



A surprising diagnosis in a patient presenting with urticaria and uncontrolled asthma symptoms: A rare case of pleomorphic adenoma in the trachea

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■ ABSTRACT

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Pleomorphic adenomas are the most common benign tumors of the salivary glands. Although they frequently occur in the parotid gland, they are rarely observed in the trachea. Diagnosis can be delayed because the symptoms may mimic those of asthma. In this case report, we present a case of pleomorphic adenoma in the trachea in a patient who had been experiencing intermittent urticaria for 2 years and had uncontrolled respiratory symptoms for the last 2 months despite clinical asthma treatment. This study aimed to raise awareness among clinicians about differential diagnoses by highlighting the absence of asthma and urticaria symptoms following tracheal lesion removal by thoracic surgery.

Keywords: Asthma, Urticaria, Pleomorphic adenoma, Trachea, Fixed obstruction

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■ INTRODUCTION

Urticaria and asthma are commonly encountered conditions in daily clinical practice. However, there are cases in which the symptoms of these two diseases may indicate rarer and more serious pathologies, necessitating careful differential diagnosis. Pleomorphic adenomas (PA) are benign tumors that typically arise in the salivary glands, albeit rarely observed in the trachea [1], and they may cause symptoms that might resemble those of asthma. The present case describes such a case of PA and is presented to create awareness regarding the possibility of underlying serious pathologies that can manifest with respiratory symptoms and chronic urticaria, even though it is a rare occurrence.

■ CASE REPORT

A 28-year-old female presented to our outpatient clinic of Immunology and Allergy Diseases with complaints of urticaria that had been ongoing for 2 years but had worsened in the last 2 months. She had been receiving asthma treatment for 2 years. The physical examination showed that there was stridor and wheezing. There were no signs of urticaria or angioedema during the physical examination.

The patient underwent a detailed evaluation of the etiology of urticaria. Considering the reported history of uncontrolled asthma despite treatment with high-dose salmeterol-fluticasone and montelukast, high-resolution computed tomography (HRCT) of the chest was performed. HRCT revealed a suspicious lesion in the trachea (Figure 1). Pulmonary function tests showed fixed obstruction in the flow-volume curve (Figure 2). The patient was referred to an otolaryngologist for endoscopic evaluation, which revealed no pathology. A bronchoscopy was then performed, revealing a mass lesion at the level of the second cartilage ring, obstructing 80% of the tracheal lumen (Figure 3). The tracheal lesion was excised via thoracic surgery using rigid bronchoscopy (Figure 4). The pathology report identified the lesion as PA. After the lesion was removed, the fixed upper airway obstruction observed in the flow-volume curve improved (Figure 5). A signed consent form was obtained from the patient on 28/06/2024.

■ DISCUSSION

Urticaria is characterized by hyperemic, itchy, eczematous papules or plaques on the surface of the skin and is classified

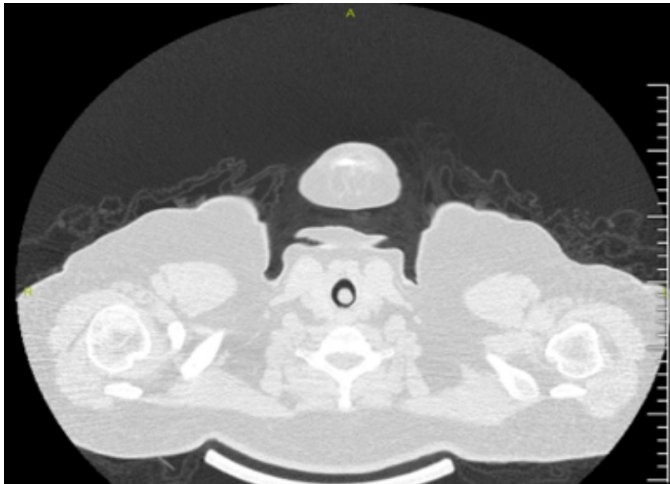


Figure 1. Appearance of a lesion occupying 80% of the tracheal lumen on high-resolution computed tomography.

