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CASE REPORT

Rare Benign Tracheal Lesions

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ABSTRACT

Breathing difficulty, change in voice and hemoptysis are symptoms that are seen with varying degrees of frequency in the routine ENT OPD. The focus of examination and investigation is usually the larynx in such cases. It is rare for the diagnosis to be a tracheal lesion.

We report three cases of uncommon benign tracheal lesions. Our aim is to highlight the presentation of such cases in a tertiary care center, and the relevant modalities of management.

Keywords: Trachea, Benign, Granular cell tumor, Rhinosporidiosis, Capillary hemangioma, Hemoptysis, CT scan.

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INTRODUCTION

Tracheal lesions are rarely seen in a routine ENT set-up. They account for 2% of all upper respiratory tract tumors.[1] Malignant neoplasms are rarer, being less than 1% of all malignancies.[2] One retrospective analysis of primary tracheal tumors, over a period of 25 years, has reported 14 cases, of which there was only one benign lesion.[3] They often present with symptoms mimicking upper airway infections. Any patient presenting to the OPD with foreign body sensation in throat, persistent cough, breathing difficulty, hemoptysis or stridor has to be investigated with tracheal lesions also as a possibility. Being a tertiary referral center, we had the opportunity to manage such cases over a period of time. We report three cases of rare benign tracheal lesions, which were managed by us, along with relevant discussion.

CASE REPORTS

Case 1

A 23-year-old female in the third trimester of pregnancy, with no known comorbidities, presented with progressive worsening of breathing difficulty for 1 month. This had worsened over the previous week preceding presentation. Clinical examination including flexible nasopharyngolaryngoscopy revealed no lesion in the nasopharynx, hypopharynx or larynx. Both vocal cords were mobile. Chest auscultation revealed expiratory wheeze.

X-ray film of the chest (posteroanterior view) and soft tissue neck (lateral view) were found to be normal. CT scan of the neck with contrast was done. This showed an intraluminal tracheal mass with the wall intact (Fig. 1). It was decided to surgically intervene. Tracheostomy was done for ventilation, followed by endoscopic excision of the mass. Histopathological examination of the specimen was reported as a granular cell tumor (Fig. 2). She then underwent cesarean section. She was asymptomatic at discharge. Check bronchoscopy was done 1 month postoperatively which did not show any evidence of residual lesion. She was then lost to follow-up. On enquiry after few months, it was reported that she was HIV positive and had died due to related complications. It is assumed that she may have been in the window period during her preoperative investigation for this surgery.

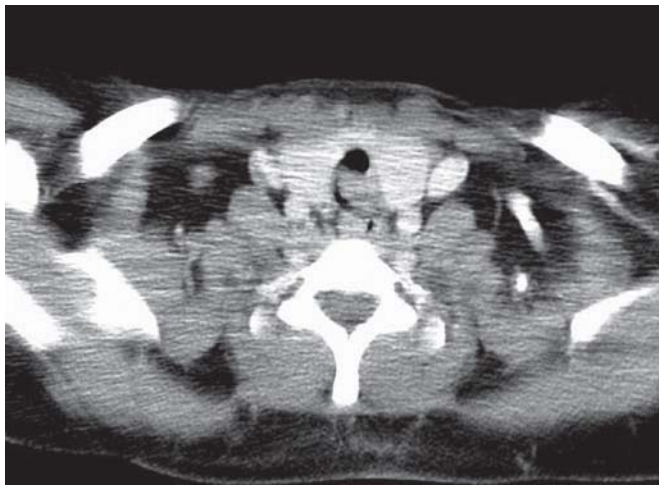

Fig. 1: Axial contrast CT image of the thorax showing a soft tissue mass in the distal trachea obliterating the lumen

