Long-segment tracheal stenosis: Slide tracheoplasty and a multidisciplinary approach improve outcomes and reduce costs

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Objective: Long-segment tracheal stenosis is rare, life-threatening, difficult, and expensive to treat. Management remains controversial. A multidisciplinary tracheal team was formed in 2000 to deal with a large number of children with airway problems referred for management. We review the effect of that service, comparing the era before and after the establishment of the multidisciplinary tracheal team.

Methods: From January 1998 through January 2004, 34 patients with long-segment tracheal stenosis (21 patients with cardiovascular anomalies) underwent surgical intervention. Cardiopulmonary bypass was used in all operations. Before the multidisciplinary tracheal team, pericardial patch tracheoplasty with or without an autograft technique was the preferred method of repair. After the multidisciplinary tracheal team, an integrated care plan preferring slide tracheoplasty was initiated, correcting cardiac lesions simultaneously.

Results: Before the establishment of the multidisciplinary tracheal team, pericardial patch tracheoplasty was performed in 15 of 19 patients. Twelve patients had a suspended pericardial patch tracheoplasty, 2 (17%) of whom died late after the operation. Of 3 patients who had had a simple unsuspended patch, 2 (67%) died early after the operation. Four patients were operated on with the tracheal autograft technique, 2 (50%) dying early in the postoperative period. After multidisciplinary tracheal team formation, in the era between 2001 and 2004, 15 patients were operated on with slide tracheoplasty, and there were 2 (13%) early postoperative deaths. A significant reduction in cost and duration of stay has been shown both in the intensive care unit and the hospital.

Conclusion: Our data suggest that a formalized multidisciplinary team approach and a policy of primary slide tracheoplasty are beneficial in the management of children with long-segment tracheal stenosis.

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Long-segment tracheal stenosis (LSTS) in children is rare and potentially life-threatening, and the treatment remains controversial. Over the years, we have used a number of therapeutic options.¹ Several specialty groups were involved, with interactions based on a consultation-when-needed policy. We had evolved an ad hoc policy involving slide tracheoplasty (STP)¹ for short- and medium-length stenosis and, on the basis of the late 1990s literature,^{2,3} patch tracheoplasty (PTP) for long or very long stenoses. Stenting was used for early failure, and tracheal homograft patch repair⁴ was used for late recurrence.

We were dissatisfied with our PTP results, and thus in late 2000, we met to review our strategies. We created a multidisciplinary tracheal team (MDTT) comprising all specialties involved in caring for such patients. We took the following decisions:

1. All referrals of children with tracheal problems would be channeled through the MDTT.
2. Investigations would be carried out according to a fixed protocol.
3. STP would become our procedure of first choice and extended to include longer-segment stenosis.
4. Coexistent congenital heart defects would be fixed at the same operation.
5. PTP would become a second-string strategy. Tracheal homograft repair would be used for recurrent severe stenosis if stenting failed.
6. Postoperative surveillance would be by means of fiberoptic bronchoscopy (FOB) and bronchography. Granulations would be managed with radial balloon dilatation.
7. Members of the team would be cross-skilled (especially in FOB and stent placement) to facilitate timely follow-up in the resource-limited system in which we work.
8. Nurse liaison and lead administrator posts would be established to improve integration of care pathways and communication with patients, families, referrers, and the multiple teams involved. Shared care with referring units would be encouraged and facilitated.
9. A weekly meeting of the MDTT would be held for patient and data review.

This article is designed to define the effect of these changes, implemented in January 2001, by reporting a retrospective 6-year review of our experience for 3 years before and 3 years after the introduction of the MDTT and the change of strategies indicated above.

Patients and Methods

Era

The study took place from January 1998 through January 2004, a 3-year period on either side of the establishment of the MDTT in January 2001.

Patients

Details of the patients are summarized in Table 1. Thirty-four patients (24 male and 10 female patients) aged less than 16 years with a primary diagnosis of LSTS were treated. All patients had stenosis extending for more than two thirds of the length of the trachea and involving the carina. The median age of the patients was 131 days (range, 9–5475 days). Thirty-one patients were less than 1 year of age. There was a high incidence (21/34 [62%]) of coexistent cardiovascular anomalies. Two patients had absence of the right lung.

