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CASE REPORT

Acquired idiopathic laryngomalacia in a 12-year-old adolescent: A case report

Minsol Kim,[1] Seungwon Lee,[2] Meeyong Shin[3]

[1]Department of Emergency Medicine, Inha University Hospital, Inha University College of Medicine, Incheon; [2]Department of Otolaryngology, Soonchunhyang University Bucheon Hospital, Soonchunghyang University College of Medicine, Bucheon; [3]Department of Pediatrics, Soonchunhyang University Bucheon Hospital, Soonchunghyang University College of Medicine, Bucheon, Korea

Laryngomalacia, the most common cause of stridor in infants, is characterized by the inward collapse of soft and immature upper laryngeal cartilages during inspiration, resulting in airway obstruction at the supraglottic level. Acquired laryngomalacia is a rare condition that mainly occurs following significant neurological dysfunctions associated with cerebrovascular disease, head and neck surgery, or cervical trauma. We present a case of acquired idiopathic laryngomalacia in a 12-year-old adolescent caused by the prolapse of redundant arytenoid mucosa. The patient exhibited no neurological dysfunctions or laryngeal deformities. However, he had allergic rhinitis accompanied by high serum immunoglobulin E levels. His symptoms worsened after being infected with severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2). Furthermore, allergic rhinitis or SARS-CoV-2 infection may have worsened preexisting asymptomatic congenital or acquired laryngomalacia through neurological damage. Acquired idiopathic laryngomalacia is rale in children. In cases where children and adolescents present with a sudden onset of inspiratory stridor, it is essential to perform a laryngoscopic examination for identifying potential cases of acquired laryngomalacia. (*Allergy Asthma Respir Dis 2024;12:40-43*)

Keywords: Laryngomalacia, Adolescent, SARS-CoV-2, Allergic rhinitis

INTRODUCTION

Congenital laryngomalacia, the most common cause of neonatal and infantile stridor, accounts for 60% of laryngeal anomalies in children. It typically manifests within the first 2 weeks of life and worsens progressively over 6 months; however, gradual improvement can occur. Children commonly exhibit inspiratory stridor, with severe cases often accompanied by cyanosis, choking, feeding difficulties, and failure to thrive.[1] Conversely, acquired laryngomalacia is a relatively uncommon cause of stridor in children. Peron et al.[2] revealed that redundant aryepiglottic fold is a novel cause of stridor based on 7 reported cases. Acquired laryngeal collapse has been extensively studied in elderly patients.[3-5] Most cases are associated with central nervous system injuries, head and neck tumors, or previous head and neck surgery.[6]

We present a case of acquired idiopathic laryngomalacia in a 12-year-old adolescent, which occurred following severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2) infection and was caused by the prolapse of redundant arytenoid mucosa.

CASE REPORT

A 12-year-old boy with a sudden onset of inspiratory stridor and dyspnea was admitted to the pediatrics department of a tertiary care hospital. He had been experiencing a mild intermittent cough for approximately 1 month, which did not improve with antibiotic treatment. The patient contracted a SARS-CoV-2 infection 10 days before his admission, which resulted in a worsened cough and the development of inspiratory stridor accompanied by chest tightness. Despite initial treatment with systemic corticosteroids and inhaled beta-agonist bronchodilator, the symptoms did not improve. The symptoms were not associated with exercise or sleep. About 1 year ago, the patient experienced temporary inspiratory stridor, which resolved spontaneously. In addition, he had a history of allergic rhinitis but no other significant medical conditions, anomalies, or history of foreign body aspiration. The patient complained of cough, difficulty in breathing, and chest tightness, but there was no fever and decreased in food intake. Physical examination revealed an increase in heart rate (130 beats/min) and respiratory

Correspondence to: Meeyong Shin https://orcid.org/0000-0003-3699-8741
Department of Pediatrics, Soonchunhyang University Bucheon Hospital, Soonchunghyang University College of Medicine, 170 Jomaru-ro, Wonmi-gu, Bucheon 14584, Korea
Tel: +82-32-621-5114, Email: smy0218@schmc.ac.kr