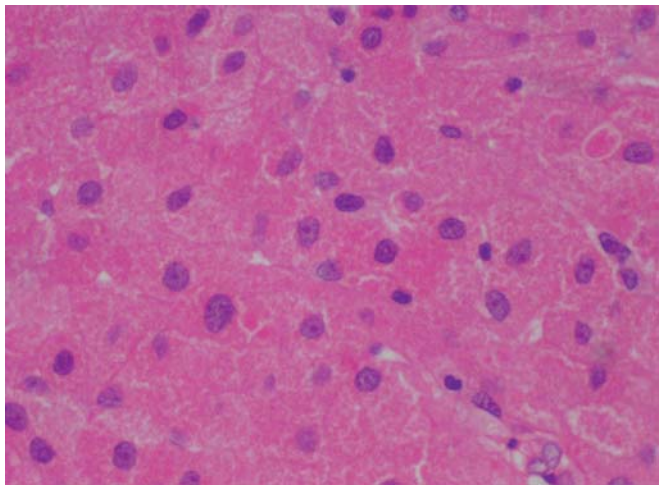

Fig. 2: Photomicrograph of the tumor showing round polygonal spindle cells with mild-to-moderate nuclear atypia and with eosinophilic cytoplasm and vesicular nuclei (H&E, × 400)

Case 2

A 41-year-old male presented with complaints of breathing difficulty of gradual onset and change in voice since 1 month. He had a past history of recurrent rhinosporidiosis with multiple surgeries elsewhere over the past 20 years. Details of surgeries were not available. The last excision was apparently about 3 years prior to this presentation. He did not have any nose-related complaint in the interim. Clinical examination of the nose and throat was normal. Indirect laryngoscopic examination was clear. Radiological evaluation (CT thorax) revealed a large mass in the middle third of trachea almost completely obstructing its lumen (Fig. 3). Bronchoscopy revealed a corresponding lesion, reddish irregular mass 2.5 cm proximal to the carina (Fig. 4).

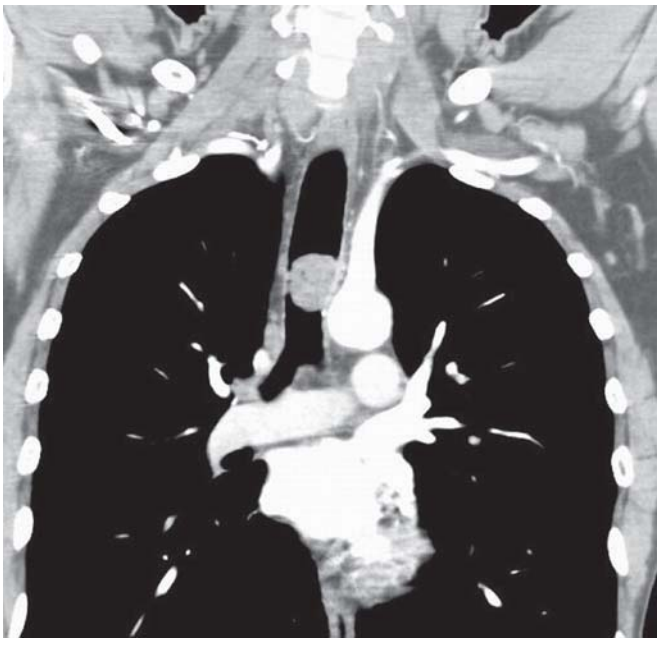

Fig. 3: Coronal CT image of the thorax showing a soft tissue mass in the distal third of tracheal lumen

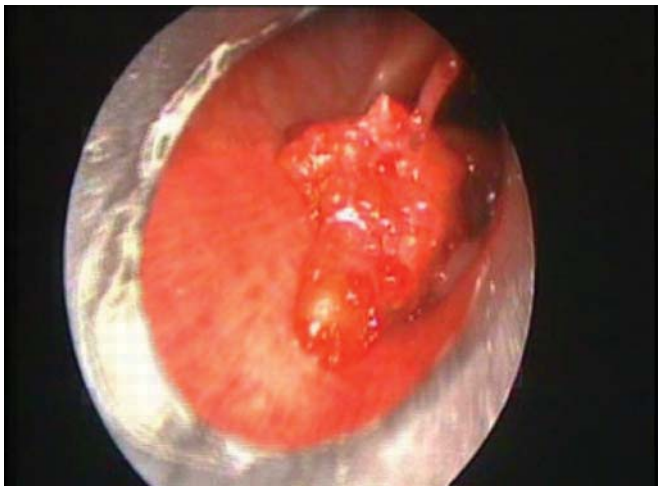

Fig. 4: Bronchoscopic view showing a reddish irregular mass 2.5 cm proximal to the carina

