

## Case Report

## Lobular Capillary Hemangioma of the Trachea

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As a polypoid form of capillary hemangioma, lobular capillary hemangioma (LCH) most commonly occurs on the cutaneous and mucosal surfaces and is frequently benign in children. Tracheal LCH is a rare benign tumor in adults, with hemoptysis being one of the most serious forms of presentation. A definite diagnosis of LCH depends on its histopathology. In clinical practice, however, radiological characteristics are an important component for making a suggestive diagnosis. We present a case of tracheal LCH and describe its features on computed tomography, pathology, and differential diagnosis. A review of the relevant literature is also provided.

**Keywords:** Hemoptysis, lobular capillary hemangioma, trachea

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**Introduction**

Lobular capillary hemangiomas (LCHs) are benign, typically painless tumors that occur on the skin and mucosal surfaces. In these lesions, the capillaries display a distinctive lobular arrangement in an edematous, fibroblastic stroma. Histopathologically, LCH was previously termed “pyogenic granuloma”, although it is neither induced by bacterial infection nor a true granuloma.<sup>1</sup> The term “lobular capillary hemangioma” was introduced to describe these lesions more accurately.<sup>2</sup>

In about 25% of patients with tracheal neoplasms, especially malignant tumors, hemoptysis will be present.<sup>3</sup> Almost all tracheal tumors can be diagnosed by radiologic examination and endoscopy. In this article, we describe a case of LCH of the tracheal mucosa, its differential diagnosis, and its findings on computed tomography (CT). We also review several relevant studies from the literature.

**Case report**

A 64-year-old man, in good health until experiencing an episode of cough with white sputum of 3 days duration (about 10 mL total) was admitted to our hospital with bloody sputum and recurrent hemoptysis that lasted for a few days (about 20 mL total). He responded poorly to medical treatment with antimicrobials. He had no prior history of foreign body aspiration, dyspnea, dysphagia, hoarseness, trauma, intubation, or airway endoscopy.

On physical examination, the patient’s lungs and heart showed no abnormalities. Routine examinations of the ear, nose and throat were unremarkable. Detailed investigations for tuberculosis yielded negative results. Hematologic and clinical laboratory test results were within the normal ranges, and there were no abnormal findings on chest X-ray.

Axial CT images of the chest in the lung window showed a

polypoid hyperdense tumor (Figure 1A). Spiral CT with three-dimensional reconstruction revealed a polypoid tracheal tumor in the left anterolateral wall of the main trachea. No calcification or fat density shadow in the tumor was detected on CT. The maximum diameter of the mass was about 4 mm. The density of the mass was uncertain because of the small tumor size on plain CT. Nevertheless, a homogeneously marked enhancement was observed after contrast injection, with an average density of 161 HU (Figure 1B).

Bronchoscopy revealed a polypoid tracheal tumor, 0.3 to 0.4 cm in size, with a hyperemic overlying mucosa. Histologic examination revealed numerous capillaries arranged in a lobular pattern, separated by an edematous fibrous stroma accompanied by mild inflammatory changes (Figure 1C). A diagnosis of polypoid LCH was made. While under general anesthesia, the patient underwent endoscopic excision of the mass with flexible biopsy forceps. There was no recurrence after a follow-up of about 8 months, as evidenced by CT and endoscopy.

**Discussion**

LCH is a common polypoid form of capillary hemangioma that is often found on the skin and oral mucosa.<sup>4</sup> There are multiple reports of LCH in the nasal cavity, tongue, conjunctiva, penis, duodenum, and colon.<sup>1,2,4-9</sup> LCH is more common in children, but relatively rare in adults.<sup>10</sup> The pathogenesis of LCH is not well-defined, but postulated etiologies include previous trauma, hormonal shifts, viral oncogenes, and infection, among others.<sup>11</sup>

Hemoptysis and airway obstruction are the most common symptoms of patients with tracheal LCH. An accurate diagnosis of this condition requires CT and bronchoscopy. The CT features of tracheal LCH are not typical, but LCH lesions usually have a homogeneously marked enhancement after administration of intravenous contrast agent. Pathological diagnosis of this disease can be made from the appearance of numerous capillaries arranged in a lobular pattern, separated by fibrous stroma and accompanied by mild inflammatory changes. In the present case study, the spiral CT finding raised suspicion for LCH and permitted the early diagnosis of the tumor.

