CASE REPORT

'Nocturnal noises': an isolated stridor in an adolescent

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SUMMARY

An 18-year-old man presented to primary care with a 2-year history of exclusively nocturnal 'noisy breathing'. He was otherwise asymptomatic. He had never smoked and was previously healthy. Spirometry showed a severely obstructive picture with forced expiratory volume in 1 s (FEV1) 1.87 L (44% predicted), forced vital capacity (FVC) 4.0 L (80%) and FEV1/FVC ratio of 47%. A diagnosis of asthma was suspected and a trial of inhaled bronchodilators and corticosteroids was initiated. Failure to improve symptoms led to referral to the Respiratory Clinic, where his mother replayed a recording of the 'noisy breathing' on her mobile phone. Subsequent examination revealed a stridor on expiration. Flow volume loop showed a plateau of the expiratory limb, consistent with intrathoracic upper airway obstruction. CT of the thorax revealed a massively dilated oesophagus, filled with food residue, reflecting an achalasia, causing lower tracheal compression. He is now being considered for a myotomy procedure.

BACKGROUND

Achalasia presenting with an isolated stridor is rare. To the best of our knowledge this is the only reported case of achalasia presenting solely with stridor in an adolescent.

CASE PRESENTATION

An 18-year-old male student was seen in primary care with a 2-year history of 'noisy breathing', exclusively nocturnal, which was recorded by his mother on her mobile phone.

He had never smoked, was previously healthy and otherwise asymptomatic.

A diagnosis of asthma was made, but there was no response to 8 months of therapy with inhaled bronchodilators and corticosteroids. He was then referred for specialist assessment of ongoing symptoms.

INVESTIGATIONS

On attendance at the respiratory clinic, the mother replayed her recording to the attending physician, which demonstrated a high-pitched monophonic expiratory sound.

Spirometry showed a severely obstructive picture with forced expiratory volume in 1 s (FEV1) 1.87 L (44% predicted), forced vital capacity (FVC) 4.0 L (80%) and FEV1/FVC ratio of 47%. Flow volume loop showed an abnormal plateau of the expiratory limb, typical of an intrathoracic upper airway obstruction (figure 1). Chest radiograph was unremarkable.

Following Respiratory Clinic assessment and the finding of a stridor, he subsequently underwent a CT of the thorax. This demonstrated a massively dilated oesophagus throughout its length and filled with food residue, from which tracheal compression was evident (figure 2).

Oesophagogastroscopy demonstrated a dilated oesophagus, with no stricture or obstructing mass lesion and oesophageal manometry was consistent with a hypertensive lower oesophageal sphincter, which did not relax on swallowing. These investigations confirmed a diagnosis of achalasia.

TREATMENT

Various treatment options were considered, including botulinum toxin injection into the lower oesophageal sphincter or pneumatic dilation. However, these strategies would provide only temporary benefit rather than a long-term solution. Therefore, the patient was referred for definitive treatment by surgical cardiomyotomy.

OUTCOME AND FOLLOW-UP

Achalasia is characterised by failure of relaxation of the lower oesophageal sphincter and aperistalsis of the oesophagus. It most commonly presents with

Figure 1 Flow volume loop demonstrating abnormal plateau in the expiratory limb, consistent with upper airway obstruction.

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Figure 2 CT of the thorax demonstrating a massively dilated oesophagus, filled with food debris and resultant tracheal compression.

dysphagia, but may also cause chest pain, heartburn and food aspiration. The presence of respiratory symptoms is uncommon.

Presentation with stridor due to tracheal compression is a rare manifestation, which has been reported primarily in older individuals, typically in septogenarians and octogenarians. It is usually associated with longstanding antecedent dysphagia. However, it may present acutely with life-threatening airway compromise.^{1–5}

Achalasia presenting with an isolated stridor is extremely rare, particularly in younger individuals. One case has been reported of intermittent stridor due to tracheal compression in a 14-year-old boy, in the absence of any gastrointestinal symptoms.⁶ However, this boy also had several years of dry cough at the time of presentation. This patient was also initially given treatment for suspected asthma, resulting in a delayed diagnosis and treatment.

Of note, this patient did not experience any daytime symptoms, sleep disturbance, swallowing difficulty, vomiting, regurgitation or

loss of appetite. This is unique as a case of achalasia of the oesophagus presenting solely with a stridor in an adolescent.

Learning points

- ▶ It is important for clinicians to distinguish the polyphonic wheeze of asthma from the monophonic 'wheeze' or stridor of upper airway obstruction.
- ▶ Stridor is a rare manifestation of achalasia, which can result from tracheal compression by the dilated oesophagus.
- ▶ Treatment options include calcium channel blockers, nitrates, botulinum toxin injection, balloon dilation or cardiomyotomy.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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