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CLINICAL INFORMATION

Aberrant right subclavian artery-esophageal fistula: massive upper gastrointestinal hemorrhage secondary to prolonged intubation



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KEYWORDS

Aberrant right
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Abstract Aberrant right subclavian artery-esophageal fistula is a rare but potentially fatal complication. It may be associated with procedures, such as tracheostomy and tracheal or esophageal intubation, and yields massive upper gastrointestinal bleeding difficult to identify and to control.

A high index of suspicion is essential for early diagnosis and better prognosis.

We report a rare case of a patient who survived after emergent surgical procedure for massive upper gastrointestinal bleeding secondary to *aberrant right subclavian artery-esophageal fistula* after prolonged intubation.

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PALAVRAS-CHAVE

Artéria subclávia
direita anômala;
Hemorragia maciça;
Intubação gástrica
prolongada

Fístula de artéria subclávia direita anômala com esôfago – hemorragia digestiva alta maciça secundária a intubação gástrica prolongada

Resumo A fístula de artéria subclávia direita anômala com o esôfago é uma complicação rara, mas potencialmente fatal. Pode estar associada a procedimentos como traqueostomia e intubação traqueal ou esofágica e originar hemorragia digestiva alta maciça, de difícil identificação e controle.

Um elevado índice de suspeição é essencial para o diagnóstico precoce e a melhoria do prognóstico.

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Relatamos caso raro de doente que sobreviveu após intervenção cirúrgica emergente por hemorragia digestiva alta maciça secundária a fístula de artéria subclávia direita anômala com esôfago, após intubação gástrica prolongada.

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Background and objectives

Aberrant right subclavian artery is a relatively common congenital vascular anomaly (1–2%) incidence.¹ Most patients have no symptoms, which makes early detection difficult.

The presence of aberrant right subclavian artery may have devastating consequences during procedures, such as tracheostomy, tracheal intubation, and surgical repair of thoracic aneurysms² or during prolonged esophageal intubation.³ Gastric tubes can cause erosion of the esophageal wall resulting in fistula formation with the underlying aberrant artery, causing massive gastrointestinal bleeding that is difficult to identify and control.

Case report

A male patient, 20 years old, was admitted to the intensive care unit due to severe traumatic brain injuries, thoracic and abdominal trauma, and left lower limb fracture secondary to a car accident. On admission, the patient was hemodynamically stable, with a Glasgow Coma Scale of 4. The patient was intubated with a 7.5-cuffed tube and mechanically ventilated. He also had a nasogastric tube on free drainage and urinary catheter.

At 22 days of hospitalization, the patient remained sedated with midazolam and intubated for airway protection and mechanical ventilation, as there were no conditions for spontaneous ventilation. He was hemodynamically stable and remained with urine output monitoring and nasogastric tube for enteral nutrition.

The patient developed suddenly upper gastrointestinal bleeding with significant hematemesis. The gastroenterologist contacted for urgent endoscopy revealed: "Abundant amount of blood and clots throughout the esophageal path. Bloodstream seems to come from high esophageal level. The entire gastric cavity filled with massive clot and the same happening in the duodenal level". Because of the severity of bleeding and inability to identify its origin, the patient underwent emergency surgery.

Upon entering the operating room, the patient had hypovolemic shock, marked hypotension, intense pale mucous, and decreased hemoglobin values from 12.3 g dL⁻¹ to 6.8 g dL⁻¹.

He underwent balanced anesthesia with sevoflurane, fentanyl, and rocuronium for surgical intervention. Massive transfusion was initiated with rapid infusion of colloid and crystalloid solutions and blood products via central venous

catheter placed in the right femoral vein and two catheters, 14 G and 16 G, in both upper limbs.

