CASE FOR DIAGNOSIS

Stridor in an infant

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Case history

A 7 month old male presented with stridor from the age of 6 weeks. He was one of identical twins born at 38 weeks gestation following an otherwise uncomplicated pregnancy and delivery. He remained completely well until 6 weeks of age when the isolated symptom of stridor began gradually, increased in loudness until 10 weeks of age and then remained constant until presentation. It was present at all times, worse on exertion and not influenced by body position nor feeding. The child had remained well throughout and was thriving with no episodes of fever, feed refusal nor swallowing difficulties. There was no history of ingested nor inhaled foreign body. A clinical diagnosis of laryngomalacia had been made by several medical practitioners.

On examination he was well looking with normal growth. He had biphasic stridor that was louder on expiration, rough in character and associated with mild indrawing of intercostal muscles. The chest was normally shaped and clear to auscultation. Examination was otherwise normal. The unusual character of stridor and age of onset of symptoms prompted further investigation.

His chest radiograph was normal with no apparent airway narrowing. Barium swallow showed no obvious abnormality but was limited owing to technical difficulties. Flexible fiberoptic bronchoscopy was performed. This was followed by spiral computed tomography (CT) of the chest, (fig. 1) and a bronchogram (fig. 2).

Fig. 1. – Axial computed tomographic scan (with contrast) of the chest at the level of the second thoracic vertebra.

Fig. 2. – Bronchogram: lateral view of trachea obtained following direct instillation of 1 mL Isovue 300 into the trachea. A nasogastric tube is in place.

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BEFORE TURNING THE PAGE, WRITE DOWN YOUR INTERPRETATION OF THE FIGURES, YOUR DIAGNOSIS, ALTERNATIVE DIAGNOSES AND SUGGESTED TREATMENT

Interpretation

Bronchoscopy

Bronchoscopy (image not shown) revealed a slit-like, antero-posterior narrowing of the mid-trachea that began ~4 cm below the larynx and extended over a length of 4 cm, with normal overlying mucosa. The narrowed region showed no increase in diameter with application of continuous positive airway pressure (CPAP) of 20 cmH₂O pressure. The distal trachea, carina, major and segmental bronchi were all normal.

Computed tomography

Figures 1 and 3 show that the trachea (T) is compressed and displaced to the right by a bi-loculated gas filled mass (M) sited anterior to the collapsed oesophagus (O). The mass had the dimensions of 1.5 cm. (antero-posterior) by 2 cm (transverse) by 4 cm (cranio-caudad). A radiolucent line (D) is seen posteriorly within the mass. This represents a flat plastic disc in transverse section. There was minimal contrast enhancement. No communication between the mass and either the trachea or oesophagus was demonstrated.

Bronchogram

The bronchogram (fig. 2) shows antero-posterior bowing and narrowing of the trachea over a length of 4 thoracic vertebrae. A naso-gastric tube is seen posteriorly within the oesophagus. A soft tissue mass separates trachea from oesophagus. No connection between the trachea and mass is demonstrated.

Diagnosis: "Tracheal compression by a mediastinal abscess secondary to oesophageal perforation by a flat plastic disc"

Clinical course

The radiolucent line (D) on the CT was not recognized prior to surgery. The lesion was interpreted as representing a foregut duplication that had an intermittent or valve-like connection with the oesophagus or airway. The authors

Fig. 3. – Identical image to figure 1 (computed tomographic scan of the chest) with explanatory labels. T: trachea; M: mass, with two gas filled cavities; D: plastic disk; O: oesophagus. See text (*Interpretation* section) for explanation.

were not able to demonstrate this connection prior to surgery using contrast studies of the oesophagus and trachea. Because of the ongoing stridor and risk of infection, surgical removal was attempted. An approach *via* a right thoracotomy was planned on the basis that this would give good exposure to the potential attachment between lesion and upper oesophagus and/or trachea. At exploration no lesion could be identified, although there was evidence of chronic inflammation in the upper mediastinum. One week later exploration *via* the left neck revealed a diverticulum, encased in fibrous tissue, extending from the left side of the oesophagus. On opening this lesion a flat plastic disc lying parallel to the oesophageal wall was identified and removed (fig. 4). The disc was probably a counter from a children's game. The dome of the cavity was excised and the oesophageal wall repaired. Histologic examination of excised tissue showed a chronic abscess with no evidence of a mucosal lining, indicating that the diverticulum probably resulted from perforation of the oesophagus by the foreign body, rather than being congenital in nature.

