

Acquired Idiopathic Laryngomalacia Treated by Laser Supraglottic Laryngoplasty

Ai Kawamoto,¹ Yukio Katori,¹ Yohei Honkura,¹ Masaki Ogura,¹
Yoshitaka Takanashi² and Toshimitsu Kobayashi²

¹Division of Otorhinolaryngology, Sendai City Hospital, Sendai, Miyagi, Japan

²Department of Otorhinolaryngology and Head and Neck Surgery, Tohoku University Graduate School of Medicine, Sendai, Miyagi, Japan

Laryngomalacia is the most common cause of stridor in neonates and infants, where the soft cartilages and tissues surrounding the upper larynx collapse inward during respiration. On the other hand, acquired idiopathic laryngomalacia in adults is quite rare, but should be borne in mind for differential diagnosis of upper airway distress. Allergic factors may cause airway distress, but have not been highlighted previously as the background of laryngomalacia. In this report, we describe two patients with acquired idiopathic laryngomalacia with reference to allergic rhinitis and high serum levels of immunoglobulin E. The first patient was a 16-year-old female who presented with inspiratory stridor and dyspnea due to attachment between the epiglottis and bilateral arytenoids, and the second patient was an 18-year-old male who also presented with inspiratory stridor due to attachment between the epiglottis and posterior pharyngeal wall. The respiratory function of both patients was within the normal range but the inspiratory stridor interfered with daily life. Laryngomicrosurgery was performed in both patients using a CO₂ laser to remove the arytenoid mucosa in the first patient, and to remove the tip of the epiglottis in the second. Both patients were followed up while receiving oral anti-allergic agents. Laser supraglottic laryngoplasty to remove the vibrating excess tissue was effective for resolving the symptoms. However, recurrence occurred three times in the first patient, and inferior turbinotomy to improve nasal respiration was useful for diminishing the symptoms.

Keywords: acquired laryngomalacia; allergic rhinitis; inspiratory stridor; serum immunoglobulin E; supraglottic laryngoplasty

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Introduction

Laryngomalacia is the most common cause of stridor in infants, where the soft, immature upper laryngeal cartilage collapses inward during inspiration, creating airway obstruction at the supraglottic level. This form of congenital laryngomalacia (CL) usually shows complete resolution in most children by the age of 12 to 18 months. However, other forms of laryngomalacia can also appear in later life, such as exercise-induced laryngomalacia (EIL) in later childhood (Smith et al. 1995) and acquired laryngomalacia (AL) (Gessler et al. 2002). Among the three types of laryngomalacia, AL is rare but causes continuous supraglottic airway narrowing. Most cases of AL are related to neuromuscular disorders resulting in weakness of the muscles of the larynx and/or pharynx, and improvement can be obtained by supraglottic laryngoplasty or resolution of the

underlying neurologic disorder. However, a few reviews have documented acquired idiopathic laryngomalacia (AIL) in otherwise healthy patients (Gessler et al. 2002; Siou et al. 2002; Echternach et al. 2008). The pathophysiological background of AIL is still unclear, and more information is needed in order to devise appropriate treatments. A recent report (Lim and Li 2011) has suggested that allergic factors can also create laryngeal airway distress and vocal cord dysfunction by increasing irritation of the throat and leading to excess inspiratory effort. Allergic factors may also influence AIL, but have not been pointed out previously as part of the background of laryngomalacia. In this report, we present two patients with AIL with special reference to allergic rhinitis and high serum immunoglobulin E levels. Both patients were treated by supraglottic laryngoplasty using a carbon dioxide laser and administration of anti-allergic agents.

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Correspondence: Yukio Katori, M.D., Ph.D., Division of Otorhinolaryngology, Sendai City Hospital, 3-1, Shimizukoji, Wakabayashi, Sendai 984-8501, Japan.

