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Case Report

A case of tracheal tumor with unusual bronchoscopic presentation: A case report

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ABSTRACT

Primary tumor of the trachea and bronchial tree is rare. Primary tracheobronchial malignancies make up about 0.1% of all pulmonary tumors. The survival rate depends on several factors which include the malignant potential of the tumor, the patient's comorbidities, and the location of the tumor. The index case is a 60-year-old woman who had 7 months history of globus with an associated history of central chest discomfort and cough of 3 months duration. There was a positive history of dysphagia to solid meals, odynophagia, dyspepsia, nausea, excessive belching, bloating, and regurgitation. These symptoms led to the diagnosis of gastroesophageal reflux disease. However, the persistent symptom of globus and worsening cough necessitated her referral to the chest clinic. Chest radiograph revealed opacity in the left upper lobe. Chest computerized tomographic scan showed a solitary, lobulated, and calcified soft-tissue mass in the apicoposterior segment of the left upper lobe with loss of volume. Bronchoscopy showed multiple planes of non-glistering submucosa lesions which were biopsied. The histology report revealed invasive adenocarcinoma of the airway. The surgical resection of tumors with stents was done and she was discharged home 2 weeks later. However, the patient refused adjunct chemotherapy. She is alive and well with complete resolution of symptoms. Early presentation, availability of investigative tools, and prompt treatment may improve survival in cases of trachea tumors as seen in the index case. A high index of suspicion is also needed to make a prompt diagnosis of trachea tumors, especially in patients presenting with globus.

Keywords: Adenocarcinoma, Bronchus, Globus, Trachea

INTRODUCTION

Primary tumor of the trachea and bronchial tree is rare. These tumors are in most cases malignant and are usually at an advanced stage at presentation.[1] The most common malignant tumors of the trachea are adenoid cystic carcinoma (ACC), squamous cell carcinoma, mucoepidermoid carcinoma (MEC), and carcinoid/neuroendocrine tumors, while the most common benign airway tumor is squamous papilloma (associated with human papilloma virus types 6 and 11).[1]

Primary tracheobronchial malignancies make up about 0.1% of all pulmonary tumors.[2] The most common types are MEC and ACC.[3] The actual prevalence of tracheobronchial tumors is difficult to determine.[4] The survival rate of rare airway tumors depends on several factors which include the malignant potential of the tumor, the patient's comorbidities, and location of the tumor as well as access to treatment modalities.[5]

Patients often present with dyspnea, cough, wheezing, hemoptysis, and stridor. Hemoptysis is usually associated with squamous cell carcinoma and often leads to early diagnosis. However, wheezing and stridor occur more often in the adenoid cystic variant. When dysphagia and hoarseness of voice are present, they indicate advanced disease.[6]

Benign tumors are usually localized and are usually treated with surgical resection with a very low recurrence rate.[5,6] Tumors that are treated with endoscopic excision have varying degrees of recurrence, but re-excision is usually feasible. Imaging and bronchoscopy are usually used to survey for possible recurrence.[5,6]

The outcome of malignant tumors varies from one to another and it depends on where it is located, if the lymph node is involved and if there is a mediastinal invasion of vital organs.[7]

CASE REPORT

A 60-year-old woman presented in the gastroenterology clinic with a feeling of a lump in her throat (globus), central chest discomfort, and productive cough of 7- and 3-month durations, respectively. Globus was of sudden onset and associated with dysphagia and odynophagia. She also had occasional dyspeptic symptoms, nausea, excessive belching, bloating, and regurgitation often relieved by antacid. There was no significant weight loss and no hemoptysis. She had a uvulectomy 3 days before the presentation. No family history of cancers.

On examination, she was audibly wheezing with flaring of alae nasi. Other general examination findings were essentially normal. Oxygen saturation was 90% in room air. She was tachypneic (respiratory rate was 22 cycles/min), and the trachea was central with normal chest excursion bilaterally and vesicular breath sounds global. An initial diagnosis of gastroesophageal reflux disease was made to exclude acid peptic disease and the patient was placed on medications. However, the upper gastrointestinal endoscopy done showed moderate hyperemia of the lower one-third of the esophageal mucosa, short tongues of Barrett esophagus, and mildly inflamed and nodular gastric antrum. Histology of gastric and esophageal tissues revealed chronic active gastritis.

