Case Report



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TANAFFOS

# Tracheal Lobular Capillary Hemangioma: A Rare Localization

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**Background:** Lobular capillary hemangioma is a benign vascular tumor commonly found within the skin and upper respiratory mucosa and has rarely been reported within the trachea. The first case was reported by Irani et al. in 2003 and since then, less than 20 cases have been described. That's why the characteristics and treatments remain relatively unknown.

**Case Presentation:** A 53-year-old woman was symptomatic of recurrent episodes of hemoptysis associated with paroxysmal dyspnea. Physical examination, routine blood investigations, and chest x-ray were normal. The flexible bronchoscopy showed a polypoid bleeding lesion arising from the right lateral wall of the middle third of the trachea. Tumor biopsy was not performed considering an eventual bleeding risk. Computed tomography scanning showed a vascular, endotracheal budding tissue process without peritracheal or distant extension. A rigid bronchoscopy was performed for diagnostic and therapeutic purposes. A 10-millimeter bronchoscope was used. A rigid coring technique was performed to remove the tumor. A minimal bleeding was completely controlled after diode laser treatment. There were no complications during or after the procedure. Pathology revealed no malignancy and the diagnosis of lobular capillary hemangioma was confirmed. At a 6-month follow-up, the patient was asymptomatic and the endoscopic control did not show any tumor recurrence.

**Conclusion:** The lobular capillary hemangioma is a benign tumor rarely observed in the trachea. Clinical features are not specific and the short-term prognosis depends on tumor size. Considering its benign nature, tumor removal by interventional bronchoscopy should be proposed as the first-line treatment.

**Keywords:** Hemoptysis; Tracheal tumor; Dyspnea; Bronchoscopy

## INTRODUCTION

Lobular capillary hemangioma (LCH) previously known as pyogenic granuloma is a benign vascular tumor commonly found within the skin and upper respiratory mucosa (1). It occurs most frequently in children and young adults (2). It has rarely been reported within the trachea (3). The first case of tracheal LCH was reported by Irani et al in 2003. Since then, fewer than 20 cases have been described, which is why the characteristics and treatments remain relatively unknown (1).

## CASE SUMMARIES

A 53-year-old woman was referred to our institution with recurrent episodes of hemoptysis over the previous three months. She had chronic rhinitis, wheezing, and paroxysmal dyspnea for two years treated as asthma. She

has not reported any other symptoms. The patient was a nonsmoker with no history of airway instrumentation or foreign body aspiration. Physical examination, routine blood investigations, and chest x-ray were normal.

To explore hemoptysis, a flexible bronchoscopy was performed; it showed a polypoid bleeding lesion arising from the right lateral wall of the middle third of the trachea. Its diameter was about 5 millimeters. This tumor was causing a 30% obstruction of the tracheal lumen during inspiration and an 80% obstruction during expiration, with no other significant findings (Figure 1). Tumor biopsy was not performed considering an eventual bleeding risk.

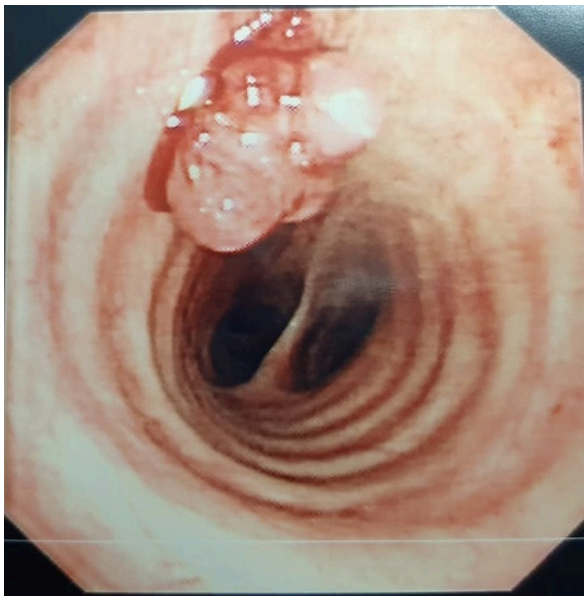

**Figure 1.** Endoscopic view of the tumor

Computed tomography (CT) scanning demonstrated a vascular endotracheal budding tissue process without peritracheal or distant extension. It measures 6 x 4 millimetres in the axial plane and extends over 6 millimetres in height (Figure 2).

A rigid bronchoscopy was performed for diagnostic and therapeutic purposes. A 10-millimeter bronchoscope was used. A rigid coring technique was performed to remove the tumor. A minimal bleeding was completely controlled after diode laser treatment. There were no complications during or after the procedure. The microscopic examination revealed tracheal mucosa with a respiratory underlying. It contained a benign vascular proliferation made of small capillaries arranged into lobules within the chorion (Figure 3). These features were characteristic of LCH.

At 6-month follow-up, our patient was asymptomatic and the endoscopic control did not show any tumor recurrence (Figure 4).