e-mail entsendai@yahoo.co.jp

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Clinical report

Patient 1

A 16-year-old female patient was admitted to a local clinic because of sudden onset of inspiratory stridor and dyspnea during usual exercise at high school. She had a history of allergic rhinitis but no other diseases or anomalies. She was initially diagnosed as having bronchial asthma, and was treated by systemic administration of corticosteroid and inhalation of a beta agonist bronchodilator. However, the symptoms did not improve and she was referred to a general hospital. There was no sign of acute inflammation in the larynx and her body temperature and blood cell counts were normal. During inspiration, the mucosa in the bilateral arytenoid region dropped into the glottic area and became attached to the epiglottis, causing stridor and supraglottic airway distress (Fig. 1A). Phonation was normal but conversation was disrupted by the inspiratory stridor. The patient complained of breathing difficulty, but the saturation ratio of arterial oxygen was 98 ~ 100% on room air. Respiratory function, in terms of lung capacity (LC, 3.07 L) and forced expiratory volume% in 1 second (FEV1, 88.7%) was within the normal range. The patient's symptoms improved when she consciously performed nasal breathing, and also diminished when she was asleep. Neck CT examination demonstrated neither stenosis of the upper air way or deformity of the laryngeal cartilages. Brain MRI and neurological parameters were normal. In accordance with the presence of allergic rhinitis, a high level of serum IgE (1,360 IU/ml; normal level <170 IU/ml) was detected by radioimmunosorbent test (RIST). A radioallergosorbent test (RAST) demonstrated strong reactions against house dust, mites, candida and Japanese cedar.

Oral administration of corticosteroid (prednisolone, 20 mg/day) and inhalation of beclomethazone were tried initially for two weeks with the aim of reducing the swelling of the vibrating arytenoid mucosa, but the symptoms were remained unchanged. Psychiatric counseling and advice to maintain nasal breathing also failed in diminishing the symptoms. The inspiratory stridor and sensation of breathing difficulty continued for eight weeks and interrupted the patient's daily life, for example during exercise at high school and playing a wind instrument, and therefore the surgical therapy was indicated.

Two months after onset, removal of the arytenoid mucosa and incision of the aryepiglottic fold were carried out bilaterally by laryngomicrosurgery using a carbon dioxide laser (Fig. 1B). Oral administration of levocetirizine dihydrochloride, a third-generation non-sedative antihistamine agent, and pranlukast hydrate, an anti-leukotriene agent, together with a corticosteroid nasal spray were started after the operation. The stridor was diminished after surgery without vocal or swallowing deficits. Three months later, however, the stridor and the sensation of breathing difficulty recurred. During inspiration, the mucosa in the interarytenoid region moved towards the glottis and

vibrated (Fig. 1C). A second operation was performed to ablate the mucosa of the left arytenoid and the interarytenoid region (Fig. 1D) and the symptoms were diminished thereafter. Six weeks after the second operation, the stridor recurred again due to vibration of the mucosa in the right arytenoid region (Fig. 1E). The stridor was less severe than in the previous episodes, and there was no breathing difficulty. A third operation was performed to ablate the mucosa in the vibrating region (Fig. 1F) and the stridor was relieved. Although all the mucosa in the arytenoid region was removed or ablated, the stridor and sensation of breathing difficulty recurred at three months after the third operation. At this time, the mucosa in the bilateral arytenoid region dropped in a glottic direction again and vibrated (Fig. 1G). A fourth operation was performed to broadly ablate the mucosa in the bilateral arytenoid region (Fig. 1H). To improve nasal respiration in a background of allergic rhinitis, inferior turbinotomy and laser evaporation of the nasal mucosa were carried out simultaneously. The patient was followed up with continuous administration of the anti-histamine agent, anti-leukotriene agent and corticosteroid nasal spray, and so far the symptoms of AL have not recurred during six months of follow-up after the final surgery (Fig. 1I).

Currently, LC and FEV1 are normal and no inspiratory stridor has appeared. No complications of phonation or deglutition were evident either before or after the surgical procedures.

Patient 2

An 18-year-old male patient presented with inspiratory stridor that had been evident for several years. He had no dyspnea but complained of serious throat irritation and stridor. He had a history of allergic rhinitis but no other diseases or anomalies. Although the glottis was well opened during expiration (Fig. 2A), a sound was produced due to attachment between the epiglottis and posterior pharyngeal wall during inspiration (Fig. 2B). Glottic closure and phonation were normal (Fig. 2C). The patient was able to control the extent of the attachment, but could not avoid it, or the stridor. The symptom did not appear during sleep. CT and neck X-ray examinations showed no evidence of upper airway stenosis. There was no evidence of acute inflammation, and blood parameters were normal without a high level of serum IgE (1637 IU/ml) in the RIST. The RAST demonstrated strong reactions against house dust, mites, cat and dog hairs, orchard grass and Japanese cedar. Respiratory function in terms of LC (4.51 L) and FEV1 (90.5%) was within the normal range, but the patient had severe inspiratory stridor when talking and listening a quiet sound, and therefore the surgical therapy was indicated.