TABLE 1. The age, sex, and associated cardiovascular anomalies of patients before and after the establishment of the multidisciplinary tracheal team in 2001

	Pre-MDTT (n = 19)	Post-MDTT (n = 15)
Median age at operation, wk (range)	18.7 (1.3–104.2)	14.42 (1.8–782.1)
Sex (male/female)	13/6	11/4
Associated anomalies (n)	11	10
PA sling	7	6
ASD	1	1
VSD	1	1
PDA	1	1
A-P window	1	—
Trisomy 21	2	—
Absent right lung	1	1
DORV	—	1
Subaortic stenosis	—	1
Coarctation of aorta	—	1
Type II common arterial trunk, truncal valve dysplasia, IAA	—	1
Esophageal atresia	1	—

MDTT, Multidisciplinary tracheal team; PA, pulmonary artery; ASD, atrial septal defect; VSD, ventricular septal defect; PDA, patent ductus arteriosus; A-P, aorta-pulmonary; DORV, double-outlet right ventricle; IAA, interrupted aortic arch.

Methods

A retrospective chart review was undertaken. The patients were divided for reporting into 2 groups: before the MDTT and after the MDTT.

For the pre-MDTT group, no fixed protocols existed, and care was individualized. Surgical intervention was carried out according to published methods.^{2,3,5,6} Postoperatively, endotracheal intubation was used as a temporary stent until the patch was judged, both subjectively and on FOB, to have become firm.

For the post-MDTT group, care was more structured. STP was carried out according to standard techniques.^{1,7,8} Patients were intubated for a minimum of 5 days, after which they underwent FOB and bronchography to define timing for extubation. Intravenous antibiotics were given for 7 days after the operation. FOB and bronchography were repeated after 2 weeks and 6 months and thereafter as needed.

Statistical Analysis

Data were entered into a Statview (version 5.0; SAS Inc, Cary, NC) spreadsheet for calculation of descriptive statistics and further analysis. Data between groups were compared with unpaired *t* tests.

Results

Before the MDTT (1998–2001), pericardial PTP was performed on 15 of 19 patients (Figure 1). Twelve patients had a suspended pericardial PTP, 2 (17%) of whom died late after surgical intervention (see below). Of 3 patients who had had a simple unsuspended patch, 2 (67%) died early

Figure 1. The fate of children with LSTS managed from 3 years before to 3 years after the establishment of the MDTT in the year 2001.

TABLE 2. The median, range, and *P* values for the durations of stay in the intensive care unit, the high-dependency unit, and the hospital and the duration of endotracheal intubation, with ICU, HDU, and total hospital costs

	Pre-MDTT, median (range)	Post-MDTT, median (range)	<i>P</i> value
Duration of stay in ICU (d)	28 (5–130)	7 (2–54)	.03
Duration of endotracheal intubation (d)	17.8 (0.7–40.4)	5.2 (1.7–54)	.09
Duration of stay in HDU (d)	17 (1–130)	5 (1–26)	.78
Total duration of stay in hospital (d)	59 (5–243)	21.5 (10.4–55)	.01
ICU cost (£ sterling)	41,208 (8585–223, 210)	12,019 (6868–92,718)	.03
HDU cost (£ sterling)	13,209 (777–101,010)	3885 (777–20,202)	.07
Total cost of hospital stay (£ sterling)	56,331 (8585–324,220)	18,401 (13,648–92,718)	.01

MDTT, Multidisciplinary tracheal team; ICU, intensive care unit; HDU, high-dependency unit.

after the operation. In one patient a fistula from the innominate artery to the trachea formed. This was repaired, but the patient died later of sepsis and patch dehiscence. The second patient had untreatable distal malacia and granulations, causing fatal respiratory insufficiency.

Four patients underwent repair by means of the tracheal autograft technique. Two (50%) died early postoperatively. The first had esophageal atresia and hypoplastic left lung. Autopsy revealed solid lung and severe distal airway malacia, despite satisfactory tracheal reconstruction. The second patient had trisomy 21 and multiple ventricular septal defects. This patient died of an unrepairable cardiac abnormality. After the formation of the MDTT (2001–2004), 15 patients underwent STP, and 2 (13%) died early postoper-

atively. Both had severe additional congenital cardiac malformations, and both had treatment withdrawn despite widely patent major airways, the first for severe distal bronchomalacia (autopsy revealed complete absence of bronchial cartilage) and the second for truncal valve regurgitation and multiple ventricular septal defects.

Intensive care unit stay was significantly shorter in the post-MDTT group, as was the total hospital stay (Figure 2). Duration of endotracheal intubation and stay in the high-dependency unit were not statistically different (Table 2).

