Case Report

Rigid bronchoscopic management of acute respiratory failure in a 30-year-old woman

Karan Madan, Ashesh Dhungana, Neha Kawatra Madan[1], Anant Mohan, Vijay Hadda, Rakesh Garg[2], Deepali Jain[1], Randeep Guleria

Departments of Pulmonary Medicine and Sleep Disorders, [1]Pathology and [2]Onco-Anaesthesia, All India Institute of Medical Sciences, New Delhi, India

ABSTRACT

A 30-year-old woman presented with a history of progressive shortness of breath, cough, and hoarseness. Stridor was audible on examination. Chest X-ray showed normal lung fields and contrast-enhanced computed tomography thorax showed lower tracheal occlusion with endoluminal growth. Diagnostic flexible bronchoscopy demonstrated multiple whitish glistening nodules over both vocal cords and lower tracheal occlusion by whitish nodular growth. In view of critical central airway obstruction, rigid bronchoscopy and excision of the lower tracheal growth were performed. Histopathological examination of the excised specimen demonstrated features of squamous papillomas. A diagnosis of respiratory papillomatosis was established. On follow-up surveillance bronchoscopy, there was a gradual spontaneous regression of the residual lesions, and the patient remains currently asymptomatic 1 year since the procedure.

KEY WORDS: Bronchoscopy, central airway obstruction, human papilloma virus, respiratory papillomatosis, rigid bronchoscopy

Address for correspondence: Dr. Karan Madan, Department of Pulmonary Medicine and Sleep Disorders, All India Institute of Medical Sciences, Ansari Nagar, New Delhi - 110 029, India. E-mail: drkaranmadan@gmail.com

INTRODUCTION

Respiratory papillomatosis is a disorder characterized by multiple wart-like lesions arising from the upper aerodigestive tract. It is more common in children and has a more aggressive course than in adults.[1] Laryngeal involvement is most common, and other sites of involvement are oral cavity, esophagus, bronchi, and lung parenchyma. Only 5% of cases have tracheal involvement, and majority of these show concurrent laryngeal papillomas.[2] More than 90% cases are caused by the human papillomavirus (HPV) of type 6, 11, 16, and 18.[3]

Presentation with predominant tracheal involvement and with respiratory failure at the time of presentation is unusual. Here, we present a case of a 30-year-old woman presenting with central airway obstruction (CAO) by respiratory papillomatosis who was successfully managed by urgent rigid bronchoscopy and excision of the growth.

CASE REPORT

A 30-year-old woman presented with a history of progressive shortness of breath for 7 months. Dyspnea was insidious in onset and progressed rapidly over the last month, incapacitating the patient in her daily activities. It was associated with cough with expectoration of scant mucoid sputum, noisy breathing, and wheezing for last 1 month. Patient had also noticed hoarseness of voice for the last 6 weeks. There was no history of hemoptysis, facial puffiness, distended neck veins, chest pain, or difficulty

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in swallowing. There were no constitutional symptoms and her past and personal history was unremarkable. She was a lifetime nonsmoker, a homemaker, and had no occupational exposure.

On general physical examination, heart rate was 116/min and blood pressure was 126/78 mm Hg. Stridor was audible which was predominantly expiratory. Patient was tachypneic (respiratory rate - 28/min) and use of accessory muscles of respiration was visible. Oxygen saturation while breathing room air was 92%. On respiratory system examination, expiratory stridor was audible. Rest of the systemic examination was unremarkable. Blood investigations revealed normal hemogram, liver and kidney function tests. Two-dimensional (2D) echocardiography was normal. Arterial blood gas analysis showed respiratory alkalosis and hypoxemia. Chest X-ray showed normal lung fields; however, there was a doubtful luminal narrowing of the lower trachea [Figure 1a]. Contrast-enhanced computed tomography thorax showed the presence of endoluminal soft tissue growth arising from the lower tracheal wall (approximately 2 cm above the carina) causing near complete occlusion of the lower trachea [Figure 1b]. Diagnostic flexible bronchoscopy (using pediatric bronchoscope, 2.8 mm) demonstrated multiple whitish glistening nodules over both vocal cords and lower tracheal occlusion by whitish nodular growth.

In view of critical CAO and impending respiratory failure, urgent rigid bronchoscopy and excision of the lower tracheal growth were performed. Under general anesthesia, mechanical coring of the tracheal growth was performed using rigid bronchoscope tubes of increasing sizes and tumor fragments were removed [Figure 2a]. There were no procedural complications, and airway patency was successfully achieved. Histopathological examination of the excised specimen demonstrated numerous finger-like projection of stratified squamous epithelium supported by connective tissue stroma [Figure 2b]. P-16 immunohistochemical (IHC) staining was positive [Figure 2c]. Diagnosis of respiratory papillomatosis (HPV related) was established.

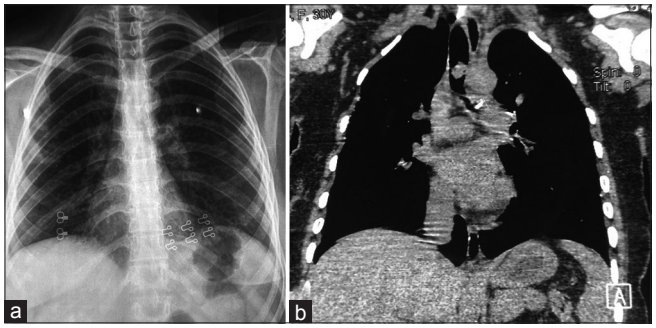

Figure 1: (a) Posteroanterior chest radiograph is essentially normal. (b) Computed tomography thorax demonstrating a large globular endoluminal soft tissue causing near complete occlusion of the lower trachea

Following rigid bronchoscopic excision, follow-up surveillance flexible bronchoscopy at 3, 6, and 12 months postprocedure showed gradual spontaneous resolution of the residual tracheal and vocal cord papillomas and patient remains currently asymptomatic.