Laryngomicrosurgery was carried out to reduce the length of the epiglottis using a carbon dioxide laser, and approximately 10 mm of the epiglottis tip was removed (Fig. 2D). After the operation, the glottic opening was preserved during the expiratory period (Fig. 2E) and the attach-

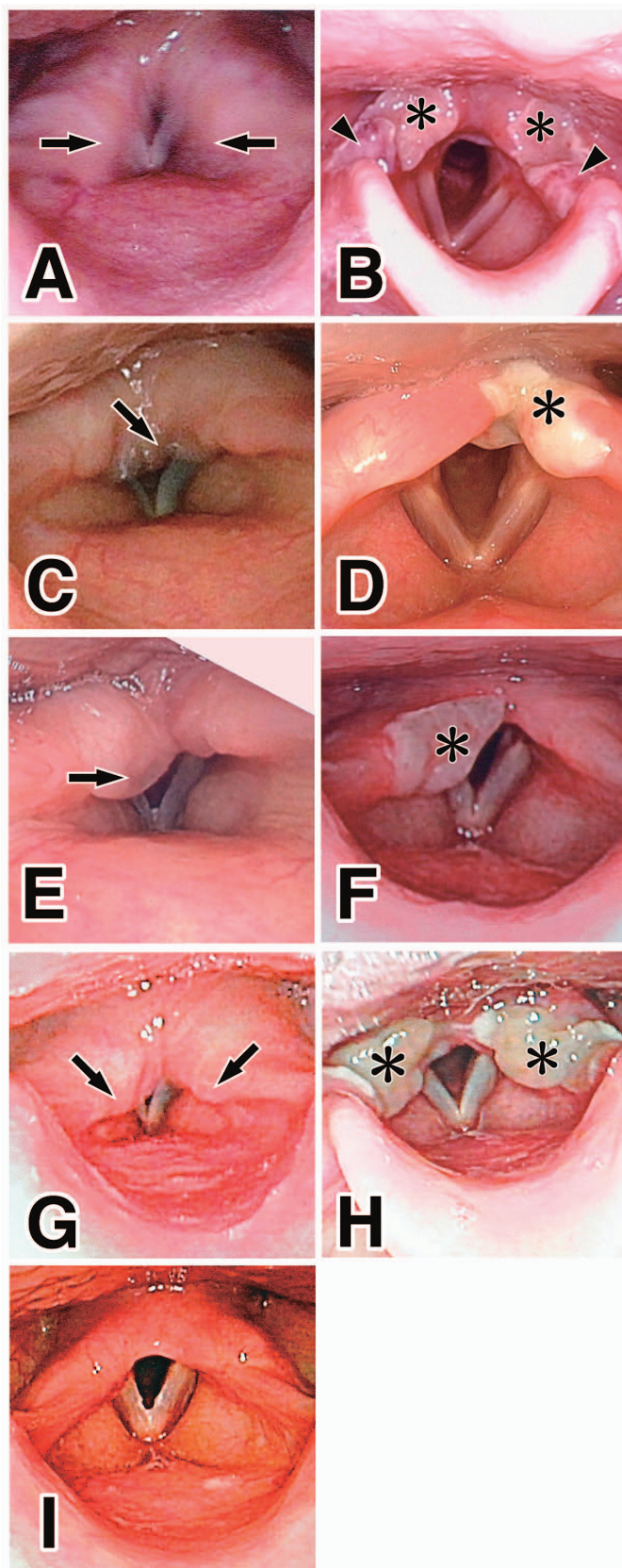


Fig. 1. Fiberoptic findings in the larynx of patient 1.

All photographs were taken during deep inspiration.

A. At the first admission, the bilateral arytenoid regions dropped towards the glottis (arrows) and became attached to the epiglottis, creating vibration and airway distress.

B. After removal of the bilateral arytenoid mucosa (asterisks) and cutting of the aryepiglottic folds (arrowheads) in the first operation, the symptoms disappeared.

C. Three months after the first operation, inspiratory stridor and airway distress recurred with vibration of the interarytenoid region (arrow).

D. Mucosa of the left arytenoid and interarytenoid regions was ablated using a CO₂ laser in a second operation (asterisk) and the symptom was diminished.

E. Six weeks after the second operation, inspiratory stridor recurred with vibration of the right arytenoid region (arrow).

F. The mucosa in the vibrating region was ablated in a third operation (asterisk) and the stridor was relieved.

G. Three months after the third operation, the mucosa of the bilateral arytenoid region again dropped towards the glottis (arrows) and vibrated.

H. A fourth operation was carried out to broadly ablate the bilateral arytenoid regions (asterisks), and the symptoms disappeared.