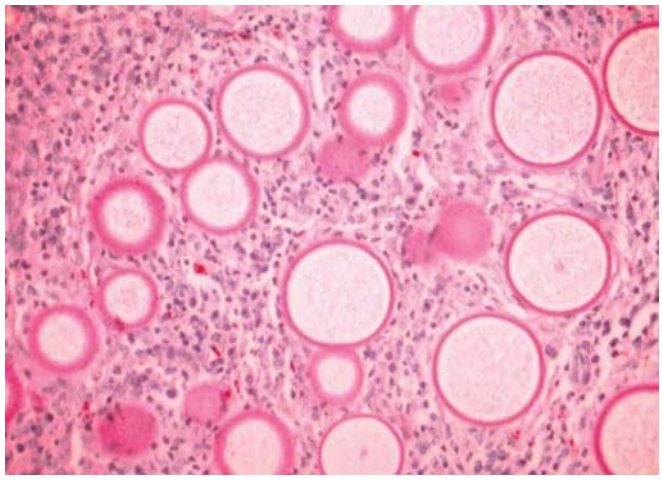

Fig. 5: Photomicrograph of biopsied polyp showing classical sporangia of rhinosporidium (PAS stain, 100×)

Laser excision was done. Histopathology was reported as rhinosporidiosis (Fig. 5). He is on dapsone treatment and follow-up since then, and has had no further recurrence till 2 years postoperative.

Case 3

A 45-year-old woman presented with multiple episodes of hemoptysis over the past 4 months. Clinical examination did not reveal any lesion in the nose, throat or larynx. Hemoptysis in an otherwise healthy young woman with no obvious pathology prompted a thorough imaging. Spiral CT with three-dimensional reconstruction of the neck and mediastinum was done which revealed a small intraluminal tracheal mass (Fig. 6). The patient underwent endoscopic excision of the mass under general anesthesia. A cherry-red tracheal mass, 10 × 5 mm, was seen arising from the right anterolateral wall of the trachea in the upper one-third. Excision biopsy of the mass was done. Histopathology was reported as polypoid lobular capillary hemangioma (Fig. 7). Patient was followed-up for 1 year without any recurrence, as evidenced by endoscopy.

DISCUSSION

Tracheal tumors, especially primary type, are uncommon. It is estimated that only 2.7 new cases occur per million per year.[4] Therefore, only a few centers have acquired expertise in their treatment.

Although primary tumors can be found anywhere in the trachea, the proximal and distal third are most frequently affected. In adults, most of these are likely to be malignant, whereas in children, the majority is benign. Benign lesions are usually well circumscribed, round, soft and smaller than 2 cm.[4] On chest CT scanning, they typically do not extend through the tracheal wall. The preferred treatment for primary tracheal tumors is surgical excision.

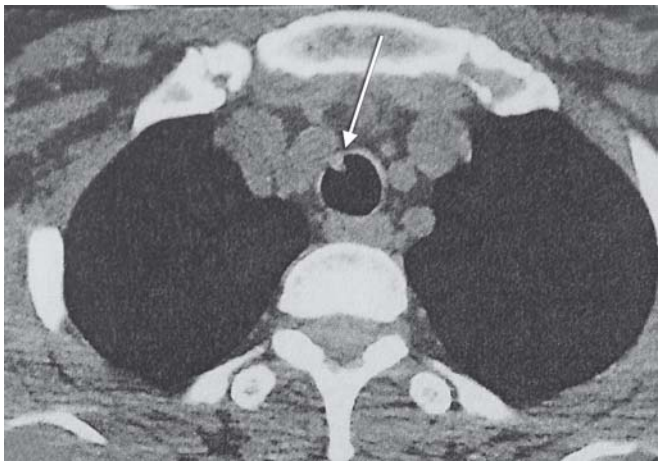

Fig. 6: Axial CT scan image showing a pedunculated, densely enhancing mass, 2 × 1 cm, arising from the right anterolateral wall of trachea (arrow)