Intensive care unit costs and total hospital costs were both significantly lower for the post-MDTT patients. The change in high-dependency unit costs approached statistical significance (Table 2).

Figure 2. Box and whisker plots showing the difference in (a) duration of stay (DOS) in the intensive care unit (ICU), (b) duration of intubation, (c) duration of stay in the high-dependency unit (HDU), and (d) the total length of hospital stay.

Costs of postoperative morbidity are also important. In the pre-MDTT group a significantly higher percentage of patients (58% vs 7%) had recurrent stenosis after surgical intervention. These costs amounted to £20,800 and £4550, respectively (Figure 3). In the pre-MDTT group significant granulations were detected in 12 patients, 11 of whom were stented. In the post-MDTT group only one patient, who received a temporary stent, had moderate granulations.

Before the MDTT, the average cost of treatment per patient ($n = 19$) was £77,333. After the MDTT, the average cost per patient ($n = 15$) was £26,219. This represents a savings per patient of £51,114.

The median follow-up for the 26 survivors was 38 months (range, 2-57 months). There have been 2 late deaths (8 and 38 months postoperatively), both in the pre-MDTT group, from severe distal tracheobronchomalacia.

Discussion

In our hands the combination of STP and multidisciplinary teamwork has been associated with improved and more predictable results, reduced morbidity, and simplified management while reducing cost. We judge STP to be a better procedure (lower mortality, less stenting, shorter postoperative stay, and fewer granulations) than PTP. Furthermore, the team approach has maximized the overall efficiency of the management process.

Do our data justify these inferences? To answer this, we need first to consider the appropriateness of both the deci-

sion to hold and the outcomes of our consensus meetings in late 2000. As previously reported,¹ we had used a wide variety of surgical repairs for LSTS. Treatment had been individualized but was heavily influenced by the most recent literature. We had been seduced by the results from Backer,² Brown,³ and their associates describing autograft and PTP, which seemed sensible for patients with long-segment stenoses extending into the bronchi.

The published literature was not definitive, principally because of small numbers, single-center reporting, and the large number of available surgical procedures. Nevertheless, others had concluded that STP was the preferred option.^{2,7-14} We have become confident that STP can be used for almost all cases of LSTS, even those extending into the bronchus. Since 2001, further supportive data have been published.¹⁵⁻²⁰ There seems little doubt that reconstruction of the trachea with vascularized tracheal tissue should be superior to the use of devascularized, nontracheal tissue.

The surgeon has been seen as central to outcomes, but we were well aware that individual proficiency in a particular task is only one facet of effective team performance.²¹ Thus the MDTT was modeled on previous successes in, for example, transplantation and extracorporeal membrane oxygenation. The unstructured approach to our patients before 2000 lent itself to such rationalization. Several blocks to efficiency were identified: difficulty in integrating specialty groups and coordinating admissions, liaising over intensive care

Figure 3. Box and whisker plots showing the differences in (a) intensive care unit (ICU) cost, (b) high-dependency unit (HDU) cost, and (c) total cost.

bed availability, establishing shared care arrangements, arranging and performing FOB (only performed by the respiratory physician [CW] and the cardiothoracic surgeon [MJE] before 2000), and endoscopic tracheal stenting (only performed by the interventional radiologist [DR] before 2000). Most problems were resolved by establishing liaison and administration posts, written protocols, care pathways, and the weekly meeting. The FOB and stenting issue was more complex. We decided to implement cross-skills by training others in these tasks. During the latter part of 2000 and throughout 2001, MJE learned to implant stents and to perform balloon dilations and bronchography, and DR and CM (radiographer) learned to do FOB. Consequently, it became much easier to arrange follow-up or urgent investigations, contributing to the reduction in hospital stay and cost. Others²² have shown that providing cross-skills improves team performance.

The MDTT has had other benefits. For example, there has been increased referral of patients with many different airway problems from a progressively wider area. This has increased our patient population, accelerated our learning curve, and unified policies supported by regular audit. We therefore recommend centralization of care and an MDTT approach for pediatric patients with major airway problems. We are very proud to be members of the MDTT and believe we have achieved what Morgan and colleagues²³ described as the 7 dimensions of teamwork: communication, adaptability, cooperation, acceptance of suggestions or criticism, giving suggestions or criticism, commitment to a common goal, and team spirit.

Limitations

There are several limitations to this study:

1. It represents a small, short, single-center experience.
2. The design of the study precludes formal comparison of strategies and is neither prospective nor randomized.
3. The follow-up period is very short. Current perceptions of good outcome might be dispelled by time.
4. The effect of era and learning curve might account for the study findings.