I. Although scar formation was evident in the arytenoids and aryepiglottic folds, no vibration or airway distress remained after surgery.

ment of the epiglottis to posterior pharyngeal wall during inspiration disappeared (Fig. 2F). The patient was followed up with administration of the anti-histamine agent, anti-leu-

kotriene agent and a corticosteroid nasal spray. Glottic closure was preserved at phonation (Fig. 2G), and the inspiratory stridor disappeared and has not recurred during a

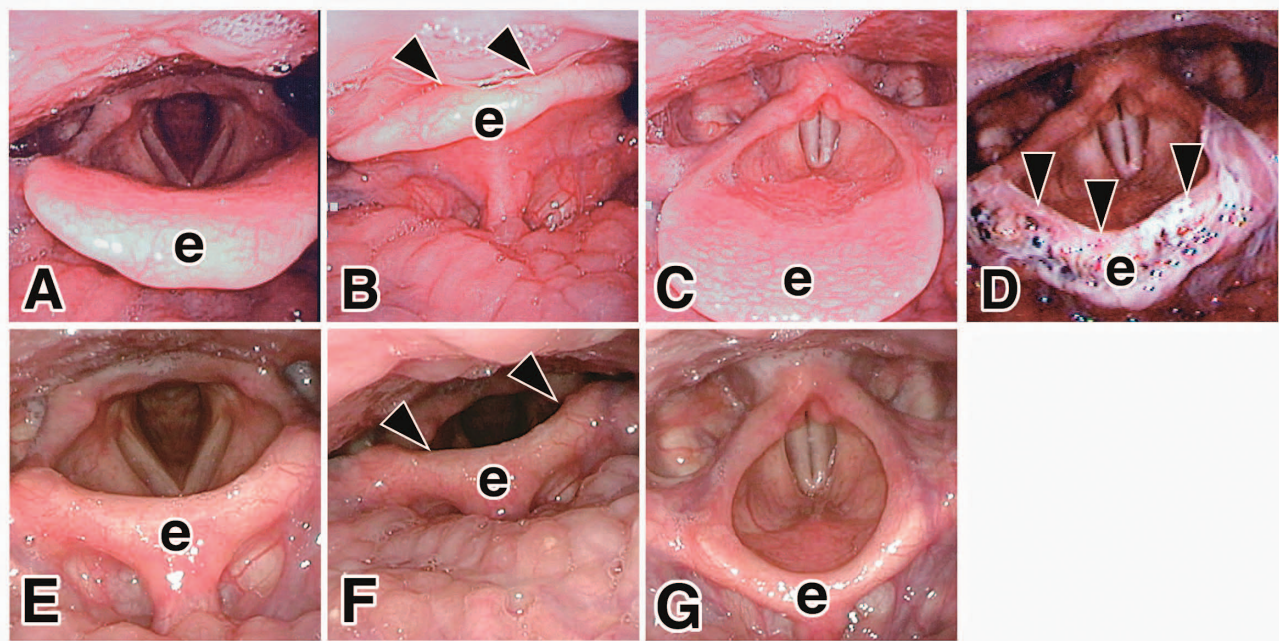


Fig. 2. Fiberoptic findings of the larynx in patient 2.

A and E were taken during expiration. B and F were taken during deep inspiration. C, D and G were taken during phonation.

A, B and C. At the first admission, the epiglottis (e) moved in the backward direction and became attached to posterior wall of the pharynx during inspiration (arrowheads), creating vibration. The movement of the arytenoid upon phonation was normal. D. Approximately 10 mm of the tip of the epiglottis (e) was removed using a CO₂ laser (arrowheads). E, F and G. Three months after surgery. The epiglottis (e) was shortened and did not attach to the posterior wall of the larynx during deep inspiration (arrowheads). Glottic closure was normal and no saliva pooling was observed.

follow-up period of eight months. No problems related to phonation or deglutition were evident either before or after surgery.

