CASE REPORT

A case of tracheal pleomorphic adenoma misdiagnosed as asthma

Mamoru Takahashi*, Takahumi Yorozuya, Yuki Miyasaka, Kentaro Kodama, Takumi Yoshikawa, Tetsuya Taya, Yuki Mori, Kimiyuki Ikeda, Satsuki Miyajima, Hirofumi Chiba and Hiroki Takahashi

Department of Respiratory Medicine and Allergology, Sapporo Medical University School of Medicine, Sapporo 060-8543, Japan

*Correspondence address. Department of Respiratory Medicine and Allergology, Sapporo Medical University School of Medicine, Sapporo 060-8543, Japan. Tel: +81-11-611-2111; Fax: +81-11-613-1543; E-mail: tmamoru@sapmed.ac.jp

Abstract

A 51-year-old woman had an incidental finding of a tracheal tumor during oesophagogastroduodenoscopy following the diagnosis of asthma for 2 months. A computed tomography scan revealed a 15-mm tumor in the subglottis. Endoscopic resection was performed safely, and pleomorphic adenoma was diagnosed histologically. The patient's condition was satisfactory 30 months after the procedure. Tracheal pleomorphic adenoma is rare and may be misdiagnosed as asthma. If the tumor is large, surgery may be required; however, endoscopic polypectomy may be effective if the tumor is small. Therefore, early diagnosis of tracheal pleomorphic adenoma is important. At the first visit, the flow–volume curve suggested upper airway obstruction, which should have raised the suspicion of an upper airway obstruction. In patients with suspected asthma, early pulmonary function testing is needed to substantiate asthma diagnosis and prevent an alternative diagnosis being missed.

INTRODUCTION

Benign tracheal tumors are rare, and pleomorphic adenoma is very rare [1]. The main symptoms are chronic cough and stridor [2]. These are similar to the symptoms of asthma, and tracheal pleomorphic adenoma is often misdiagnosed as asthma. Most patients undergo treatment for asthma for a long time until they receive a correct diagnosis [3]. If the tumor is small, it can be removed safely, and therefore early diagnosis is important.

CASE REPORT

A 51-year-old non-smoking woman was referred to our hospital due to cough and wheezing at night for 2 months. She had no remarkable past history or comorbidities. Her physical findings were wheezing in the anterior lung fields. Her chest X-ray and respiratory function tests were normal. She was diagnosed with...
A polyp was subsequently found by chance in the subglottis by esophagogastroduodenoscopy during a thorough medical check-up. A chest computed tomography (CT) scan showed a round tumor (major axis, 15 mm) in the periphery 30 mm from the glottis (Fig. 1). We could hear stridor in the trachea and neck. Laboratory tests and tests for tumor markers, including carcinoembryonic antigen and squamous cell carcinoma antigen, were unremarkable. A pulmonary function test performed at the first visit showed a forced expiratory volume in 1 second (FEV1) of 2.95 L (126.9% of predicted) and a forced vital capacity (FVC) of 3.73 L (133.9% of predicted). The FEV1/FVC ratio was 79.1%, and the shape of the flow–volume curve suggested upper airway obstruction (Fig. 2A). A whole-body contrast-enhanced CT scan found no additional abnormalities. Bronchoscopic examination showed a tumor occluding ∼80% of the tracheal lumen, with a smooth surface and capillary dilatation (Fig. 3A). Since the tracheal tumor was smaller than the tracheal diameter and there was thought to be no risk of tracheal obstruction due to resection, we performed bronchoscopic electrosurgical snaring for diagnosis and treatment. We resected the tumor completely and removed it from the traechae with forceps (Fig. 3B and C). Immediately after the procedure, her symptoms improved significantly. The post-operative course was uneventful. Histological examination revealed pleomorphic adenoma. Histological examination of the resected specimens included abundant epithelial and stromal components (Fig. 4A). The epithelial components contained glandular structures (Fig. 4B). A pulmonary function test performed 2 months after the procedure showed an FEV1 of 3.11 L (133.5% of predicted) and an FVC of 3.80 L (153.7% of predicted). The FEV1/FVC ratio was 81.8%, and the shape of the flow–volume curve was improved to normal (Fig. 2B). Thirty months after the procedure, the patient’s condition was satisfactory, and no abnormal findings were observed by chest CT scan.

