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Case Report

An uncommon diagnosis behind a common respiratory symptom: Lobular capillary hemangioma of the trachea in a pediatric patient ☆

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ABSTRACT

Lobular capillary hemangioma (LCH) is a benign vascular tumor that is usually found in the skin and mucous membranes. However, tracheal LCH is rare and presents with non-specific respiratory symptoms such as chronic cough, hemoptysis or airway obstruction, which often lead to misdiagnosis. This case report describes a six-year-old girl who was initially misdiagnosed with hyperreactive airway disorder due to persistent coughing and respiratory distress. Further evaluation via CT scan and bronchoscopy revealed a tracheal mass, which was confirmed as LCH following histopathological analysis. Successful treatment involved an excisional biopsy, which resolved the symptoms. This case highlights the importance of considering tracheal masses such as LCH in pediatric patients presenting with refractory respiratory symptoms and emphasizes the need for advanced imaging and endoscopy techniques to ensure accurate diagnosis and effective management.

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Introduction

Lobular capillary hemangioma (LCH) is a benign vascular tumor that was previously incorrectly named 'pyogenic granuloma'. This is an inappropriate term as the tumor is neither infectious in origin nor a true granuloma [1]. It is most found in the skin and upper respiratory mucosa, but its occurrence in the trachea, particularly in children, is rare [1,2]. Due to non-specific symptoms such as chronic cough, dyspnea and hemoptysis, and the rarity of tracheal masses, diagnosis is often delayed and confused with more common respiratory conditions such as asthma or bronchitis. Herein, we present a case of tracheal LCH in a 6-year-old girl that was initially overlooked. This case emphasizes the critical importance of including tracheal mass such as LCH in the differential diagnosis of pediatric patients who exhibit recurrent respiratory symptoms particularly when conventional therapies fail to alleviate patient's symptoms

Case presentation

A previously healthy six-year-old girl was admitted to the emergency department of a tertiary children's hospital with a two-month history of chronic cough and dyspnea, which had progressively worsened over the preceding two weeks. She had no significant medical history, including no prior episodes of foreign body aspiration, hoarseness, trauma, hospitalization, intubation, or medication use prior to the onset of symptoms.

On initial evaluation, her physical examination revealed no abnormalities, with normal lung auscultation and stable vital signs (including oxygen saturation). Routine hematological and biochemical investigations were within normal limits. Imaging studies, including Waters' view and frontal chest X-rays, showed no abnormalities. The patient was empirically managed with salbutamol (2 puffs every 6 hours) and fluticasone (1 puff every 12 hours) inhalers for suspected hyperreactive airway disease, which resulted in partial symptomatic improvement. She was discharged on maintenance inhaler therapy.

However, seven days later, she presented with a worsening cough, respiratory distress and inspiratory stridor upon examination.

Given the progression of respiratory symptoms and the onset of inspiratory stridor despite normal chest X-ray findings, contrast-enhanced CT scans of the neck and chest were performed. The chest CT scan was unremarkable, but the contrast-enhanced neck CT scan revealed an 8.5 × 6.5 × 6.5 mm well-circumscribed, homogeneously mildly enhancing soft tissue mass arising from the left lateral wall of the upper trachea, approximately 2 cm below the vocal cords. The lesion had smooth margins and showed no evidence of local invasion or calcifications, causing luminal narrowing estimated at 70% (Fig. 1).

Subsequent flexible bronchoscopy revealed a well-defined, round, pedunculated mass with erosion of overlying mucosa in the proximal trachea, which was causing significant luminal narrowing.

Taking into account the risk of bleeding from the mass and the limitations of rigid bronchoscopy for endoscopic removal, as well as the uncertainty surrounding the exact nature of the lesion and the potential for recurrence, the multidisciplinary team decided to proceed with surgical excision of the mass. This approach was chosen to establish a definitive histopathological diagnosis and to achieve complete removal of the mass, given its size and location, which carried risks of future airway compromise. Histopathological examination of surgical specimen revealed capillary sized vessels in lobular patterns covered by ulcerative mucosa surrounded by scattered inflammatory cells (Fig. 2)

The patient was discharged one week after surgery in good clinical condition. During the subsequent 10-month clinical and endoscopic follow-up period, there was no evidence of lesion recurrence.