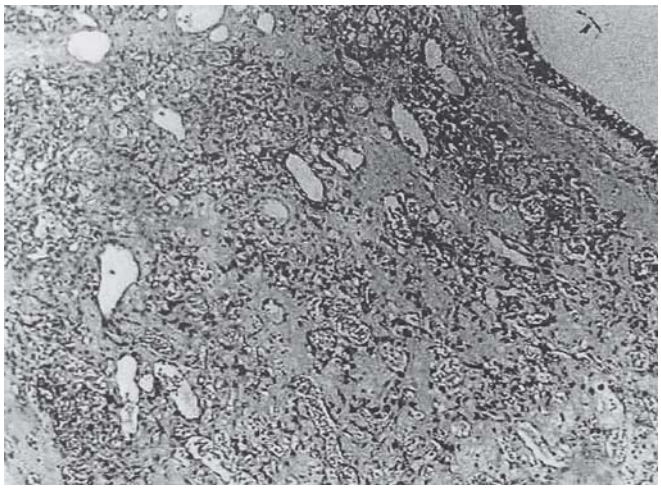

Fig. 7: Photomicrograph of the polypoidal tissue lined by pseudostratified ciliated columnar epithelium with vascular proliferation of capillary channels (H & E, 200×)

Granular cell tumor (GCT): This was first described in 1926 by Abrikosoff.[4] It was found to be neurogenic in origin, probably from the Schwann cell. These tumors can develop throughout the body. Approximately 50% occur in the head and neck region and 10% in the larynx. However, the trachea is rarely affected. In that way, our first case is quite rare. In case of tracheal involvement, the cervical portion is the most common location.[4] In the present case, the distal trachea was involved. Only sporadic cases of tracheal GCT and GC myoblastoma have been reported, with no reports from India.[5-7] An article published in 1999 cites 31 cases overall, in English language literature on the subject.[8] The management was by the well-established methods of CT scan diagnosis and endoscopic excision. Usually, these cases have a good prognosis. However, our patient had HIV-related mortality. We could conjecture on the association of the GCT with HIV status, although no evidence could be found. It has been stated that pulmonary GCTs occur in HIV cases; we could not find any specific case report.

Rhinosporidiosis: This is a chronic granulomatous disease caused by the fungus *Rhinosporidium seeberi* which predominantly affects the mucous membrane of the nose and nasopharynx. Other sites of involvement include skin, conjunctiva, oropharynx, lips, palate, uvula, trachea, bronchi, penis, vagina and vulva. The largest number of cases have been reported from India and Sri Lanka.

Our patient presented with an isolated tracheal rhinosporidial lesion. Local spread of rhinosporidiosis, due to direct spillage of spores from the nasopharynx into the larynx or trachea, is a known event, although rare.[9] However, the usual presentation tends to be recurrent nasal and/or nasopharyngeal masses with further lesions below. A medline search of English literature showed many reports of tracheal and bronchial rhinosporidiosis. However, only two similar previous reports of isolated tracheal rhinosporidiosis. One of these was very similar to our case.[10] The other was an isolated primary tracheal lesion.[11] The site of the lesion and the potential for bleeding and aspiration make the management a challenge. CT scan and the use of laser helped in our case.

Capillary hemangioma: Typically, this benign vascular lesion occurs in the subglottis of the child. Cavernous hemangioma has been reported in the adult larynx.[12] So, the third case in the present series is quite uncommon. In case of tracheal involvement, hemoptysis can be a serious presentation.[13] If detected early, these tumors can be excised endoscopically with minimal morbidity, as happened in our case. Very few reports could be found in literature search.[14,15] The present case has been published as a case report.[16] We have included it here as part of the series of rare tracheal lesions that presented to us. In addition, this case demonstrated the importance of having a high index of suspicion to diagnose it. It also highlighted the modality of spiral CT with 3D reconstruction.

CONCLUSION

This report is an attempt to bring to light the rare cases that can be seen in the setting of a tertiary referral center. The three tracheal lesions encountered were of disparate pathologies and were successfully managed.

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