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Case Report

Stridor, Snoring and Sleep Apnea in a Three-Year-Old Girl: Report of a Patient with Follicular Lymphoid Hyperplasia

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Abstract

The present study describes a three-year-old girl who was referred to us due to stridor and snoring from one week of age and progressive worsening of sleep apnea two months before our visit. Gastroesophageal reflux disease was ruled out due to being unresponsiveness to treatment. Laryngomalacia was also ruled out because the symptoms and signs were aggravated by passing the time. The presence of a vascular ring was also overruled by echocardiography. Direct laryngoscopy and bronchoscopy were performed to investigate the vocal cord paralysis, glottic stenosis, airway foreign body, adenotonsillar hypertrophy, nasopharyngeal mass, and tracheal stenosis as the etiology. A tonsillar mass was found by them which was confirmed by CT scan. The surgical excision of the mass resulted in the complete resolution of the signs and symptoms. The pathologic studies of the resected mass found it to be follicular lymphoid hyperplasia (FLH), the most common benign lymphoid reaction of the nasopharynx area.

Keywords: Follicular Lymphoid Hyperplasia; Sleep Apnea; Snoring; Pediatric; Tonsillar Mass.

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Introduction

Being considered a frequent sign of upper airway obstruction, stridor and snoring in children have remarkable clinical importance in the diagnosis, treatment, and follow-up of patients with upper airway obstruction (1, 2). Stridor shows the presence of turbulent airflow resulting from partial airway obstruction, and snoring shows the presence of excessive air turbulence. Stridor could be supralaryngeal, supraglottic, glottic subglottic, and tracheal according to the level of the obstruction (1, 3). The snoring could be caused by space-occupying lesions such as adenotonsillar hypertrophy, obstructed nasal airway, flaccid pharyngeal muscles, and excessive length of the soft tissue in the pharyngeal airway (1). Adenotonsillar hypertrophy is considered the most common etiology of snoring and stridor. Benign or neo-

plastic lymphoid proliferation could involve head and neck region compartments, including the nasopharynx, nasal and paranasal sinuses, and salivary glands. The nasopharynx is abounding with lymphoid tissue. The lymphoid tissue of this compartment is tantamount to that of the gastrointestinal tract and is considered a part of the mucosal-associated lymphoid tissue system. The most common benign lymphoid reaction of this site is follicular hyperplasia. Follicular lymphoid hyperplasia (FLH) is not a fully understood entity (4). This lesion is formed due to the dense lymphocyte infiltration within the overlying epithelium, submucosa, and lamina propria. Lymphocyte proliferation is reactive to unknown non-specific antigen stimulus, most commonly antigen of persistent infection (5). This study is going to present a three-year-old girl admitted due to stridor, snoring, and sleep apnea from one week of age, with the first presentation of tonsillar mass, which was diagnosed as an FLH mass.

Case Presentation

A 3-year-old girl was referred to our institution due to sleep apnea and snoring. She was the first offspring born by cesarean section delivery following an uneventful pregnancy. Snoring was present from one week of age and aggravated by feeding. Cyanosis, cough, and asphyxia-like episodes occurred during the first days of life following the feeding. The sleep apnea started two months prior and had worsened progressively; the patient woke up several times at night with remarkable sweating. Gastroesophageal reflux disease (GERD) was suspected to be the cause of snoring first, and Ranitidine was administered. The symptoms were not alleviated, and Ranitidine was discontinued one year later. The next diagnosis was made as laryngomalacia, and she was advised to perform a medical follow-up. The symptoms were worsened besides the appearance and worsening of the sleep apnea despite being more than 18 months old. The patient was referred because of worsened snoring and increased asphyxia-like episodes.

