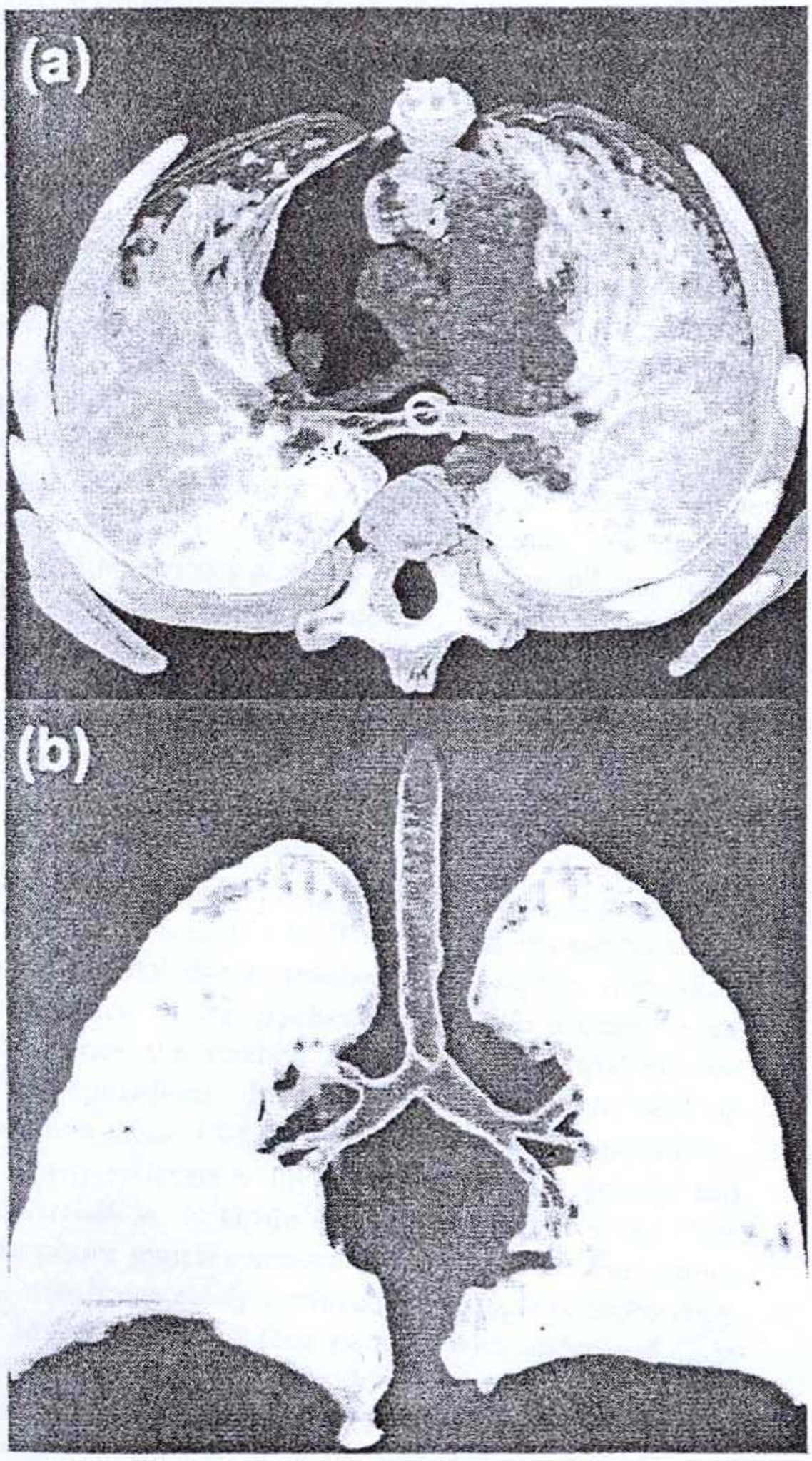

Fig. 3. Postoperative 3D computed tomography of a 6-month-old baby after slide tracheoplasty for long-segment tracheal stenosis.

4. Discussion

Congenital tracheal stenosis can be a life-threatening disease, especially in cases involving the long-segment of the trachea [12]. If the stenosed segment is short, it is best treated with resection and end-to-end anastomosis [2–5]. However, it has been suggested that resection of more than 30% of the tracheal length may lead to excessive anastomotic tension followed by recurrent stenosis or fatal separation [5,6].

When patients developed symptoms early in infancy, surgical repair may be a considerable challenge because of the small size of the airway, which is easily obstructed by postoperative edema [13]. Furthermore, it has been reported that the infant trachea has a lesser tolerance for anastomotic tension than the adult trachea [6]. It has also been suggested that long-segment tracheal stenosis is often associated with other anomalies such as pulmonary vascular sling, intracardiac lesion, and right-sided aortic arch compared to short-segment stenosis [15–17]. For these reasons, the management of long-segment tracheal stenosis in infancy still reports significant morbidity and mortality rates [11]. In our current series, operative deaths occurred in two out of eight patients, resulting in an early mortality rate of 25%.

