Paediatric Airway Reconstruction: A Preliminary Study

Z Amin, MMed (ORL-HNS)*, S A H Suzina, MMed (ORL-HNS)**

Department of Otorhinolaryngology-Head & Neck Surgery, *Kulliyyah of Medicine, International Islamic University Malaysia, Jalan Hospital Campus, 25100 Kuantan, Pahang, **School of Medical Sciences, Hospital Universiti Sains Malaysia, 16150 Kubang Kerian, Kelantan, Malaysia

SUMMARY

To set the foundation for developing a centre for airway reconstruction, we performed a retrospective database review of patients operated at a tertiary-care university hospital. Over the past 3-year period from 2004 onwards, five paediatric cases of airway reconstruction procedures were performed. All cases had a two stages laryngotracheal reconstruction (TSLTR) for laryngotracheal stenosis (LTS). All patients were children below 15 years and the mean age was 9 years. Only one patient had a Grade IV Myer-Cotton[1] stenosis, the rest all had Grade III stenosis. Three out of four of the Grade III stenosis patients were successfully decannulated within one year, the other one died of causes unrelated to LTS. The grade IV patient was still under follow-up and surgery was done only recently. This paper highlights the complexity of managing LTS in the paediatric age group and recommends the use of LTR with rib graft as a choice for the management of LTS.

KEY WORDS:
Paediatric, Laryngotracheal reconstruction, Laryngotracheal stenosis

INTRODUCTION

Laryngotracheal stenosis (LTS) remains one of the most complex and difficult problems facing the laryngologist. LTS is defined as narrowing of the airway, either congenital or acquired, that may affect the glottis, subglottis and/or trachea. Acquired causes such as trauma to the airway, either from an intubation injury or from an external source like blunt trauma or tracheostomy account for the majority of cases.

Although evaluation and treatment options continue to evolve, the precise identification of the location and choosing the appropriate procedure for its correction are the two main challenges in cases of LTS. Reconstruction of the airway has rapidly evolved since the last 30 years after Fearon and Cotton[2] introduced the concept of cartilage augmentation and laryngotracheal reconstruction (LTR). Since then, there have been numerous publications reporting surgical outcomes for the various operative procedures for airway reconstruction.

LTR is a relatively new operation conducted in Hospital Universiti Sains Malaysia (HUSM), beginning only in the last two and a half years since May 2004. In Malaysia, this operation is also at an infant stage and there were not many papers published to date regarding the series of LTR in Malaysia and in South-East Asia region in general. LTR can be classified into two broad categories: augmentation procedures and cricotracheal resection anastomosis (CTR). In HUSM, we are more familiar with augmentation procedures in that all of our LTR cases performed so far were using this technique.

OBJECTIVE

To analyse the procedures of paediatric airway reconstruction performed at HUSM namely the technique, success rate, complications and to relate with the series done elsewhere. This review is targeted as a personal learning curve for better management of our patients in the future as a centre for airway reconstruction.

MATERIALS AND METHODS

Retrospective clinical records review was done on all cases of LTR performed in HUSM since May 2004 until November 2006. The data included demographic information on each patient, causes and severity of subglottic stenosis, other mode of treatment besides LTR, details of the surgery performed and the intra-operative findings. Information on the number of procedures/surgeries performed, post-surgical follow-up, complications and the date of decannulation for all patients was made available. Supplementary information was also obtained from the same surgeon who conducted all the operations and added to the data taken. An inclusion criteria for age is defined as below the age of 15 years at the time of operation.

The surgical technique used for LTR here was a cartilage augmentation procedure. This involves increasing the subglottic and tracheal lumen diameter by placing a homogenous cartilage graft into the edges of a surgically created split in the anterior cricoid and upper tracheal cartilages.

RESULTS

A total of five paediatric cases of LTR were performed since May 2004 in HUSM. The average number of paediatric LTR cases was two per year. The age range of the patients at the time of airway reconstruction was from 14 months to 13 years and the average age was nine years. All of the subjects were male except for one, and all were Malays.

