

# Treatment strategies in the management of severe complications following slide tracheoplasty in children<sup>†</sup>

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## Abstract

**OBJECTIVES:** This study focuses on the different surgical and endoscopical treatment alternatives when dealing with severe complications after slide tracheoplasty (STP).

**METHODS:** Retrospective study of patients with symptomatic congenital tracheal stenosis (CTS) admitted to a single institution, between January 1997 and January 2013, surgically treated by means of STP. The following variables were evaluated: demographics, preoperative tracheal stenosis characteristics, associated anomalies and outcome measures.

**RESULTS:** Cohort included 14 patients (8 males and 6 females) with a mean age of 8.7 months when treated (range, 1–43 m). Eleven patients (78%) showed a long segment CTS (>30% of total tracheal length) and 9 (64%) had associated cardiac or great vessel anomalies (left pulmonary artery sling). Three patients (21%) showed severe postoperative complications that required significant airway reintervention: tracheal resection of a restenotic segment, laser division with balloon dilatation of a residual stenosis and placement of a biodegradable endotracheal stent in an extensive tracheal narrowing. All patients are in good clinical condition with a mean follow-up of 6.3 years (range, 2 months to 16 years).

**CONCLUSIONS:** STP has become the procedure of choice when dealing with CTS. Although it shows clear advantages compared with other surgical techniques, severe and difficult to manage complications may occur. Surgeons involved in their treatment should be familiar with diverse surgical and endoscopical procedures. Biodegradable airway stenting is a new and promising technique when long and severe post-surgical tracheal stenosis is present.

**Keywords:** Tracheal stenosis • Slide tracheoplasty • Bronchoscopy • Biodegradable airway stent • Children

## INTRODUCTION

Congenital tracheal stenosis (CTS) is a very uncommon obstructive malformation of the airway usually caused by complete tracheal rings [1]. Although CTS is classically associated with significant mortality, increasing experience with different surgical and endoscopic techniques has remarkably improved the outcome of these patients. Slide tracheoplasty (STP), initially described by Tsang *et al.* [2] in 1989, has become the surgical procedure of choice for the treatment of CTS by the majority of tracheal teams and paediatric airway units all over the world [3–5]. This technique has been thoroughly described in the literature and basically consists of transection of the tracheal stenotic segment at its midline followed by a vertical incision on the posterior aspect of the upper tracheal segment and another incision on the anterior surface of the inferior segment. Then, the two tracheal flaps are slid together for an anastomosis (Fig. 1). The placement of both vertical incisions can

be modified, if judged convenient, according to the specific features of the stenosis or the presence of an associated left pulmonary artery sling [6]. Although STP has demonstrated to be a highly effective and versatile technique in the treatment of CTS and its diverse morphological variants, severe and difficult to manage postoperative complications may occur.

The aim of this study is to focus on the different surgical and endoscopical treatment alternatives when dealing with severe complications after STP.

## MATERIALS AND METHODS

A retrospective review of paediatric patients with symptomatic CTS undergoing STP reconstruction in our institution, from January 1997 to January 2013, was performed. Approval for this study was obtained from the investigation committee. Hospital and office charts were reviewed to obtain pertinent information regarding: demographics, preoperative CTS characteristics, associated anomalies and outcome measures. Information was

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complete in all but one patient for whom long-term follow-up was not possible.

Patient management including preoperative evaluation, patient selection and operative technique used have been described in previous reports from our unit [3]. STP was performed using interrupted absorbable sutures under cardiopulmonary bypass in every case. Associated vascular anomalies were corrected simultaneously when present.

The main outcome variable was a severe postoperative complication that required surgical or endoscopic treatment (more than two balloon dilatations, airway stenting or laser therapy). These complications were classified into three main categories: (i) tracheal restenosis at the anastomosis causing symptoms and leading to reintervention; (ii) incomplete tracheal reconstruction with a residual stenosis not addressed at the initial surgical procedure; and (iii) significant anastomotic dehiscence that required either reoperation or stenting.