Only 7 cases of LCH of the tracheal mucosa have been previously reported in the literature. The average age for patients with

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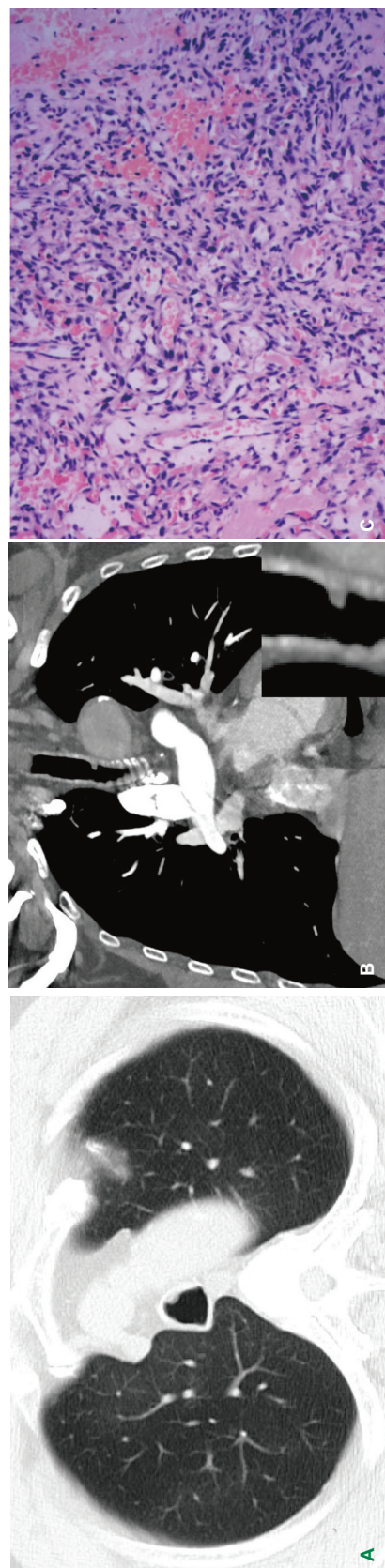
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**Table 1.** Summary of case reports regarding the clinicopathological features of tracheal LCH.

Author	Age (y), M/F	Symptoms	Location	No.	Tumor size (cm)	Treatment	Prognosis
Irani, et al. <sup>16</sup>	72 F	Cough, hemoptysis	3 cm below the vocal cords	1	0.3–0.2	Endoscopic excision	Good (1 y)
Madhumita, et al. <sup>3</sup>	40 F	Foreign body sensation, hemoptysis	Right anterolateral wall of the upper third of the trachea	1	1 × 0.5	Endoscopic excision	Good (1 y)
Porfyridis, et al. <sup>17</sup>	17 M	Hemoptysis	Left anterolateral wall of the upper third of the trachea	>1	0.4	Endoscopic excision	Good (1 y)
Chawla, et al. <sup>18</sup>	62 M	Hemoptysis	Right wall of the distal trachea	1	ND	Endoscopic excision and laser therapy	ND
Udoji, et al. <sup>19</sup>	55 M	Cough, hemoptysis	Left lateral wall of the distal trachea	1	0.4 × 0.5	Cryoprobe	Good (3 mo)
Amy, et al. <sup>11</sup>	22 M	Cough, hemoptysis	Left posterior wall, 3 cm from the carina	1	1.5–1	Electrocautery	Good (ND)
Shen, et al. <sup>15</sup>	35 M	Cough, bloody sputum	Left lateral wall of the proximal trachea	1	1.5 × 2.0	Brachytherapy	Good (2 y)
Present case	64 M	Cough, hemoptysis	Left anterolateral wall of the trachea	1	0.4–0.3	Endoscopic excision	Good (8 mo)

ND = not determined.

