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Aryepiglottoplasty for laryngomalacia: 100 consecutive cases

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Abstract

A retrospective review of the notes of 100 consecutive patients who had undergone aryepiglottoplasty for laryngomalacia, at Great Ormond Street Hospital for Children, was undertaken. Fifty-six were male, 44 female and 47 were under three months of age. Indications for surgery were oxygen desaturation below 92 per cent and feeding difficulties causing failure to thrive. Forty-seven patients had other pathology contributing to their airway compromise or feeding problems. Improvement in stridor after one month was achieved in 86/91 (94.5 per cent) being abolished completely in 50/91 (55 per cent). Of the 25 per cent of patients whose symptoms took more than one week to resolve, 16/22 (63.6 per cent) were later found to have a serious neurological condition. Feeding was improved in 42 of 58 patients (72.4 per cent) who had a pre-operative feeding difficulty. The complication rate was low, with only five out of 86 (10 per cent) experiencing initial worsening of the airway and six per cent having aspiration of early feeds before improvement occurred.

Endoscopic aryepiglottoplasty remains the operation of choice for patients with severe laryngomalacia, however, in the presence of neurological disease surgery is less likely to be successful.

Key words: Larynx; Airway Obstruction; Surgical Procedures, Operative; Endoscopy

Introduction

Laryngomalacia is the most common cause of neonatal and infantile stridor and is responsible for 59.8 per cent of congenital laryngeal abnormalities presenting with airway obstruction.[1] The supraglottic larynx collapses into the airway during inspiration. McSwiney *et al.*[2] described three variations of the normal supraglottic anatomy which predispose to laryngomalacia: (1) a long, curled (omega-shaped) epiglottis which prolapses posteriorly on inspiration, (2) short aryepiglottic folds, and (3) bulky arytenoids capable of prolapsing forwards on inspiration. The endoscopic appearance is often a combination of these features.

In its most mild form the stridor can be heard only during exertion and sometimes when asleep; 90 per cent of cases fall into this category, with normal feeding and weight gain. No intervention is needed and the stridor usually resolves by the age of two years. In 10 per cent of patients severe malacia results in stridor at rest. This is often postitional, being worse when supine, with recession and sometimes pectus excavatum.

Feeding is frequently affected. The child takes longer to complete a feed and may interrupt feeding to breathe. Increasing severity generates high negative intrathoracic pressures which may result in reflux with choking, aspiration and recurrent chest infections. Oxygen desaturation may occur during exertion and feeding, but also in severe cases when asleep and even at rest. Very severe untreated cases may develop cor pulmonale[3] that may even prove fatal. Co-existing congenital airway and non-airway pathology is well acknowledged. This most commonly includes tracheomalacia and tracheobronchomalacia, which both may be primary, or secondary to anomalous compressing vessels.

These symptoms are usually of great concern to both parent and doctor and may result in ventilatory insufficiency with oxygen desaturation, and a need for airway support, particularly if an upper respiratory tract infection supervenes. In severe cases, there is failure to thrive due to inadequate dietary intake, coupled with increased energy expenditure owing to the greater work of breathing.

In the last few years, the operation of endoscopic aryepiglottoplasty (syn. epiglottoplasty, supraglottoplasty) has attempted to address the primary pathology. The short aryepiglottic folds are incised, resulting in epiglottic release and the redundant supra-arytenoid mucosa and sub-mucosa is trimmed ensuring that an intact inter-arytenoid mucosal bridge is left at the posterior commissure to prevent

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TABLE I
GRADE OF STRIDOR

Grade of stridor	Clinical features
1	Mild inspiratory stridor
2	Severe stridor and/or oxygen desaturation below 92%
3	Pre-operative intubation required

inter-arytenoid scarring. The procedure may be performed using standard sharp micro-dissection techniques, or a CO_2 laser. This study presents our results with this proedure and examines the clinical features of those patients for whom the procedure did not provide an early cure.

Materials and methods

The case notes of 100 consecutive patients who underwent aryepiglottoplasty surgery at Great Ormond Street Hospital for Children, London, England, were reviewed. The first 12 cases were those previously reported by Jani *et al.*[4] The patients were all diagnosed as having significant laryngomalacia requiring definitive treatment. Indications for endoscopic aryepiglottoplasty were plasma oxygen desaturation below 92 per cent and/or failure to thrive on standard growth measurement charts due to feeding difficulties. Subjects were identified from hand-written operating-theatre registers kept within the ENT theatre suite. These were cross-referenced with computer-held coding data kept since 1992, and also with duplicate copies of the hand-written operation records filed separately from the in-patient notes.

