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Laryngotracheal reconstruction and cricotracheal resection in children: Recent experience at Great Ormond Street Hospital

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ABSTRACT

Background: Surgery for paediatric airway stenosis is constantly evolving. Surgery is the primary treatment modality via either an open or endoscopic approach. The objective of this study was to review the results of laryngotracheal reconstruction (LTR) and cricotracheal resection (CTR) procedures performed at Great Ormond Street Hospital over the past 10 years.

Methods: All patients who underwent open airway reconstruction surgery from January 2000 to December 2010 were included in this study. Patients treated entirely endoscopically were excluded. The data was collected using the electronic operating theatre database and the discharge summary database.

Results: Complete data was available for 199 patients who underwent open airway reconstruction from January 2000 to December 2010. The procedures included single stage LTR (57, 28.6%), two stage LTR (115, 57.7%), single-stage stomal reconstruction (14), single-stage CTR (8) and two-stage CTR (5). The diagnoses at the initial airway endoscopy were laryngeal web (22), subglottic stenosis (151), posterior glottic stenosis (9), suprastomal collapse (15), supraglottic stenosis (1) and tracheal stenosis (1). For those with subglottic stenosis, the stenosis was grade 1 in 1 patient, grade 2 in 26 patients, grade 3 in 117 patients and grade 4 in 6 patients. At the completion of intervention 175/199 (87.9%) patients reported improvement in their symptoms. Amongst the subglottic stenosis group, post LTR success was achieved in 100% with grade 1 stenosis, 92.3% with grade 2 stenosis, 88.1% in grade 3 stenosis and 83.3% in grade 4 stenosis. Of the two-stage LTR procedures, 100/115 (86.9%) had their tracheostomy removed and 15/115 (13.1%) have failed decannulation. Of the single-stage LTR group, 50/57 (87.7%) patients were better both on airway examination and symptomatically postoperatively. Of the single-stage stomal reconstruction group, 13/14 (92.8%) were better symptomatically and on airway examination. Patients who underwent single-stage CTR had a better airway on examination and were symptomatically improved in all cases (8/8). For the patients who underwent two stage CTR, the tracheostomy was removed in 3/5 (60%) and retained in 2/5 (40%). For the whole group, 15/199 (7.5%) patients underwent a revision LTR. On further analysis, revision LTR was required in 4/57 (7.1%) single-stage LTR, 9/115 (7.8%) two-stage LTR, 1/5 (20%) two-stage CTR and 1/8 (12.5%) single-stage CTR. In this study complications occurred in 13/199 (6.5%).

Conclusions: Subglottic stenosis in children needs to be approached on the basis of the nature and severity of stenosis and the individual patient's general health. Good outcomes are achieved with both LTR and CTR. Good results are obtained both with single-stage and two-stage LTR, but restenosis remains a problem. An individual approach is required for treatment of paediatric airway stenosis to achieve good final outcomes. The overall success rate has increased only marginally in our institution over the last 20 years.

1. Introduction

The management of laryngotracheal stenosis (LTS) forms a significant part of the practice of the paediatric otolaryngologist. Surgery is the primary treatment modality via either an open or endoscopic approach. The two most common open procedures are laryngotracheal reconstruction (LTR) and partial cricotracheal resection (CTR). LTR involves splitting the cricoid cartilage and expanding the framework with a cartilage graft. Partial CTR involves segmental excision of the stenotic segment, preserving the posterior cricoid plate, and an end-to-end anastomosis [1]. The management of paediatric airway reconstruction has changed considerably since its introduction in the 1970s.

The purpose of this study is to present the results of surgery for LTS procedures performed at Great Ormond Street Hospital over the past 10 years, and contrast them with our previous 10-year series from the 1980s [2–4].

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2. Methods

This study was conducted in the Department of Paediatric Otolaryngology at Great Ormond Street Hospital for Children. All the patients who underwent open airway reconstructive surgery from January 2000 to December 2010 were included in this study. Patients treated entirely endoscopically were excluded, as were anterior cricoid split procedures. The data was collected using the prospective electronic operating theatre database and the discharge summary database. The information collected included patient demographics, co-morbidities, severity of stenosis, level of stenosis, reason for the procedure, the nature of the operation, operative details, complications and the final outcomes. The subglottic stenosis was graded according to the Myer–Cotton grading system using an endotracheal tube to size the airway [5]. The outcomes measured included rate of decannulation for those patients who had a tracheostomy before the reconstruction, and airway endoscopy findings and symptomatic improvement for those who did not have a tracheostomy prior to the reconstruction.