Secondary outcome variables were clinical status at follow-up and mortality. No statistical analysis has been performed due to the small number of patients in the series.

## RESULTS

Fourteen patients (8 males and 6 females) with a mean age of 8.7 months when operated (range, 1–43 m) were included in this study. Eleven patients (78%) showed a long segment CTS (>30% of total tracheal length) and a right upper lobe tracheal bronchus was present in 5 (35%). A left pulmonary artery sling was detected in 9 cases (64%). Three patients (21%) showed severe postoperative complications that required surgical or endoscopic treatment. One of each corresponded to one of the three different categories that have been described previously. They represent the core of this manuscript and are described in detail next.

The other 11 patients did not show significant post-surgical complications and remain asymptomatic from a respiratory standpoint. There was no mortality in the series with a mean follow-up of 6.3 years (range, 2 months to 16 years).

### Patient 1

This boy was operated at the age of 6 months because of long segment CTS (4 cm long, 3 mm diameter) and left pulmonary artery sling. The surgical procedure was uneventful but extubation was not possible until 5 weeks after the operation due to mucosal oedema, granulation tissue and secretions. During this period of time, the endotracheal tube was positioned above the reconstructed trachea and the patient was maintained on pressure support ventilation. Fiberoptic bronchoscopy through the endotracheal tube showed no

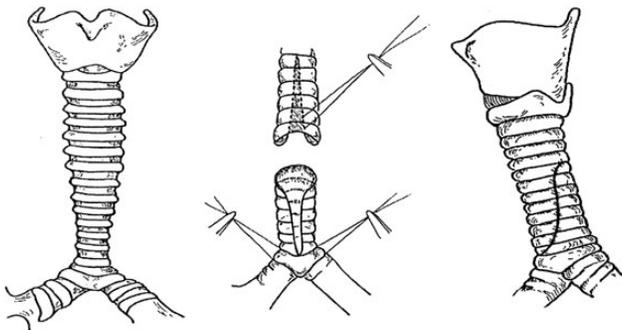


Figure 1: The slide tracheoplasty technique.

tracheal dehiscence and was useful for clearing secretions. Before discharge, a moderate restenosis was observed during bronchoscopy. This lesion was located in the mid-trachea and the proximal trachea and the carina looked healed and normal (Fig. 2). The stenosis was initially dilated with a 6 mm balloon catheter with good response. The patient developed respiratory symptoms in the following weeks and so further endoscopic balloon dilatation was deemed necessary. Dilatation was performed every 3–5 weeks, with balloon diameters ranging from 6 to 9 mm, when the patient became symptomatic. Mitomycin C (0.8 mg/ml) was topically used on two occasions after dilatation. A self-expanding nitinol stent, 8 mm diameter × 30 mm length (Silmet Novatech, La Ciotat, France) was placed 7 months after the operation and provided temporary relief. Unfortunately, obstructive granulation tissue developed in the distal end of the stent and so it was removed 10 weeks after placement.

Because a sustained clinical relief could not be obtained 11 months after the initial intervention, a surgical approach was planned. Resection of the stenotic segment was carried out under cardiopulmonary bypass. This lesion was located above the carina and was 2 cm long. To perform a reconstruction with healthy tracheal tissue, the anastomosis was placed next to the carina (Fig. 3). The patient was extubated 3 days after the procedure and the postoperative course was uneventful. No further endoscopic

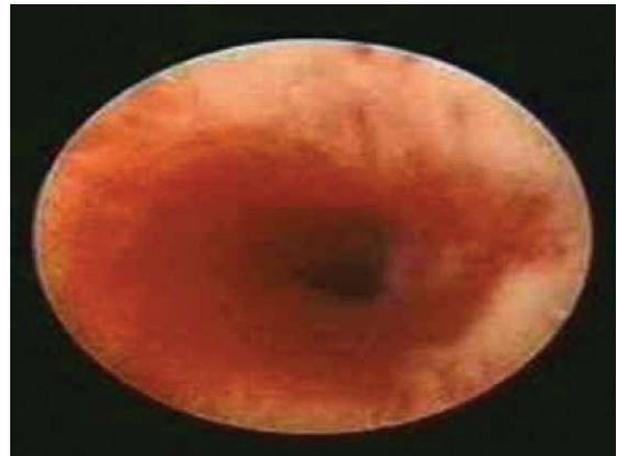


Figure 2: Bronchoscopic view of restenosis after slide tracheoplasty.