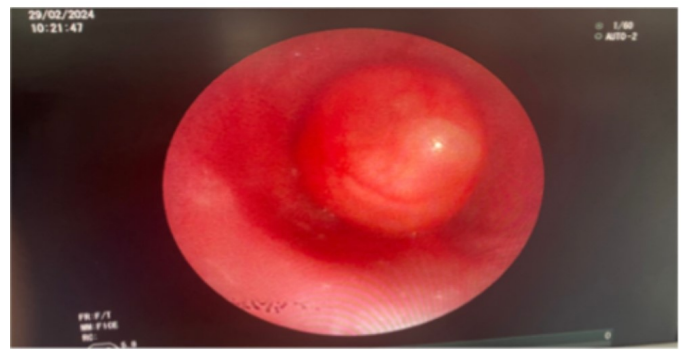


Figure 3. Intraluminal polypoid mass appearance in the trachea at the level of the second cartilage ring during bronchoscopy.



Figure 4. Removal procedure of the polypoid mass occupying 80% of the tracheal lumen at the level of the second cartilage ring.

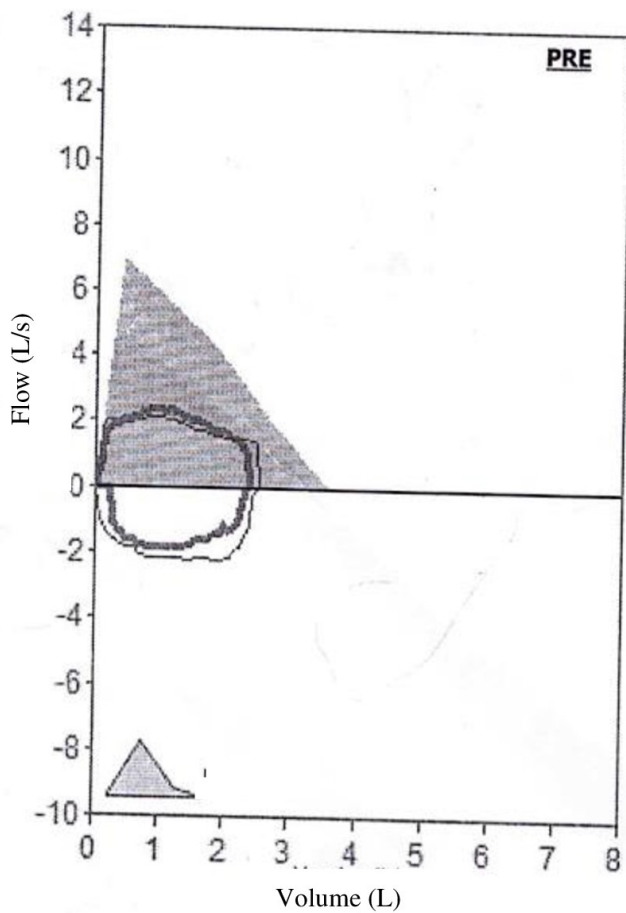


Figure 2. Flow-volume curve showing fixed upper airway obstruction, represented by the characteristic plateau in the inspiratory and expiratory curves.

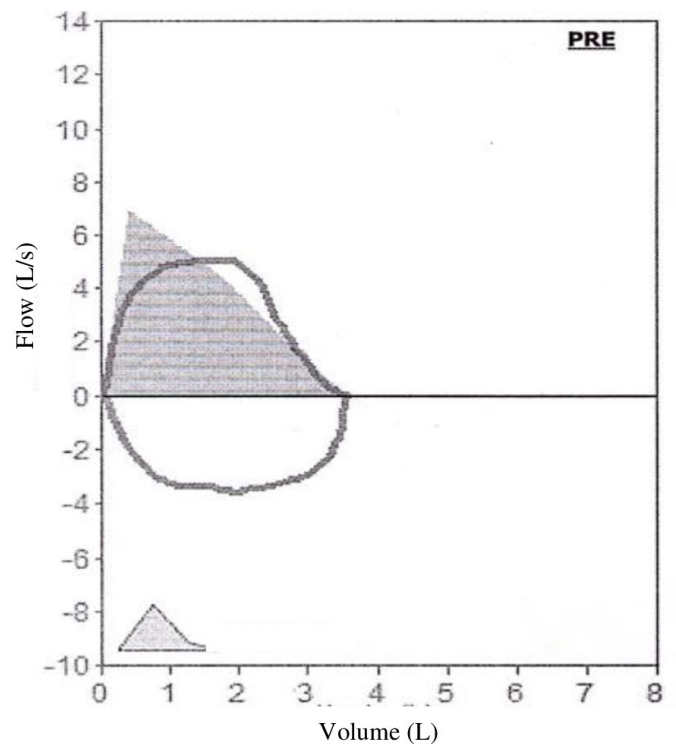


Figure 5. Resolution of fixed upper airway obstruction in the flow-volume curve after lesion removal.

