

Supraglottoplasty in neonates and infants

A radiofrequency ablation approach

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Abstract

Background: Supraglottoplasty is the mainstay of surgical treatment for laryngomalacia. A novel supraglottoplasty surgical technique is needed to achieve better efficacy. The purpose of this study was to introduce modified microscopic radiofrequency ablation supraglottoplasty (MMRAS) for the treatment of congenital laryngomalacia and to evaluate the outcome and effectiveness of this novel approach.

Methods: Seventeen children with severe laryngomalacia who underwent MMRAS were studied retrospectively. Supraglottoplasty of type III laryngomalacia was different from classical method. All the patients were kept intubated for 5 days after surgery to achieve a better epiglottal position and to avoid reoglutination of aryepiglottic folds. The patients' demographic information, symptoms, comorbidities, type of laryngomalacia, synchronous airway lesions and final outcomes were examined.

Results: The median age at the time of surgery was 3.36 months (3 months 10 days). Operative indications included feeding difficulties, noisy breathing or respiratory distress (or both), and sleep-related symptoms. The MMRAS success rate was 82.4%. Most patients were extubated successfully on postoperative day 5. The major postoperative complication was pulmonary infection which occurred in 3 cases (17.6%) and required anti-infective therapy. No perioperative deaths and no long-term complications occurred. Failures were observed in 3 (17.6%) of 17 cases, 2 patients presented with a neurological disease and required tracheostomy, 1 patient relapsed because of postoperative adhesions and later underwent revision supraglottoplasty.

Conclusions: From these results, we conclude that MMRAS is an effective and safe treatment for symptomatic laryngomalacia and has the potential to provide better breathing, feeding, and sleeping outcomes in children with severe laryngomalacia. Postoperative intubation for 5 days may result in better therapeutic outcomes. Multicenter cooperative studies of comparison between MMRAS and conventional approaches would lend further evidence-based support for this surgical method.

Abbreviations: ENT = ear, nose, and throat, FFL = flexible fiberoptic laryngoscopy, ICU = intensive care unit, MMRAS = modified microscopic radiofrequency ablation supraglottoplasty, RFA = radiofrequency plasma ablation.

Keywords: laryngomalacia, radiofrequency ablation, supraglottoplasty

1. Introduction

Laryngomalacia, a congenital anomaly of the larynx leading to inward collapse of the supraglottic airway with inspiration, is the most common cause of stridor in neonates and infants, accounting for over 60% of all cases.^[1] Several theories have been advanced to explain the etiology of the supraglottic airway collapse, including inadequate rigidity of the laryngeal cartilages, excessive laryngeal inflammation due to gastroesophageal reflux, and defective neuromuscular control of the larynx. Although the exact cause of laryngomalacia is still

unknown, the anatomic theory and the immaturity of the cartilaginous structures are widely accepted explanations.^[2] The characteristic inspiratory stridor symptom of laryngomalacia typically occurs at or shortly after birth, with an average age of presentation of 2 weeks.^[3] Although symptoms can be resolved with conservative management in most cases, patients who develop life-threatening complications—including failure to thrive, weight loss, apnea, cyanosis, pulmonary hypertension, cor pulmonale, severe respiratory distress, pectus excavatum, hypoxia/hypercarbia, and failed medical intervention—may require surgery.^[4] Tracheotomy used to be the mainstay of surgical treatment for laryngomalacia before the 1980s. After the first supraglottoplasty with otological instruments was described by Lane et al^[5] in 1984, the technique involved in this surgical procedure developed quickly. With the advancements in fiberoptic technology, transoral supraglottoplasty gained widespread acceptance and can now be performed using microlaryngeal instruments,^[6] a microdebrider,^[7] a CO₂ laser,^[8] a thulium laser,^[9] and a diode laser. Nevertheless, no differences in terms of the results obtained with these various techniques have been reported in the literature.^[10] Therefore, a novel supraglottoplasty surgical technique is needed to achieve better efficacy.

In the last decade, the technique of bipolar radiofrequency plasma ablation (RFA) has been well developed and widely accepted in clinical practice. This technique uses glow discharge plasma that produces chemically active radical species from the dissociation of water and the breaking of molecular bonds, thus causing tissue dissolution. The thermal effects of this technique can be modulated by modifying the electrode construction, limiting the local temperature to <50°C in order not to induce wound bed

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renecrosis.^[11] Coblation is currently being widely used in pediatric ear, nose, and throat (ENT) surgery and is reportedly effective, minimally invasive, and economical, but few studies have focused on the use of coblation in managing laryngomalacia.