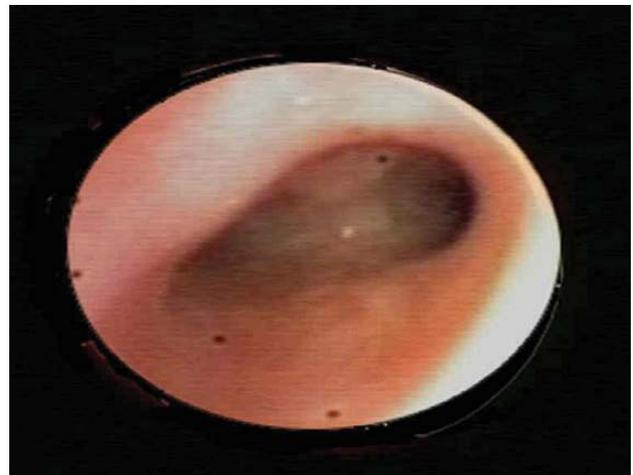


Figure 3: Bronchoscopic aspect after resection of the stenosis with the anastomosis close to the carina.

dilatations were necessary and the patient remains symptom-free and in good condition 26 months after.

## Patient 2

This boy was referred to us at 1 month of age with a diagnosis of complete atrioventricular canal defect, left pulmonary artery sling and CTS. Preoperative bronchoscopy showed a long segment stenosis extending from the mid-trachea to the carina (3.5 cm long, 2 mm diameter). STP and pulmonary artery sling correction were performed immediately after under cardiopulmonary bypass. Extubation failed in the ensuing days because of severe respiratory distress. Flexible bronchoscopy performed on the sixth postoperative day through the endotracheal tube disclosed a severe residual stenosis above the reconstructed trachea (Fig. 4). Ventilation became increasingly more difficult in the following days urging operative treatment.

Because STP had been performed very recently and the healing process was not complete, we decided to do interventional bronchoscopy instead of reoperating the patient. Endoscopic diode laser division of the stenotic rings followed by forceful balloon dilatation (4–6 mm diameter) was carried out in order to produce a controlled tracheal split (<1 cm long) in the posterior part of the residual stenotic segment. The trachea was then fully stented with an age-appropriate endotracheal tube (Portex no 3- OD 4.2 mm, Smiths Medical, St. Paul, MN, USA). The patient was kept intubated with total muscle paralysis for 2 weeks in order to achieve complete tracheal healing. Ventilation was uneventful during this period of time and no infectious complications occurred. Bronchoscopy performed at extubation showed adequate tracheal healing with mild granulation tissue and no restenosis. A follow-up bronchoscopy 3 months after the endoscopic procedure disclosed a patent trachea with an ample lumen (Fig. 5). The patient remains asymptomatic, from a respiratory standpoint, 21 months after the bronchoscopic intervention.

## Patient 3

This 2-month old girl was referred to our unit for surgical treatment. She had a diagnosis of CTS and left pulmonary artery sling. Preoperative bronchoscopy showed that the entire trachea was severely stenotic (2 mm diameter) and that both main bronchi

were slightly stenotic too. STP and left pulmonary artery reimplantation were performed under cardiopulmonary bypass. Owing to the length of the stenosis, the tracheal reconstruction had some degree of anastomotic tension even though surgical release manoeuvres were performed. Subtotal dehiscence of the anastomosis occurred and the patient was reoperated on the fourth postoperative day, performing a new STP. Again, flexible bronchoscopy disclosed anastomotic failure and so the endotracheal tube was advanced to the carina in order to stent the whole trachea and promote secondary healing. Follow-up bronchoscopy, 2 weeks after, demonstrated healing of the dehiscent trachea with severe restenosis (Fig. 6).