as acute (less than six weeks) or chronic (recurring or lasting more than six weeks) [2]. Common causes of acute urticaria include allergic factors such as infections, medications, insect bites, and foods, as well as etiologies that directly activate mast cells. Chronic urticaria, which is triggered by physical stimuli like heat, cold, exercise, pressure on the skin, water, vi-

bration, or sunlight, is called chronic inducible urticaria. If no physical stimulus is present, it is termed chronic spontaneous urticaria (CSU). CSU has been associated with various allergic causes, thyroid disorders, and autoimmune conditions; however, the relationship between CSU and malignancy remains unclear. Guidelines do not recommend malignant tumor screening unless specific symptoms or findings are present [2-4]. In cases in which urticaria is associated with malignancy, it has been theorized that proteins and hormones secreted by tumor cells may cause urticaria as a part of a neoplastic syndrome [5]. Such cases of urticaria resolve following cancer treatment [6]. However, it is crucial to note that CSU was not determined to increase the risk of malignancy, as reported by a study by Lindelöf et al. [7], which carried out long-term follow-up of 1155 Swedish patients with CSU.

A review of 29 studies involving 6462 patients with CSU identified underlying diseases that could be associated with the condition in only 105 of these patients (1.6%) [8]. Of these, 60 had urticarial vasculitis, 17 had thyroid disease, 7 had lupus, 16 had other connective tissue diseases, 3 had paraproteinemia, 4 had polycythemia vera, and 5 had various malignancies (breast cancer, acute myeloid leukemia, renal cell carcinoma, and two unspecified cancers) as well as other malignancies [8,9]. In a case report by Kartal et al. [10], a patient with urticaria was diagnosed with thyroid papillary cancer, and the regression of urticaria after thyroidectomy emphasized that the association between cancer and urticaria was not a coincidence.

According to the International Urticaria Guidelines, antihistamines (standard dose starting with 1 daily and increasing up to 4) are used as the first step in the treatment of CSU. In unresponsive cases, omalizumab (standard dose 300 mg/4 weeks, but if not sufficient 600 mg/2 weeks) is used as the second step, and in cases that do not respond to these treatments, cyclosporine (5 mg/kg dose) is used as the last step [2]. Although it is not possible to say for sure whether urticaria and PA are directly related in our case, the fact that she did not use antihistamine treatment after the removal of the tracheal lesion and did not have recurrent urticaria suggests a possible connection.

Asthma is a heterogeneous disease characterized by chronic airway inflammation. The condition is defined by symptoms such as wheezing, shortness of breath, chest tightness, and cough, which vary in frequency and time, along with variable expiratory airflow limitation [11]. The diagnosis of asthma is based on characteristic symptoms and the identification of expiratory airflow limitation via pulmonary function testing. Additional tests, including spirometry and laboratory and imaging studies, may be required to confirm asthma or exclude alternative diagnoses that could explain the respiratory symptoms. The goal of asthma treatment includes reducing exacerbations, minimizing permanent airflow limitation, and reducing medication adverse effects. In some patients, asthma control cannot be achieved despite appropri-

ate treatment, and this may necessitate the reassessment of the asthma diagnosis. In our case, HRCT was performed due to uncontrolled asthma despite the Global Initiative for asthma (GINA) step 4 treatment and to investigate the potential common underlying causes (infection, tuberculosis, bronchiectasis, malignancy) for both asthma and urticaria. The patient was diagnosed with a PA in the trachea following advanced investigations prompted by the presence of a tracheal lesion detected on HRCT.

Pleomorphic adenomas (PAs) are typically benign tumors of the salivary glands, though they are also reported in the soft palate, hard palate, upper lip, nasal septum, nasopharynx, orbital region, lower eyelid, buccal mucosa, cheek, external auditory canal, eyelid, and, very rarely, the trachea [8]. A study by Liao et al. examined 29 tracheal PA cases, finding a mean patient age of 48 years (range: 8–83 years) with no sex bias. Over half of these lesions were located in the lower or upper trachea, and the most common symptoms, depending on location, included cough, shortness of breath, stridor, and wheezing [12].

