Case Report

Tracheal Lobular Capillary Haemangioma: A Rare Benign Cause of Recurrent Haemoptysis

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1. Introduction

Tracheal tumours represent only 2% of all upper respiratory tract tumours [1]. Those in adults are usually malignant; benign tumours, including chondroma, papilloma, and fibroma most often occur in the proximal third of the trachea in adults and its distal third in children [2]. Lobular capillary haemangioma (LCH), previously called pyogenic granuloma, commonly presents on the lip, nose, oral cavity, or tongue [3, 4] but has rarely been reported within the trachea. We here report the case of a 56-year-old woman with a tracheal LCH managed by bronchoscopic resection using biopsy forceps.

2. Case Presentation

A 56-year-old Caucasian woman was referred to our institution with several episodes of mild haemoptysis over the preceding three months. She denied associated chest pain, exertional dyspnoea, dysphagia, syncope, weight loss, anorexia, fever, and rigors. There was no history of airway instrumentation or foreign body aspiration. Her medical background included laparoscopic sterilisation, oesophagitis, meningitis, and hypertension. She was a nonsmoker and consumed alcohol within normal limits.

Physical examination was unremarkable and routine blood investigations and chest radiograph were normal. Pulmonary function was satisfactory with FEV₁ 3.50 L and FVC 3.85 L. Computed tomography (CT) scanning (Figure 1) demonstrated a pedunculated lesion arising from the right tracheal wall and projecting into its lumen. Following case discussion at a multidisciplinary team meeting, patient consent was obtained for rigid bronchoscopy under general anaesthetic. A 7 mm polypoid mass with a small pedicle was identified 2 cm inferior to the vocal cords on the right tracheal wall and resected in entirety utilising biopsy forceps. Haemostasis was achieved following local application of electrocautery and adrenaline-soaked swabs, and the patient was discharged home the next day. The patient was asymptomatic at six-month and one-year follow-up with no radiological or bronchoscopic evidence of disease recurrence.

Histological examination (Figure 2) of the surgical specimen demonstrated nodular proliferation of endothelial cells and capillary-type lumina separated by mildly oedematous and inflamed stroma, suggestive of a capillary haemangioma. There was no pathological evidence of dysplasia or malignancy.

3. Discussion

Tracheal tumours account for less than 2% of all upper respiratory tract tumours [1] and are usually malignant in
The extent and size of the lesion, as well as patient age and comorbidities, require consideration prior to any therapeutic intervention for LCH. Despite their benign nature, local recurrence is common and thus surgical excision remains the treatment of choice. Nevertheless, mucocutaneous LCH is also amenable to various nonsurgical techniques including snare electrosurgery, cryotherapy, YAG laser therapy, and plaque radiation. In the present case, we were able to safely remove the culprit lesion with biopsy forceps alone and with minimal bleeding owing to its small vascular pedicle.

To our knowledge, there are only 14 reports of tracheal LCH in English medical literature to date (Table 1). Irani et al. successfully extracted a 2-3 mm LCH occurring 3 cm below the vocal cords in a 72-year-old female with flexible biopsy forceps [3]. Endoscopic techniques were employed by Xu et al. [11] and Madhumita et al. [2], who resected a 4 mm tracheal LCH in a 64-year-old male and a 1 cm tracheal LCH in a 40-year-old female, respectively. Chawla et al. combined endoscopic excision and laser therapy for distal tracheal LCH in a 62-year-old male [12]. Chen et al. utilised cryotherapy to remove a 2 cm tracheal LCH which occluded the majority of the tracheal lumen in a 14-year-old girl [13]. Cryotherapy was similarly applied for a 1.5 cm tracheal LCH near the carina in the report by Udoji and Bechara [14]. Two 4 mm lesions were excised with biopsy forceps and electrocautery in Porfyridis’s report of a 17-year-old boy with recurrent haemoptysis [6]. Electrocautery loop snaring was also used in a 22-year-old male with a 1.5 cm tracheal LCH near the carina in the report by Amy and Enrique [4], as well as in a 57-year-old male by Kalanjeri et al. [15]. Following multiple poor responses to electrocautery and argon plasma coagulation, Shen et al. [16] utilised brachytherapy successfully to control a 2 cm tracheal LCH in a 35-year-old male. Dabó et al. required photocoagulation to achieve haemostasis following significant bleeding on removal of a tracheal LCH using rigid biopsy forceps in a 51-year-old female [17]. Zambudio et al. performed arterial embolisation to control massive haemoptysis from a bleeding tracheal capillary haemangioma in a 66-year-old female with thrombocytopenia [9]. Circulatory assistance with extracorporeal membrane oxygenation was employed as a precautionary measure when debulking a large LCH in a 23-year-old pregnant female in the report by Prakash et al. [10]. Putora et al. propose this lesion occurring as a consequence of erlotinib chemotherapy in a 64-year-old patient with squamous cell lung cancer; interestingly, complete resolution of the LCH was noted on discontinuation of erlotinib and no invasive intervention was necessary [18].
Table 1: Summary of previously reported cases of tracheal lobular capillary haemangioma.

