

Supraglottoplasty for Laryngomalacia: Who Will Benefit?

Azida Zainal, Bee See Goh, Abdullah Sani Mohamed, Department of Otorhinolaryngology-Head and Neck Surgery, Faculty of Medicine, Universiti Kebangsaan Malaysia Medical Centre, Kuala Lumpur, Malaysia.

OBJECTIVE: Laryngomalacia is the most common cause of neonatal and infantile stridor. The aim of this study was to assess the outcome of surgical intervention in children with laryngomalacia.

METHODS: Between January 1998 and December 2008, 15 children with laryngomalacia underwent surgical intervention at the Universiti Kebangsaan Malaysia Medical Centre, from which only eight case notes were available. These were retrospectively reviewed for demographic data, symptoms, comorbidities, operative technique, postoperative recovery, complications, length of hospital stay including intensive care unit (ICU) care, and resolution of symptoms.

RESULTS: Patients consisted of seven males and one female. One patient underwent three procedures, resulting in a total of 10 procedures for this study. The mean age was 15.6 months (range: 2–39 months). The most common indication for surgery was severe stridor resulting in failure to thrive. Intra-operatively, all patients were found to have short aryepiglottic folds, and four also had redundant arytenoid mucosa. Supraglottoplasty was performed in 10 patients: three by cold instruments and seven by laser. Successful extubation was achieved in the operating theatre in eight patients while the other two were extubated in the ICU on the same day. Postoperative ICU nursing was required in six patients: three for up to 3 days, and three for longer periods because of medical problems. Resolution of stridor was complete in four patients, partial in one, and no difference in five. Two patients defaulted follow-up. There were no postoperative complications from the procedures. The average length of follow-up was 15 weeks (range: 12 days to 7 years).

CONCLUSION: Supraglottoplasty remains an effective method to treat severe laryngomalacia. Patients who will benefit most are those with severe laryngomalacia that is uncomplicated by neurological conditions or multiple medical problems. In our institution, early extubation is the norm, and a significant number of patients can be nursed in the normal wards and be discharged within 48 hours of the procedure. [*Asian J Surg* 2011;34(2):92–96]

Key Words: intensive care unit, laryngomalacia, stridor, supraglottoplasty

Introduction

Laryngomalacia is the most common cause of neonatal and infantile stridor, comprising 59.8% of all congenital

laryngeal diseases with airway obstruction.¹ Children usually present with inspiratory stridor; severe cases may be associated with cyanosis, choking, feeding difficulties, and failure to thrive.

Address correspondence and reprint requests to Dr Bee See Goh, Department of Otorhinolaryngology-Head and Neck Surgery, Universiti Kebangsaan Malaysia Medical Centre, Jalan Yaacob Latif, 56000 Cheras, Kuala Lumpur, Malaysia. E-mail: irenegbs@yahoo.com • Received: Dec 8, 2009 • Revised: Aug 26, 2010 • Accepted: Jan 3, 2011

© 2011 Elsevier. All rights reserved.

There are various classification systems for laryngomalacia. McSwiney et al² described three anatomical variations seen in laryngomalacia: (1) omega-shaped epiglottis, (2) short aryepiglottic folds, and (3) bulky and forwardly prolapsed arytenoids. Holinger and Konior³ used the direction of supraglottic collapse to define laryngomalacia: posterolateral (Type A), complete (Type B), or anterior (Type C). Kay and Goldsmith⁴ classified it as follows: (1) foreshortened or tight aryepiglottic folds, (2) presence of soft tissue in the supraglottis, or (3) consequence of other aetiologies, such as neuromuscular disorders.

The current standard for surgical treatment is some form of supraglottoplasty, which includes one or more of the following procedures: division of the aryepiglottic folds, debulking of the arytenoid mucosa and cuneiform cartilages, making linear incisions on the lingual surface of the epiglottis, or trimming of the lateral edges of the epiglottis. These may be performed by micro-dissection with cold instruments or with a CO_2 laser. The aim of these procedures is to widen the supraglottis to alleviate the obstruction that occurs during inspiration.

We herein present our supraglottoplasty series and discuss the outcome of surgical intervention.

Patients and methods

At the Universiti Kebangsaan Malaysia Medical Centre, every patient referred for stridor undergoes diagnostic flexible nasopharyngolaryngoscopy after the airway is stabilised. Laryngomalacia cases are stratified into mild, moderate, and severe. Mild cases are treated conservatively by observation on an outpatient basis. Moderate and severe cases are planned for surgery. The instruments used are a reflection of the availability of instruments and surgeon preference.

All supraglottoplasty cases between January 1998 and December 2008 were identified from operating theatre records. Patient case notes were then retrospectively reviewed and the following data collected: age, sex, symptoms, associated medical problems, and clinical signs of respiratory distress. Details of the procedure were also analysed in terms of method of anaesthesia, anatomical findings of the supraglottis, other laryngeal pathology, procedure performed, method used (laser or cold instruments), and laterality (unilateral or bilateral). Postoperative data included medications administered (systemic steroids, anti-reflux medication), resolution of symptoms, complications, length of stay in hospital including intensive care unit (ICU) care, further procedures required, and length of follow-up.