Whilst acknowledging the difficulties of retrospective studies with information taken from clinical notes, the recorded symptoms of both airway obstruction and feeding difficulties were graded to enable an assessment to be made of improvement or deterioration (Tables I and II). These criteria were selected as they provide clear-cut differences in the clinical state and are sufficiently simple to have been well recorded. An assessment was made at 24 hours and three days post-operatively, at discharge from Hospital; and subsequently at one month and then at three monthly intervals. The follow-up period ranged from 12 to 120 weeks (mean 17.1 weeks).

Results

The Department of Paediatric Otolaryngology at Great Ormond Street Hospital for Children performs approximately 850 microlaryngo-tracheo-bronchoscopies per annum, under general anaesthesia. On average, 50 new cases of laryngomalacia per year were encountered during the period of this study. In the study group 56 patients were male and 44 female, 47 being under three months of age (range 10 days to 11 years). Isolated laryngomalacia in an otherwise well child was present in 53 patients. Other pathology contributing to airway or feeding problems was present in 47 patients (Table III). Stridor was present at birth in 57 cases (increasing to 67 by three days of age) being inspiratory in 82 and biphasic in 10 (unrecorded in eight). Within the biphasic group, only four had lower airway pathology. Tracheomalacia or tracheobronchomalacia was found in 17. Endoscopic evidence of gastro-oesophageal reflux was found in 47, being severe in 19, and three patients had required fundoplication prior to aryepiglottoplasty. Feeding difficulties were present in 58, ranging from prolonged feeding to choking and aspirating feeds. Seven required pre-operative nasogastric feeding. Co-existing primary neurological conditions were present in 28 patients.

Due to initial concern regarding the risk of supraglottic scarring and contracture after bilateral surgery, 16 of the early cases were treated with a unilateral procedure. No benefit was demonstrated by performing a unilateral procedure, and the second side subsequently required surgery in 50 per cent (eight of 16). The mean hospital stay was 5.1 days (SD 5.9) for bilateral procedures and 11.7 days (SD 15.2) for unilateral procedures. There was a clear increase in bed occupancy due to the time interval between unilateral procedures, and this approach was soon abandoned. The mean stay for all patients with isolated laryngomalacia was 5.3 days (SD 4.8) and for patients with co-existing pathology 12.9 days (SD 11.9). The minimum post-operative follow-up was three months (mean 16 weeks).

Stridor data could be obtained on 91 patients (insufficient data = nine) (Table IV). Of the 86 children with stridor improved by surgery, 64 had done so within seven days of the operation and the remaining 22 within one month. The airway was immediately worsened in five of the 86 cases, but after intubation and intravenous steroids, these patients were extubated and they also improved. Persistent stridor was evident in five patients, but in

TABLE II
GRADING OF FEEDING DIFFICULTIES

Grade of feeding difficulty	Clinical features
1	Prolonged feeding time, interrupting feeds to breathe
2	Failure to thrive
3	Nasogastric tube feeding required

TABLE III
CO-EXISTING AIRWAY PATHOLOGY

Tracheomalacia/bronchomalacia	14
Gross posterior glottic oedema	7
Sub-glottic stenosis	7
Vocal fold palsy	6
Others	13
Total	47

TABLE IV
EFFECT OF SURGERY ON STRIDOR AT 24 HOURS POST-OP

Abolished	50
Decreased	36
Worsened	5
Insufficient data	9
Total	100

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TABLE V
EARLY POST-OPERATIVE COMPLICATIONS

Intra-operative haemorrhage	1
Initial worsening of airway/persistent stridor	10
Post-operative intubation	4
Tracheostomy	1
Aspiration of early feeds	6
Peri-operative death (non-airway related)	1
Total	23

only one of these was there persistent worsening of the airway following the procedure, a tracheostomy being required: all five of these patients later proved to have neurological hypotonic disease that was not apparent at the time of surgery. Slow resolution, taking more than one week to significantly improve, was observed in 22 of the 86 cases (25 per cent). Fourteen of these 22 were later found to have serious neurological disease, mostly hypotonic in nature. The other eight had delayed improvement due to complications of the procedure.

Entries in the medical notes referring to feeding were generally of good quality and this information was also referred to frequently in nursing records. Fifty-eight patients had significant problems with their feeding. In 42 of these, surgery considerably improved their feeding. Six remained unchanged by the procedure, but eight became worse, five patients requiring nasogastric feeding (two long-term): all five were later shown to have progressive neurological disorders.