A total of 28 procedures were performed on these five patients under general anaesthesiae, an average of 5-6 procedures per patient. Those procedures include diagnostic direct

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Corresponding Author: Zamzil Amin Asha'ari, Department of Otorhinolaryngology-Head & Neck Surgery, Kulliyyah of Medicine, International Islamic University Malaysia, Jalan Hospital Campus, 25100, Kuantan, Pahang Email: zamzilamin@yahoo.com

laryngoscopy and bronchoscopy, endoscopic dilatation, granulation tissue excision either with cold instrumentation or carbon dioxide laser, tracheostomy, LTR, decannulation and stent removal. All of the procedures were done by the same surgeon or at least done on her presence in the operating theatre.

The causes of stenosis were acquired in all cases except for one case with congenital subglottic stenosis. The acquired causes were mostly due to recurrent, traumatic endotracheal intubation and prolonged ventilation. Each subject's airway was assessed before reconstruction, and all of them had single stenosis at subglottic region, except for one child with congenital subglottic stenosis displaying at least two sites of stenosis. The shape of his cricoid cartilage was unusually triangular and besides a subglottic stenosis 5mm below the vocal folds, he had another stenotic segment at the level of the lower border of cricoid cartilage and upper first tracheal cartilage. The average length of the stenotic segment in all subjects was 11.4 mm, with a range from 5 to 30 mm. All subjects had stenosis below the vocal cord and the average distance from vocal cord was 11 mm.

All of the cases were subjected to tracheostomy prior to LTR as a life saving procedures. All subjects had at least Grade III Myer-Cotton[1] stenosis and only one of them had complete Grade IV stenosis. All cases had cartilage augmentation LTR two-stage procedure; the graft was placed at the anterior cricoid cartilage. All patients had graft taken from right costal cartilage. Stenting was only done in the patient with Grade IV stenosis. The stent used was an appropriate sized endotracheal tube, cut into the exact size to cover the length of the stenotic segment. The top end of the tube was sutured so that it becomes funnel shaped and was placed just slightly below rima glottidis. The stent was fixed in position by a stay suture that was brought to the outside of the skin.

Table I shows the overall morphology and clinical data of all subjects including the age of patients at time of operation, degree of stenosis, date of operation, date of decannulation and the overall decannulation time. Overall, the quickest decannulation time was within seven months and the longest was after 12 months with an average decannulation time of nine months. In one case, the tracheostomy could not be taken off because of underlying progressive neurological deficit causing inability of the patient to maintain airway protection. She succumbed to death due to her underlying illness two years after LTR with tracheostomy still in situ.

DISCUSSION

Since 1994, the most common grading system used to assess subglottic stenosis has been that proposed by Myer *et al.*[1], which is based on endotracheal tube sizes, which was used to grade the severity of our patients in this review. Definitive diagnosis of LTS is made via direct laryngoscopy and bronchoscopy under general anaesthesiae to closely evaluate the extent of airway narrowing. A complete diagnostic work-up must include the exploration of the entire upper airway, from the supraglottis to the carina, defining not only the site of the obstruction but also its severity and the coexistence of associated complicating factors. Vocal cord paralysis, tracheomalacia, and tracheoesophageal fistula may complicate the management of patients with LTS and should be precisely identified. Palpation of the interarytenoid region may be required to elucidate interarytenoid adhesions of the vocal fold mucosa.

Rapid assessment and securing the airway is crucial in cases of severe LTS. This either requires placement of an endotracheal tube when possible, or often in severe cases of stenosis, placement of a tracheostomy tube below the level of obstruction. In all our cases so far, we had to place tracheostomy tube as a life saving procedure because the severity of airway stenosis made intubation impossible.

Multiple options are available for the treatment of LTS. These include intralaryngeal steroid injections, endoscopic dilatations, endoscopic removal of the obstruction via laser or cold instrumentation, and open surgical techniques. A technique for the management of airway stenosis can be difficult, and this choice must be based on specific goals. The ultimate goals should be the establishment of an adequate airway that allows a patient to participate in normal daily living activities without a tracheostomy, using the minimal number of procedures needed with minimal morbidity and duration of hospitalization. Although mild to moderate airway stenosis are now successfully treated with endoscopic procedures, severe stenosis still require open reconstruction and meticulous post-operative intensive care.