Discussion

LCH of the trachea is a rare benign vascular tumor that usually affects the skin and mucosal surfaces of the upper respiratory tract. Tracheal involvement is exceptionally uncommon. The first histopathologically confirmed case of tracheal LCH

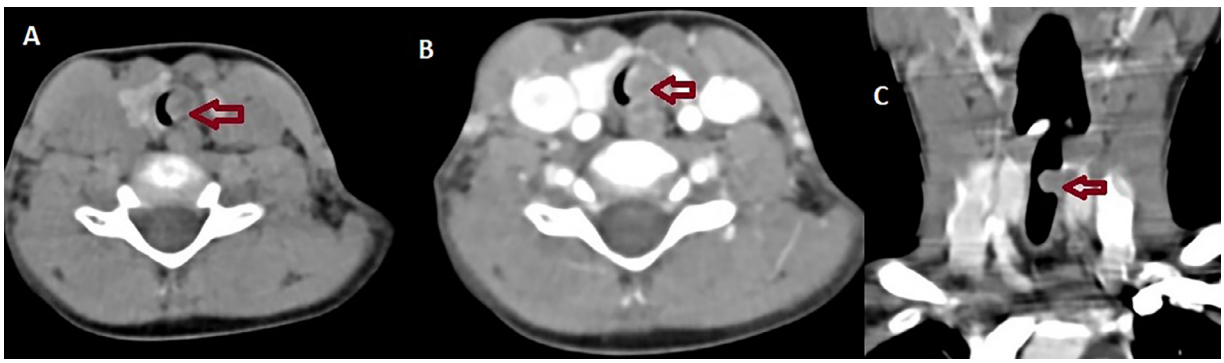

Fig. 1 – Axial non-enhanced neck Ct scan (A) and contrast enhanced neck Ct scan in axial (B) and coronal (C) views show a well-defined homogeneous enhancing polypoid intraluminal mass arising from the left lateral wall of upper part of trachea (arrow) that causes obvious luminal narrowing.

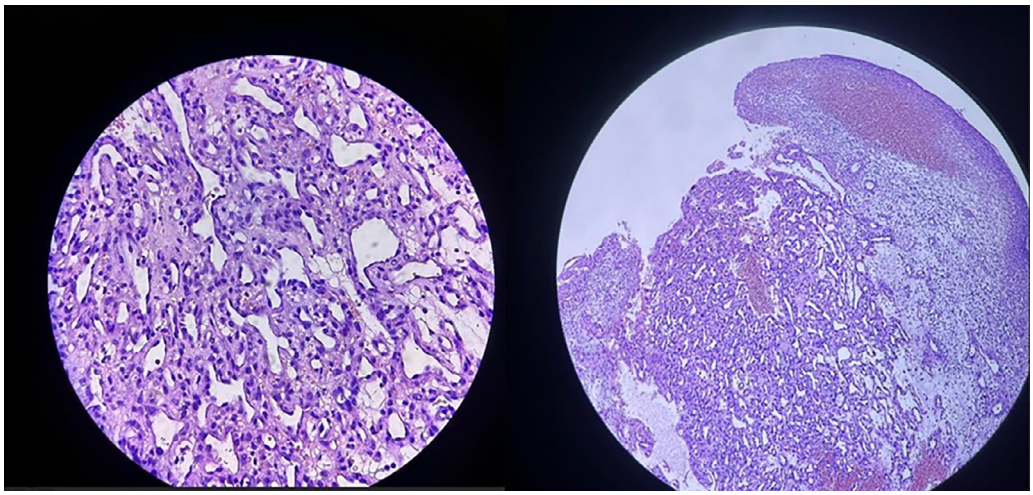

Fig. 2 – Histopathological examination demonstrated capillaries proliferation arranged in lobular patterns covered by ulcerated mucosa and accompanied by scattered inflammatory cells .(hematoxylin and eosin . A: magnification *40, B: *10).f

Table 1 – The characteristics of previously reported tracheal lobular capillary hemangioma in children.