A respiratory rate of 25 per minute, suprasternal retraction, and nasal flaring were detected during the physical examinations. On lung auscultation, stridor was evident, but the cardiac auscultation was normal. No clubbing was found, with a 5-10% weight percentile. No history of dysphagia was present. The differential diagnosis of this condition consisted of Vocal cord paralysis, glottic stenosis, laryngomalacia, airway foreign body, adenotonsillar hypertrophy, nasopharyngeal mass, GERD, vascular ring and tracheal stenosis. The laryngomalacia was ruled out because the symptoms were not resolved by passing the time, The GERD was also ruled out because the symptoms persisted despite Ranitidine administration. Echocardiography was performed to rule out the vascular ring, which showed normal cardia with mild pulmonary hypertension and no vascular ring. For another differential diagnosis, direct laryngoscopy and bronchoscopy studies were done, which revealed normal epiglottis, glottis, and bronchia. A tonsillar mass with a size of 1.5*2 cm with mass effect of glottis was evident on bronchoscopy (**Figure 1**). The head and neck computed tomography (CT) scan showed a tonsillar mass measuring 1.5*2cm, resulting in partial stenosis of the glottis. The mass was resected surgically, and pathologic examinations reported follicular lymphoid hyperplasia (FLH) (**Figures 2 and 3**). The snoring and sleep apnea were resolved thoroughly after surgical excision. The mild pulmonary hypertension found in the initial echocardiography probably occurred due to the mass effect of the mass and chronic hypoxia, which was resolved after the mass resection. The symptoms did not reoccur during the one-year follow-up, with good weight gain.

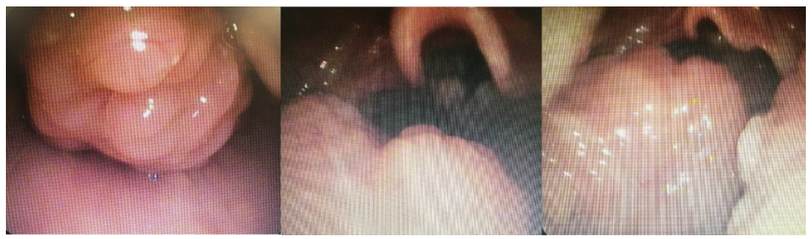

Figure 1. The bronchoscopy findings show a non-ulcerated tonsillar mass.

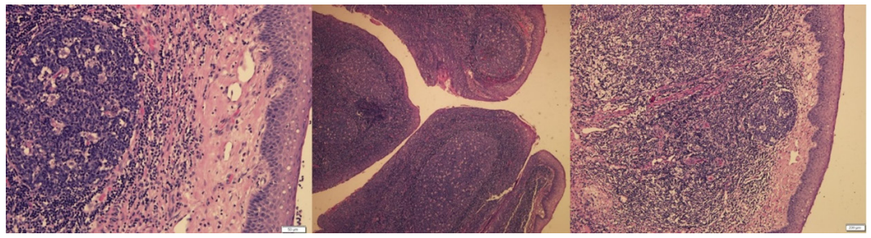

Figure 2. The pathologic study of the resected mass shows lymphoid follicles with germinal centers surrounded by a well-defined mantle zone consisting of small lymphocytes in favor of lymphoid follicular hyperplasia.

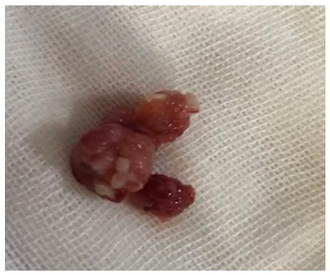

Figure 3. The resected mass.

Discussion

The present case shows our challenge regarding the approach to prolonged stridor and snoring in a 3-year-old girl. GERD was ruled out due to being unresponsive to treatment. Laryngomalacia was also ruled out because the symptoms and signs were aggravated by passing the time. The presence of a vascular ring was also overruled by echocardiography. Direct laryngoscopy and bronchoscopy were performed to investigate the vocal cord paralysis, glottic stenosis, airway foreign body, adenotonsillar hypertrophy, nasopharyngeal mass, and tracheal stenosis as the etiology. They found a tonsillar mass, which was confirmed by a CT scan. The surgical excision of the mass resulted in the complete resolution of the signs and symptoms. The pathologic studies of the resected mass found it to be FLH.

