A Rare Tumor Mimicking Bronchial Asthma

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ABSTRACT

Wheezing is a common symptom that leads us to assume most cases are obstructive airway disease. We present a case of a 45-year-old female without any comorbidities who presented with recurrent episodes of wheezing and was treated at multiple centers with escalating doses of inhaled corticosteroids and bronchodilators. She was then diagnosed with adenoid cystic carcinoma when she developed post-obstructive pneumonia and presented to our center. She underwent complete resection with no residual disease and no further requirement for any inhalers.

Keywords: Asthma, Wheeze, Bronchoscopy, Bronchoalveolar lavage

INTRODUCTION

Adenoid cystic carcinoma (ACC) is a rare malignancy most commonly arising in the salivary glands but can also occur in other locations, including the respiratory tract[1]. Primary ACC of the lung is an uncommon form of pulmonary neoplasm, accounting for 0.02-0.4% of all lung cancers[2]. It typically arises from the submucosal glands of the tracheobronchial tree and is characterized by slow growth and a propensity for perineural invasion[3]. Diagnosis is often delayed, such as in this case, which we are presenting, where the patient was mostly managed as bronchial asthma for wheezing.

CASE

A 45-year-old female, a non-smoker without any past co-morbidities, presented to the pulmonology OPD with complaints of intermittent cough and wheezing for 5 years. The cough was mostly dry but occasionally productive with whitish mucoid sputum, a teaspoonful, with no episodes of hemoptysis and no history suggestive of any diurnal or past seasonal variation. The patient also complained of wheezing, which she and her partner noticed happened after bouts of cough and after exertion, without any diurnal variation or after exposure to anything specific. There was a history of occasional shortness of breath, but non-exertional and without orthopnea and paroxysmal nocturnal dyspnea (PND). There was also no history of any constitutional symptoms of fever, weight loss, or night sweats, and no history of contact with tuberculosis or any recent travel. Her family history was not significant for any respiratory illness, and she did not claim to have any pets or birds at home.

She was treated for bronchial asthma at multiple other centers with the use of formoterol and inhaled corticosteroid inhalers with occasional salbutamol use. But even after multiple centers, dose increments, occasional oral steroids, and antibiotics, her symptoms did not resolve. All previous documents showed significant findings of wheezing in all of her examinations without any other remarkable features.

During this visit to our center, she presented with complaints of fever, cough, and increased wheezing for 7 days. The patient was started on oral beta-lactams and macrolides before presentation. She had a normal general examination, but her respiratory system examination revealed the previously stated bilateral expiratory polyphonic wheeze without any other significant findings. The initial chest x-ray was normal (Figure 1-A) with slight neutrophilia but otherwise normal blood parameters. The patient was advised to continue oral antibiotics with the addition of oral steroids and asked to follow up after 5 days.

However, after 5 days, there was a slight improvement in symptoms, but on examination, wheezing persisted. The patient was then admitted for further workup, and a repeat chest x-ray showed slight infiltration over the right upper lobe (Figure 1-B). Her blood counts were within the normal range with a normal metabolic panel. Other inflammatory markers were slightly raised, with an erythrocyte sedimentation rate (ESR) of 50 mm/hr and a C-reactive protein (CRP) of 27 mg/dl. Sputum for AFB, GeneXpert, and culture was negative. Previously done spirometry showed features of moderate obstructive airway obstruction (FEV1/FVC: 53.2%) without any reversibility. Serum IgE was within normal limits.

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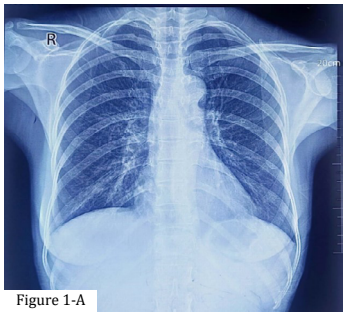
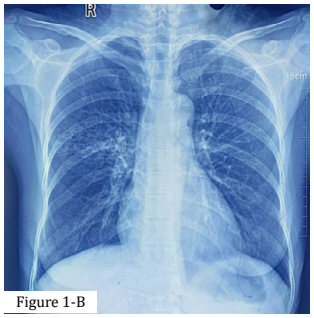


Figure 1-A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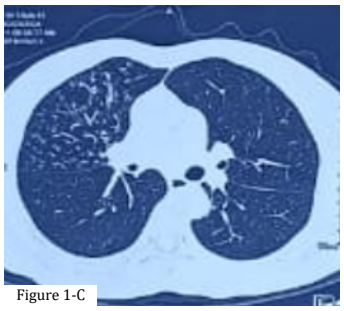
Figure 1-C

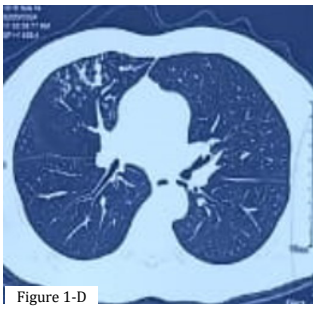
Figure 1-D

Figure 1-A. Chest X-ray prior to presentation, 1-B. Chest X-ray at current presentation showing right upper lobe infiltration. Figure 1-C, D HRCT showing diffuse centrilobular nodularities with Ground Glass Opacity (GGO) in a linear branching pattern giving a tree-in-bud appearance in the anterior segment of the right upper lobe and middle lobe.