Over the past 20 years, various surgical techniques have been suggested for the treatment of this disorder. These techniques include rib cartilage tracheoplasty, pericardial patch tracheoplasty, tracheal autograft, and slide tracheoplasty [5,7–14]. Recently, successful results have been reported in several studies [8,11,12,17,18]. Nonetheless, there still remains much debate as to the appropriate surgical procedure due to the rarity of the disease and lack of long-term follow-up data [8,12,13].

The limitation on the trachea length to be resected safely necessitates the introduction of various patch materials for tracheal augmentation. Initially, Kimura and associates [7] reported surgical repair for long-segment stenosis using

costal cartilage in 1982. In 1984, Idriss and colleagues [9] used autologous pericardium as an alternative material to costal cartilage for the same purpose. Jaquiss and associates [8] showed favorable results for rib cartilage tracheoplasty with a low rate of postoperative problems and without operative mortality. Backer and associates [18] described a notable outcome using pericardial patch tracheoplasty, which resulted in a survival rate of 83%. One advantage of cartilage graft includes its rigidity, which allows the avoidance of prolonged postoperative airway splinting by the endotracheal tube in contrast to the pericardial patch technique in which airway splinting is mandatory [8]. However, it is not easy to offer an airtight suture line using this rigid material. In contrast, pericardial patch has an advantage due to its pliability, which allows for an airtight suture line [8]. However, its collapsible feature requires the suspension of the patch to the surrounding mediastinal structures and prolonged airway stenting by prolonged intubation during the early healing periods [9,11]. Although all these advantages are desirable, the most prominent problem of patch techniques is the formation of granulation tissue. Granulation tissue arising from the mesenchymal surface of the patches requires multiple bronchoscopic debridements and other complications, such as necrosis or collapse of the patch, may occur [5,18,19]. Several studies claimed that patients who underwent rib cartilage or pericardial patch tracheoplasty suffered from granulation tissue less frequently than expected [8,12]. In our series, however, all three patients who received pericardial patch tracheoplasty developed troublesome granulation tissue without exception. Among these patients, two died of persistent respiratory acidosis caused by granulation tissue.

To solve this vexing problem, Tsang and associates proposed slide tracheoplasty, as established by Grillo [5,13,14]. In this technique, the stenotic segment is divided transversely at its midpoint, the upper stenotic segment is incised vertically posteriorly, the lower stenotic segment is incised anteriorly, then the two are slid together and sutured. After being repaired, the circumference of the trachea should be doubled, creating a nearly four-fold increase in cross-sectional area [5,13]. The greatest advantage of this technique over patch tracheoplasty is the immediate reconstruction of the trachea with native tracheal tissue [5,13]. Since the trachea is lined with normal ciliated tracheal epithelium, there is little tendency to develop granulation tissue [5,13]. Additionally, routine postoperative airway splinting is unnecessary, allowing prompt and early extubation. In Grillo's series, two of the seven slide tracheoplasty patients showed a single suture line granuloma, which was easily removed by a single bronchoscopy [13]. In our series, all four patients who underwent slide tracheoplasty showed no evidence of granulation tissue. Several authors who favor patch tracheoplasty argued that the actual advantages of slide tracheoplasty should await wider application to a larger series of young patients [8,12]. experience of slide tracheoplasty, only three infants were included in the total of eight cases [5]. In contrast, all our cases of slide tracheoplasty were performed in infancy and resulted in acceptable outcomes. When carefully executed, slide tracheoplasty has not led to the loss of viability of the trachea from interference with the tracheal blood supply and most of the lateral blood supply of the trachea can be preserved [5]. We believe slide tracheoplasty can be safely applied to long-segment tracheal stenosis, particularly in infancy.

For slide tracheoplasty, satisfactory subsequent growth was demonstrated experimentally and clinically [20,21]. Grillo and colleagues [5] showed that the repaired tracheal segment continued to grow in their clinical series. In our series, it is difficult to show that growth of the repaired trachea has been observed in patients with slide tracheoplasty because of relatively short follow-up periods. However, all three patients who survived after slide tracheoplasty underwent follow-up imaging studies, which showed that growth of the trachea could be predictable. A more deliberate study of growth potential will be needed.