DISCUSSION

Respiratory papillomatosis has a very high predilection for involvement of upper respiratory tract as compared to the lower respiratory tract or the lungs. The most common route of transmission of the infection in children is vertical during gestation or delivery. In adults, it occurs as a reactivation of latent HPV infection or is acquired by orogenital contact.

Since the vocal cords are the most common initial site of involvement, hoarseness is usually the first symptom to appear.[4] In our patient, breathlessness appeared before the onset of hoarseness indicating an earlier involvement of the trachea. In view of predominant lower tracheal involvement in our patient, expiratory stridor was more prominent. Other symptoms include chronic cough, recurrent pneumonia, dyspnea, dysphasia, and acute respiratory failure.

Surgical removal of the papilloma is preferred whenever feasible. Tracheostomy for patients with upper airway obstruction due to multiple papilloma in the upper respiratory tract has been used; however, there is also risk of distal dissemination of the infection with the procedure.[5] Since the lesions tend to be multiple, complete surgical excision is often offset by complications such as subglottic and glottis stenosis, web formation, and resulting airway

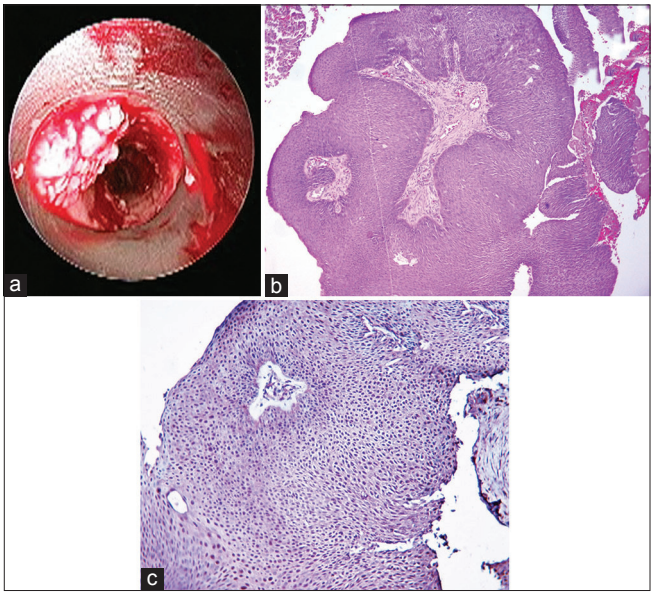

Figure 2: (a) Whitish glistening nodular exophytic endotracheal growth as visualized at the time of rigid bronchoscopic removal. (b) Histopathological examination of the removed growth demonstrating finger-like tissue fragments lined by stratified squamous epithelium showing acanthosis, papillomatosis, and mild nuclear atypia (H and E, ×40). (c) P-16 staining positivity is noted (immunohistochemical × 200)

stenosis. CO_2 laser has been used successfully to cure the disease in a small subset of patients.[6] In view of lower tracheal involvement in our patient, tracheostomy was not attempted, and rigid bronchoscopic excision was undertaken.

Rigid bronchoscopic excision not only permits better control of the airway and simultaneous ventilation but also ensures a better hemostasis and large diameter instruments to be passed through it for endoscopic procedures.[7,8] Adjuvant medical therapy after papilloma excision is indicated for recurrent lesions, multisite spread, and rapid regrowth with airway compromise.[1] Antiviral drugs such as interferon alpha, ribavirin, acyclovir, and cidofovir have been reported in case series to avoid recurrence.

The differential diagnosis for tracheal neoplasms with CAO includes a variety of benign and malignant causes. Ninety percent of tracheal tumors are malignant. Primary malignant tracheal tumor, esophageal carcinoma, and metastasis to the trachea are the most common malignant causes of CAO. Benign tracheal/airway tumors include carcinoids, hamartoma, leiomyoma, lipoma, neurogenic tumors, and inflammatory polyp.[9,10] Other rare tumors include pleomorphic adenoma, amyloidoma, fibroma, squamous papilloma, and hemangioma.

Chest radiology is rarely diagnostic in tracheal tumors as was noted in the index patient. Computed tomography thorax with 3D airway reconstruction is a useful tool to demonstrate the site and size of airway narrowing, its relationship to surrounding structures and to plan interventional bronchoscopic/surgical procedure. They can also be used for objective grading of therapeutic success and for follow-up after intervention.

The typical histology in respiratory papillomas is of finger-like projection of stratified squamous epithelium supported by connective tissue stroma. Almost all papillomas are exophytic and abnormal keratinization, papillomatosis, and basal hyperplasia are identified in 100% cases. Uncommon histopathologic features include dyskeratosis, orthokeratosis, and koilocytosis which are at the most focal. Nuclear atypia if found is almost always mild unless a malignant degeneration has occurred. HPV is epitheliotropic, and majority of respiratory tract papillomas are associated with low-risk virus types, i.e., HPV 6 and 11.[11] P-16 protein expression is a surrogate marker of HPV infection, and IHC analysis of the excised tracheal growth in our patient showed positivity confirming a diagnosis of HPV-related tracheal papillomatosis.[12]

Summary - Benign tracheal tumors are a rare cause of CAO and may cause respiratory failure necessitating urgent management. Rigid bronchoscopic excision is the preferred modality as it achieves better control of ventilation and better hemostasis.[9] Tracheal papillomatosis associated with HPV is rare in adults and have a high propensity to recur; hence, monitoring for recurrence postexcision is required.

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Conflicts of interest
There are no conflicts of interest.

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