Repeated endoscopic balloon dilatations (3.5–6 mm diameter) did not yield satisfactory results and so we decided to place a tracheal stent. Recent experience with biodegradable airway stents in Great Ormond Street Hospital in London encouraged us to try them in this particular patient. A custom-made self-expanding polydioxanone stent (5 mm diameter × 15 mm length; SX-ELLA, Czech Republic) was inserted in the distal trachea 3 months after STP (Fig. 7). It was well tolerated by the patient, with mild granulation tissue formation, and so discharge was possible soon after (Fig. 8). As expected, repeated stenting after stent absorption has been necessary on three more occasions. We have gradually increased the diameter (6–8 mm) and length (from 15 to 30 mm) of the stent in order to provide full internal support to the entire trachea. Although follow-up bronchoscopies have been necessary to dilate the stents and remove soft granulation tissue, tolerance has been satisfactory and the patient remains in good clinical condition 1 year after the first stent implantation.

## DISCUSSION

Management of CTS has evolved dramatically in the last 30 years. Conservative treatment was the rule in the 1970s and before, but increasing experience with new surgical techniques have gradually improved patient outcomes and survival in such a way that surgical correction is, at the present time, the treatment of choice for symptomatic CTS [7]. In general, surgical procedures fall into three categories: (i) tracheal reconstruction with autologous tracheal tissue (tracheal resection, STP and free tracheal autograft); (ii) tracheoplasty with non-tracheal autologous tissue (costal cartilage



**Figure 4:** Bronchoscopic view of a residual stenosis proximal to the reconstructed trachea.



**Figure 5:** Deferred tracheal healing has occurred leaving a patent lumen.



**Figure 6:** Severe tracheal restenosis due to anastomotic dehiscence.



**Figure 7:** Bronchoscopic view of a biodegradable tracheal stent immediately after placement.

and pericardial patch) and (iii) tracheal transplant (with a tissue-engineered cadaveric homograft) [8]. Although CTS represents a spectrum of lesions causing airway narrowing and it is the actual tracheal pathology and its particular anatomic features that dictate the choice of a specific surgical technique, STP has emerged as the preferred method of tracheal reconstruction at most centres worldwide [3–5, 9].

There seems to be general agreement that STP is a safe, reliable and versatile technique when dealing with CTS. Nevertheless, complications may occur although it is considered to yield lower mortality rates and fewer postoperative complications than other surgical techniques [4]. Anastomotic failure, including restenosis and

dehiscence, is the most frequent postsurgical complication. Wright *et al.* [7] reported a 35% global rate of complications after tracheal surgery in children and that 23% showed anastomotic failure. In their experience, age younger than 7 years, reoperative procedures and long segment stenosis requiring resection of more than 30% of the tracheal length predicted a higher risk of complication, especially anastomotic failure. Manning *et al.* [4] have recently published the Cincinnati experience with 80 children who underwent STP. Although their results are outstanding, they report a 5% mortality rate and 28% (23/80) of patients needed significant airway reintervention (defined as a need for more than a single endoscopic dilatation, stent placement or surgical revision). Other reports from



**Figure 8:** The same stent 5 weeks after insertion.

individual centres with smaller series have demonstrated mortalities in the 10–30% range [5, 10, 11].