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rate (30 breaths/min) with inappropriate respiratory effort and the presence of inspiratory stridor; however, no rales or wheezes were detected. He had a normal body temperature and exhibited no signs of acute laryngeal inflammation like redness or swelling of larynx in laryngoscopy. Head and neck examination revealed no abnormalities; moreover, skull, chest, and neck radiographs did not reveal any nasal, laryngeal, tracheal, or intrathoracic deformities. Hematological and biochemical findings were within the normal limits. In addition, viral and bacterial cultures yielded no growth. Pulmonary function tests, including forced vital capacity (79%) and forced expiratory volume in 1 second (86%), as well as exhaled nitric oxide levels (12 ppb), were within normal limits. Serum immunoglobulin (IgE) level (487 IU/L) was elevated in ImmunoCAP IgE test (ImmunoDiagnostics, Uppsala, Sweden) and it indicated sensitization to house dust mite (*Dermatophagoides farinae*, 87.9 kU/L) and dog dander (1.74 kU/L); however, no sensitization was observed to *Alternaria alternata* (0.03 kU/L), silver birch (0.14 kU/L), ragweed pollen (0.14 kU/L), or Western ragweed (0.27 kU/L). A neck computed tomography revealed a high-density lesion measuring approximately 0.8 cm beneath the right thyroid gland, suggestive of an exophytic right thyroid nodule. However, no lesions were identified that could explain stridor and dyspnea. Despite treatment with IV methylprednisolone 1 mg/kg/day and nebulized epinephrine 0.1 mg/kg/dose twice a day for 3 days, the symptoms did not improve. Therefore, a laryngoscopy was performed by an otolaryngologist under general anesthesia, which revealed an extensive and generalized prolapse of redundant mucosa over the arytenoid cartilage (Fig. 1A). The inspiratory stridor was caused by the prolapse of redundant soft tissue into the glottic space. Partial resection of the arytenoid mucosal hypertrophy was performed by using a potassium titanyl phosphate laser system (Laserscope; AMS, Minnetonka, MN, USA) (Fig. 1B). A mucosal biopsy was conducted under endoscopic guidance. Postoperatively, the patient was closely monitored for respiratory rate and effort in the intensive care unit for 27 hours, and levocetirizine hydrochloride 5 mg was administered orally once a day. The stridor and dyspnea resolved after the resection of redundant arytenoid mucosa; as a result, the patient was discharged after 66 hours of surgery. A follow-up endoscopic examination 7 days postoperatively revealed no scarring or limitations in arytenoid abduction during inspiration. Histological examination of the excised tissue showed normal mucosa without any signs of inflammation or congestion. The patient administered levocetrizine hydrochloride 5 mg once a day for 1 month. At the 2-month follow-up, the patient remained stridor free and exhibited no symptoms or signs of respiratory distress (Fig. 1C).

Data collection was approved by the Institutional Review Board (IRB) of Soonchunhyang University Hospital Bucheon (IRB number: 2023-05-013). The informed consent requirements for this retrospective chart review were waived by the IRB. In conducting the study, informed consent was received from the patient and his parents.

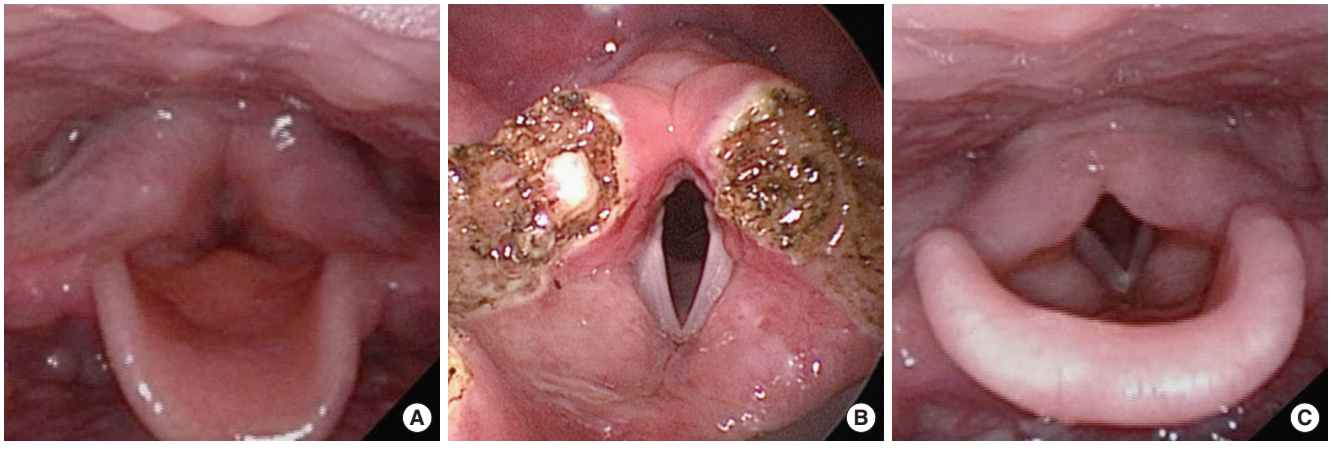

Fig. 1. Fiberoptic findings in the larynx of the patient. (A) Bilateral arytenoid redundant mucosa drops towards the glottic space during inspiration. The audible inspiratory stridor was attributed to the prolapse of redundant soft tissue into the glottis. (B) Bilateral redundant arytenoid mucosa is removed with laser laryngomicrosurgery. (C) A follow-up examination conducted 2 months after the surgery demonstrates the absence of stridor and any symptoms or signs of respiratory distress in the patient.