She was seen by an ear, nose, and throat surgeon who noticed mild tenderness in the left submandibular region on examination. The laryngoscopy done was normal. She was subsequently reviewed in the chest clinic with worsening cough, mostly at night, and early morning generalized headache. Chest examination revealed the presence of bronchial breath sound in the left upper and middle lung zones which resulted in a diagnosis of aspiration pneumonia. Her chest radiograph showed opacities in the left upper lobe and apex [Figure 1] but sputum GeneXpert was negative for *Mycobacterium tuberculosis*. She was treated with oral antibiotics for 7 days. Chest computerized tomography scan done subsequently showed a solitary soft-tissue mass in the left lung with lobulated outline and multiple calcific densities with a popcorn appearance in the apicoposterior segment of the left upper lobe and loss of volume. No invasion of adjacent bone was noted [Figure 2].

Bronchoscopy and biopsy revealed papillomatous lesions in the trachea, left main bronchus, and right main bronchus with extrinsic lesions compressing the left main stem/upper lobe bronchus [Figures 3 and 4]. Histology of the biopsy sample showed clusters and solitary atypical cells with foamy/vacuolated cytoplasm and hyperchromatic nuclei within a husky background [Figure 5a] suggestive of invasive adenocarcinoma. The immunohistochemistry showed negative immunostaining to novel aspartic proteinase of the pepsin family A (Napsin A) [Figure 5b], pan-cytokeratin (pan-CK) [Figure 5c], and thyroid transcription factor-1 [Figure 5d].

The results of other laboratory tests done were all essentially normal.

She was jointly reviewed by the cardiothoracic surgeon and the chest physician 22 days after first presentation and a diagnosis of Bhattacharyya Stage 3 tumor (T3N0) was made. The surgical resection of tumors was done and she was discharged home 15 days later. She was scheduled for adjunct chemotherapy following surgical resection, which she declined. She is, however, alive and well with complete resolution of symptoms.

DISCUSSION

This case presentation showed that trachea tumors are uncommon and the description of a new case, especially in African, enhances the understanding of this disease as it accounts for <1% of all malignancies.[8] The bronchoscopic

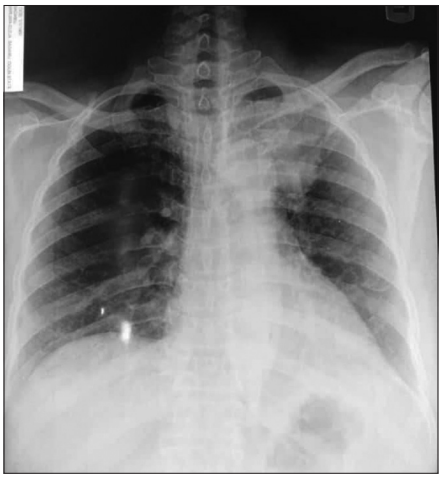

Figure 1: The plain chest radiograph showing opacities in the left upper lobe and apex.

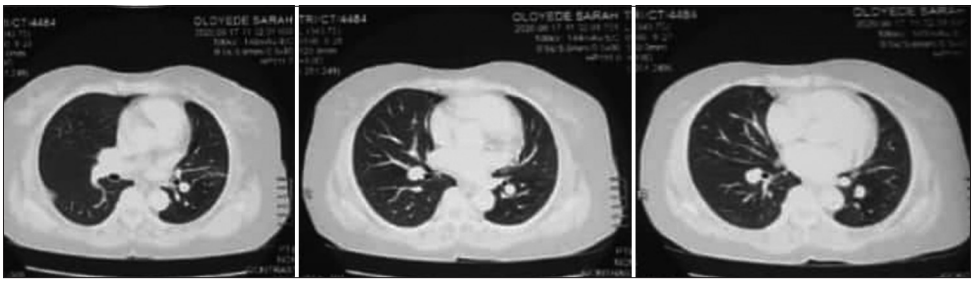

Figure 2: Chest CT scan (axial view) showing a solitary left upper lung soft-tissue mass with a lobulated outline in the apicoposterior segment of the left upper lobe associated with loss of volume in the affected segment.

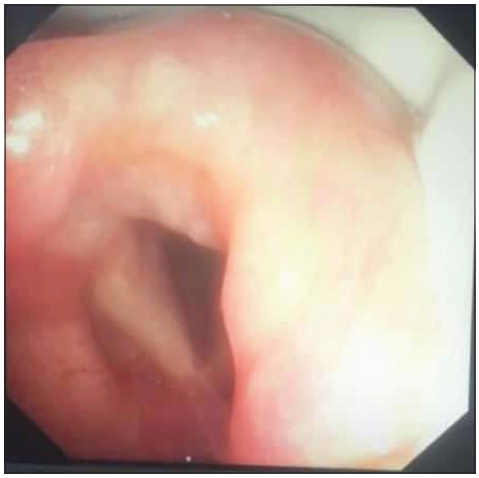

Figure 3: A lesion compressing the left main stem/upper lobe bronchus.