DISCUSSION

Pleomorphic adenoma is the most common benign neoplasm of the salivary glands. The adenomas are composed of neoplastic myoepithelial cells intermingled with neoplastic ducts and stroma [4]. They progress slowly and can produce significant
morbidly and, rarely, death. They have been reported to have the possibility of malignant transformation [8]. Primary benign tracheal tumors are rare, and pleomorphic adenoma of the trachea is extremely rare. Gaissert et al. reported 164 cases of salivary gland tumors of tracheobronchial origin at a single center during a 34-year period. Of these, 137 (83.5%) were adenoid cystic carcinomas, 21 (12.8%) were mucoepidermoid carcinomas and only 3 (1.8%) were pleomorphic adenomas [1].

The symptoms of tracheal tumors are stridor, coughing and dyspnea, which are similar to asthma symptoms [5]. In particular, stridor can sometimes be misinterpreted as wheezing.

Stridor is a whistling sound caused when breathing on inspiration, indicating obstruction of the trachea or larynx [6]. Tracheal tumors are difficult to detect by chest X-ray. Because asthma is common in patients who have wheezing and no abnormalities on chest X-ray, tracheal tumors are often misdiagnosed as asthma. If a patient does not improve with asthma treatment, CT scan may find a tracheal tumor [5]. There are reports of patients who were diagnosed with tracheal tumors 2–5 years after the diagnosis of asthma [2,3]. In our patient, careful observation of the breath sounds and pulmonary function tests may have indicated correct diagnosis earlier. We initially could not distinguish stridor from wheeze in breath sounds, but we could hear stridor with careful auscultation. Other reports of tracheal tumors often describe that wheeze is heard instead of stridor in breath sounds but does not give much detail [2,3]. Breath sounds should be carefully auscultated because they are basic physical findings that could differentiate tracheal tumors from asthma. The pulmonary function test, including the flow–volume curve, is a low-cost test compared to a CT scan and is an easy-to-perform examination in patients with suspected asthma. The type of airway obstruction can be estimated from the form of the flow–volume curve. Generally, peak expiratory flow and maximal expiratory flow at 75% reflect obstructive changes of the upper airway, and maximal expiratory flow at 50% and 20% reflect obstructive changes of the peripheral airways. The upper airway obstruction pattern is characterized by loss of the peak and flat portions of the curve. However, in practice, we may overlook the upper airway obstruction pattern because the peak expiratory flow may be affected by the patient’s technique [7]. In this case, if we had observed the form of the flow–volume curve carefully at the first examination, we could have performed a CT scan and found the tumor. A standard treatment for tracheal pleomorphic adenoma has not been established. The electrosurgical snare is one of the safest and most effective interventional techniques [8,9]. However, in the case of large tumors, endoscopic resection may carry a high risk.

Early diagnosis of tracheal tumors is important to initiate correct therapies and prevent the treatment of a misdiagnosed mimic, such as asthma. In patients with suspected asthma, we should carefully auscultate. Flow–volume curves should be carefully inspected, and quality-assured spirometry may be required to aid accurate diagnosis.

ACKNOWLEDGEMENTS
The authors would like to thank Enago (www.enago.com) for the English language review.

CONFLICT OF INTEREST STATEMENT
None declared.

FUNDING
None.

ETHICAL APPROVAL
No approval required.

CONSENT
Written informed consent was obtained from the patient for the publication of this case report.

GUARANTOR
Mamoru Takahashi.
7. Handa H, Huang J, Murgu SD, Mineshita M, Kurimoto N, Colt HG, et al. Assessment of central airway obstruction using impulse oscillometry before and after interventional bronchoscopy. Respir Care 2014;59:231–40 https://doi.org/10.4187/respcare.02094.

8. Bolliger CT, Sutedja TG, Strausz J, Freitag L. Therapeutic bronchoscopy with immediate effect: laser, electrocautery, argon plasma coagulation and stents. Eur Respir J 2006;27:1258–71 https://doi.org/10.1183/09031936.06.00013906.

9. Matsubara M, Yasuo M, Tanabe T, Tsushima K, Urushihata K, Yamamoto H, et al. Pleomorphic adenoma with an endobronchial resection. Intern Med 2008;47:1117–20 https://doi.org/10.2169/internalmedicine.47.0853.

10. Sugiyama M, Yoshino I, Shoji F, Hamatake M, Yohena T, Osoegawa A, et al. Endotracheal surgery for leiomyoma of the trachea. Ann Thorac Cardiovasc Surg 2009;15:206–8 https://doi.org/10.1510/icvts.2007.152280.