Postoperative recovery was complicated by airway obstruction that necessitated endotracheal intubation for a period of 77 h. The patient was discharged 7 days after the operation and remains well without stridor nor apparent difficulties with swallowing.

Discussion

The relative prominence of the expiratory component of the stridor suggested tracheal narrowing at an intrathoracic site. Bronchoscopy confirmed this, and suggested extrinsic compression. The radiological features confirmed a mass between oesophagus and trachea that contained gas. The fact that the patient was well, indicated that an abscess with gas producing organisms was unlikely, and that a

Fig. 4. – Circular flat plastic disk found lying within a pseudodiverticulum of the upper oesophagus. Dimensions were 15.0 mm in diameter and 1.0 mm in thickness. The disc was probably a counter from a children's game.

connection between the mass and either trachea or oesophagus (or both), was present. The provisional diagnosis prior to operation was a developmental anomaly of the primitive foregut, most likely an oesophageal duplication cyst. Bronchogenic cyst, and spontaneous abscess with perforation into oesophagus or trachea were also considered. Foreign body was not considered likely owing to the young age at onset of stridor. Other lesions that may present as a superior mediastinal mass at this age (*e.g.* neuroblastoma) were considered unlikely as they would not contain gas, and would not normally be positioned between the oesophagus and trachea.

The clinical, operative and histologic findings of this case suggest that the infant ingested the plastic disc at ~6 weeks of age and that it lodged in the upper oesophagus, which it subsequently perforated. The resulting chronic abscess lead to airway compression and stridor. Whilst the circumstances that lead to ingestion of the disc are completely unknown, it is speculated that it may have been given to the patient by one of his three older siblings. There was no evidence whatsoever that the ingestion was an act of deliberate harm.

Chronic stridor in infancy is most commonly due to laryngomalacia but may be the result of more serious pathology [1]. If the typical clinical features of laryngomalacia are present then detailed investigation is not usually required. In this case the more prominent expiratory component of the stridor (indicating an intrathoracic site of obstruction) and the late age at onset of symptoms were inconsistent with laryngomalacia and lead to further investigation [1]. Biphasic stridor in particular, is rarely seen in laryngomalacia and suggests the need for further investigation when seen in a child with chronic stridor.

Bronchoscopy is the gold standard for investigation of chronic stridor. It reliably detects narrowing of the larynx, subglottis, trachea and major bronchi. Tracheal narrowing is an uncommon cause of stridor caused by tracheomalacia, intrinsic narrowing (tracheal stenosis) or extrinsic compression by masses or aberrant vascular structures (*e.g.* vascular ring) [1]. In all cases of tracheal narrowing extrinsic compression must be considered and further imaging is frequently required. The normal chest radiograph and repeatedly normal oesophograms in this case highlight the limitations of these modalities in detecting extrinsic compression to the airway. This is particularly true when pathology is located at the thoracic inlet, involves diffuse anterior compression rather than a focal constriction of the oesophagus or where the mass is not directly applied to the oesophagus.

Magnetic Resonance Imaging (MRI) or CT are frequently considered once tracheal narrowing is demonstrated. MRI is superior to other modalities in preoperative assessment of vascular ring and is used when this pathology is suspected [2, 3]. CT is used when the oesophogram does not suggest vascular ring, as it is more readily available and is adequate in assessing for mass lesions.

Foreign body was not seriously considered in the preoperative differential diagnosis despite being visible in retrospect on the CT. This highlights the difficulties involved in diagnosis of radiolucent foreign bodies in children [4, 5]. Between 17% and 30% of oesophageal foreign bodies in children are asymptomatic [4]. Delayed diagnosis may result in serious complications including airway obstruction, oesophageal perforation, mediastinal infection, tracheo-oesophageal fistula, oesophago-aortic fistula, pneumomediastinum and spondylodiscitis [4].

In summary, oesophageal foreign body should be considered in the differential diagnosis of stridor at all ages, including infancy. Whilst chest radiograph and barium meal are of value in the diagnostic assessment of stridor, bronchoscopy remains the gold standard and should be used where clinical features are not typical for a benign aetiology.

Keywords: Foreign bodies, oesophageal perforation, respiratory sounds

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