The purpose of this study is to introduce modified microscopic radiofrequency ablation supraglottoplasty (MMRAS) for the treatment of congenital laryngomalacia and to evaluate the outcome and the effectiveness of this novel approach.

2. Materials and Methods

2.1. Ethical standards

This study was approved by local ethical committee. All procedures performed in this study involving human participants were in accordance with the ethical standards of the institutional and national research committee and with the 1964 Declaration of Helsinki and its later amendments or comparable ethical standards. Informed consent was obtained from all individual participants included in the study.

3. Patients

Seventeen children with severe laryngomalacia who underwent MMRAS were studied retrospectively from January 1, 2016 to December 31, 2016 at the Department of Otorhinolaryngology (head and neck surgery) of the Shanghai Children's Hospital at Shanghai Jiao Tong University. The patients' demographic information, symptoms, comorbidities, type of laryngomalacia, synchronous airway lesions, and final outcomes were examined.

4. Diagnosis

A presumptive clinical diagnosis of laryngomalacia was made based on the classic symptoms of inspiratory stridor worsened by feeding, agitation, supine positioning, or crying.^[12] Transnasal flexible fiberoptic laryngoscopy (FFL) was carried out under surface anesthesia in all cases to confirm the diagnosis of laryngomalacia and to obtain a dynamic view of the airways, including the vocal cord function. As has been reported in the literature, severe cases that require surgical intervention in the form of supraglottoplasty were defined as severe stridor with tachypnea, dyspnea, and hypoxia, usually present as moderate to severe laryngeal obstruction with 3 depression signs. Other symptoms, such as aspiration pneumonia, feeding difficulties, and failure to thrive, were also sought but were not required for inclusion.^[13] Each child was then evaluated with laryngeal contrast-enhanced computed tomography (CT) to make a comprehensive evaluation of the airways and to rule out synchronous lesions, such as lingual root cysts, subglottic hemangioma, tracheomalacia, or laryngeal neoplasms. The supraglottic anatomic abnormality of each patient was categorized according to Olney et al.^[3] Type I consists of prolapse of the mucosa overlying the arytenoid cartilage. Type II consists of foreshortened aryepiglottic folds. Type III consists of posterior displacement of the epiglottis.^[3]

5. Surgical technique

All operations were conducted by the same surgeon using general anesthesia. A suspension laryngoscope was positioned to expose the structures to be ablated. The procedure was performed using bipolar RFA under a microscope, and surgical procedures were somewhat modified from those reported in the literature.^[3,8] Therefore, this procedure was termed MMRAS. The precise surgical technique was based on the type of laryngomalacia. Surgical correction of type I involved the ablation of redundant