During exploratory laparotomy and anterior gastrostomy, it was not possible to identify the source of bleeding. The gastroenterologist was again contacted to perform another endoscopy which identified: "Ulcer with copious bleeding at the proximal esophagus level, at a distance of 20 cm from the incisors". The option was a temporary closure of gastrostomy and abdomen, and an exploratory right posterolateral thoracotomy was performed, which showed arterio-esophageal fistula. Due to the need for differentiated surgical treatment for arterial fistula correction, a cardiothoracic surgeon was contacted.

Left thoracotomy was performed with identification of controlled rupture of aberrant right subclavian artery. Right re-thoracotomy with ligation of the aberrant right subclavian artery, esophagography, and abdominal closure were performed.

To control massive hemorrhage, 18 units of erythrocyte concentrate, 16 units of fresh frozen plasma, two pools of platelets, prothrombin complex, and fibrinogen were administered, in addition to colloid 500 mL and crystalloid 1500 mL.

Surgery lasted 8 h. At the end of surgery, the patient was transported to the intensive care unit, intubated and on mechanical ventilation monitored.

The patient survived, evolved favorably and was transferred to the Department of Surgery. He was discharged one month and a half after this occurrence and was followed in outpatient visits of surgery, physical medicine, and rehabilitation.

Discussion

The most common anomaly of the aortic arch is the aberrant right subclavian artery, with an incidence of 1–2%.¹ There is a predominance of females (65–72%).⁴ The aberrant right subclavian artery forms an incomplete vascular ring, emerging from the descending aorta and obliquely crosses the mediastinum toward the right armpit. In 80% of the cases, it lies posterior to the esophagus (as in the clinical case described); in 15% of the cases, it lies between the esophagus and the trachea; and in 5% of the cases, it lies anterior to the trachea.⁵

Most patients have no symptoms. However, a small proportion may manifest gastrointestinal symptoms (dysphagia) and, rarely, respiratory symptoms may be present.⁶

It is extremely rare in the occurrence of bleeding by communication between the esophagus and anomalous right subclavian artery. There are few cases reported after prolonged esophageal intubation.^{3,7}

The anatomic proximity to the esophagus and, eventually, to the trachea renders the aberrant subclavian artery vulnerable to extrinsic compression by gastric, tracheal or vascular tubes.⁵

The relatively high incidence of this anomaly and the common use of gastric tubes in hospitals increase the possibility of this complication.

Bleeding caused by esophageal fistula and aberrant right subclavian artery is manifested by sudden massive hemorrhage with massive fresh arterial blood hematemesis, often followed by an asymptomatic period of variable duration, and a period of sudden and often fatal hemorrhage.^{3,8} Clinical signs, such as precordial pain or sentinel hemorrhage, which are present in 50% of the cases of aorto-esophageal fistula, are rarely present in this situation.⁵

Faced with massive bleeding, the priority is to ensure and protect the airway by tracheal intubation. In our patient, this procedure was not necessary, as he was already intubated and on mechanical ventilation, which may have improved his prognosis. Subsequently, the priority is to control the bleeding.

Early performance of endoscopy is essential for diagnosis and exclusion of other potential causes of gastrointestinal bleeding. However, in most cases, massive bleeding prevents a conclusive endoscope examination. The diagnosis of the source of bleeding is most often made intraoperatively as a laparotomy and/or exploratory thoracotomy, as happened in this case. Only the recognition of this situation will allow the establishment of immediate surgical treatment, with decreased hemodynamic changes resulting from massive hemorrhage.

Patient's prognosis depends on early diagnosis and surgical repair.⁵

Diagnostic difficulty in patients with massive hemorrhage secondary to fistula between aberrant right subclavian

artery and esophagus determines the high mortality of this complication.³

A high index of suspicion is essential for this condition early diagnosis.⁷

This case report aims to raise awareness among anesthesiologists for the presence of this congenital anomaly, rarely identified, but potentially responsible for serious and/or fatal complications associated with acts as simple as nasogastric or tracheal tube placement.

Conflicts of interest

The authors declare no conflicts of interest.

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