Single-stage laryngotracheal/cricotracheal/stomal reconstruction was defined by the patient not having a tracheostomy tube in place at the conclusion of the procedure. This therefore includes patients who had their tracheostomy removed at the time of the surgery, and those who did not have a tracheostomy preoperatively. All these single-stage LTR/CTR/stomal reconstruction patients had a nasotracheal tube left in place at the end of the procedure for around 7 days and were monitored on the intensive care unit. Two-stage laryngotracheal/cricotracheal reconstruction was defined by the patient having a tracheostomy tube at the end of the procedure. All of these patients had a tracheostomy in place pre-operatively. The majority of patients undergoing two-stage LTR had a stent sited perioperatively held in place by a prolene transfixion suture. The stent was custom-made using an age-appropriate ivory portex endotracheal tube and removed endoscopically on average 4–6 weeks after placement.

3. Results

Complete data was available for 199 patients (122 male, 77 female) who underwent open airway reconstruction from January 2000 to December 2010. The procedures included single-stage LTR (57, 28.6%), two-stage LTR (115, 57.7%), single-stage stomal reconstruction (14), single-stage CTR (8) and two-stage CTR (5) (Fig. 1). The diagnoses at the initial airway endoscopy were laryngeal web (22), subglottic stenosis (151), posterior glottic stenosis (9), suprastomal collapse (15), supraglottic stenosis (1) and tracheal stenosis (1). For those with subglottic stenosis, the stenosis was grade 1 in 1 patient, grade 2 in 26 patients, grade 3 in 117 patients and grade 4 in 6 patients. The vast majority (159/199, 79.9%) of these patients had significant co-morbidities. The associated comorbidities in this varied group included cerebral palsy, chronic lung disease, CHARGE syndrome, tracheomalacia, bilateral and unilateral cord palsy, Fallot's tetrology, craniofacial abnormalities, reflux disease, Fraser syndrome, Di George syndrome, Larsen syndrome, velocardiofacial abnormality, laryngeal cleft, developmental delay, cricoarytenoid joint fixation, cerebral ischaemia, cardiac abnormalities and dwarfism. Sixty-seven were born prematurely, ranging from 23 weeks to 31 weeks at birth. The age at the time of operation varied from 9 months to 16 years. Most of the patients (173/199) were between 1 and 5 years of age at the time of operation. Amongst these 19 patients were less than 1 year age at time of operation, 49 patients between 1 and 2 years at operation, 33 between 2 and 3 years, 37 between 3 and 4 years and 35 between 4 and 5 years at the time of operation. In this group of patients, 160/199 (80.4%) had a tracheostomy prior to undergoing laryngotracheal reconstruction. The pathology was congenital in 47 and acquired in 152/199 patients.

Amongst the 57 single-stage LTR's, 11 (19.3%) had a tracheostomy reinserted at some stage after the procedure. One of the 14 single-stage stomal reconstructions and one of the 8 single-stage CTR's had the tracheostomy reinserted. Overall 13 of the 79 (16.5%) patients who underwent single-stage procedures had a tracheostomy reinserted.

Costal cartilage was the primary choice of graft material in this series. Of all the LTR/stomal reconstructions (186), the reconstruction was done with an isolated anterior graft in 91 cases, both anterior and posterior grafts in 61 cases, an anterior graft and posterior split in 26 cases and an isolated posterior graft in 8 cases.

In two-stage LTR/CTR procedures a custom-made stent (fashioned from an ivory portex endotracheal tube) was used in 113 of the 120 cases. A nasotracheal tube functioned as a stent in the single-stage procedures. No stent was used in 2 of the 5 two-stage CTR's and in 5 of the 115 two-stage LTR's. The stents were left in situ for between 3 and 8 weeks, with the majority used for 6 weeks. Nearly all of the patients who underwent two-stage LTR required further surgical intervention following their initial open operation, the common procedures being excision/laser ablation of a suprastomal granuloma (26 patients), removal of granulations (13 patients), balloon dilatation (29 patients) and closure of a tracheocutaneous fistula (31 patients).

