Official Case Reports Journal of the Asian Pacific Society of Respirology
Respirology Case Reports
OPEN ACCESS

Laryngomalacia presenting as severe uncontrolled asthma

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Keywords
Laryngofiberscopy, laryngomalacia, severe asthma, stridor, supraglottoplasty.

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Received: 5 November 2017; Revised: 26 February 2018; Accepted: 3 March 2018; Associate Editor: Hiromasa Inoue.

***Respirology Case Reports*, 6 (5), 2018, e00316**

doi: 10.1002/rcr2.316

Key message

Congenital laryngomalacia is the most common cause of stridor in infants and usually resolves without therapy by 12–18 months of age. However, a recent study found that laryngomalacia may leave structural and functional traces with increased risk of later respiratory symptoms, suggesting that late-onset laryngomalacia may represent long-term consequences of milder or even undiagnosed forms. Unusual cases demonstrated that inspiratory stridor developed subsequent to upper respiratory tract infections. The lack of airway hyperresponsiveness in adulthood also raised questions regarding the diagnosis of childhood asthma. Laryngomalacia should be distinguished from severe asthma.

Video

The European Respiratory Society/American Thoracic Society severe asthma guidelines state that asthma diagnosis should be confirmed at the first stage. Here, we present a case of laryngomalacia that was followed up as severe asthma at other hospitals.

A 22-year-old woman with a history of allergic rhinitis, atopic dermatitis, and sea food allergy was referred to our hospital for the management of severe uncontrolled asthma. She had a history of childhood asthma and subsequent remission. At the age of 20 years, her asthma relapsed and had not been controlled with adequate asthma treatment, including inhaled corticosteroid and long-acting β2 agonist, leukotriene modifier, and anti-IgE. Asthma Control Test and Asthma Control Questionnaire scores were 17 and 2.0, respectively. The diurnal variability of peak expiratory flow rate (morning/evening, L/min) was 57.1% (250/450). Pulmonary function tests revealed forced vital capacity (FVC) of 2.34 L (81.3% of predicted) and forced expiratory volume in 1 sec (FEV_1) of 2.15 L (81.4% of predicted), and the fractional exhaled nitric oxide level was 14.5 ppb. Total serum IgE was 443 IU/mL, and specific IgE was positive for house dust mite, *Alternaria*, and Japanese cedar and cypress. Tiotropium bromide was added to her medications; however, her asthma condition did not change without acute exacerbation.

Three months later, subsequent to an upper respiratory tract infection, the patient developed inspiratory stridor, which was audible without a stethoscope. A semi-urgent laryngofiberscopic examination revealed an inward prolapse of the bilateral flaccid arytenoid mucosa during inspiration, leading to the diagnosis of laryngomalacia (Video S1, Supporting Information). Her condition was not very critical; systemic corticosteroids were given, showing minimal improvement. After an elective laser supraglottoplasty, her symptoms notably improved (Video S2). The postoperative airway hyperresponsiveness test using methacholine was negative. She is being followed up without any asthma medications.

Congenital laryngomalacia is the most common cause of stridor in infants and is caused by the collapse of supraglottic structures during inspiration [1]. It is considered a benign and self-limiting condition and usually resolves without therapy by 12–18 months of age. However, a recent study found that laryngomalacia may leave structural and functional traces with increased risk of later exercise-induced respiratory symptoms and laryngeal obstruction. It suggested that late-onset laryngomalacia may represent long-term consequences of milder or even undiagnosed forms of congenital laryngomalacia [2]. Unusual young adult cases have been reported, showing that inspiratory stridor developed subsequent to upper

respiratory tract infections and required surgical intervention with supraglottoplasty [3,4]. These findings suggest that this patient might have had undiagnosed laryngomalacia, which became obvious later in life. The lack of airway hyperresponsiveness also raised questions regarding the diagnosis of childhood asthma. Laryngomalacia should be included in the list of diseases that can masquerade as severe asthma.

Disclosure Statement

Appropriate written informed consent was obtained for the publication of this case report and accompanying images.

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Supporting information

Additional Supporting Information may be found in the online version of this article at the publisher's web-site: http://onlinelibrary.wiley.com/doi//suppinfo.

Video S1. Preoperative laryngofiberscopic findings showing a downward vibration of the bilateral flaccid artenoid mucosa during inspiration.

Video S2. Postoperative findings showing no vibration of the mucosa.