Our patient, a 28-year-old female, presented with stridor, shortness of breath, and wheezing caused by a near-complete obstruction in her upper trachea. PA lesions generally progress over 5.5 months to 10 years; our patient had experienced respiratory symptoms and urticaria for two years. After the lesion's removal, her respiratory symptoms resolved, and the wheezing noted on physical examination completely disappeared. During a 5-month follow-up period, she experienced no recurrence of urticaria.

■ CONCLUSION

Our case is significant for three reasons: First, the patient presented with urticaria and was diagnosed with a tracheal mass. Second, the symptoms and findings caused by the tracheal mass may have led to a misdiagnosis of asthma, potentially delaying treatment. Third, the PA was found in the trachea, a rare location for this type of tumor. Although PA is a benign tumor, the resolution of urticaria symptoms after excision in our case suggests a possible relationship between PA and urticaria, possibly through unknown tumor-related factors. This case highlights the importance for physicians to reassess patients with asthmatic symptoms who do not respond to advanced treatments, as this is a crucial step in differentiating such rare pathologies.

Informed Consent: Written informed consent was obtained from the patient for the publication of the case report.

Peer-review: Externally peer-reviewed.

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■ REFERENCES

1. Gaissert HA, Mark EJ. Tracheobronchial gland tumors. *Cancer Control*. 2006 Oct;13(4):286-94. doi: 10.1177/107327480601300406. PMID: 17075566.
2. Zuberbier T, Abdullatif AH, Abuzakouk M, et al. The international EAACI/GA2LEN/EuroGuiDerm/APAAACI guideline for the definition, classification, diagnosis, and management of urticaria. *Allergy*. 2022 Mar;77(3):734-766. doi: 10.1111/all.15090.
3. Powell RJ, Du Toit GL, Siddique N, et al. BSACI guidelines for the management of chronic urticaria and angio-oedema. *Clin Exp Allergy*. 2007; 37:631. doi: 10.1111/j.1365-2222.2007.02678.x.
4. Tedeschi A, Girolomoni G, Asero R, AAITO Committee for Chronic Urticaria and Pruritus Guidelines. AAITO Position paper. Chronic urticaria: diagnostic workup and treatment. *Eur Ann Allergy Clin Immunol*. 2007; 39:225.
5. De P, Abbasi R, Senadhira T, et al. Urticaria and large cell undifferentiated carcinoma of lung. *DOJ*. 2005;11(3). doi: 10.5070/D39gk2f593.
6. Daniel B, Murrell D. Chronic Urticaria Resolving after Resection of Mucinous Breast Cancer. *J Clin Exp Dermatol Res*. 2010;1(103):2. doi:10.4172/2155-9554.1000103.
7. Lindelöf B, Sigurgeirsson B, Wahlgren CF, et al. Chronic urticaria and cancer: an epidemiological study of 1155 patients. *Br J Dermatol*. 1990; 123:453. doi: 10.1111/j.1365-2133.1990.tb01449.x.
8. Kozel MM, Bossuyt PM, Mekkes JR, et al. Laboratory tests and identified diagnoses in patients with physical and chronic urticaria and angioedema: A systematic review. *J Am Acad Dermatol*. 2023;48(3)409-16. doi: 10.1067/mjd.2003.142.
9. Chen YJ, Wu CY, Shen JL, et al. Cancer risk in patients with chronic urticaria: a population-based cohort study. *Arch Dermatol*. 2012 Jan;148(1):103-8. doi: 10.1001/archdermatol.2011.682.
10. Kartal O, Abdullah B, Ramazan E, Mustafa G, Zafer C, Osman S. Acute urticaria associated with thyroid papillary carcinoma: a case report. *Ann Dermatol*. 2012 Nov;24(4):453-4. doi: 10.5021/ad.2012.24.4.453.
11. Global Initiative for asthma (GINA). Global strategy for asthma management and prevention; 2023 (Available at: www.ginasthma.org. Accessed on: 17.07.2023).
12. Liao, Qian-Nuan, et al. "Pleomorphic adenoma of the trachea: A case report and review of the literature. *World J Clin Cases*. 2020 Dec 6;8(23):6026-6035. doi: 10.12998/wjcc.v8.i23.6026.