Author	Age/sex	Presenting symptom	Tumor Size and location	Treatment	Follow up
Porfyridis et. al	17 y/male	Cough+ hemoptysis	Two 4mm nodules arising left lateral of trachea	Electrocauterization	12months,nl bronchoscopy
Chen et.al	14y/female	Cough+ hemoptysis	15*20 mm pedunculated mass attached anterior wall of trachea	cryoablation	3months,nl bronchoscopy
Wang et.al	10y/female	Cough + vomiting	4*10 mm sessile mass of posterior tracheal wall	KTP laser ablation	4months,nl bronchoscopy
Luo et.al	8y/female	Cough+ hemoptysis	7mm pedunculated mass adherent to left wall of trachea	Surgical resection	No information is available
Tao et.al	11y/male	hemoptysis	4/9 *4/6*4/6 mm pedunculated mass of rt wall of trachea	Bronchial artery embolization and bronchoscopic removal	No information is available
Our case	6y/female	Cough and stridor	8.5 × 6.5 × 6.5mm pedunculated mass arising of left lateral wall of trachea	Surgical resection	11months,nl bronchoscopy

was reported in a 72-year-old woman presenting with recurrent hemoptysis, for which a 2 × 3 mm proximal tracheal mass was removed by bronchoscopy [1]. The pathogenesis of LCH remains poorly understood, with several theories having been proposed, including local trauma, angiogenic imbalance, infections, hormonal influences and genetic factors affecting vascular regulation [2,3]. To date, only five pediatric cases of tracheal LCH have been documented in the literature. The characteristics of these patients, who ranged in age from 8 to 17 years, are summarized in Table 1 [2,4–7].

When evaluating tracheal masses, clinicians must consider a broad differential diagnosis that includes congenital lesions such as infantile hemangiomas, which demonstrate a unique natural biological course and characteristic imaging features during their proliferative phase, as well as acquired lesions such as post-intubation granulomas or cysts. Other rare neoplasms must also be considered, ranging from benign tumors such as squamous papilloma and inflammatory myofibroblastic tumors to malignant entities such as carcinoid tumors, mucoepidermoid carcinomas and metastases. As imaging findings and clinical symptoms and signs of tracheal masses are often non-specific, histopathological examination is essential for a definitive diagnosis. [8–10].

The management of tracheal LCH presents unique challenges. Although complete surgical resection is considered the gold standard in most cases, endoscopic approaches have been attempted, despite the significant risks involved, including hemorrhage (particularly with lesions measuring over 5 mm), transmural injury and high recurrence rates [5–7,10,11]. Limitations of endoscopic management include the requirement for repeated anesthesia, contraindications in

cases involving extraluminal extension or severe airway compromise, and potential complications such as post-procedural atelectasis. In selected cases, alternative approaches such as trans arterial embolization may be considered [2,10].

In our case, the decision was made to surgically remove the mass due to its size, which led to significant airway obstruction, and the uncertainty surrounding its nature, as well as the lack of access to rigid bronchoscopy. This was a successful outcome. The patient experienced no postoperative complications, and bronchoscopy at the 10-month follow-up revealed no signs of recurrence.

Conclusion

This presentation emphasizes the critical role of advanced imaging and endoscopic techniques in diagnosing and managing rare tracheal lesions, such as LCH.

Early diagnosis and treatment are essential to prevent complications and ensure the best possible outcomes for children presenting with atypical respiratory symptoms.

Patient consent

An informed consent was taken for the patient's parent for the publication of this case report.

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