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Review article

Treatment outcomes of supraglottoplasty for pediatric obstructive sleep apnea: A meta-analysis

Chia-Fan Lee [a, 1], Wei-Chung Hsu [b, c, 1], Chia-Hsuan Lee [d], Ming-Tzer Lin [c, e, **], Kun-Tai Kang [d, *]

[a] Speech Language Pathologist, Child Developmental Assessment and Intervention Center, Taipei City Hospital, Taipei, Taiwan, ROC
[b] Department of Otolaryngology, National Taiwan University Hospital, Taipei, Taiwan, ROC
[c] Sleep Center, National Taiwan University Hospital, Taipei, Taiwan, ROC
[d] Department of Otolaryngology, Taipei Hospital, Ministry of Health and Welfare, New Taipei City, Taiwan, ROC
[e] Department of Internal Medicine, Hsiao Chung-Cheng Hospital, New Taipei, Taiwan, ROC

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ABSTRACT

Objective: To comprehensively review changes in sleep parameters and the success rate of supraglottoplasty for treating obstructive sleep apnea (OSA) in children. In particular, to elucidate treatment modalities and factors affecting treatment outcomes in children with both laryngomalacia and OSA.
Methods: The study protocol was registered on PROSPERO (CRD42015027053). Two authors independently searched databases including PubMed, MEDLINE, EMBASE, and the Cochrane Review database. The keywords were "supraglottoplasty," "laryngomalacia," "OSA," "polysomnography," "child," and "humans." Supraglottoplasty served as the primary treatment for OSA or secondary treatment for persistent disease after previous surgeries. Subgroup analyses were conducted for children receiving supraglottoplasty as the primary or secondary treatment for OSA, and for children with and without comorbidities.
Results: Eleven studies with 121 patients were analyzed (mean age: 3.7 years; 64% boys; mean sample size: 11 patients). After surgery, the mean differences between the pre- and postoperative measurements were a significant reduction of 8.9 events/h in the apnea–hypopnea index (AHI) and an increase of 3.7% in minimum oxygen saturation ($MinSaO_2$; $P < 0.05$). The overall success rate was 28% according to a postoperative AHI <1 and 72% according to an AHI <5. Children receiving supraglottoplasty as the primary treatment had significantly younger ages (0.6 vs 6.4 years $P < 0.001$) than those receiving supraglottoplasty as the secondary treatment, but the outcomes were similar (33% vs 19% for a postoperative AHI < 1, $P = 0.27$; 77% vs 61% for a postoperative AHI < 5, $P = 0.233$). Moreover, children with comorbidities, compared with those without, had a similar success rate according to a postoperative AHI <1 (25% vs 21%, $P = 0.805$) and postoperative AHI <5 (62% vs 84%, $P = 0.166$).
Conclusions: Supraglottoplasty is an effective surgery for AHI reduction and $MinSaO_2$ increase in children with OSA and laryngomalacia. However, complete resolution of OSA is not achieved in most cases, and factors affecting treatment outcomes in these children require future studies.

Contents

* Corresponding author. Department of Otolaryngology, Taipei Hospital, Ministry of Health and Welfare, No.127, Siyuan Rd., Xinzhuang Dist., New Taipei City, Taiwan, ROC.
** Corresponding author. Department of Internal Medicine, Hsiao Chung-Cheng Hospital, New Taipei, Taiwan, ROC.
E-mail addresses: lightpool2010@gmail.com (M.-T. Lin), kang.kuntai@msa.hinet.net (K.-T. Kang).
[1] Chia-Fan Lee and Wei-Chung Hsu contributed equally as the first author to this article.

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1. Introduction

Laryngomalacia is a disease characterized by the collapse of redundant supraglottic tissues sucked into the laryngeal inlet, resulting in the presence of intermittent stridor during inspiration [1]. Laryngomalacia was first described by Jackson in 1942 [2]. Laryngomalacia is the leading cause of congenital stridor in infants, accounting for 60%–70% of cases [3–5]. Congenital laryngomalacia usually appears within 2 weeks after birth and resolves within 24 months [3,4], and is often considered self-limiting; the majority of children are managed conservatively. However, approximately 5%–20% of children with severe or refractory disease require more aggressive intervention [5]. Furthermore, the condition may also appear in patients older than 2 years without a history of prior disease. This condition is called late-onset laryngomalacia [6]. Occult or state-dependent laryngomalacia presents with stridor in children, limited to sleep or exercise [7–9]. Clinical features and treatment modalities differ between congenital and noncongenital laryngomalacia [9,10].

Several indications for surgery in children with laryngomalacia have been raised, including severe dyspnea, failure to thrive, cyanosis, cor pulmonale, and sleep apnea syndromes [11–18]. Surgical modalities for laryngomalacia have evolved over the years, from open tracheostomy to transoral endoscopic techniques [11,12]. Supraglottoplasty addresses the specific anatomic abnormalities involved in each individual case and has been shown to be both safe and effective [13,14]. Thus, supraglottoplasty is currently the primary form of surgical treatment for laryngomalacia [13–15]. Supraglottoplasty can be performed using cold knife [16], microdebrider [17,18], or CO_2 laser techniques [19], depending on surgeon training and preference; each technique produces a similar outcome [5]. Laryngomalacia may present as an isolated disease or may be associated with other comorbidities, such as gastroesophageal reflux disease, craniofacial anomalies, and neurocognitive disease [5]. A systematic review by Preciado and Zalzal reported a high risk of surgical failure and aspiration complications in patients with associated medical comorbidities, compared with those with isolated disease [20]. This finding should be considered for predicting outcomes in children undergoing supraglottoplasty.

Associations between laryngomalacia and obstructive sleep apnea (OSA) in children have increasingly received attention [21,22]. OSA in children includes a spectrum of respiratory disorders characterized by upper airway collapse during sleep [23]. Untreated OSA in children is associated with adverse cardiovascular [24], neurocognitive [25], and somatic growth consequences [26]. The pathophysiology of childhood OSA is mainly caused by enlarged adenotonsillar tissues [27,28]; therefore, adenotonsillectomy is the first-line therapy for pediatric OSA worldwide [29–34]. Increasingly, laryngomalacia is recognized as a risk factor for childhood OSA and a potential cause of residual disease after adenotonsillar surgery in children [21,35,36]. Therefore, children with both laryngomalacia and OSA may require supraglottoplasty. However, treatment modalities in these children have not been well studied [35–38]. A recent meta-analysis by Camacho et al. revealed that supraglottoplasty alleviates OSA in children [38]. However, factors affecting treatment outcomes in children with both laryngomalacia and OSA are not thoroughly understood [39].

The main purpose of this study was to elucidate (1) changes in sleep parameters (i.e., the apnea-hypopnea index [AHI] and minimum oxygen saturation [$MinSaO_2$]) after supraglottoplasty; (2) the overall success rate (i.e., postoperative AHI < 1 and AHI < 5) for supraglottoplasty; (3) the difference in surgical outcomes between children receiving supraglottoplasty as the primary treatment and those receiving it as the secondary treatment after previous surgeries; and (4) the difference in surgical outcomes between children with and those without comorbidities.

2. Methods

2.1. Search strategy

Study protocol was registered on PROSPERO (CRD42015027053). This meta-analysis was conducted according to PRISMA statement and the recommendations of the Meta-