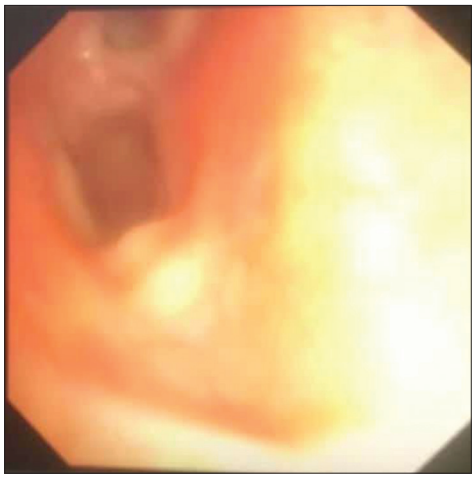

Figure 4: Multiple glistering lesions on the lateral and posterior walls of the right main bronchus.

findings were also uncommon and contrast with previous descriptions.[8] The majority of primary trachea tumors present with severe airway obstruction.[9] A major dilemma in tracheal tumors is the modes of presentation. Findings showed a wide range of symptom profiles and chest radiographic findings. Cough, obstructive symptoms, and hemoptysis were the most common clinical features.[9] This finding was, however, different from this index case in which the dominant clinical presentations were globus and chest discomfort. This may be explained by the early diagnosis of the tumor in this patient. The early diagnosis has been found to be the important factor affecting overall outcome in patients with trachea tumors.[10] Our patient did not present with hemoptysis and we did not find any blood in the airways and the tumor was not bleeding.

Tracheal tumors are treatable and curable when diagnosed early.[8] In most cases, however, insidious onset of symptoms and lack of investigative tools needed for diagnosis often lead to delay in diagnosis, making these potentially treatable disease difficult to manage causing preventable deaths. Our approach to this case was timely which probably led to the preservation of the life of this patient. The choice of bronchoscopy and transbronchial biopsy as an investigative tool rather than transthoracic biopsy is to limit anticipated complications that may arise from the transthoracic biopsy procedure.[6] The presence of dysphagia and odynophagia often indicates advanced disease with a poor prognosis.[9] Dysphagia was seen in the index case. However, we strongly believe that the patient presented early as adenocarcinomatous tumors of the trachea are highly infiltrative and often present with hemoptysis, obstructive features, dysphagia, and odynophagia as described in the literature.[10]

The best mode of treatment for tracheal tumors with a good hope of achieving a cure is surgical resection.[6,10] However, radiotherapy and chemotherapy can be used if the patient cannot tolerate surgery or as an adjunct to surgical resection.[2] Laser therapy can also be used but it is often associated with a small risk of perforation but this is not available for use in Nigeria. Surgical resection was used as the mode of treatment for the index patient with a good outcome. As was the case in our index case, it is very important to make a diagnosis early and obtain a definite histological subtype. The index patient could have benefited from additional chemotherapy as an adjunct to surgical resection which could have further improved survival. Clinical, radiographic, and bronchoscopic presentations were critical in tumor management. We

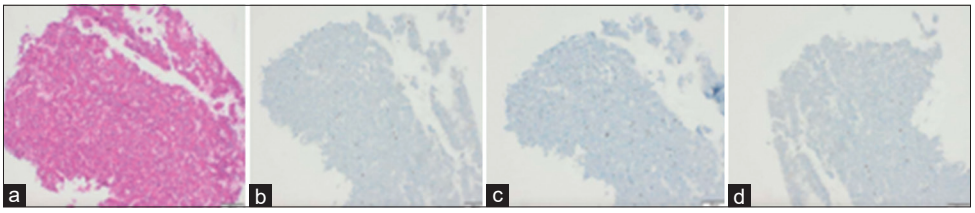

Figure 5: The histology slides of biopsy sample showing (a) clusters and singly occurring atypical cells with foamy to vacuolated cytoplasm and vesicular to hyperchromatic nuclei (hematoxylin and eosin stain), negative immunostaining to (b) novel aspartic proteinase of the pepsin family A (Napsin A), (c) pan-cytokeratin (pan-CK), and (d) thyroid transcription factor-1.

advise physicians to take seriously complaints of globus and investigate appropriately.

CONCLUSION

Trachea tumors are uncommon cancers affecting the windpipe (trachea). They do not produce symptoms on most times until they have grown enough to cause obstruction. Trachea tumors are treatable and curable when diagnosed early.

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Authors' contributions

OJO and MS were involved in the literature search, drafting, and critical review of the manuscript.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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