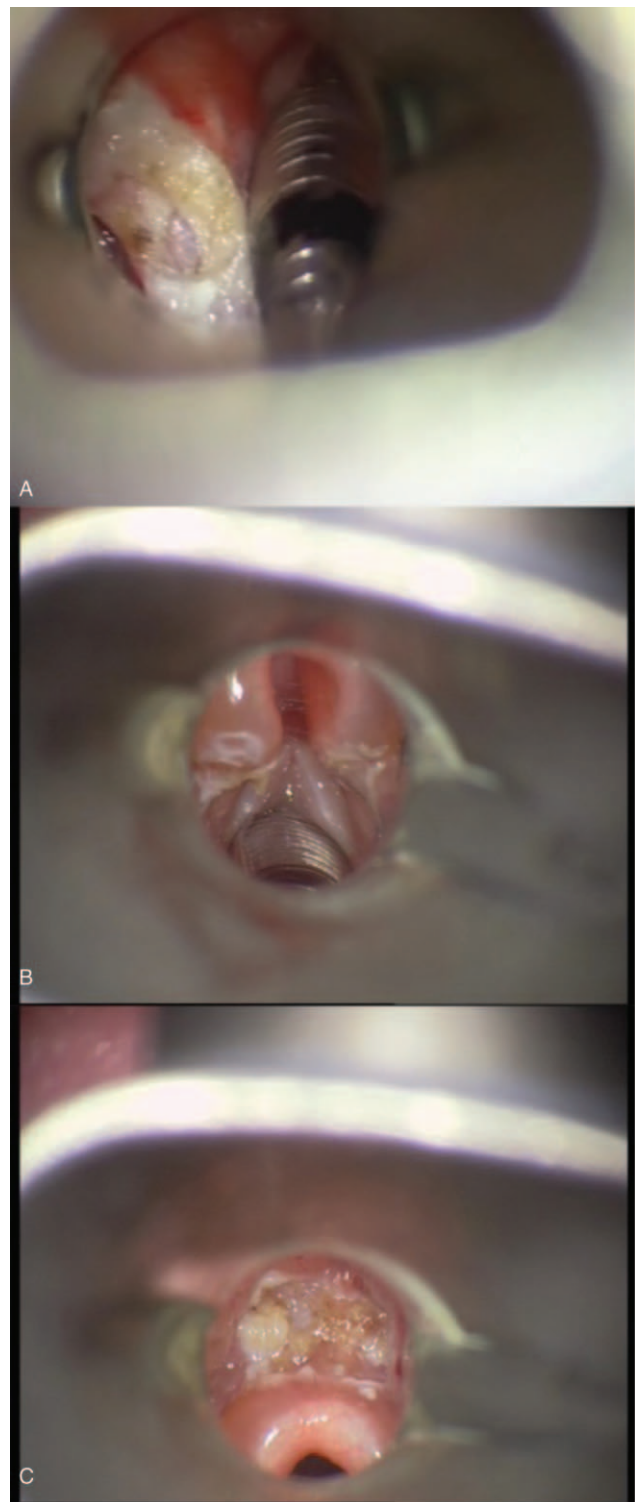


Figure 1. (A) Type I laryngomalacia, ablation of redundant mucosal tissue over the arytenoids. (B) Type II laryngomalacia, ablation of the shortened aryepiglottic folds and/or ablation of the lateral edge of the epiglottis. (C) Type III laryngomalacia, ablating wound surfaces on the base of the tongue.

mucosal tissue over the arytenoids. Type II was treated by ablation of the shortened aryepiglottic folds and/or ablation of the lateral edge of the epiglottis. Type III was corrected by ablating wound surfaces on the base of the tongue and epiglottic vallecula using an intratracheal cannula to hold the epiglottis in its normal position of clinging to the tongue base without sutures (Fig. 1). All patients

Table 1**Characteristics of patients (n=17).**

Age in days, median (range)	
Presentation of symptoms	16 days (birth: 2 months 23 days)
Diagnosis with endoscopy	3 months 10 days (19 days–10 months 21 days)
Sex, n (%)	
Male	7 (41.2)
Female	10 (58.8)
Gestational age, wk; mean (SD)	36.8 (2.088)
Birth weight, g; mean (SD)	3242 (393.089)
Presenting symptoms, n (%)	
Stridor	17 (100)
Dyspnea	12 (70.6)
Hypoxia	8 (47.1)
Feeding problems	8 (47.1)
Failure to thrive	5 (29.4)
Aspiration pneumonia	6 (35.3)
Type of laryngomalacia, n (%)	
Type I	7 (53.8)
Type II	14 (82.4)
Type III	7 (41.2)
Mixed type	10 (58.8)
Comorbidities, n (%)	
Synchronous airway lesions	3 (17.6)
Neurological	1 (5.9)
Syndrome/genetic disorders	1 (5.9)
None	12 (70.6)

SD=standard deviation.

were kept intubated for 5 days after surgery in the intensive care unit (ICU) to achieve a better epiglottal position and to avoid reoglutination of the aryepiglottic folds.

6. Results

Seventeen children with severe laryngomalacia who underwent MMRAS were studied retrospectively from January 1, 2016 to December 31, 2016 at the Department of Otorhinolaryngology

of the Children's Hospital of Shanghai at Jiao Tong University. The patients consisted of 7 males (41.2%) and 10 females (58.8%). The median age at surgery was 3.36 months (3 months 10 days) (range 19 days to 10 months 21 days).

FFL was done in all 17 cases. Shortened aryepiglottic folds, type II according to Olney's classification, were found in 14 cases (82.4%). In 10 cases (58.8%), endoscopy showed a combination of >1 type. Three cases had secondary airway lesions in the form of lingual root cysts. Symptoms appeared in 13 cases (76.5%) during the first 3 weeks of life, and in 10 cases (58.8%), stridor was noted to be present at birth. At the time of diagnosis, all children presented with stridor. In addition, 8 children (47.1%) presented with feeding difficulties, 5 (29.4%) with failure to thrive, and 8 (47.1%) with hypoxia. Six children (35.3%) presented with aspiration pneumonia, and no children presented with cor pulmonale or pulmonary hypertension. Table 1 shows the characteristics of the study subjects.