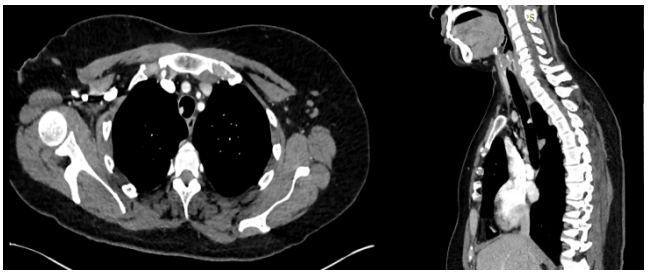

**Figure 2.** CT Scan view of the tumor

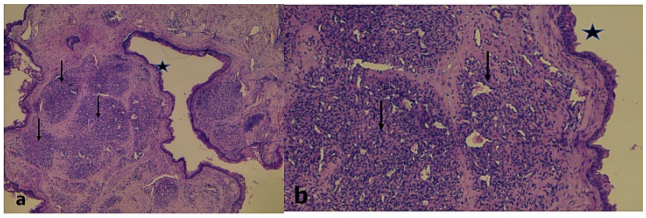


**Figure 3.** a/ Microscopic view of a benign vascular proliferation (arrow) arranged into lobules, within a tracheal wall lined by a respiratory epithelium (star) (HEx250), b/ Higher magnification showing small capillaries (HEx400)

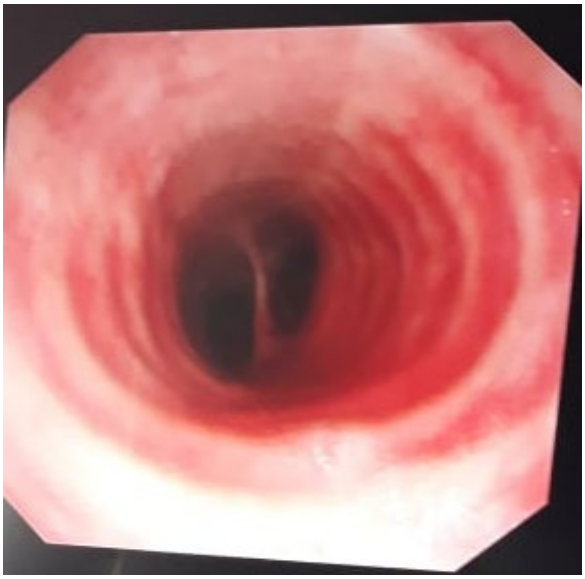

**Figure 4.** Endoscopic view of the trachea after tumor removal

## DISCUSSION

LCH is a benign lesion that occurs on the skin and mucosal surfaces. It is rarely found within the trachea. Mills et al. reported 639 cases of vascular lesions of the upper airway; LCH was found in 273 cases without any tracheal localization (4). The first case of histologically proven LCH of the trachea was reported by Irani et al. in 2003 (1). According to many studies, LCH is more common below 18 years in males, whereas in females it is more common during reproductive age, similar to our patient (4,5). Several factors have been implicated such as previous trauma, hormonal disruption, Bartonella infection, viral oncogenes, production of angiogenic factors, and cytogenetic abnormalities (6). Certain drugs can also be involved. Putora et al. reported the case of a 64-year-old patient with squamous cell lung cancer treated by erlotinib chemotherapy (7). A tracheal LCH was diagnosed during treatment. A complete resolution of the lesion was noted after stopping erlotinib and no invasive intervention was necessary (7). Most theories on pathogenesis revolve around LCH as a hyperplastic, neovascular response to an angiogenic stimulus with the imbalance of promoters and inhibitors (8).

Among clinical features described in the literature, hemoptysis was the most frequent symptom: generally, of low abundance like the case of our patient, but one case of massive hemoptysis in a 66-year-old female with thrombocytopenia was reported requiring arterial embolization (9). Other symptoms were reported such as cough, dyspnea, and foreign body sensation (5). Chest pain is an uncommon symptom (5).

Para clinical investigations such as chest x-ray and computed tomography findings are not specific and may be inconclusive and thus diagnosis of LCH is confirmed by the bronchoscopy with biopsy (3). Considering the potential risk of bleeding, as it is with vascular tumors, it is highly suggested to have interventional bronchoscopy available during the removal.

Many effective treatment modalities have been reported such as snare cautery, excision biopsy, and plaque radiation (1,10). However, the preferred treatment has yet to be established because of the limited number of cases. In the literature, among 15 patients treated for tracheal LCH, a treatment based on interventional bronchoscopy was performed in 10 patients (3). In fact, the extent and the size of the lesion, as well as the patient's age and comorbidities must have been considered (3). In our case, LCH was successfully treated with rigid bronchoscopy and laser therapy.

The long-term prognosis of the LCH depends essentially on the risk of local recurrence. Malignant transformation has not been reported but the localized recurrence after excision and satellite spread of the lesions are known phenomena (1). Therefore, bronchoscopic control is recommended.

## CONCLUSION

The LCH is a benign tumor rarely observed in the trachea. Clinical features are not specific and the short-term prognosis depends on tumor size. Considering its benign nature, tumor removal by interventional bronchoscopy should be proposed as a first-line treatment.

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