DISCUSSION

Laryngomalacia, the most common cause of stridor in infants, is characterized by the inward collapse of soft and immature upper laryngeal cartilages during inspiration, resulting in airway obstruction at the supraglottic level. Congenital laryngomalacia resolves completely by 12–18 months. However, other forms of laryngomalacia, such as exercise-induced and idiopathic acquired laryngomalacia, can manifest later in life.[7,8] Our case was classified as acquired laryngomalacia based on the age of onset and the absence of exercise-related symptoms. Acquired laryngomalacia is a rare condition that mainly occurs following significant neurological dysfunctions associated with cerebrovascular disease, head and neck surgery, or cervical trauma.[2,3,6] Idiopathic acquired laryngomalacia, which lacks neurological dysfunctions and laryngeal deformities, is uncommon in children and adults. To our knowledge, this is the first reported case of acquired idiopathic laryngomalacia in children within the South Korea.

Allergic factors can cause glottic narrowing and vocal cord dysfunction.[9] Previous research has reported cases of acquired laryngomalacia in patients with allergic rhinitis and elevated serum IgE levels. These patients underwent laser supraglottic laryngoplasty and inferior turbinectomy and were treated with the antihistamine agent, antileukotriene agent and corticosteroid nasal spray.[10] Most adolescent cases of acquired laryngomalacia are associated with allergic rhinitis, elevated serum IgE levels, and sensitization to aeroallergens.[10] Our patient exhibited high serum IgE levels and strong sensitization reactions to non-seasonal allergens, such as house dust mites and dog dander. The patient underwent laser microsurgery and received continuous administration of the antihistamine agent and anti-leukotriene agent, resulting in no recurrence of symptoms. Further studies are required to better understand the relationship between acquired laryngomalacia and allergic rhinitis.

Acquired laryngomalacia has been reported in 2 patients with obstructive adenotonsillar hypertrophy who experienced worsened stridor following adenotonsillectomy. Of these patients, one underwent supraglottoplasty, whereas the other achieved resolution of airway obstruction through racemic epinephrine and positional adjustments. Progressive alterations of airway dynamics due to obstructive adenotonsillar hypertrophy can result in elevated inspiratory pressures, leading to the development of an acquired form of supraglottic edema and supraglottic prolapse. However, our patient did not experience adenotonsillar enlargement, which indicates no association with secondary laryngomalacia.[5]

Our patient had a mild intermittent cough for a month, and his shortness of breath worsened after being infected with the SARS-CoV-2. In addition, the stridor observed was attributed to the prolapse of redundant arytenoid mucosa. Given the short time interval between stridor onset and SARS-CoV-2 infection, causality cannot be inferred. Furthermore, SARS-CoV-2 infection may have worsened preexisting asymptomatic congenital or acquired laryngomalacia through neurological damage. However, the relationship between SARS-CoV-2 and idiopathic acquired laryngomalacia remains unclear. Therefore, further investigation is needed to better understand this association.

Supraglottoplasty, originally described in 1922, has emerged as the surgery of choice to treat laryngomalacia.[11] Depending on the specific anatomical and functional abnormalities, different techniques are preferred. In patients with abnormally shortened aryepiglottic folds, resection using microscissors, lasers, or microdebrider is recommended.[11-13] Furthermore, in patients with redundant arytenoid mucosa, supraglottoplasty, which involves resection of the aryepiglottic folds, excess mucosa, and lateral surface of the epiglottis, is recommended.[14] However, if the obstruction is caused by epiglottis inhalation, it is recommended to consider partial epiglottectomy or glossoepiglottopexy.[15-17] The choice of surgery for acquired laryngomalacia is similar to that for congenital laryngomalacia. The recommended surgery for irreversible acquired laryngomalacia involves the resection of redundant mucosa of the arytenoids using endoscopic laser surgery. Laser technology in endoscopic procedures enables precise tissue removal while maintaining homeostasis and minimizing postoperative edema. This technique is safe and effective, with no complications.[6] In our case, the patient exhibited generalized prolapse of redundant mucosa over the arytenoid cartilage. Therefore, laser supraglottoplasty was performed, which involves the excision of hypertrophied arytenoid mucosa.

We report the first case of acquired idiopathic laryngomalacia in children within the South Korean. In cases where children and adolescents present with a sudden onset of inspiratory stridor, it is essential to perform a laryngoscopic examination to identify potential cases of acquired laryngomalacia.

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