HRCT chest was done, which showed centrilobular nodules and ground glass opacities over the right lung field with fibrobronchiectatic changes over the right upper lung field (Figures 1-C and 1-D). In view of persistent symptoms, the patient was planned for bronchoscopy and bronchoalveolar lavage (BAL) assessment. However, during bronchoscopy, a polypoidal growth causing partial luminal obstruction was noted in the right intermediate bronchus with normal underlying segments. A biopsy from the specimen was taken, and BAL fluid for analysis was collected. BAL GeneXpert for tuberculosis was normal with no growth in cultures. A biopsy of the growth revealed findings suggestive of adenoid cystic carcinoma.

The patient then underwent evaluation for any metastatic disease and preoperative workup, and as the patient had limited disease, she underwent lobectomy with clear margins and without any perineural invasion. Postoperatively, at 6 months follow-up, she is now asymptomatic without any wheezing and the need for any inhalers.


Figure 2-A

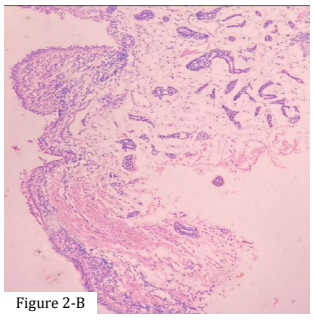
Figure 2-B

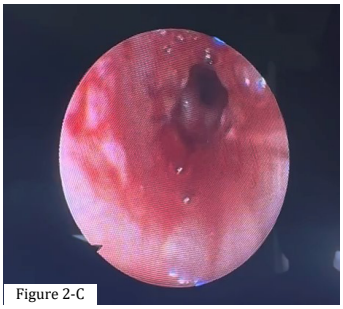
Figure 2-C

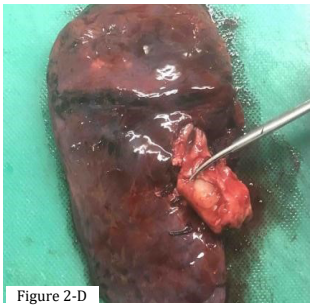
Figure 2-D

Figure 2-A The right intermediate bronchus exhibits growth in the bronchoscopy image. Figure 2-B Biopsy of the growth showing bronchial tissue infiltrated by cords, aggregates, and tubules of basaloid cells with a small amount of cytoplasm and oval hyperchromatic nucleus with mucoid materials. Figure 2-C Post-resection anastomosis bronchoscopy. Figure 2-D Resected specimen following right upper lobectomy.

DISCUSSION

Adenoid cystic cancer is a malignant neoplasm arising mostly from the salivary glands but may also arise from the submucosal glands, in the tracheobronchial tree and may mimic asthma or chronic bronchitis due to airway obstruction[4]. In a study done on 40 patients by Molina et al., the presenting symptom was cough (70%), followed by dyspnea (51.7%), wheezing (38.3%), obstructive pneumonia (30%), hemoptysis (28.3%), and fever (16.7%)[5]. The median time to diagnosis from symptom is around 4-15 months, showing the indolent nature of the disease[6].

Surgical resection is the mainstay of treatment, especially in limited disease. Although late local recurrence is a feature, excellent long-term palliation is commonly achieved with complete or incomplete resection[7]. 5-year survival rates in patients who underwent surgical resection have been shown to be around 87%, while for those with unresectable tumors, it is around 33%[8].

CONCLUSION

Adenoid cystic carcinoma is a rare and indolent malignancy that poses diagnostic challenges. Patients may present with wheezing when treated with bronchodilators, but if an adequate response is not seen, further testing should be sought, such as bronchoscopy, as CT may not reveal endobronchial lesions, as in this case. There is good potential for complete surgical resection, but long-term surveillance for late recurrence is essential.

FUNDING

None

INFORMED CONSENT STATEMENT

The informed consent was obtained from the patient.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

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CONFLICT OF STATEMENT

None

REFERENCES

1. Moran CA, Suster S, Koss MN. Primary adenoid cystic carcinoma of the lung. A clinicopathologic and immunohistochemical study of 16 cases. Cancer. 1994 Mar;73(5):1390–7.
2. Thornton G, Sivarajan L, Gillespie C, Corbridge T. Primary Pulmonary Adenoid Cystic Carcinoma: A Case Report. Chest. 2017 Oct;152(4):A671.
3. Travis WD, Travis LB, Devesa SS. Lung cancer. Cancer. 1995;75(1 Suppl):191–202.
4. Jang S, Patel PN, Kimple RJ, McCulloch TM. Clinical Outcomes and Prognostic Factors of Adenoid Cystic Carcinoma of the Head and Neck. Anticancer Res. 2017 Jun;37(6):3045.
5. Molina JR, Aubry MC, Lewis JE, Wampfler JA, Williams BA, Midthun DE, et al. Primary salivary gland-type lung cancer: Spectrum of clinical presentation, histopathologic and prognostic factors. Cancer. 2007 Nov 15;110(10):2253–9.
6. Hu MM, Hu Y, He JB, Li BL. Primary adenoid cystic carcinoma of the lung: Clinicopathological features, treatment and results. Oncology Lett. 2015;9(3):1475–81.
7. Maziak DE, Todd TRJ, Keshavjee SH, Winton TL, Van Nostrand P, Pearson FG, et al. Adenoid cystic carcinoma of the airway: Thirty-two-year experience. J Thorac Cardiovasc Surg. 1996;112(6):1522–32.
8. Tan X, Xu T, Shen W, Ai C, Zhang W, Tang X, et al. Primary pulmonary adenoid cystic carcinoma: A clinicopathological study of 64 patients. Thorac Cancer. 2024 Feb;15(5):386–93.