Despite these limitations, we judge our current protocols for the management of LSTS to be working. The MDTT is functioning well, and our results with STP are consistent with those in the literature. The patients who have succumbed since the introduction of the MDTT have done so either for cardiac reasons or because of severe distal malacia and not because of failed airway repairs.

Ideally, there would be specifically designed prospective randomized studies. In reality, the available patient population for such studies is too small, even in our own busy and active unit. We simply have to apply common sense to the data to suggest best practice.

Conclusions

Our data suggest that children with LSTS benefit from the formalized multidisciplinary team approach and a policy of primary STP.

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Discussion

Dr Hermes C. Grillo (Boston, Mass). Zeng and Goldstraw described slide tracheoplasty in 1989, with success in one of their 2 patients. In 1994, after our short report of success in 4 consecutive patients, the method became quite widely applied around the world, and the advantages over patch tracheoplasty seemed evident. I can only say "seemed." The trachea is immediately reconstructed with tracheal tissue. The result is complete airway stability at the end of the operation and complete respiratory epithelial lining right at the outset. As a result, granuloma formation in the suture lines is negligible. Postoperative intubation and ventilation, we have found, is generally unnecessary in these patients.

By 2001, we had accumulated a small series of 8 consecutive successful repairs, to which we have added a few more. By that time, we found there were 22 patients reported worldwide, and therefore the experience was growing. Slide tracheoplasty, I think, continued to show clear advantages over previous patch procedures, and you have pointed out their complexity, complications, failures, and mortality.

You and your colleagues have added 15 more cases, and the organization of the multidisciplinary team, I believe, might or might not have contributed to, as you have also stated today, improved results over the patch. I say that because the prior experiences that I have just listed suggest that the substitution of what seems to be a superior procedure—and I realize that is a matter of opinion—is most likely the principal factor rather than the team. Unfortunately, the rarity of these lesions and, at the same time, these excellent surgical experiences in the last few years really militate against randomized or prospective series, as you have indicated.

I would say that the need for a special team would seem to us to be dictated by institutional imperatives more than anything else. In our hospital, for example, congenital stenosis is treated by general thoracic surgeons who have special expertise in tracheal surgery. They are experienced in diagnostic and interventional bronchoscopy and direct the preoperative and postoperative care. Therefore it is a unitary kind of care. The specialized radiologists and anesthesiologists, of course, play a role, and congenital cardiac surgeons participate as needed for each individual patient. The results have been excellent thus far, as indicated.

Also, just to follow up on your last comment, the long-term follow-up in this small series with the smaller patients has been excellent. They do grow, and they grow very nicely without symptoms.

I certainly agree that the anomaly is so rare and so surgically challenging that patients would indeed be best treated in regional centers. And it is preferable to treat patients where there is expertise in this type of operation and airway surgery. I am afraid that is very unlikely to occur in the United States for a number of obvious reasons.

I only have one minor question, and it is procedural. In our limited experience, 5 of the 8 patients were extubated in the operating room, and 3 of the 8 were extubated at 1, 3, and 8 days, respectively. What is the need for 5 days of routine ventilation with these patients who have essentially a normal airway when you finish the operation?

Dr Elliott. Thank you very much, Dr Grillo. It was intimidating delivering the paper in front of you and having you ask the question.

I agree with all of your comments, of course, because I think they reflect the philosophy that we have described.

The issue about the period of ventilation was, like all the other decisions we took at the beginning of the team, a consensus decision. It did not have a particular value other than that. I am certain that some of the patients could have been extubated earlier, and therefore perhaps we need to develop a protocol for that. However, I do have a caveat, perhaps related to the patient population that we see, which has a very disproportionate load of associated congenital heart disease. There is a subset that seems to have been exposed to, by the very repair of their proximal tracheal stenosis, very severe distal bronchomalacia, as though they have "auto-PEEP" by the presence of a long-segment stenosis, which, when it is corrected, exposes severe distal malacia.

Four of the patients who died in this series had this condition. Furthermore, we now have another patient in exactly the same position in the intensive care unit since these data were drawn together. I am very concerned that we do not yet have a mechanism for identifying this subset. Thus we have been cautious about early extubation until we can be more certain about the presence or absence of that distal malacia.