Comorbidities were present in 5 patients (29.4%): 3 (17.6%) had lingual root cysts, 1 presented with atelencephalia, and 1 had spinal muscular atrophy. The median age at surgery of the 5 children with comorbidities was 5.74 months (5 months, 22 days). No anesthesiological complications were found. One patient was accidentally extubated within 24 hours after the operation. Thirteen patients (76.5%) were extubated after 5 days in the ICU, and 3 patients (17.6%) were intubated up to 8 days to treat postoperative pulmonary infection.

Success was achieved in 14 of the 17 patients (82.4%), with resolution of the symptoms. A well-tolerated intermittent stridor without labored breathing persisted in most of the cases (13/14; 93%). Postoperative pulmonary infection occurred in 3 cases (17.6%) and required anti-infection therapy. No long-term complications occurred. Three patients (17.6%) were considered as failures and needed further intervention (Table 2.)

One of these was a 25-day-old boy. Stridor with tachypnea, hypoxia, and feeding difficulties presented shortly after his birth. The persistence of shortened aryepiglottic folds and redundant mucosa over the arytenoid region was successfully managed by

Table 2**Failures of supraglottoplasty cases: symptoms, surgical procedure, postoperative status, and further treatment.**

Patient	Comorbidities	Symptoms	Laryngomalacia type	Surgical procedure	Postoperative status	Further treatment
1	None	Stridor, hypoxia and feeding difficulties	I, II	Ablation of arytenoid mucosa, ablation of the aryepiglottic folds, and extubation within 24 hours	Symptoms improved significantly after extubating, relapse of severe stridor in 1 month, and aryepiglottic folds reshortened	Revision supraglottoplasty, extubation after 5 days, follow-up >6 months, and symptoms improved significantly
2	Fetal macrosomia atelencephalia	Stridor, hypoxia, apneas, feeding difficulties, and failure to thrive	I, II	Ablation of arytenoid mucosa, ablation of the aryepiglottic folds, and extubation 5 days later	Symptom of stridor improved significantly, hypoxia and apneas disappeared, feeding difficulties persisted, recurrent severe stridor and apneas, failure to thrive after 1 month, upper airway collapse, and supraglottic mucosal swelling	Tracheotomy was recommended and parents refused to accept any further medical practices
3	Spinal muscular atrophy	Stridor, feeding difficulties, failure to thrive, recurrent aspiration pneumonia, motor delays, and low muscle tone	I, II	Ablation of arytenoid mucosa, ablation of the aryepiglottic folds, and extubation 7 days later	Symptom of stridor improved significantly, feeding difficulties, aspiration, and low muscle tone persisted	Confirmed type I spinal muscular atrophy after genetic diagnosis, required comprehensive multidisciplinary care, and parents refused to accept any further medical practices

MMRAS. It was planned that this infant was to be kept intubated in the pediatric ICU (PICU) for 5 days, but he was accidentally extubated within the first 24 hours after the operation. As no tachypnea or hypoxia was found, we decided not to intubate again. When the patient was discharged from the hospital, his symptoms of laryngeal obstruction improved significantly. However, the infant was found to have severe stridor recurrence at the 1-month follow-up. Aryepiglottic folds appeared reshortened by FFL. A revision supraglottoplasty was carried out, and the patient was left intubated in the ICU for 5 days. In the following 6-month follow-up period, no stridor symptoms or reshortening was observed.

The second patient was a 3-month-old girl who presented with stridor, hypoxia, apneas, feeding difficulties, and failure to thrive. Upon her birth by cesarean section, she was diagnosed with fetal macrosomia, weighing 4 kg. She was also diagnosed with atelencephalia; cranial sonography showed dilated bilateral cerebral ventricles and subependymal effusion on the right side. In the 3 months after her birth, the stridor persisted, hypopneas with episodes of oxygen desaturation up to 78% were recorded, and she gained only 500 g. The FFL showed a serious prolapse of the mucosa overlying the arytenoid cartilage and shortened aryepiglottic folds with an omega-shaped epiglottis. MMRAS was performed under general anesthesia. The stridor improved significantly and the hypoxia and apneas disappeared soon after extubation; however, the feeding difficulties persisted. This patient was followed up 1 month after surgery and had recurrent severe stridor and apneas; she only weighed 4.8 kg. An endoscopy of the upper airway showed a persistent collapse and mucosal swelling of the supraglottis. Tracheotomy was recommended, but unfortunately her parents refused to accept any further medical treatment.