All patients in our series were treated with the augmentation LTR. The original two-stage laryngotracheal reconstruction (TSLTR) was introduced in 1972 by Fearon and Cotton[2]. Single-stage laryngotracheal reconstruction (SSLTR) was first reported by Prescott[3]. Both types of LTR expand the airway lumen with a cartilage graft. With SSLTR, the tracheostomy is closed during the procedure and the reconstructed airway is stented with an endotracheal tube (ETT). With TSLTR the tracheostomy was kept after the first stage and later decannulated after some time in a second stage procedure. Logistic regression analysis revealed no statistically significant differences in overall extubation rates for either the SSLTR or TSLTR[4]. However, a review of cases with SSLTR in Great Ormond Street Hospital in 1999 showed that the procedure is associated with higher incidence of extubation complications e.g. bleeding, reintubation, or subcutaneous emphysema and higher risk of failure, particularly in children with prematurity and low body weight[5]. No significant correlation was found between the number of day's stented and the reintubation rate or the postoperative tracheostomy rate in SSLTR procedure[6].

Comparison of data between procedures and between reports remains difficult, as the definition of success tend to differ from study to study. One of the most common outcome measures identified is the decannulation rate for patients with preoperative tracheostomy tubes. By this measure alone, airway expansion grafting procedures have demonstrated high levels of success. Cotton *et al.*[7] reported decannulation rates above 90% for grade II and III lesions and above 70% for grade IV lesions. Great Ormond Street Hospital in London demonstrated decannulation rates with grade II and III lesions of approximately 80% and with grade IV lesions of 50%[8]. An update of the Cincinnati group's data in 2000 increased their overall decannulation rate for SSLTR to 96%[9].

Data from Iowa in 2004 continued to support this positive outcome, as grade II and III lesions treated with expansion grafting showed a nearly 100% decannulation rate and a single patient with a grade IV lesion who was treated with expansion grafting also attained decannulation[10]. We also experienced the same success rate in our cases of grade III stenosis, when all were decannulated within one year from the LTR, apart from one case with underlying medical illnesses who was not able to maintain airway protection without tracheostomy. On the other hand, the prognosis of our grade IV patient remains to be seen, as he had LTR performed recently. His latest endoscopic examination was encouraging, we were able to remove the stent and the graft seemed to be well taken up with minimal granulation tissue seen. He was planned for decannulation at that time but had to postpone due to patient's domestic problem.

Decannulation rates alone are often inadequate as a measure of success to LTR. Many of the patients who ultimately attained decannulation did so only after undergoing multiple procedures over long periods of time and this is infrequently discussed in the reporting literature. Few available studies do offer data on this subject demonstrated the need for multiple open surgical expansion procedures in 20-54% of grade III [10,11] and 70%-80% of grade IV stenosis[11,12]. As each procedure certainly requires long periods of healing with operative morbidities, these operative events represent long treatment times over many months and sometimes years. Although the ultimate goal of an adequate airway is frequently achieved, the data showed that we are still less successful in meeting our secondary goals of a single procedure and minimal treatment times.

Average number of procedures in many oversea series was less than two procedures per patient [4,11], much lower if compared to our series of 5.6 procedures per patient. While this may reflect the levels of experience between the centres, it could also be due to our inclusion of all procedures done under general anaesthesiae albeit for simple procedures like direct laryngoscopy assessment. Another explanation is that our series only included more severe cases of grade III and above, which undoubtedly carry a more difficult recovery and higher complications from the surgery.

Not many series discussed in detail regarding the decannulation time, particularly because there are many factors influencing it rather than the surgery itself. Gavilan *et al.* [4] in their 60 series of paediatric LTR found the median decannulation time between operation and decannulation was one and a half years, with a range of nine days to more than 12 years. Average decannulation time for our small sample study was nine months. In our cases, we found that parental anxiety and lack of awareness and knowledge of the child's problem, together with living far away from hospital influence the delay in decannulation in most of the patients.

The commonest and a particularly frustrating complication we encountered in all of our patients was the development of granulation tissue at the edges of reconstructed cartilage. This can be troublesome for both patients and surgeon, and was the cause for repeated procedures under general anaesthesia to be corrected. We have tried application of mitomycin C to the area of reconstructed cartilage to reduce granulation tissue formation and, despite overwhelming recommendations by many authors; we found that the problem still occurred in all our patients after some time. We postulated that age factor may play a role in that older patient tends to have more problem with the granulation tissue formation. This is supported by a larger series study by Gavilan *et al.* [4] who found that age is the only parameter with a significant effect on the success rate among patients who completed LTR. The study further states that factors other than age, gender, aetiology of the stenosis (congenital or acquired), preoperative diameter of the stricture, length of the stenotic segment, and site of stenosis (isolated versus combined) had no significant effect on the outcome of the procedure. Despite lack of significance, they stated that a large number of failures appeared in the group of stenosis produced by acquired causes, longer stenotic segment (more than 20 mm) and in combined stenosis.