Surgical technique

The standard anaesthetic technique used was general anaesthesia with endotracheal intubation. A paediatric laryngoscope was positioned in the vallecula and suspended. An operating microscope was used. Jet ventilation was used in two patients. Laser was available during the latter part of our study period, and for who underwent the use of laser, a CO_2 laser system was mounted on the operating microscope. An examination under anaesthesia was first performed by examining the supraglottis, glottis, subglottis, and trachea to confirm the diagnosis of laryngomalacia and rule out associated airway anomalies. Redundant arytenoid mucosa (Figure 1) was removed with a microdebrider or ablated with the laser. Aryepiglottoplasty was performed for short aryepiglottic folds (Figure 2) using fine grasping forceps and laryngeal scissors (Figure 3)



Figure 1. Redundant arytenoid mucosa.



Figure 2. Tight aryepiglottic folds.



Figure 3. Aryepiglottoplasty with cold instruments.

or the laser (Figure 4). The extent of the procedure depended on the location of redundant tissue prolapsing into the airway, from resection of arytenoid mucosa to division of aryepiglottic folds or both. Haemostasis was carried out using adrenaline-soaked strips of ribbon gauze. Intravenous dexamethasone was routinely administered during the procedure.

Results

Between January 1998 and December 2008, 15 patients underwent supraglottoplasty at the Universiti Kebangsaan Malaysia Medical Centre. Because of missing or destroyed records, we were able to review only eight cases.

Of these, seven were male and one was female. Their ages ranged from 2 months to 39 months (mean age: 15.6 months). One patient underwent three procedures, giving a total of 10 cases. Neurological problems were present in three patients (two cerebral palsy and one global developmental delay) and cardiorespiratory problems in two (one patent ductus arteriosus and one atrial septal defect with hyperactive airway), and one patient had gastrooesophageal reflux.

The diagnosis of laryngomalacia was made preoperatively from the clinical history and flexible laryngoscopic examination. In all patients, the indication for surgery was moderate to severe stridor that affected the child's growth. Feeding difficulties and severe aspiration in neonates can be a presenting symptom of laryngomalacia even without stridor; however, none of the patients in our series fell into this category. Six patients had stridor with failure to thrive, and two had feeding difficulties (one had prolonged feeding and one had regurgitation and was gasping for breath during feeding). This last



Figure 4. Aryepiglottoplasty with laser.

patient was confirmed to have gastro-oesophageal reflux disease pre-operatively. Of note, symptoms suggestive of gastro-oesophageal reflex were poorly documented, and pH monitoring is not routinely performed at our centre.

There were 10 cases of supraglottoplasty; a CO_2 laser was used for seven, and cold instruments were used for three. All were carried out electively. The operative findings, according to the McSwiney classification were as follows: three patients had isolated Type 2 laryngomalacia (short aryepiglottic folds), one had a combination of Types 1 and 2 laryngomalacia (omega-shaped epiglottis and short aryepiglottic folds), and four had a combination of Types 2 and 3 laryngomalacia (short aryepiglottic folds and redundant arytenoid tissue).

All eight patients had division of the aryepiglottic folds. Redundant mucosa was ablated with the laser in two patients (one unilateral and one bilateral) and microdebrided in one patient (bilateral).

Successful extubation was achieved in the operating theatre in eight patients, while the other two were extubated in the ICU on the same day. Three patients required re-intubation because of respiratory distress (two within a few hours postoperatively and one on the 1st postoperative day). They were diagnosed with aspiration pneumonia, septicaemia, and bronchopneumonia, respectively. One patient was re-admitted with supraglottitis 9 days after being discharged. He was treated conservatively with antibiotics and nebulisers with a good outcome.

Of the 10 cases, 4 had complete resolution; 3 of these 4 patients within 24 hours and 1 patient on the 5th postoperative day. Partial resolution was achieved in one patient.

There was no difference in stridor in four patients. The child who had septicaemia was nursed in ICU for 11 days. Upon discharge, there was no improvement in the stridor. Interestingly, he had no comorbidities. Unfortunately, he did not return for follow-up. One child had a history of repeated admissions with aspiration pneumonia and multiple medical problems (atrial septal defect, hyperactive airway disease, congenital hypothyroidism, and Down syndrome). Following failed extubation, he underwent direct laryngoscopy and bronchoscopy, which revealed a tongue of granulation tissue consistent with intubation trauma. There was no difference in his stridor upon discharge. He was admitted again for pneumonia 2 months later. One month after being discharged, he died at home. The cause of death was unknown because the family declined a post-mortem examination.

One of the children with cerebral palsy developed bronchopneumonia postoperatively. There was no difference in the stridor upon discharge 1 month after the procedure. He was re-admitted 2 months later with respiratory distress which required a tracheostomy. Direct laryngoscopy was performed; findings were normal. This child still has a long-term tracheostomy. The other child with cerebral palsy had three procedures. The first was performed at 1 year of age and the second at 2 years of age; neither improved her stridor or failure to thrive. She underwent another procedure at 8 years of age and had complete resolution. After being discharged on the 1st postoperative day, she defaulted follow-up.