Dr Vaughn A. Starnes (*Los Angeles, Calif*). Could you define a long segment for me? I mean, we have seen patients from the subglottic region down to the carina and actually some recent children onto the left bronchus. Given the extensive nature of the disease in some children, where can you apply this particular technique of slide tracheoplasty?

Dr Elliott. First question first. I do not know of a good definition of long-segment tracheal stenosis, but my colleague, Dr Kocyildirim, is presenting what I hope will be such a definition at the American Society of Radiology in Charleston later this week on the basis of bronchographic measurements.

For the purposes of this population of patients, we have defined long-segment stenosis as more than two thirds of the trachea. We agree that many of those do extend on to the bronchus; in fact, that was why we migrated toward patch tracheoplasty in the 1990s.

I do not now think that a slide tracheoplasty needs to be limited at all. Some of these patients have had carina-to-right-main-bronchus slide tracheoplasties without any problem. I was worried about devascularization of the trachea, but in the small baby it really does not seem to be a major issue.

Dr Carl L. Backer (*Chicago, Ill*). I would like to congratulate Martin and his colleagues on their excellent results with infants with long-segment congenital tracheal stenosis. Our center has had a strong interest in these patients for many years, and our series now includes more than 60 infants and children. I have a few comments and questions.

I believe the question that Martin has asked is as follows: Is it the team, is it the technique, or is it both? In trying to answer this question, I hate to disagree with Dr Grillo, but I believe that the team approach is a significant key to your success. Our institution has essentially had a de facto tracheal surgery team, and we have been very careful to limit the number of people who actually care for these critically ill patients. I agree with and encourage the creation of dedicated teams to take care of these patients.

The second part of the question concerns the technique. We are all searching for the Holy Grail of the ideal operation for long-

segment congenital tracheal stenosis. I would submit, even if you look just at our little area of the Midwest, that in the hands of dedicated teams, different techniques have led to similar results. Dr John Brown has done 23 pericardial tracheoplasties with 3 deaths. Dr Chuck Huddleston uses the cartilage tracheoplasty, which I have used as a rescue operation. He reported on 10 patients with 1 death. Our group now has 19 autografts with 3 deaths. Finally, Dr Peter Manning, who is right behind me, reported 11 slide tracheoplasties with 2 deaths. That is just in the Midwest. At these 4 institutions, you have 4 different techniques with almost identical results to yours with the slide tracheoplasty.

I have 3 questions. The first question relates to the technique of the operation. I assume that you used cardiopulmonary bypass in all of these operations?

Dr Elliott. All of them had bypass.

Dr Backer. My second question relates to a paper presented at the Southern Thoracic Surgical Society that recommended the use of extracorporeal membrane oxygenation (ECMO) postoperatively in these patients. Did you use ECMO postoperatively in any of your patients, and what do you think about that strategy?

Dr Elliott. We used ECMO in none postoperatively and 3 preoperatively. We have used ECMO as a resuscitative maneuver for patients when they cannot be ventilated. The other marginally ventilatable patients have been managed with helium ventilation, which can keep a child going for quite a long time until you can get them onto some form of support.

Dr Backer. My final question relates to the tracheal homograft. It was based on your very encouraging results with that technique that we started using the tracheal autograft technique in 1996. I wondered if you could make a few comments about your use of the tracheal homograft for rescue operations.

Dr Elliott. We still use tracheal homograft repair for severe recurrence. Two patients in the patch tracheoplasty series have ended up with secondary operations with tracheal homograft repair. We have relegated tracheal homograft repair, if you like, to a salvage operation and for patients referred from elsewhere who have recurrent severe stenosis. We always counsel the patients who we have either stented or who arrive with stents that they might in the future require further tracheal surgery, which would probably be with a tracheal homograft.

Dr Peter B. Manning (*Cincinnati, Ohio*). This is just an excellent series. As we have discussed, our experience mirrors yours. We have had 29 tracheal reconstructions using bypass, and the last 22 have been slides, and we have been very pleased with the results. It is a very versatile technique.

Your experience is probably like ours; when you start to get a name for yourself with this, you start to get more complex patients. We found the slide to be very versatile, even in patients who have been operated on before, either with tracheal reconstructions or with a tracheostomy. Would you comment about your experience with both of those problems? If you have seen them, how do you deal with the tracheostomy if a patient already has one?

Dr Elliott. Well, if they come with a tracheostomy, then we would try and get everything fixed at the same time and take the tracheostomy out at the operation.

And yes, we use the slide tracheoplasty if people have had operations, including one of Richard's [Jonas] old patients.