Anastomotic failure seems to be the most feared and frequent complication after tracheal surgery, including STP. Herein, we describe 3 patients with postoperative anastomotic complications resulting in severe stenosis. Although this outcome was the same in the 3 cases, the management strategy was different in each one according to their specific characteristics. In Patient 1, restenosis was probably due to ischaemia of part of the tracheal reconstruction. In this setting, endoscopic balloon dilatation is usually accepted as the most appropriate treatment [3, 5]. A sustained long-term clinical improvement was not achieved and so reoperation was considered almost 1 year after the initial surgical procedure. Tracheal resection of the stenotic, poorly vascularized, segment with end-to-end anastomosis between healthy tracheal tissues was definitely successful. This strategy could not be used in Patient 2 because STP had been performed very recently and tracheal healing was not complete. On the other hand, it was a different type of lesion though it was a residual proximal stenosis not addressed during STP. This complication can be considered a surgical technical error more than a true postoperative complication. In any case, the stenosis prevented an adequate ventilation of the patient, requiring immediate treatment. Although there is very little experience with endoscopic techniques as a primary treatment of CTS, we decided to try endoscopic laser division followed by balloon dilatation in this particular case. The rationale of this technique is to produce a limited tear or split in the posterior part of the complete tracheal rings with subsequent deferred healing [12–14]. Performing a complex endoscopic procedure in such a small airway is challenging and not without risks, but we believed that it was the best chance for this highly compromised patient. Although the result has been satisfactory, surgical reconstruction continues to be our treatment of choice for CTS.

Patient 3 showed a large anastomotic dehiscence that recurred after a redo procedure. Tracheal full-length stenosis can be corrected by STP but anastomotic tension is recognized as the most relevant risk factor resulting in surgical failure. Again, balloon

dilatation was ineffective when dealing with very severe tracheal narrowing. Stenting emerged as the best option in this setting for the maintenance of tracheal lumen patency. Our unit is experienced on airway stenting and we have inserted more than 60 devices in children with severe airway stenosis or malacia [15]. Available metal or plastic stents did not seem to be the most adequate in this particular case and so we tried a biodegradable stent for the first time. Vondryš *et al.* [16] have reported their initial experience with this type of stent in 4 patients. Their results encouraged our multidisciplinary team in the decision of using this new type of stent. Biodegradable stents have been originally developed for oesophageal, intestinal, urological, biliary and vascular stenosis [16]. Polydioxanone is a monofilament, absorbable material widely used as a suture in surgery. It is a biodegradable, semicrystalline polymer belonging to the polyester group. The polydioxanone stent is manufactured from a single wire and it shortens slightly when fully expanded. It has a degree of shape memory, tends to coil and degrades by random hydrolysis with the resultant products being harmless [17]. The degradation time in the airway has not been precisely defined but it seems 15 weeks after implantation it has dissolved completely [16]. In our experience, it was well tolerated by the tracheal mucosa with only mild granulation tissue formation. Tracheal patency has been achieved with this type of stent due to its radial force, although, as predicted, repeated stenting has been necessary. Nevertheless, we have been able to progressively increase the stent size, from 5 to 8 mm diameter, as the child grew. In our opinion, although the patient remains stent-dependent, several substantial advantages result from this approach: (i) the patient is alive and ventilator independent; (ii) her respiratory status has progressively improved over time; (iii) fewer biodegradable stent-related complications have occurred compared with other type of stents; and (iv) we buy time until the stenosis gets stabilized or other therapeutic alternatives, such as tracheal replacement, become available [18].

In conclusion, management of children with CTS remains challenging. STP seems to be the preferred surgical technique due to its effectiveness, reliability and versatility. Nevertheless, severe complications may occur and diverse endoscopic and surgical

alternatives must be considered in the setting of a multidisciplinary team approach.

**Conflict of interest:** none declared.

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## APPENDIX. CONFERENCE DISCUSSION

**Dr D. Mathisen (Boston, MA, USA):** This is a very difficult group of patients, small individuals, small airways, with a very narrow margin for error. So these are very difficult problems to deal with. I have a few questions. You mentioned three

patients who had less than 30% involvement of their trachea. Is there ever an indication for an end-to-end resection, or do you use slide tracheoplasty for any congenital stenosis in an infant that's symptomatic?