FLH is also known as pseudolymphoma, nodular lymphoid tissue, benign lymphoid hyperplasia, and reactive lymphoid hyperplasia. Different organs, such as skin, orbit, nasopharynx, larynx, lungs, gastrointestinal tract, breast, spleen, pancreas, and liver, could be affected by this condition (6, 7). It is more common in children and young adults, showing follicular pattern and B cell proliferation caused by humoral immune reactions (8). Pellettiere et al. in 1980 reported hoarseness, throat fullness, and dysphagia in a 62-year-old female (9). The lesion on indirect laryngoscopy resembled neoplasia, involving the false vocal cords. Two direct laryngoscopies and one open approach treated her successfully (9). Al-Saleem et al. in 1970 reported a 65-year-old woman with an extensive supraglottic laryngeal lesion, progressive dysphonia, and dysphagia (10). Salked, in 1955, reported a 58-year-old man with dysphagia, intermittent dysphonia, and a large mass attached to the posterior aspect of the left arytenoid cartilage by a broad pedicle on direct laryngoscopy (11). In 2020, Bandino et al. reported that a 66-year-old female presented with intermittent dysphonia and voice hoarseness (12). Pathologic evaluations revealed lymphoid nodules with reactive follicles and epithelial crypts lined by stratified squamous epithelium with no cytological atypia, dysplasia, or malignancy, considered to be ectopic tonsillar lymphoid tissue (12). All studies mentioned above reported FLH lesions in adult populations; however, the present case was a three-year-old girl who had become symptomatic from one week of age. Acar et al. in 2011 reported a 10-year-old boy with upper airway obstruction and infection due to excessive lingual

tonsil and adenoid tissue hyperplasia plus bilateral multiple cervical lymphadenopathies (13). On indirect laryngoscopy, a mass at the root of the tongue descending on both sides all the way to the larynx level was found. An excessively enlarged adenoid tissue filling the nasopharynx was detected at nasal endoscopy. Lingual tonsillectomy and an adenoidectomy, besides the lymphoid tissue resection, were performed under general anesthesia (13). The pathologic study of the resected lymphoid tissue showed reactive follicular hyperplasia (13). In the current study, the FLH mass was found to be tonsillar, causing stridor, snoring, and sleep apnea due to its stenotic nature. All symptoms were resolved following the surgical excision without any recurrence.

From the histologic point of view, FLH includes multiple well-circumscribed lymphoid follicles with the demarcation of the germinal center and mantle zone. Most of these germinal centers are hyperplastic, with both small and large lymphoid cells. The monotonous aspect of lymphoid cells could be mistaken for follicular lymphoma (13, 14). The average age of the affected individuals is 61 years, ranging between 38 and 79, with female prominence (female to male ratio: 3.2:1) (6, 14). The lesion is often presented as a unilateral, slow-growing, and non-ulcerated mass (14, 15). Local surgical excision is the treatment of choice for FLH. The recurrent lesion is rare, and few patients need further interventions. The reported recurrence rate of FLH is almost 16.7% (14). Prolonged follow-up is mostly satisfying without malignant changes. The age of the current case was three years, and the patient responded well to the surgical excision. The one-year follow-up was acceptable without the recurrence of the lesion.

Conclusion

FLH should be suspected in clinical settings of the presence of a mass in the nasopharynx, resulting in symptoms such as chronic, progressive, and resistant-to-treatment stridor, snoring, and sleep apnea. Accurate histologic investigation of the mass and clinical evaluations is necessary for the correct diagnosis in time. It is better to perform a long-term follow-up to rule out the occurrence of malignancies, although no neoplastic alterations have been reported up to the present.

Conflict of Interest

The authors declare that they have no conflict of interest.

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