There is little agreement on the use of cardiopulmonary bypass. Many surgeons have found it very useful to achieve excellent operative exposure of the trachea [8,12,18,22,23]. Several authors also recommended that cardiopulmonary bypass should be available as a backup, particularly for small infants or patients with complex cardiac anomalies [8]. On the contrary, others have preferred to avoid bypass [7,13, 14,24]. Grillo and associates suggest that the conduct of the operation is simplified by avoiding cardiopulmonary bypass [5,13]. In our series, seven of the eight patients received operation under cardiopulmonary bypass. A patient, in whom we did not use cardiopulmonary bypass, died of anastomosis dehiscence, which may be referred to as a technical failure. For us, cardiopulmonary bypass is a useful tool to facilitate the dissection of the trachea without being disturbed by equipment for distal airway maintenance. We feel that it is not necessary to avoid the use of CPB in the face of the risks, if CPB time falls within an acceptable and safe range.

In our series, five patients did not undergo tracheal surgery because they had no airway symptoms at the time of cardiac repair. Those patients required no ventilatory support preoperatively and their airway abnormalities were incidentally diagnosed during the anesthetic induction for the repair of cardiac anomalies. Those cases of tracheal stenosis tended to be less severe compared to the operation group. However, the size of the stenosis, per se, did not seem to be an accurate indicator for tracheal surgery. Although these patients did not receive any correction of their airways, all of them are surviving well without symptoms. Based on these observations, we suggest that the presence of significant respiratory symptoms is the more important factor in deciding to perform tracheal surgery than the degree of stenosis itself.

In summary, surgical repair of long-segment congenital

rates. Every patient who underwent pericardial patch tracheoplasty suffered from troublesome granulation tissue. As slide tracheoplasty provided relatively good results in the short and mid-term follow-up periods, it seems to be a preferred method for the treatment of long-segment congenital tracheal stenosis.

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Appendix A. Conference discussion

Dr J. Hasse (*Freiburg, Germany*): You demonstrated the problems with patch plasty. Now, there was a recent publication on the patch plasty with the aid of the anterior wall of the esophagus which was quite convincing, and I think that was a Japanese group. They published 5 cases of pediatric or neonatal ones and 3 adults. They had no mortality with this technique. The only handicap was that in the few adults, they had in 2 cases some swelling problems, which resolved after a while, but they had no mortality and good end results. Do you know about that article?

Dr Kim: Actually, I don't remember that article but I think there are a couple of other papers where they achieved good results with patch tracheoplasty. I agree with those findings.

Dr A. Tcherveniakov (*Sofia, Bulgaria*): This is really very rare but a very severe pathology. Have you considered in your practice to perform anastomosis of the trachea with the left main bronchus and reimplantation of the right main bronchus in the left bronchus as one opportunity for plasty substitution to do this repair?

Dr Kim: Do you mean when there is stenosis in the main bronchi?

Dr Tcherveniakov: No, when there is stenosis in the trachea, to perform the anastomosis with the left main bronchus and the trachea and reimplantation of the right bronchus in the left main bronchus.

Dr Kim: Yes, that may be an option for short stenosis. But, with long-segment stenosis, I don't think that it is a good option.

Dr P. Macchiarini (*Hannover, Germany*): You have a very high mortality which, in my opinion, is unacceptable for this cohort of patients. Those who deal with tracheal surgery in pediatric patients know that the problems are in between what you have told us and the reality. For instance, you don't mention about stenting a patch tracheoplasty.

Dr Kim: Stenting?

Dr Macchiarini: A stent inside the trachea when you do a patch tracheoplasty. This would avoid all the granulation tissue and the mortality that you had. And please try to avoid comparing a slide tracheoplasty with a patch tracheoplasty because the indications are completely different.

Dr Kim: I'm not so clear about what the difference is for the indication between slide tracheoplasty and patch tracheoplasty. In my opinion, I don't think there is any difference. Could you clarify what the difference is for the indication?

Dr Macchiarini: With a slide tracheoplasty, you need to do it if you have a long congenital tracheal stenosis with or without vascular or cardiovascular problems. The patch tracheoplasty could eventually be a good choice but only if you use a stent inside for those patients who have a long congenital tracheal stenosis without pulmonary artery

slings or other malformations, but we can discuss that further if you want.

Dr Kim: I would not agree with that. In our series, there were cases where we did perform slide tracheoplasties even after cardiac operation. I mean, for a patch tracheoplasty, we did have cases without any cardiac problems at all, as well as cases with cardiovascular problems. So I don't think that there is any difference. I think it's just a matter of how you treat the patient. I accept that we do have a high mortality rate, but our series is a chronologic experience. At first, we started with slide tracheoplasty without cardiopulmonary bypass, through the low collar incision. It was a difficult operation, which would be a different opinion from what Dr Grillo had told us. Unfortunately, however, we failed and the patient died. That is the reason we switched to the patch tracheoplasty. The thing that we experienced, however, was that with the patch tracheoplasty, there was a high complication rate related to the granulation tissue. After that, we again changed to slide tracheoplasty with cardiopulmonary bypass through a median sternotomy. In the 3 recent cases, we had no mortality and no complication related to the tracheal surgery itself.