CASE REPORT

All that wheezes is not asthma

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SUMMARY

A young woman was admitted with respiratory failure. Prior to her admission, she had been treated for clinical bronchial asthma for about 2 years for recurrent wheezing. Endotracheal intubation was difficult. A fibroptic bronchoscopy, while the patient was ventilated, revealed a central tracheal tumour compromising >90% of the tracheal lumen. The tumour was cored out during rigid bronchoscopy while ventilating the patient through tracheostomy. Histopathology of the tumour was suggestive of adenoid cystic carcinoma.

BACKGROUND

Tracheal tumours are uncommon and present with non-specific symptoms of cough and wheezing. Not infrequently this leads to an erroneous diagnosis of bronchial asthma.¹ Many of these cases are treated with anti-asthma medications such as bronchodilators and inhalational steroids for long before the cause of the symptoms is unravelled. We herewith present a young woman who had a history of wheezing and eventually presented to the emergency in respiratory failure, and the symptoms were found to be related to an obstructing tracheal tumour. The case re-emphasises the admonition that 'all that wheezes is not asthma.'

CASE PRESENTATION

A 25-year-old nomadic female was admitted to the accident and emergency department with respiratory distress. Clinical examination revealed a severely dyspnoeic, thin built young woman in grade 2-3 encephalopathy with severe central cyanosis. Her pulse was 114/min, respirations were 42/min and blood pressure 110/70 mm Hg. Accessory muscles of respiration were active and examination of the chest revealed stridor and bilateral wheeze with grossly diminished air entry on either side of the chest. Other general physical and systemic examination was unremarkable. Her medical history revealed that she had been treated with inhaled bronchodilators and steroids for wheezing for 2 years for which she had been labelled as bronchial asthma without a documentation on spirometry. In addition, her history disclosed occasional and intermittent streaking of sputum with blood. She denied a history of chest pain, inhalation of a foreign body, any recent loss of weight, pedal oedema or any abdominal swelling.

INVESTIGATIONS

A diagnosis of a life-threatening bronchial asthma was made. An arterial blood gas analysis revealed a PaCO₂ of 61 mm Hg with an oxygen saturation of 84% on FiO₂ 0.8. She was immediately put on

Figure 1 Near total occlusion of the tracheal lumen on CT.

high-flow oxygen at 10 l/min via face mask with bag reservoir, parenteral steroids and nebulised levo-salbutamol. The patient continued with respiratory failure and after about 30 min, mechanical ventilation was necessitated. Intubation with a 7.5 size tube was difficult and a 6.0 tube was used instead. The patient's respiratory failure improved. Haemoglobin was 10.5 g/dL, total leucocyte count $8.8 \times 10^9/L$ with 82% neutrophils and 15% lymphocytes and platelets $217 \times 10^9/L$. The serum biochemical parameters were normal. A radiograph of the chest showed hyperinflated lung fields.

On day 2, a high-resolution-CT (HRCT) of the airways and chest revealed a tracheal growth causing near total occlusion of the tracheal lumen, just below the vocal cords (figure 1). A fibroptic bronchoscopy was planned and the patient was tracheostomised and ventilation carried out through a cuffed tracheostomy tube. Fibroptic bronchoscopy was performed that revealed a walnut-shaped, polypoid vascular lesion arising from the anterolateral wall of the trachea, located about 1.5 cm from the vocal cords. The lesion was causing >90% occlusion of tracheal lumen. (figure 2) Biopsy was obtained.

TREATMENT

The patient's general condition and the respiratory failure improved completely on ventilation administered through a tracheostomy. On day 4, a rigid bronchoscopy was performed under general anaesthesia and the tumour eviscerated completely.

OUTCOME AND FOLLOW-UP

Postoperative period was uneventful. The patient was taken off the ventilator, the tracheostomy tube removed and discharged after 7 days of her admission. The histopathological examination of the tumour biopsy as well as the surgical specimen revealed features of adenoid cystic carcinoma (figure 3). The patient was lost to follow-up after discharge.

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Figure 2 The appearance of the tumour on bronchoscopy.

DISCUSSION

Tracheal tumours account for <1% of all malignancy with the majority being malignant.² The most commonly encountered type is the squamous cell carcinoma followed by adenoid cystic carcinoma.³ Other histologies, especially benign are even rarer than the malignant histologies.⁴

It is common for tracheal tumours to be misdiagnosed as bronchial asthma due to the presentation of cough and wheezing with episodes of dyspnoea. The mimicking symptoms often lead to a delay in the diagnosis and a time lag of 2 months to 2 years was reported in a series from Toronto General Hospital.⁵

An important accompaniment of tracheal tumours is stridor that is seen in up to one-fifth of the cases.⁶ The constellation of breathlessness, wheezing, stridor, haemoptysis and voice change, termed as the 'tracheal syndrome' may present in up to 85% of patients with tracheal neoplasms.⁷ Wheezing in tracheal obstruction is typically described as inspiratory as against the predominantly expiratory wheezing in bronchial asthma. Orthopnoea and paroxysmal dyspnoea may also manifest secondary to the change in position of the tumour with the change of posture by the patient as well as accumulation of secretions within the trachea during sleep.⁸ Obstruction to the trachea during inspiration occurs by a 'ball-valve' mechanism, resulting in stridor,

Figure 3 The histopathological features suggestive of adenoid cystic carcinoma.

whereas the tracheal lumen gets obstructed during expiration by a 'check-valve' mechanism that results in wheezing and hyperinflation.⁸

Various features that suggest a tracheal neoplasm as the cause of symptoms include a rather recent onset of wheezing which is paroxysmal, more pronounced during inspiration, poorly responsive to bronchodilators and steroids and relieved by change in body position. Accompanying haemoptysis and orthopnoea or paroxysmal nocturnal dyspnoea in the absence of a cardiovascular disease also strongly suggest a non-asthmatic aetiology to wheezing.⁸ Any wheezing that is relieved or precipitated by a change in body position should lead to a suspicion of obstructing lesion of the trachea.⁸

Upon spirometry a linear relationship between volume and time, a flat-topped flow-volume loop and a low peak flow relative to forced expiratory volume in 1 s (FeV1) may point towards a tracheal obstruction.⁹ There is no bronchodilator response as against the characteristic increase of FeV1 in bronchial asthma. Radiography and CT imaging help document the tumour. Bronchoscopy is not only diagnostic but can be performed with curative intent, especially in benign lesions such as lipomas and neurilemmomas.

Adenoid cystic carcinomas are generally well-circumscribed lesions that are smooth in appearance. They grow slowly and patients have been known to survive up to 15 years even with pulmonary metastasis.¹ Surgical resection with reconstruction is the best mode of treatment with radiotherapy being the next option in patients who cannot tolerate surgery. Some cases may require adjuvant chemotherapy, which is employed after these procedures. Laser therapy and/or tracheal stents are employed for palliation.

Learning points

- ▶ Patient with wheezing may not have underlying asthma.
- ▶ Stridor and haemoptysis accompanying wheezing suggest a non-asthmatic aetiology of wheezing.
- ▶ Wheezing or breathlessness that improves with change in body position should alert a physician to a tracheal obstruction.
- ▶ Spirometry may aid in the diagnosis of fixed airway obstruction.

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