Pleomorphic Adenoma of the Subglottis Mistreated as Chronic Obstructive Pulmonary Disease, Report of a Case

Cai Chang, MD, Sun Chenchao, Dong Lei, and Li Yuping

Upper airway obstruction due to subglottic mass can be misdiagnosed. We report the case of a 66-year-old man who was treated for chronic obstructive pulmonary disease (COPD) before a diagnosis of pleomorphic adenoma of the subglottis was made. According to the history of chronic cough and exertional dyspnea, he was treated with inhaled corticosteroids for COPD. Bronchoscopy and computed tomography (CT) revealed a mass occupying the subglottic cavity. After the excision operation, all symptoms disappeared. Histological evaluation revealed the diagnosis of pleomorphic adenoma. This case report emphasizes that not all chronic cough and dyspnea are attributable to COPD.

Key words: pleomorphic adenoma, chronic obstructive pulmonary disease, bronchoscopy, diagnosis

Introduction

Pleomorphic adenoma, relatively the most common benign tumor of major salivary glands, is rare in the trachea and lung. The majority of salivary gland tumors are benign, and about 70% are pleomorphic adenomas.1 To our knowledge, few patients with pleomorphic adenoma of the trachea have been previously reported in the literature.2–4 The clinical differential diagnosis may include various tumors, such as leiomyoma, osteochondroma, adenoid cystic carcinoma and squamous cell carcinoma. The majority of pleomorphic adenomas in the trachea are benign and slow growing. They are believed to arise from tracheobronchial gland epithelium. We present a case of pleomorphic adenoma of the subglottis and the clinical course. This case report provides some information about the diagnosis of pleomorphic adenoma of the subglottis.

Case Presentation

A 66-year-old man was admitted to our hospital with a ten-day history of fever, blood-streaked sputum and dyspnea. The patient was managed with antibiotics in another hospital five days prior to our admission. The chest computed tomography (CT) revealed a large patchy infiltration shadow in the left lower lobe. Because symptoms were not improved, he was referred to our hospital.

On admission, mild respiratory distress was noted
with a respiratory rate of 24/min. But inspiratory stridor was not detected. The rest of the physical examination was unremarkable. The arterial blood gas without oxygen inhalation revealed pH: 7.364, PaO$_2$: 85.9mmHg, PaCO$_2$: 39.0mmHg. The first diagnosis was presumed to be community acquired pneumonia and chronic obstructive pulmonary disease (COPD), and the routine treatment was given initially. But the symptoms were not relieved five days later. The clinical diagnosis was judged to be suspected. The bronchoscopy examination was performed in order to obtain pathogenic bacteria specimen and exclude possible airway obstruction.

Amazingly, an unhoped finding was discovered. Bronchoscopy revealed a fleshy polyloid mass in the subglottic site originating from the membranous part, with obstruction of the lumen. The bronchoscope could be carefully inserted through the lumen, and no other tracheobronchial lesions or extrinsic compressions were found (Fig. 1A and 1B). Biopsy specimens of the tumor surface were not diagnostic. An urgent chest and neck contrast-enhanced CT with multiplanar reconstruction was undertaken. The CT images showed a sharply margined intraluminal mass of 1.5 × 1.4 × 1.2cm in diameter, with partial enhancement in the anterior part, and moderate to severe occluding of the airway (Fig. 1C and 1D).

The smooth surface and localized nature of the mass were consistent with a benign tumor, such as carcinoids and pleomorphic adenoma. Treatment is aimed at relieving airway obstruction with minimum adverse effects. After comprehensive consultation with specialists and the family, the tracheostomy and open surgical resection were regarded as a preferred treatment option.