**Dr Antón-Pacheco:** Yes, indeed, we use resection and reconstruction for short segment CTS. But really in recent years slide tracheoplasty is not more complicated to do in short segments than resection and reconstruction, and it's a very versatile technique. So probably we are switching from the end-to-end anastomosis, the classic anastomosis for short CTS, to the slide tracheoplasty for short segment, too, yes. But we used to do it before.

**Dr Mathisen:** In your presentation you mentioned that those patients who had more than two dilations were considered failure of the procedure, and it represented one of the patients in your series, the implication being that the other 11 patients had some dilations. Could you just share with us how often dilation is required following slide tracheoplasty?

**Dr Antón-Pacheco:** Yes. After slide tracheoplasty sometimes you find this figure 8 lumen and it's not a stenosis, it's just that the trachea is remodelling. So sometimes you need one or maybe two balloon dilations to get the full size of the trachea and to correct this figure 8 trachea. So this is the minor complication that we have found in some of the cases which has been treated with one or two balloon dilations.

**Dr Mathisen:** The one patient that you resected, it looked like from your drawing that the stenosis was below the slide segment; is that correct?

**Dr Antón-Pacheco:** No, it was in the slide segment.

**Dr Mathisen:** In that particular case, were you concerned about the blood supply when you resected that segment, did it interfere with the blood supply in any way?

**Dr Antón-Pacheco:** Well, in fact, it was a very ischaemic segment, it was a scar. So we resected this tissue, this scar, and we performed an end-to-end anastomosis between healthy tissue.

**Dr Mathisen:** And then in the individual in whom you used this biodegradable stent (and I realize the experience is limited), in the year that you have followed that patient, do you see any compromise of the stent, has it started to resorb, has its diameter diminished because it is starting to lose its integrity?

**Dr Antón-Pacheco:** Yes. In fact the stent begins to reabsorb completely between the 13th and the 15th week. So in this period of time you hardly see the stent, you only see some sutures, pieces of suture, and it begins to disappear. So it's a problem with a child that we have to keep going on and replacing the stent every three or four months for the moment. We don't know the future. I would like to know the future of this child, but we are sure that she is alive because of this stenting procedure. Although we keep going and we keep placing new stents, maybe the stenosis will stop, will be controlled with growing or with the stents, or maybe we are buying time for another procedure like tracheal replacement.

**Dr Mathisen:** And then finally, one can infer from your presentation that there is a different technique you used to use for congenital stenosis. Could you share with us what that technique was. And one would assume, since you've chosen the slide tracheoplasty, that you think the results with the slide tracheoplasty are superior to what you used to do.

**Dr Antón-Pacheco:** Well, in fact, we perform slide tracheoplasty for nearly every case with CTS. In some cases of very short congenital tracheal stenosis we perform the classical resection and anastomosis. And our results are much better indeed with slide tracheoplasty than the ones we used to have with cartilage graft tracheoplasty or pericardial graft tracheoplasty 20 years ago. We didn't do very much of those cases in the old days, but we did five or six cases, I can't remember, and the results were much poorer than the ones we have with slide tracheoplasty.

**Dr E. Rendina (Rome, Italy):** I have one quick question. In the case that Dr Mathisen referred to, the resection which was very close to the carina, do you use an approach through sternotomy or you can reach that point through cervicotomy? And the second question, you didn't mention any recurrent laryngeal nerve problem. Did you have any, or not?

**Dr Antón-Pacheco:** Well, concerning the first question, the procedure is performed by a full sternotomy and cardiopulmonary bypass in cooperation with our cardiovascular colleagues. I mean, unless the stenosis is in the neck or very near the neck. But if the stenosis is, as it usually is, in the distal trachea or the distal part of the trachea, always full sternotomy and cardiopulmonary bypass. And, yes, we had one case of laryngeal paralysis, unilateral, but it resolved spontaneously some months after the procedure.