

CASE REPORT

A case of tracheal pleomorphic adenoma misdiagnosed as asthma

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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

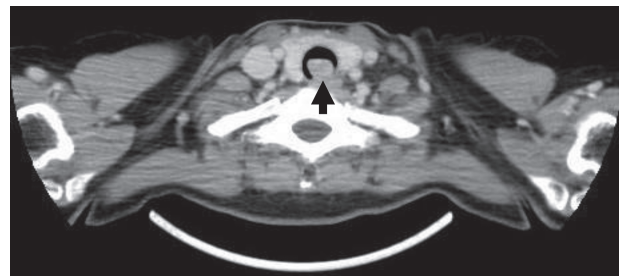


Figure 1: Chest CT findings demonstrated a round tumor arising from the membranous portion of the trachea (arrow).

were wheezing in the anterior lung fields. Her chest X-ray and respiratory function tests were normal. She was diagnosed with

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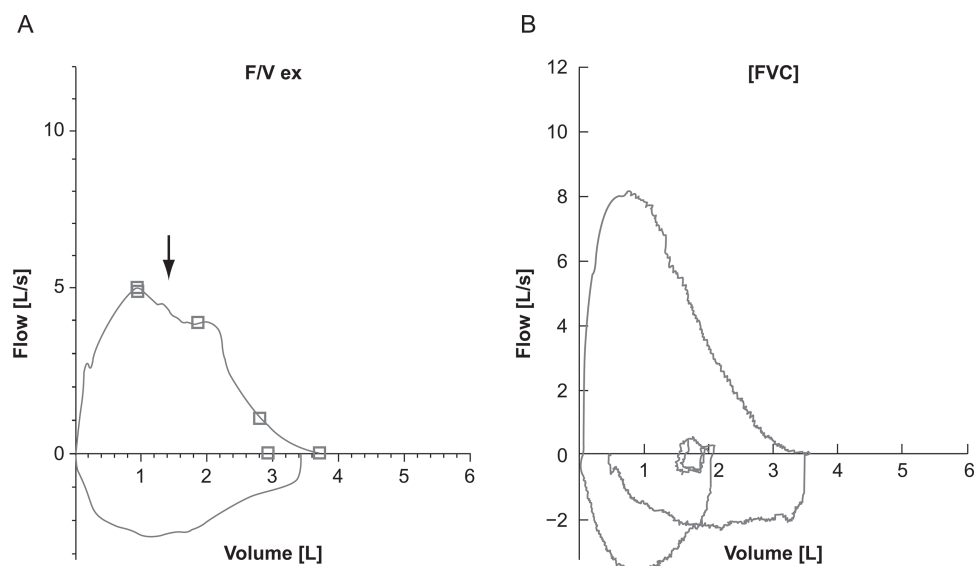


Figure 2: (A) At the first visit, the flow-volume curve showed flattening of the peak (arrow). (B) After intervention, the flow-volume loop showed improvement of the flattening.

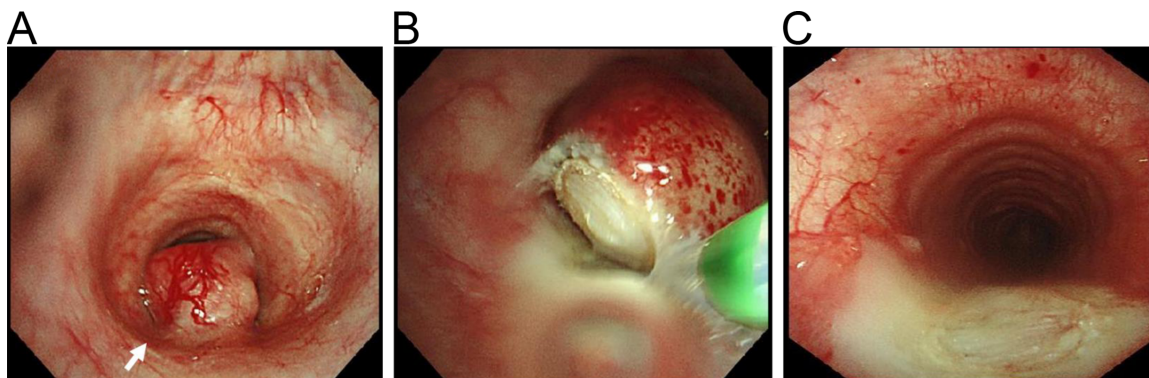


Figure 3: Bronchoscopic findings. (A) The smooth-surfaced tumor is shown in the periphery 30 mm from the glottis and occupies most of the tracheal lumen (arrow). (B) The tumor has just been resected by electrocautery snaring. (C) After snaring, the tumor was resected almost completely and removed from the trachea with forceps.

asthma based on her clinical history and underwent inhaled corticosteroid treatment for 2 months.

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 electrocautery snaring for diagnosis and treatment. We resected

the tumor completely and removed it from the trachea 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

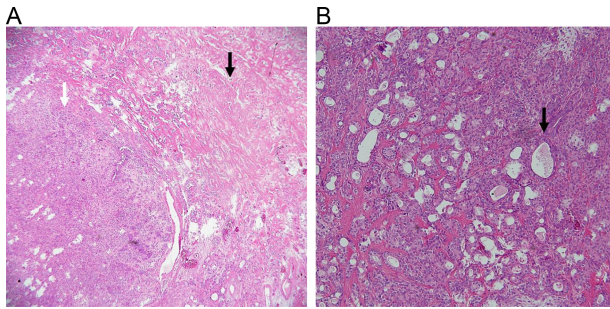


Figure 4: (A) Histological examination of the resected specimens showed both epithelial (white arrow) and stromal (black arrow) components (hematoxylin and eosin staining, $\times 40$). (B) The epithelial components contained glandular structures (arrow) (hematoxylin and eosin staining, $\times 200$).

morbidity and, rarely, death. They have been reported to have the possibility of malignant transformation [4]. 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 electrocautery 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.

[10]. 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.

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CONFLICT OF INTEREST STATEMENT

None declared.

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ETHICAL APPROVAL

No approval required.

CONSENT

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

GUARANTOR

Mamoru Takahashi.

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