Discussion

Laryngomalacia is classified into three conditions: 1) CL, 2) EIL and 3) AL. Of these, AL has been rarely reported, and most cases have occurred after significant neurological dysfunctions resulting from cerebrovascular disease, head and neck surgery, or cervical trauma (Peron et al. 1988; Kletzker and Bastian 1990; Mima et al. 1996). Age-related changes to the form of the larynx, and to the shapes and distancing of the laryngeal cartilages and hyoid bone, have also been considered to cause AL (Sawatsubashi et al. 2010). Gastroesophageal reflux disease (GERD) has also been suggested to be related to laryngomalacia (Gessler et al. 2002), especially in EIL. The two cases presented here were classified as AL on the basis of age at onset, but were not typical because there were no obvious neurological dysfunctions, laryngeal deformity or clinical findings of GERD. During the time when symptoms were evident, both of the patients breathed mostly orally with strong effort during inspiration. They were able to reduce the symptoms intentionally by nasal breathing and/or pursed lip breathing, which suppresses strong inspiration. Previous reports of acquired laryngomalacia (Chetty et al. 1994; Chan et al. 2012) have pointed out the sleep apnea syn-

drome (SAS) in AL patients. Although neither of the present two patients had been examined for SAS by polysomnography, they did not show symptoms of laryngomalacia or sleep apnea and snoring during sleep before or after the treatment. The loss of symptoms during sleep also suggested that the narrowing of the supraglottic airway was not due to neurological dysfunction, but rather to strong inspirational effort.

Both patients showed high levels of serum IgE and strong reactions against non-seasonal allergens, notably house dust and mites. A recent report has suggested that allergic factors can accelerate glottic narrowing and cause distress during breathing as a form of vocal cord dysfunction (Lim and Li 2011). That report also described the effectiveness of nasal breathing and pursed-lip breathing to reduce excess inspiration and relieve glottic airway distress. In the present cases, it is hypothesized that allergic factors may have increased hypersensitivity and irritation in the airway, leading to a sensation of throat narrowing, thus triggering increased effort on inspiration. To reduce the possibility of strong oral inspiration, treatments for allergic rhinitis were carried out in both patients: inferior turbinotomy and medication in patient 1 and medication in patient 2. In patient 1, initial corticosteroid administration improved the symptoms of nasal allergy, but as they were severe, the nasal obstruction persisted. Inferior turbinotomy in the final operation was effective for relieving the nasal obstruc-

tion, and thus for ameliorating the symptoms. In patient 2, inspiratory stridor was associated with strong throat irritation, and this was relieved by administration of an anti-histamine agent after surgery.

Although the backgrounds of laryngomalacia varied among the present patients and those reported previously, supraglottic laryngoplastic surgery was similarly effective. Kay and Goldsmith (2006) have classified laryngomalacia into three types on the basis of pathophysiologic processes and discussed various approaches for surgical repair. Type 1 is characterized by a foreshortened or tight aryepiglottic fold. Type 2 is defined by the presence of redundant soft tissue in the supraglottis. Type 3 is designated for cases with other etiologies, such as underlying neuromuscular disorders. Patient 1 was classified as having type 2, and the mucosa of the arytenoid and aryepiglottic fold regions was removed and/or ablated according to the method described by Gessler et al. (2002), which succeeded in relieving the symptoms initially. Werner et al. (2002) and Awan et al. (2004) have reported patients with laryngomalacia due to isolated posterior displacement and the effectiveness of the epiglottopexy for those with severe laryngomalacia. Patient 2 had a condition similar to that reported by them, and thus may have benefited from epiglottopexy. However, we selected to shorten the tip of the epiglottis, according to the method described by Echternach et al. (2008), as the surgical procedure is simple, and this proved temporarily effective. We reduced the length of the epiglottis by approximately 10 mm, and neither aspiration nor hoarseness were evident after surgery.

Because the results of respiratory function tests were normal in both cases, the indications for surgical therapy and evaluation of outcome were judged on the basis of endoscopic findings, the extent of stridor and subjective breathing complaints. From the viewpoint of symptom improvement, supraglottic laser laryngoplasty was successful in the short term. However, recurrence of symptoms in patient 1 suggested that the efficacy of laryngoplastic laser surgery was insufficient to ensure long-term relief. In the final operation for patient 1, not only laryngoplasty but also additional inferior turbinotomy was carried out to improve nasal respiration, and this has relieved the symptoms up to the time of writing. Furthermore, supportive therapy using anti-allergic agents, which are expected to reduce allergic irritation of the throat, may help to avoid recurrence of AIL in both cases.

Conclusion

Two patients with AIL associated with allergic rhinitis and a high serum IgE level were treated by laser supraglottic laryngoplasty to remove vibrating excess tissue in the

larynx, followed by administration of an anti-allergic agent. This combined treatment was effective for resolving inspiratory stridor and breathing difficulty in the short term. However, the first patient suffered three recurrences. Inferior turbinotomy to improve nasal respiration combined with surgery were useful for diminishing the symptoms. In the treatment of AIL, evaluation and amelioration of allergic factors, especially nasal respiration, appears to be important.

Conflict of Interest

There is no conflict of interest to declare.

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