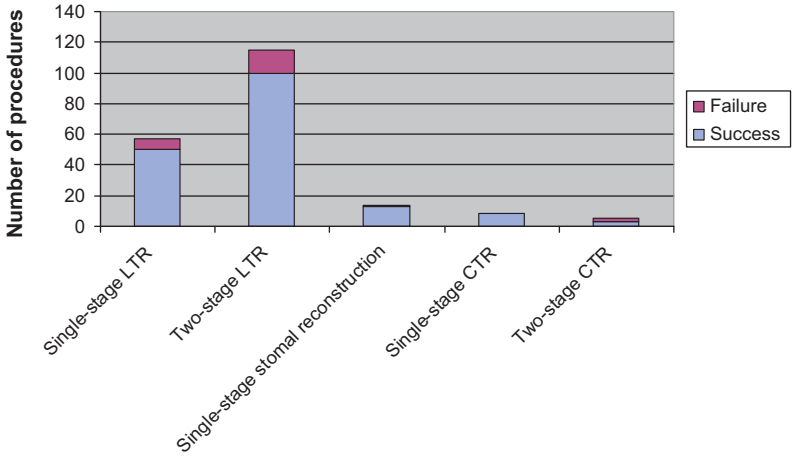

Fig. 1. Summary of results of all procedures.

Outcomes were assessed on the basis of symptomatic improvement, airway assessment and tracheostomy decannulation. For the whole group, at the completion of treatment 175/199 (87.9%) patients reported improvement in their symptoms, while 24/199 felt there was no improvement. Endoscopic airway assessment showed improvement in 176/199 patients, no change in 20/199 patients and a deterioration in 3/199 patients. 162/199 patients had a tracheostomy before airway reconstruction procedure: of these 140/162 (86.4%) were decannulated and 22/162 have not been decannulated. Objective assessment of voice before and after reconstruction is difficult in this group because of the young age of these patients and their multiple co-morbidities. Meaningful detailed statistical analysis was not possible in this group of patients as this group of patients is very heterogeneous and no subgroups could be formed because of the varying co-morbidities. As a result to be able to get outcomes using the whole patient group, the outcomes have been kept very simple and broad that can be applied to all these patients.

For 151 patients with subglottic stenosis, the outcomes according to the grade of stenosis were analysed (Fig. 2). For the two patients with grade 1 stenosis, both had improvement in symptoms as well as on airway examination (100% success). Of the 26 patients with grade 2 stenosis, symptomatic and endoscopic airway improvement was noted in 24 patients (92.3%) while the other 2 did not improve. For 117 patients with grade 3 stenosis, symptomatic and airway improvement was seen in 103 patients (88.1%). Amongst the group of 6 children with grade 4 stenosis, 5 (83.3%) had subjective and objective improvement.

Of the two-stage LTR procedures, 100/115 (86.9%) had their tracheostomy tube removed and 15/115 (13.1%) failed decannulation. In this group of patients 110/115 patients had stent placed postoperatively. Amongst these 2 patients had stent for 2 weeks, 14 patients had stent for 4 weeks, 68 for 6 weeks, 12 for 8 weeks and 4 for 12 weeks. The duration of stenting was the decision of the operating surgeon considering the nature of original pathology and the operation performed. There was no correlation between the duration of stenting and the outcomes, but detailed statistical analysis was not possible because of the variation amongst the patient groups. In the decannulated group 61/100 (61%) had the tube removed within 6 months of the LTR while in 39/100 decannulation was delayed beyond 6 months. The common reasons for a prolonged time to decannulation were suprastomal granuloma or collapse, tracheomalacia and re-stenosis. On analysing further the 15 children who failed decannulation, 5 of them had a congenital laryngeal web in addition to subglottic stenosis. Of these 8/15 had both an anterior and posterior graft indicating severe disease to start off. The reasons for failed decannulation included recurrence of the original pathology (10), graft prolapse (1) and suprastomal collapse (4).

Of the single-stage LTR group, 50/57 (87.7%) patients were better both symptomatically and on airway examination postoperatively. No symptomatic improvement was achieved in 7/57 (12.3%) patients and on airway examination 6/57 were same as before and one was worse. Analysing further these 6 children who were not successful, there was no single underlying cause to explain this. All these failed children had recurrence of stenosis/scarring.