Gastro-oesophageal reflux (GOR) was recorded as being a possible diagnosis in 86 patients, subsequently being positively diagnosed in 28. (Insufficient recorded information existed in the remaining 14/100 case notes, there being no mention of GOR. There is, however, no reason to conclude that these patients suffered significantly from it.) A barium swallow investigation was performed on 49 (57 per cent) patients, GOR being identified in 16 of these (54 per cent). Six patients had a clinical diagnosis made but their barium swallow results failed to demonstrate any GOR. One case of GOR was diagnosed by pH probe, and an unverified clinical diagnosis was made on six others from the medical history alone. The recorded follow-up information was generally unhelpful and poorly recorded, although one patient subsequently required a Nissen's fundoplication, and four patients were discharged on anti-reflux medication to their local hospitals for management of ongoing reflux problems. The only long-term complication was a clinically unimportant inter-arytenoid scar which occurred in one of the first patients in this series. There have been no long-term problems from this cause identified. There was one fatality due to cardiorespiratory failure in a child with multiple congenital defects in whom the cause of death was unrelated to the upper airway. Post-operative complications were mostly minor and are listed in Table V.

Discussion

In the past tracheostomy was the safest option for the most severe cases of laryngomalacia, although this procedure carries a recognized morbidity and a small but significant mortality. Although some authors have advocated hyomandibulopexy for severe laryngomalacia,[5] the majority of operations currently performed are endoscopic procedures on the supraglottic region. The first endoscopic procedure was undertaken by Iglauer in 1922[6] and involved partial epiglottic resection. In 1984 Lane[7] described excision of redundant supraglottic tissue and in 1985 Seid[8] described incision of the aryepiglottic folds. The majority of reported cases involve one or both of the latter, although Solomons and Prescott[9] describe epiglottopexy and partial epiglottectomy for variations in supraglottic anatomy causing supraglottic collapse.

TABLE VI
PREVIOUSLY PUBLISHED SERIES OF EPIGLOTTOPLASTY FOR LARYNGOMALACIA

Authors	Number of patients	Neuromuscular deficit	Gastro-oesphageal reflux	Outcome measures
Zalzal *et al.*[10]	10	–	–	Complete resolution of symptoms 90% Resolution after revision surgery 10%
Solomons and Prescott[9]	11 (including 1 epiglottopexy)	18.1%	–	Improvement in 82%
Hollinger and Konior[11]	13	–	23%	Stridor improved in 92% Feeding improved in 70%
Povolonski *et al.*[12]	39	–	43.6%	Complete relief of symptoms in 76.9% Improved symptoms in 17.9%
Jani *et al.*[4]	12	–	75%	Stridor improved in 92% Feeding improvement in 70%
McLurg *et al.*[13]	24	16.7%	42%	Complete resolution of symptoms in 71%
Kelly and Gray[14]	18 (unilateral)	–	44%	Second side procedure required in 17% after which 94% good relief of symptoms
Roger *et al.*[15]	115	8.8%	68%	Complete regression of symptoms 53% Much better in 36%
Remacle *et al.*[16]	21	–	–	Normalization in 38.1% Improvement in 19.0%

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A review of the current English language literature reveals nine series of aryepiglottoplasty for laryngomalacia containing 10 or more patients for whom the outcome of surgery is described (summarized in Table VI). It can be seen from the variety of outcome measures quoted that direct comparison of results is difficult. Stridor appears to improve in around 90 per cent of patients with complete resolution in the majority. Feeding can be expected to improve in about 70 per cent.

We acknowledge that an unusually high proportion of patients attending our department at Great Ormond Street Hospital for Children have co-existing airway and medical pathology, due to the super-specialist tertiary referral nature of the hospital's work. Of the 100 cases, 53 had isolated disease, and all of these did very well following surgery. Twenty-five had complications, but these were mostly minor: persistent post-operative problems occurred only in those with co-existing neurological disease. This was usually of a hypotonic nature. Surgery only worsened the airway in one case, resulting in the need for a tracheostomy, and this patient also had severe progressive degenerative neurological disease. Of the series described (Table VI), several authors have observed poor results in children with pre-existing neurological or neuromuscular deficits.[9,11,13,15] It is likely that the failure of surgery in these cases is because the primary cause of the feeding and breathing difficulties is the neurological deficit rather than the laryngeal pathology.

Conclusions

Our experience confirms that endoscopic aryepiglottoplasty is an effective and safe procedure in a specialized unit. In those patients for whom aryepiglottoplasty fails to relieve the airway and feeding difficulties associated with laryngomalacia it is important to consider a primary neurological disorder.

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