The third patient was a 9-month-old girl with stridor, who started experiencing feeding difficulties 2 weeks after birth. Over the next 8 months, she failed to thrive and suffered from recurrent aspiration respiratory tract infections. Meanwhile, she presented with neuromotoric delays and low muscle tone but normal intelligence development, and a head CT scan was normal. The FFL showed a serious prolapse of the mucosa overlying the arytenoid cartilage and shortened aryepiglottic folds. MMRAS was performed, and she was extubated 7 days later because of pneumonia. After extubation, the symptom of stridor improved significantly, but the feeding difficulties, aspiration, and low muscle tone persisted. This child was confirmed to be suffering from type I spinal muscular atrophy confirmed by genetic tests.^[14] Her parents refused further treatment after being informed that there was presently no cure for spinal muscular atrophy.

7. Discussion

Laryngomalacia is the most common cause of stridor in neonates and infants, with a reported incidence of over 60%,^[11] and 10% to 20% of children with laryngomalacia require further intervention.^[4] Since the first supraglottoplasty was described by Lane et al^[5] in 1984, endoscopic supraglottoplasty with various kinds of instruments has become an effective treatment for this condition. Many new technologies facilitated the development of supraglottoplasty. Supraglottoplasty using lasers and microdebriders is reportedly associated with less blood loss and a lower risk of laryngeal edema compared to conventional surgical methods. However, the cost of lasers and microdebriders is much higher than that of classical microsurgery instruments. In

addition, lasers require special precautions to avoid the risk of fire during surgery.^[15] Radiofrequency ablation (RFA) technology is currently being widely used in pediatric ENT surgery, such as tonsillectomies, adenoidectomies, and the removal of pediatric congenital vallecular cysts.^[16] A recent study suggested that RFA may be an effective endoscopic tool for the treatment of pediatric airway stenosis and can be an additional tool in the endoscopic armamentarium of airway surgeons.^[17] Tissue can be removed rapidly without significant bleeding because of the unique ablative property of high-energy plasma. It is believed that the risk of collateral tissue damage and airway fire in RFA airway operations may be significantly reduced compared to CO₂ laser operations, as the local temperature can be limited to <50°C.^[18,19] RFA is also relatively easier for a surgeon to master compared to using the CO₂ laser. However, few studies have focused on the use of RFA in managing laryngomalacia.

Supraglottoplasty is reported to have a success rate of 70% to 100%.^[8,20–22] In this study, the success rate is 82.4%. Even with local airway comorbidities, this operation can lead to good results. In our study, lingual root cysts were the most common comorbidity. In some cases, the cysts were not easy to find with FFL before surgery because a cyst may squeeze the epiglottis towards the glottis, making the condition look like type III laryngomalacia. However, a preoperative upper airway CT scan can help to discover it. The surgical protocol is different with the existence of the cyst. Ablation of the surface tissues of the epiglottic vallecula is not necessary if the normal epiglottic anatomical position is resumed after excision of the lingual root cyst. Ablation of redundant mucosa over the arytenoids and/or shortened aryepiglottic folds and/or the lateral edge of the epiglottis may be carried out if needed in these cases. However, neurological comorbidities were an important reason for failure. Two patients in our study had neurological comorbidities and

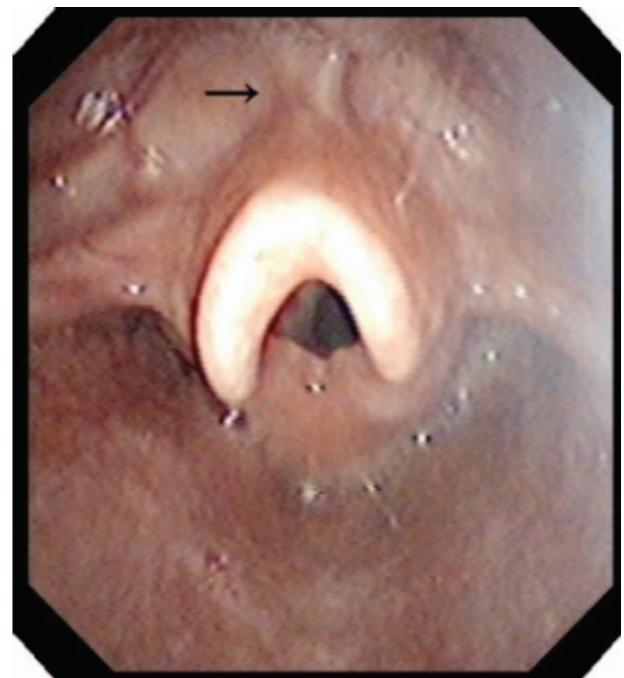


Figure 2. “→” showing the local conglutination area between wound surfaces of the epiglottis and the tongue base, this helps to maintain the wanted anatomical position without using sutures in the operation and prevent the epiglottis from collapsing into the glottis during inspiration.

