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Case Report
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A Young Sergeant with Long Standing Stridor

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Abstract

We are reporting a case of unusual presentation of stridor in an adult man, previously labeled as a case of bronchial asthma, who was identified to have an Adenoid Cystic Carcinoma of trachea (ACC). This resulted in chronic progressive upper airway obstruction and subsequent airway stridor. Following proper diagnostic workup that includes bronchoscopy and histopathological examination of tissue biopsied, the involved tracheal segment was surgically resected followed by reconstruction and involved field radiotherapy.

Keywords: Adenoid cystic carcinoma; Stridor; Pulmonology; OPD

Case Presentation

A 34 year old traffic warden, non smoker with no relevant personal history presented in Pulmonology OPD with complaints of worsening of noisy breathing for the last two years. He was alright about 12 years back when he felt this noisy breathing first time and he visited local physician. He was labeled as a case of "Bronchial Asthma" and advised some treatment that he quit just after taking two to three doses as he got relief. Since then he felt similar issues intermittently and took some home remedies to control this symptom. From last 3 years, this issue (noisy breathing/ stridor) worsened and he visited ENT specialist. He was labeled as a case of "chronic sinusitis" despite he had no nasal symptoms and advised treatment accordingly. He remained symptomatic and presented to general physician, who, after examining him, advised a CT SCAN chest (Figure 1a). With that CT scan, he presented in Pulmonology OPD. On examination he had audible stridor with no other significant findings. CT scan, that he was carrying, showed mass at level of upper trachea with marked luminal narrowing. He was admitted and scheduled for bronchoscopy that showed "multi-lobulated mass in the upper segment of trachea" (Figure 1b), that revealed "Adenoid cystic carcinoma of trachea" on histopathology (Figure 2a). Thoracic surgeon was consulted and advised to get CT scan neck and chest with reconstruction images that was done (Figure 2b) and was referred for surgery. Resection of involved segment was done and rest was reconstructed. He remained on surgical floor for 5 days and discharged with advised to get radiotherapy.

He visited our OPD, breathing comfortably without any stridor. He was under radiotherapy currently and resumed his duties.

Discussion

Adenoid Cystic/adenoid cystic carcinoma or cylindroma of the trachea is a relatively rare occurrence. Adenoid cystic carcinoma is second to squamous cell carcinoma among tracheal malignant neoplasms, and has a nature of long clinical course and less malignant potential. ACC of trachea is reported. In 5th decade without any gender predilection; bur, interestingly, our patient was in his thirties at the time of diagnosis; and smoking has nothing to do with incidence of ACC [1]. Surprisingly, many patients with ACC of trachea remained on anti-asthma treatment for long time same as in presenting case and had frequent visits to emergency department due to poorly controlled symptoms.

Commonest site for metastasis of ACC is pulmonary system but even after metastasis it remained asymptomatic for long time [1]. To detect loco-regional involvement, extension and either resectable or not, CT-scan is a favorite tool. Bronchoscopy has its own importance, as it allows tissue sampling for proper diagnosis and to assess local features/extension [2]. Surgery with or without radiotherapy is the available treatment options. Primary resection of tumor through surgery followed by end to end anastomosis is the best treatment. ACCs are incompletely resected usually due to narrow tissue plans. The complete resection rates ranged from 42% to 57% [3]. For cases, who remained unresectable due to its local invasion, and when surgical clearance is not satisfactory radiotherapy is recommended. ACC of trachea recur locally is common and occurs at an average of 51 months after

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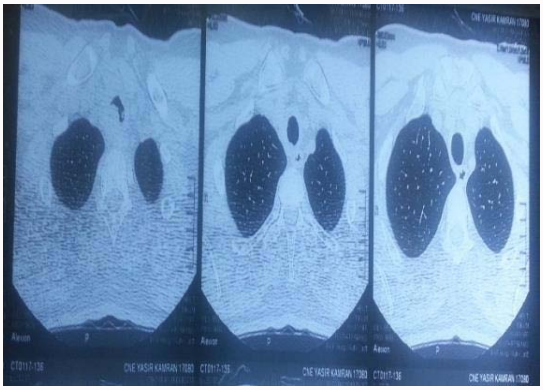

Figure 1a: CT scan image showing narrowing of tracheal lumen, irregular shape.

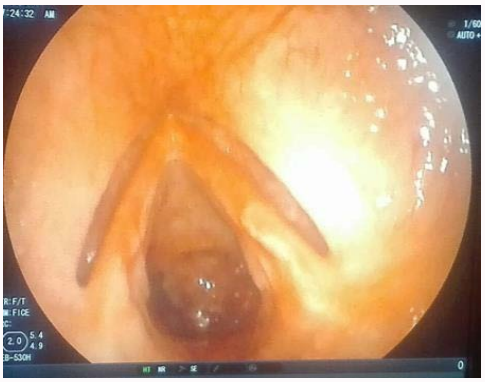

Figure 1b: Bronchoscopic view of the tracheal mass.

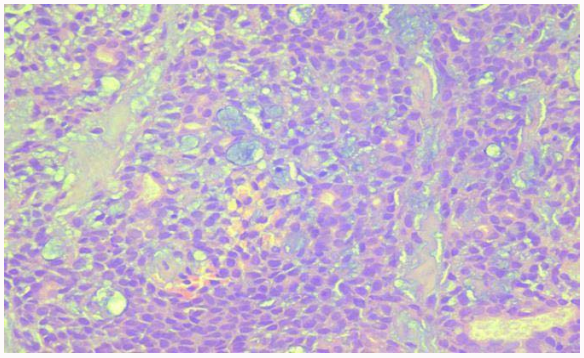

Figure 2a: Histopathological image of biopsied specimen showing cystic adenoid carcinoma features.

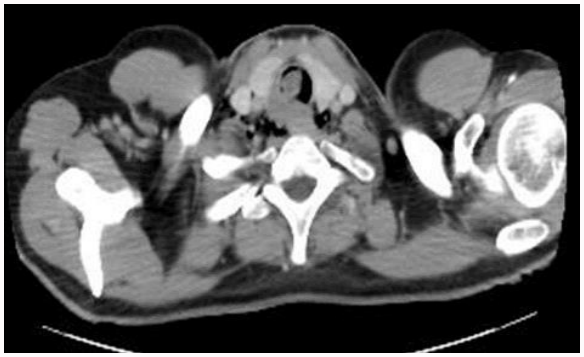

Figure 2b: CT scan neck with reconstruction images showing irregular narrowing of tracheal lumen with growth.

the primary treatment [4]. The overall survival rate is 52% to 91% (5 years) and 29% to 76% (10 years) respectively [5]. Similar case was also reported by our team previously in a young lady without any risk factors, who had favorable outcome like this case [6].

Conclusion

Adenoid cystic carcinoma is a rare confounder to diagnosis and treatment of diseases that usually presents with wheezing, and most common is bronchial asthma. Proper clinical history, clinical examination and investigations guide to early diagnosis and prompt treatment for ACC of trachea. CT scan neck and chest with reconstruction images and bronchoscopic local examination is the key to diagnosis. Proper surgical clearance followed by radiotherapy is available options.

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