FATAL HEMORRHAGE DUE TO TRACHEAL-ESOPHAGEAL-AORTIC FISTULA IN A PATIENT WITH DOUBLE AORTIC ARCH

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ABSTRACT
We report a case of a 18-year-old male with double aortic arch who underwent surgery for bleeding from a left bulbar cavernous angioma of the medulla oblongata. A tracheostomy tube was positioned but after several days, the patient died because of a tracheoesophageal fistula with left aortic arch erosion due to the decubitus of the tube cuff.

KEYWORDS double aortic arch; tracheostomy contraindication; hemorrhage

Case report
Double aortic arch (DAA) is an uncommon vascular malformation that consist in a duplication of the aorta that create a vascular ring around trachea and esophagus. Commonly there is a larger dominant right arch and a smaller, hypoplastic left aortic arch in front of the trachea/esophagus. The two arches join to form the descending aorta that is usually on the left side but sometimes we can find it right-sided or in the midline [1]. This condition is diagnosed and treated in childhood for tracheal or esophageal compression.

We report a case of a 18-year-old male that developed right hemiplegia, nystagmus, and dysarthria, associated with dysphagia and alteration of the cough reflex due to the bleeding of a left bulbar cavernous angioma of the medulla oblongata.

He underwent surgery in a major neurosurgical center with a successful control of bleeding, but on the third postoperative day, he required a tracheostomy because the symptoms did not recover.

In the next four days, he developed respiratory failure, and CT scan showed bilateral pneumonia with right pleural effusion and a double aortic arch (Fig. 1a, b, d). He had an infection due S. Aureus and S. Pneumoniae. A chest drain was placed in the right hemithorax, and antibiotic therapy was started.

In the 24th postoperative day, due to the persistence of pulmonary infection, he was transferred to our hospital where he underwent to thoracoscopic right empiemectomy and pulmonary decortication.

After the procedure, the patient did not have complications, and he had improvement of general and respiratory conditions.

In the 27th, the patient developed massive hemoptysis and htememesis that required cardiopulmonary resuscitation and blood transfusion. The bleeding was controlled by positioning of Sengstaken Blakemore tube in the upper esophagus.

The bronchoscopy did not show active bronchial hemorrhage, and the CT scan of the thorax showed the esophageal-tracheal space occupied by the tracheostomy tube cuff. The gastroscopy showed the presence of the tracheostomy cuff into the esophageal lumen (Fig. 1c) and blood into the stomach. A second bronchoscopy, carried out after the removal of the tracheal cannula, showed a tracheal mucosal irregularity, no clear signs of fistula and restart of bleeding. A new tracheal tube was positioned, and the patient was transferred to a cardiovascular department where he died due to fatal bleeding.

An autopsy revealed a tracheoesophageal fistula and left aortic arch erosion.
Fig 1. a,b) CT scan showing the double aortic arch; c) the tracheostomy cuff into the esophageal lumen; d) 3D CT scan reconstruction showing the double aortic arch.
Fatal erosion hemorrhages in patients with tracheostomy tube usually affect the innominate artery; in this case report, the erosion affected the anomalous aortic arch. The presence of double aortic arch is a contraindication for positioning of a tracheostomy tube due to the ischemic action and the decubitus of the cuff on the tracheal, esophageal and aortic wall.

Glossary of abbreviation:
Double aortic arch: DAA; Computed Tomography: CT; S. Aureus: Staphylococcus Aureus; S. Pneumoniae: Streptococcus Pneumoniae

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References