Another area worth discussing is regarding stenting following TSLTR. There is no hard and fast rule acceptable and opinion varies regarding this in different studies. In our grade III stenosis, we did not stent all our patients and all patients were successfully decannulated except for one with grade IV stenosis requiring stenting. Choices for stenting vary; among the popular one is by using a Montgomery T-tube that is maintained in place for 1 to 2 years[13]. Airway stents, which are supposed to counteract strictures, also seem to promote the development of secondary stenoses[14]. One author described the adaptation of a Montgomery T tube to incorporate an uncuffed fenestrated Shiley tracheostomy tube (combined trache-stent) in management of complex subglottic stenosis, and claimed successful decannulation after six weeks with minimal granulation tissue [13].

In general, successful decannulation rate was reported to be higher in cricotracheal resection anastomosis (CTR) than augmentation LTR [9,14]. However, the CTR can only be used when the stenotic lesion is well below the vocal cord. As it is vital to preserve the vocal folds, in cases where the stenotic segment extends to the vocal folds, an augmentation LTR needs to be performed. Among the significant complications of LTR and CTR include infection particularly from Respiratory Syncitio Virus (RSV) bronchiolitis, restenosis of the subglottic larynx and delayed healing caused by reflux[14]. Therefore it is proposed that patients should be treated empirically for reflux until the subglottic larynx has healed and all patients should have a preoperative tracheal culture and receive culture-directed perioperative antibiotics. Intraoperatively, an airtight seal of the cartilage graft should also be obtained[14].

Considering that we are still at an infantile stage of developing a centre for airway reconstruction, our overall results are encouraging. In the future, we may have to consider doing a more technically challenging posterior cricoid split and grafting, especially in cases of Grade IV stenosis which may reduce the incidence of re-stenosis and reduce overall total number of procedures per patient. Also, since the worldwide results of CTR are proven superior, continuous development of trained personnel in this technique is also crucial. It goes without saying that multidisciplinary involvement of anaesthesiologists, respiratory physicians, paediatricians, intensivists, speech pathologist and physiotherapists is mandatory in airway reconstruction.

Table I: Morphologic and clinical data of all subjects

	Case 1	Case 2	Case 3	Case 4	Case 5
Age at operation	6 years	20 months	13 years	14 months	5 years
Sex	Male	Male	Female	Male	Male
Race	Malay	Malay	Malay	Malay	Malay
Degree of stenosis	Grade IV	Grade III	Grade III	Grade III	Grade III
Type of stenosis	Acquired	Congenital	Acquired	Congenital	Acquired
Length of stenotic segment	30 mm	5 mm	10 mm	5 mm	7 mm
Distant of upper part of stenotic segment to vocal cord	10 mm	15 mm	10 mm	10 mm	10 mm
Operation date	3/8/2006	7/3/2005	29/5/2004	11/4/2005	16/8/2005
Stenting	Yes	No	No	No	No
No. of procedures performed	7	6	4	6	5
Decannulation date	Not yet	4/10/2005	Died prior to decannulation	5/11/2006	20/8/2006
Decannulation time	-	7 months	-	9 months	1 year

CONCLUSION

The challenge of airway stenosis continues to pose a formidable task to the attending otorhinolaryngologists. Our patient population will continue to increase and become more complex with the advancement of intubation and ventilation technique to improve patient survival. Decision-making regarding choices of treatment in LTS must be individualized, but must also be based on a set of goals that result in an adequate airway in the quickest and safest way possible. LTR remains a challenging set of procedures in which multiple operations may be required to achieve eventual extubation or decannulation. Patients with Myer-Cotton grade III or IV disease continue to represent a significant challenge, and refinements of techniques continue to develop to address this. A centre for airway reconstruction must include trained surgeons and good multidisciplinary collaboration. As our surgical options and techniques expand, continued evaluation of our approaches will be critical.

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