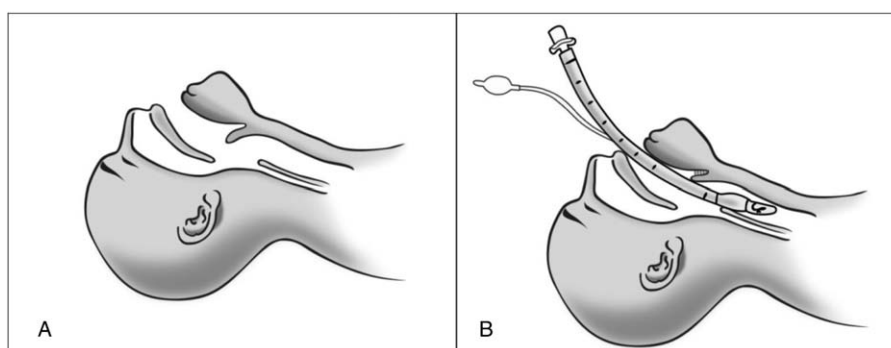


Figure 3. Intubation up to 5 days facilitates a better position of the epiglottis to avoid re-conglutination of aryepiglottic folds, and it may hold the epiglottis in its normal position clinging to the base of the tongue.

were defined as failures due to their unresolved feeding problems. Despite this, we believe that the surgical interventions in these cases were worth doing, as the patients' respiratory symptoms were partially relieved. Our experience was consistent with International Pediatric ORL Group consensus recommendations that neurological disease is not a contraindication to supraglottoplasty; however, the benefit of improving airway obstruction must be weighed against the risk of worsening.^[13]

Management of the perioperative period, particularly the postoperative period, plays an important role in the success of MMRAS. However, it is suggested that wound and postoperative reactions in RFA surgery were significantly milder compared to techniques using cold instruments.^[11] Edema of supraglottic tissues should be paid attention to after supraglottoplasty to avoid laryngeal obstruction. In our experience, too early an extubation may lead to hypoxemia and carbon dioxide retention. The general attitude in our clinic was to admit the postoperatively intubated children to the ICU and to leave them intubated for at least 5 days. Antireflux therapy and glucocorticoids were given to alleviate edema and to prevent the formation of excess granulation tissue. Another reason for intubation up to 5 days is to achieve a better epiglottal position and to avoid re-conglutination of aryepiglottic folds. Postoperative adhesion of the wound surface may cause aryepiglottic fold re-conglutination and may result in symptom recurrence. In our study, a patient with mixed-type laryngomalacia was found to have severe stridor recurrence at the 1-month follow-up. This patient was accidentally extubated within 24 hours after surgery. Aryepiglottic folds were found to be reshortened by FFL. We hypothesize that this was caused by the re-conglutination of aryepiglottic folds. After a revision supraglottoplasty and 5 days' extubation, no stridor symptoms or reshortening was observed in the subsequent 6-month follow-up period. In addition, an intratracheal cannula was used to hold the epiglottis in its normal position clinging to the tongue base. We hypothesize that the formation of a local conglutination area within 5 days between wound surfaces of the epiglottis and the tongue base can help to maintain the desired anatomical position without using sutures in the operation and to prevent the epiglottis from collapsing into the glottis during inspiration (Figs. 2 and 3). However, a relatively long intubation may cause aggravation of pulmonary infection. In this study, 3 patients (17.6%) were intubated up to 8 days because of postoperative pulmonary infection. We need to find a suitable method to shorten the intubation time in future studies.

8. Conclusion

From these results, we conclude that MMRAS is an effective and safe treatment for symptomatic laryngomalacia and has the potential to provide better breathing, feeding, and sleeping outcomes in children with severe laryngomalacia. Postoperative intubation for 5 days may result in better therapeutic outcomes. Multicenter cooperative studies of comparison between MMRAS and conventional approaches would give further evidence-based support for this surgical method.

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