<table>
<thead>
<tr>
<th>Author</th>
<th>Age (years), M/F</th>
<th>Tumour size</th>
<th>Tumour location</th>
<th>Treatment</th>
<th>Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>Madhumita et al. [2]</td>
<td>40, F</td>
<td>10 × 5 mm</td>
<td>Upper third of right anterolateral tracheal wall</td>
<td>Endoscopic resection</td>
<td>Good at 1 year</td>
</tr>
<tr>
<td>Imani et al. [3]</td>
<td>72, F</td>
<td>2-3 mm</td>
<td>3 cm above vocal cords</td>
<td>Endoscopic resection</td>
<td>Good at 1 year</td>
</tr>
<tr>
<td>Amy and Enrique [4]</td>
<td>22, M</td>
<td>10–15 mm</td>
<td>3 cm above carina on left posterior tracheal wall</td>
<td>Electrocautery</td>
<td>Good</td>
</tr>
<tr>
<td>Porfyridis et al. [6]</td>
<td>17, M</td>
<td>4 mm</td>
<td>Upper third of left anterolateral tracheal wall</td>
<td>Endoscopic resection</td>
<td>Good at 1 year</td>
</tr>
<tr>
<td>Zambudio et al. [9]</td>
<td>66, F</td>
<td>Occluding 30–40% of airway</td>
<td>Between first and third tracheal rings</td>
<td>Embolisation</td>
<td>Good at 1 year</td>
</tr>
<tr>
<td>Prakash et al. [10]</td>
<td>23, F</td>
<td>20 × 40 mm</td>
<td>Posterior tracheal wall</td>
<td>Endoscopic resection with extracorporeal membrane oxygenation</td>
<td>Good</td>
</tr>
<tr>
<td>Xu et al. [11]</td>
<td>64, M</td>
<td>3-4 mm</td>
<td>Left anterolateral tracheal wall</td>
<td>Endoscopic resection</td>
<td>Good at 8 months</td>
</tr>
<tr>
<td>Chawla et al. [12]</td>
<td>62, M</td>
<td>Unknown</td>
<td>Distal right tracheal wall</td>
<td>Endoscopic resection and laser therapy</td>
<td>Unknown</td>
</tr>
<tr>
<td>Chen et al. [13]</td>
<td>14, F</td>
<td>15–20 mm</td>
<td>Lower third of anterior tracheal wall</td>
<td>Cryotherapy and argon plasma coagulation</td>
<td>Good at 3 months</td>
</tr>
<tr>
<td>Udoji and Bechara [14]</td>
<td>55, M</td>
<td>4 × 5 mm</td>
<td>Distal left lateral tracheal wall</td>
<td>Cryotherapy</td>
<td>Good at 3 months</td>
</tr>
<tr>
<td>Kalanjeri et al. [15]</td>
<td>57, M</td>
<td>Occluding 70% of airway</td>
<td>Posterior middle tracheal wall</td>
<td>Electrocautery</td>
<td>Unknown</td>
</tr>
<tr>
<td>Shen et al. [16]</td>
<td>35, M</td>
<td>15–20 mm</td>
<td>Lateral wall of proximal left main bronchus</td>
<td>Brachytherapy</td>
<td>Good at 2 years</td>
</tr>
<tr>
<td>Dabó et al. [17]</td>
<td>51, F</td>
<td>Unknown</td>
<td>Lower third of left lateral tracheal wall</td>
<td>Endoscopic resection and laser photocoagulation</td>
<td>Good at 27 months</td>
</tr>
<tr>
<td>Putora et al. [18]</td>
<td>64, M</td>
<td>Unknown</td>
<td>Distal tracheal wall</td>
<td>Spontaneous remission on cessation of erlotinib for lung cancer</td>
<td>Good</td>
</tr>
<tr>
<td>Present case</td>
<td>56, F</td>
<td>7 mm</td>
<td>2 cm below vocal cords on right tracheal wall</td>
<td>Endoscopic resection and electrocautery</td>
<td>Good at 1 year</td>
</tr>
</tbody>
</table>

In conclusion, LCH is a benign lesion rarely found within the trachea. Common presenting features may include recurrent haemoptysis, cough, and wheeze. Symptomatic lesions are usually amenable to direct evaluation and removal via interventional bronchoscopic techniques.

### Competing Interests
The authors declare that there are no competing interests regarding the publication of this paper.

### Consent
Written informed consent was obtained from the patient for publication of this paper.

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References


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