Of the four children who experienced feeding difficulties, two had documented improvement and weight gain after the procedure. Unfortunately, documentation was lacking in the other two patients.

Three patients were discharged the day after procedure, and another three were discharged on the 2nd day. The mean follow-up duration was 15 weeks (12 days to 7 years).

Discussion

Laryngomalacia is a self-limiting condition that usually resolves by 2 years of age.⁵ Endoscopic procedures on the supraglottis are currently the mainstay of treatment for this condition. The surgical method used for supraglot-toplasty at our centre reflects the equipment available at the time of surgery. Laser is now the preferred method because it is associated with less bleeding.

It is difficult to directly compare outcomes of surgery because many different outcome measures have been

used,⁶ including the degree of resolution of stridor (partial or complete), "improvement", need for a tracheostomy, number of subsequent procedures required, and weight gained. Stridor is also difficult to standardise because of the subjectivity of its assessment. The optimal endpoint for assessing the resolution of stridor following intervention is not clear: is it within a few hours of the procedure, 1 week later, or 1 month later? Weight percentile charts have also been used as a primary outcome measure.⁷ In our series, improvement was documented in five (50%) patients. Of the remaining patients, two had associated neuromuscular conditions and one had multiple medical problems.

The neuromuscular immaturity theory, which is currently the most accepted theory for the development of laryngomalacia, states that laryngeal hypotonia occurs because of delayed neuromuscular control of the larynx. Evidence of this is seen in previous studies^{8,9} in which there was poor response to surgery in patients with cerebral palsy and other neurological conditions. In our series, neither child with cerebral palsy improved with surgical intervention. The patients who seemed to benefit were those with simple laryngomalacia uncomplicated by neurological conditions.

The following complications have been reported in previous studies: intra-operative haemorrhage, worsening of stridor, postoperative intubation, aspiration, supraglottic stenosis, granulomas, and death.^{6,10} Kuo et al⁸ found that choking was a common symptom and that 66.7% of these patients had aspiration pneumonia. They recommended increasing the duration of nasogastric feeding to avoid these complications. We had one case of aspiration pneumonia; however, this child had previous admissions for aspiration, and it is difficult to conclude whether surgery was a contributory factor.

There is no consensus on postoperative care for patients undergoing supraglottoplasty. Some units electively ventilate their patients for 24–48 hours while other units practice early extubation with no complications. In our practice, extubation in the operating theatre is the norm. The decision for ICU nursing must be made on a caseto-case basis depending on the patient's comorbidities. Patients with no co-morbidities tend to have uneventful recoveries.

Our study had two limitations. Because this was a retrospective study, it was difficult to obtain a detailed assessment of symptoms, especially postoperatively. Objective measures, such as weight gain, were also not available in most of the case notes.

Conclusion

Supraglottoplasty remains an effective method to treat severe laryngomalacia. Patients who will benefit most are those with severe laryngomalacia that is uncomplicated by neurological conditions or multiple medical problems. At our centre, early extubation is the norm and a significant number of patients can be nursed in the normal wards and be discharged within 48 hours of the procedure.

References

- 1. Cotton RT, Richardson MA. Congenital laryngeal anomalies. *Otolaryngol Clin North Am* 1981;14:203–18.
- 2. McSwiney PF, Cavanagh NP, Languth P. Outcome in congenital stridor (laryngomalacia). *Arch Dis Child* 1977;52:215–8.
- 3. Holinger LD, Konior RJ. Surgical management of severe laryngomalacia. *Laryngoscope* 1989;99:136–42.

- 4. Kay DJ, Goldsmith AJ. Laryngomalacia: a classification system and surgical treatment strategy. *Ear Nose Throat J* 2006;85: 328-31, 336.
- Roger G, Denoyelle F, Triglia JM, et al. Severe laryngomalacia: surgical indications and results in 115 patients. *Laryngoscope* 1995;105:1111–7.
- Toynton SC, Saunders M, Bailey C. Aryepiglottoplasty for laryngomalacia: 100 consecutive cases. *J Laryngol Otol* 2001;115: 35–8.
- Whymark AD, Clement A, Kubba H, et al. Laser epiglottopexy for laryngomalacia. *Arch Otolaryngol Head Neck Surg* 2006;132: 978–82.
- Kuo-Sheng L, Bo-Nien C, Cheng-Chien Y, et al. CO₂ laser supraglottoplasty for severe laryngomalacia: a study of symptomatic improvement. *Int J Pediatr Otorhinolaryngol* 2007;71: 889–95.
- Senders CW, Navarrete EG. Laser supraglottoplasty for laryngomalacia: are specific anatomical defects more influential than associated anomalies on outcome? *Int J Pediatr Otorhinolaryngol* 2001;57:235–44.
- Denoyelle F, Mondain M, Gresillon N, et al. Failures and complications of supraglottoplasty in children. *Arch Otolaryngol Head Neck Surg* 2003;129:1077–80.