When the penny drops...

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DESCRIPTION

A 1-year 5-month-old boy presents with a 1-day history of severe biphasic stridor. He had multiple previous admissions for recurrent lower respiratory tract infections. On his last admission, a month earlier, he was seen for noisy breathing, where laryngomalacia was suspected and confirmed on non-anaesthetic upper airway scope. His neonatal history was uneventful, but his mother noticed noisy breathing shortly after birth. She had a normal antenatal ultrasound with a structurally normal heart.

On the chest X-ray an oval, metallic, foreign body, representing a coin, was noted to be located posterior to the trachea and beyond the thoracic inlet (figure 1), and therefore presumed to be lodged in the upper third of the oesophagus. No parenchymal abnormality or air-trapping was noted.

Clinically the severity of the stridor was out of keeping with a foreign body in the oesophagus. Bronchoscopy before removal confirmed severe compression of the posterior membrane of the middle part of the trachea. The coin was removed with a rigid oesophagus scope with no signs of ulceration or fistula.

After removal of foreign body (FB) the trachea was noted to be displaced to the left on the chest X-ray and narrowed by a presumed right-sided aortic arch or a double aortic arch with right-sided dominance.

Postoperatively he still had stridor that improved but the bronchoscopy was repeated after a week because of ongoing noisy breathing.

Bronchoscopy confirmed 75% narrowing of the middle part of the trachea more prominent from the right with significant pulsation from both anterior and posterior, in keeping with a double aortic arch (video 1).

Video 1 Flexible bronchoscopy video 1 week after removal of the coin. Bronchoscopy was performed with an Olympus 3.5 mm scope demonstrating a prominent and large epiglottis with severe mid-tracheal narrowing more prominent from the right side. There is both anterior and posterior prominent pulsation present. Beyond the area the trachea and bronchi is normal. The bronchoscopy appearance is suggestive of a double aortic arch with a dominant right arch.

Contrast enhanced axial CT scan confirmed the presence of a double aortic arch with a right dominance (figure 2A,B). The three-dimensional (3D) Volume rendered reconstructions demonstrated the double arch more elegantly and confirmed the dominance of the right arch component (figure 2C). In addition, the 3D reconstructions demonstrated the location and extent of the tracheal narrowing as well as the implicated double aortic arch (figure 2D).

A left-sided thoracotomy was performed in the fourth intercostal space. The mediastinal pleura was opened over the descending aorta. The anterior aortic arch and the ductus arteriosus was identified. The left

Figure 1 (A) Posterior anterior and (B) lateral radiograph combination in a 1-year 5-month-old boy demonstrates an oval metallic foreign body, representing a coin lodged beyond the thoracic inlet and posterior to the trachea, likely in the upper third of the oesophagus. (C) Follow-up frontal radiograph demonstrates the trachea to be deviated to the left (arrow) and compressed from a presumed right aortic arch/right dominant double aortic arch (AA).

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Figure 2 Post contrast CT scan in a 1-year 5-month-old boy. (A,B) Sequential axial CT slices at the level of the aortic arch demonstrating the double aortic arch with a dominant right-sided component (short thick arrows) and smaller left arch encircling (long thin arrows) the trachea and oesophagus. (C) Three-dimensional (3D) volume rendered reconstruction from the same CT scan viewed from posterior demonstrates the descending aorta (DA) formed by the union of the smaller left (thin long arrow) and dominant right (short thick arrow) components of the double aortic arch and encircling the trachea (T), which is narrowed. (D) 3D volume rendered reconstruction from the same CT scan with vascular structures removed and viewed from anterior, demonstrates the tracheal compression from both right and left (arrows)—right more severe corresponding to the dominant right arch.

carotid artery originated from the anterior arch. The distal part of the anterior arch connected to the ligamentum arteriosum and to the descending aorta looked atretic and this part of the arch was tied. The ligamentum arteriosum was also tied. The ductus was then divided, and the anterior arch divided relieving the compression on the trachea and the oesophagus.

Follow-up bronchoscopy showed significant improvement of the airway compression with mild malacia present.

Consent was given by the patient's parents for publication of this case.

Double aortic arch is one of the most common vascular abnormalities causing significant airway compression.¹ Hommel described the first postmortem case in 1737, with the first

documented surgical repair only being carried out by Robert Gross in 1947.²

This case demonstrates that noisy breathing can have more than one cause as this child had both laryngomalacia and double aortic arch. The chest X-ray was not normal, with a right-side arch and narrowing of the trachea above the carina which is indicative of airway compression.

The combination of oesophagus FB and double aortic arch have been rarely reported.^{3,4}

Contributors PG and LM was involved in the management of the patient. JJ was the surgeon involved. SA was responsible for the radiology. All four were involved in creating the manuscript.

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Learning points

- Noisy breathing in a young child may have more than one aetiology including foreign body aspiration and needs a complete endoscopy investigation.
- Chest X-ray findings of double aortic arch may be subtle but if the trachea is not completely visible and especially if right aortic arch is present, the diagnosis must be considered.
- Oesophagus coin ingestion have mild airway symptoms, if symptoms are severe another diagnosis or complication must be considered.

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