**Figure 1.** (A-B) Chest CT (lung window). (A) CT scan showed a polypoid tumor (white arrow) in the trachea. (B) CT scan with three-dimensional reconstruction revealed a small tracheal tumor in the left anterolateral wall of the trachea (white arrow). (C) Histological examination revealed numerous capillaries arranged in a lobular pattern. (H&E, 10×).

this tumor, according to the published literature and the present case study, is 46 years. Six of the previously reported cases were solitary lesions, and one was multifocal. LCHs of the tracheal mucosa are typically small lesions, with a maximum size <20 mm on average, and a tumor diameter ranging from 0.2 to 2.0 cm. Patients are more commonly male,<sup>12</sup> and they usually present with hemoptysis and cough. The CT features are characteristic. A homogeneously marked enhancement is observed after contrast injection, with an average density >100 HU. Attenuation is usually homogeneous.

Many effective treatment modalities have been reported for LCH of the tracheal mucosa, including snare cautery, excision biopsy, plaque radiation, and laser surgery.<sup>13–15</sup> Recurrence of skin and mucosal LCHs after local therapy is well-known; however, neither recurrence nor malignant degeneration has been reported with LCH of the tracheal mucosa. In our patient, the tracheal LCH was removed with biopsy forceps. Subsequently, the patient was asymptomatic. Table 1 summarizes the clinicopathological features of tracheal LCHs in all reported cases, including ours.

In adults, the most frequent causes of hemoptysis are tuberculosis, infectious diseases, malignant tumors, cardiovascular disorders, and other inflammatory diseases. Although rare, LCH should be considered as a possible benign cause of hemoptysis and cough. If the soft-tissue shadow disappears or deforms after episodes of coughing in a chest CT scan, then the possibility of a mucus plug pseudotumor should be considered. However, it is important to differentiate mucus plug pseudotumors from tracheal tumors (e.g., adenoma, hamartoma, lipoma, pulmonary carcinoma, etc.).

Tracheal adenomas are relatively common benign tracheal tumors that may show significant, but slower, enhancement compared to LCHs after the administration of intravenous contrast. Hamartomas are rarely found in the trachea; only 10 cases of tracheal hamartoma have been reported in the literature.<sup>20</sup> Cytologically, they are characterized by fibromyxoid stroma, cartilage, bronchial cells, adipose tissue, and bone; however, tracheal hamartomas consist mostly of fat tissues. They can be differentiated by the CT value of adipose tissue and bone in the lesion.

Primary tracheal lipomas are remarkably rare, as they most often involve the main bronchial stems.<sup>21</sup> Pathologically, the primary tracheal lipoma grossly presents as a well-circumscribed, thinly encapsulated, rounded, pale yellow mass composed of mature adipose tissue by microscopic observation. A homogeneous and well-circumscribed lesion of lipid density can be revealed on CT imaging. It is not enhanced when contrast agent is used, owing to a lack of soft tissues.

Finally, pulmonary carcinoma is often accompanied by irritating dry cough, chest pain, emaciation, and other symptoms. Pulmonary carcinomas generally have a significant infiltration of the trachea wall. They can also show local extension into the surrounding tissues and between the cartilaginous rings of the trachea.

In general, a definite diagnosis of tracheal LCHs depends on its histopathology. However, our case with contrast enhancement >100 HU gave a suggestive diagnosis of tracheal LCH. Our case study may provide a reference for clinicians.

### Conflict of Interest

The authors declare that they have no competing interests.

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