Of the single-stage stomal reconstruction group, 13/14 (92.8%) were better symptomatically and on airway examination, and one patient had no improvement and had to have the tracheostomy re-opened.

Patients who underwent single-stage CTR had a better airway symptomatically and on examination in all cases (8/8). For the patients who underwent two-stage CTR, decannulation was achieved in 3/5 (60%) and failed in 2/5 (40%). The reason for failure in these cases was excessive granulations.

For the whole group, 15/199 (7.5%) patients underwent a revision LTR. On further analysis, revision LTR was required following 4/57 (7.1%) single-stage LTR's, 9/115 (7.8%) two-stage LTR's, 1/5 (20%) two-stage CTR's and 1/8 (12.5%) single-stage CTR's.

In this study complications occurred in 13/199 (6.5%). These were graft prolapse (9), pneumothorax (1), haematoma at the rib graft donor site (1), keloid (1) and excessive granulations (1).

4. Discussion

Laryngotracheal stenosis primarily occurs at the level of the subglottic and can be either congenital or acquired. The incidence of acquired subglottic stenosis following neonatal intubation is reported as <1% [6] and is usually a result of prolonged endotracheal intubation. The use of a large-diameter endotracheal tube even for a short period can lead to ischaemic ulceration and scarring. Poor fixation of the tube allowing movement also causes injury. Acquired subglottic stenosis is the predominant aetiology requiring surgery. Congenital subglottic stenosis is the second most common congenital laryngeal anomaly after laryngomalacia [1].

Measures taken to prevent subglottic stenosis include use of an uncuffed tube placed via a nasal route which reduces potential tube movement to a minimum. Injury to the airway is less likely with a small tube which allows a small leak on positive pressure ventilation. Good nursing care and hygiene are paramount [7].

Surgical correction of laryngotracheal stenosis was first attempted by Negus [8] using laryngofissure and dermal grafting in adults. Rethi [9] reported splitting the cricoid ring anteriorly and posteriorly in adults. Aboulker (1966) introduced a stent which could be wired into the tracheostomy tube [1]. Grahne [10] first described application of the Rethi procedure to children. At Great

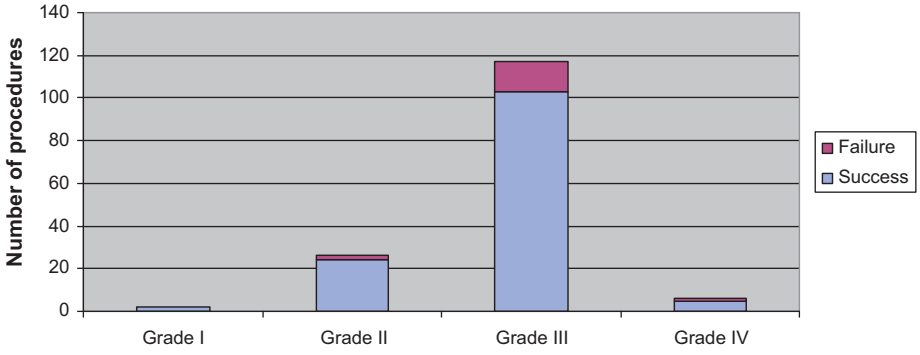

Fig. 2. Grade specific outcomes in subglottic stenosis.

Ormond Street Doig et al. [11] reported using a cartilage graft positioned in an anterior cricoid split to expand the cricoid ring and then Evans and Todd [12] described laryngotracheoplasty, which involved splitting the trachea and cricoid using a castellated incision and resuturing it in an open fashion over a stent. Cotton, unaware of Doig's publication in the German literature, separately introduced the laryngotracheal reconstruction with cartilage grafting (1978) after earlier animal work with Fearon and Cotton [13], and developed it into the preferred technique for framework expansion. In the 1970s and 1980s laryngotracheoplasty and laryngotracheal reconstruction were the main surgical procedures and Cotton and Evans [14] reported a 5-year follow up of laryngotracheal reconstruction in children. The next development was single-stage laryngotracheal reconstruction using an endotracheal tube as an airway and stent [15,16]. In 1993, Monnier et al. [17] published the first series of successful cricotracheal resections in children.