A horizontal neck incision was made in the neck extension position. The first two tracheal rings were incised, and the mass was apparently located in the subglottic cavity. The tumor was dissected bluntly from
Pleomorphic Adenoma of the Subglottis

the posterior wall of the subglottis through the vertical tracheostomy (Fig. 2A). Intraoperative frozen sections of the margins showed no tumor cells. The final histologic examination revealed a cellular tumor, which was mainly composed of a small to moderate sized glandular structure. The lining oval-shaped or cuboidal epithelial cells, both epithelial and myoepithelial cells included, varied from single layer to multi-layers. Some portion of the tumor showed myxoid stroma, and chondroid stroma was absent in this case. The tumor was clearly cut from adjacent normal tracheal tissue. Immunohistochemical staining was positive for cytokeratin, smooth muscle actine (SMA) and S100 protein, which confirmed the myoepithelial component of the tumor. These findings were compatible with pleomorphic adenoma (Fig. 2B–2D), a very rare finding in the subglottis. A repeat bronchoscopy two months after the operation demonstrated a smooth airway with no appreciable scarring. This patient is alive and well by follow-up.

Discussion

Primary pleomorphic adenoma (mixed tumor), although common in the head-neck area, is rare in the trachea and lung.5) There is no definitive incidence of pulmonary pleomorphic adenoma in the English literature. From the previous reports, it is believed to have contributed to 1% of lung malignancies and between 2% to 9% of all pleomorphic adenomas.6) Histopathologic examination reveals an epithelial component mixed with myxoid and chondroid matrices, and a morphologic diversity can be noted in a single tumor. Immunohistochemical stains prove to be positive for various cytokeratins, S100 protein, vimentin, and SMA. This describes the “mixed” nature of the tumour. Pleomorphic adenoma may resemble aggressive epithelial tumours because of the high cellularity and lack of a stromal component, which can lead to misdiagnosis as a carcinoma. The tumor may occur in the respiratory tract via minor bronchial gland epithelium.7) It does
have a group of symptoms from the obstruction of airway, such as cough, sputum production, dyspnea, and stridor. Approximately many cases may be misdiagnosed, leading to aggressive treatment. Kokturk N, et al. and Aribas OK, et al. respectively reported that the patient with pleomorphic adenoma of the trachea was misdiagnosed as asthma. Because of the rarity and histological variants of the tumor, surgical resection seems to be the reasonable choice.

The goal of the debriefing is to learn lessons and make process improvements that can help us to avoid the misdiagnosis and unnecessary treatment in the future. First, this case emphasizes the fact that not all wheezing and dyspnea are attributable to common causes, such as COPD and asthma. The diagnosis of a slowing-growing tracheal tumor is usually delayed in many cases because the trachea lumen has to be compromised before any localizing signs or symptoms appear. Also, the symptoms can mimic those of more common conditions. It is critically important to probe the cause-effect relationship between the medical presentations and clinical diagnosis. Failure to respond to general therapy suggests that the primary diagnosis should be suspected. In this report, a smoking history with a suggestive onset of symptoms, use of inhaled corticosteroids, recurrent hospitalization for COPD, and consistent findings on physical examination led the physicians initially to postulate a misdiagnosis of COPD. We think that the subglottic mass might have been there, half or one year ago, during the retrospective review of the whole medical course. Because the spirometric measurement prior to operation revealed a forced vital capacity (FVC) 88.1% of predicted, a forced expiratory volume in 1s (FEVI) 105.6% of predicted, and FEV1/FVC 94.7%, without any airway obstruction. The treadmill test was negative too. Our attention was only focused on the superficial phenomenon of symptoms, so that we had overlooked the basic essence of this disease for a long time. The conventional CT scan of chest and trachea should be recommended while facing this kind of case. And endoscopic examination is the best way to evaluate the obstruction. The histopathology in combination with immunohistostaining will be effective to make the correct diagnosis.

Conclusion

The patient with upper airway obstruction presents a great challenge to the judgments and skills of the physician. It is therefore important to bear in mind that the upper airway obstruction can lead to dyspnea in the establishment of a definitive diagnosis. There are many reasons for upper airway obstruction such as infection, angioedema, foreign bodies, and tumors. We suggest consideration of this diagnosis if the patient has a similar obstruction as a presenting complaint. In view of the rare occurrence and recurrent case report, long-term follow-up with endoscope is necessary.

Informed Consent

The Hospital Committee of Medical Research Ethics approved the report.

Written, informed consent was obtained from the patient for publication of this case report and any accompanying images. Details that might disclose the identity of the patient in this report were omitted.

References