In general LTR with costal cartilage grafting is the mainstay of surgical management for grade 2 and mild grade 3 subglottic stenosis. Mild stenosis can be treated with an anterior graft and isolated posterior scarring or stenosis with a posterior graft. Severe stenosis needs both anterior and posterior grafts. In general LTR is a less complex and extensive procedure than CTR because tracheal mobilisation is not required. For grade 4 stenosis and severe grade 3 stenosis, the preferred option is CTR. By definition, in a two-stage LTR, the tracheostomy tube is kept in place at the conclusion of the procedure and the patient is decannulated several weeks later. Single-stage LTR allows immediate decannulation at the time of the reconstruction, or avoidance of tracheostomy altogether.

LTR with autologous cartilage grafts has been demonstrated to be an effective and reliable technique for the surgical management of subglottic stenosis. Good results have been reported from many different institutions around the world [18–21]. Generally two-stage LTR is advocated in children with complex multilevel stenosis, significant neurologic deficits, significant lung disease or cases where reintubation is anatomically difficult (e.g. craniofacial anomalies) [22]. Some authors perform two-stage LTR in cases with severe stenosis [20,23] although many others advocate partial cricotracheal resection in selected cases with grade III/IV subglottic stenosis where the disease is separated from the glottis by an adequate margin [1,24].

Following the reconstruction, the endotracheal tube functions as the stent in a single stage LTR, while a stent is placed in situ after a two stage LTR. The optimal duration of stenting is unknown. A detailed analysis has been reported by Hartley et al. [25], where no correlation was found between the duration of stenting and the outcomes in terms of reintubation or tracheostomy. Similarly in our study, no correlation was found between the duration of stenting and the final outcome, as the outcome was influenced by multiple other factors in individual patients.

Paediatric airway stenosis presents with varying degrees of severity and significant comorbidities. The choice of operation for reconstructing the airway must be considered carefully by the surgeon depending upon which modality is considered likely to achieve safe and expeditious surgical reconstruction in each individual patient.

The final results of laryngotracheal reconstruction are generally expressed in terms of decannulation rates. Most of the patients who have a tracheostomy undergo surgery primarily to get rid of the tracheostomy, and thus decannulation does become an important outcome measure. For those patients who did not have a tracheostomy before the LTR, the outcomes were based on the improvement in their symptoms and the airway endoscopy findings before and after the procedure.

The previous 10-year review of airway reconstruction at Great Ormond Street Hospital in the 1980s reported on 149 operations, of which 75 were laryngotracheoplasties and 74 were laryngotracheal reconstructions with an overall success rate of 83% [2]. This current 10-year review reports on 199 airway reconstructions at the same department since 2000. Laryngotracheoplasties were not performed at all in this current series, but CTR was used for some of the most severe stenosis, and adjuvant endoscopic treatment was widely employed (especially balloon dilatation), with an overall success rate of 87%. It is interesting that increasingly sophisticated surgical techniques have made little difference to the overall success rate in our institution.

Many studies have been published looking at the outcomes of airway reconstruction. Saunders et al. [26] reported an overall decannulation rate of 91.4% after single-stage LTR and 61.8% after two-stage LTR. Hartnick et al. [27] and Agrawal et al. [23] also reported higher operation-specific decannulation rates with single-stage LTR than two-stage LTR. This has been explained on the grounds that children undergoing two-stage LTR have a more severe disease and have more medical co-morbidities. Single-stage LTR is considered the procedure of choice for uncomplicated subglottic stenosis, especially if there is no pre-existing tracheostomy. On detailed analysis of failures in this very heterogeneous group, there was not a single common factor but multiple factors that led to the reconstruction not being successful.

As shown in other studies [23,26,27] children undergoing two-stage LTR had significantly worse disease to start with. However there was no statistically significant difference in final outcomes between single-stage (87.7%) and two-stage LTR (86.9%) in this study.

5. Conclusions

Subglottic stenosis in children needs to be approached on the basis of the nature and severity of the stenosis and the individual patient's general health. Good outcomes are achieved with both LTR and CTR, and with both single-stage and two-stage LTR. Surgery must be tailored to the needs of each individual patient. The overall success rate has increased only marginally in our institution over the last 20 years.

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