CASE REPORT

JORNAL **VASCULAR** BRASILEIRO
ISSN 1677-7301 (Online)

Primary aortoesophageal fistula: case report

Fístula aorto-esofágica primária: relato de caso

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Abstract

An aortoesophageal fistula is an abnormal communication between the aorta and the esophagus, causing potentially fatal upper gastrointestinal bleeding. The objective of this article is to report a successful case of treatment with a single aortic endovascular approach and conservative treatment of the esophagus in a case of aortoesophageal fistula. An 81-year-old patient was admitted with signs of massive upper gastrointestinal bleeding and, after tests, an aortoesophageal fistula was diagnosed. Endovascular treatment was chosen and performed successfully. The patient was discharged after 9 days in hospital and remained in outpatient follow-up until the condition completely resolved. Early diagnosis is extremely important, since this is a fatal condition if left untreated. It is hoped that this report contributes content of relevance to the scientific community.

Keywords: esophageal fistula; gastrointestinal bleeding; endovascular procedures.

Resumo

Fístula aorto-esofágica é uma comunicação anormal entre a aorta e o esôfago, causadora de hemorragia digestiva alta potencialmente fatal. O objetivo deste trabalho é relatar um caso de sucesso na abordagem endovascular aórtica única e tratamento conservador do esôfago em fistula aorto-esofágica. A paciente de 81 anos foi admitida com sinais de hemorragia digestiva alta volumosa e, após realização de exames, diagnosticou-se uma fístula aorto-esofágica. Optado pela realização de tratamento endovascular, sendo bem sucedido, a paciente recebeu alta após nove dias de internação e manteve-se em seguimento ambulatorial até a resolução total do quadro. O diagnóstico precoce é extremamente importante, uma vez que se trata de uma patologia fatal na ausência de tratamento. Espera-se agregar conteúdo pertinente para comunidade científica.

Palavras-chave: fístula esofágica; hemorragia gastrointestinal; procedimentos endovasculares.

How to cite: Moreschi LK, Gomes HH, Vasconcelos CN. Primary aortoesophageal fistula: case report. J Vasc Bras. 2023;22:e20230126. https://doi.org/10.1590/1677-5449.202301262

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Financial support: None.
Conflicts of interest: No conflicts of interest declared concerning the publication of this article.
Submitted: August 17, 2023. Accepted: October 19, 2023.

The study was carried out at Hospital Norte Paranaense (HONPAR), Arapongas, PR, Brazil.
Ethics committee approval: 5.927.474 (CAAE: 61186622.4.0000.8017).

INTRODUCTION

Aorto-enteric fistulas are abnormal communications between the aorta and the gastrointestinal tract. In the majority of cases, this communication is with the duodenum and involves the esophagus in just one fifth of cases.[1] It is a disease with a rare cause and annual incidence of 0.007 per million[2] that is potentially fatal, with mortality of around 77% in patients who undergo treatment.[3] The first case to be diagnosed and described was in 1818 and the first report of successful treatment was published in 1980.[4]

Aortic fistulas can be primary or secondary. The first type is defined as a spontaneous communication between the primitive aorta and adjacent vessels or organs, while the second type involves prior aortic or esophageal surgery.[5] Clinical status varies depending on the etiology of the case, but the clinical triad of aortoesophageal fistulas described by Chiari comprises the most prevalent signs of chest pain, sentinel hemorrhage, and massive bleeding after a period with no symptoms.[6]

Since it is so rare, diagnosis demands a high index of suspicion, particularly when the fistula is primary. The initial test for diagnosis of upper digestive hemorrhages is upper digestive endoscopy (UDE), but it has a low detection rate for aortoesophageal fistula, because bleeding must have been recent.[7] The second examination for investigation is computed tomography (CT) which in turn is capable of identifying masses that may be identified as aneurysms or possible tumors.[8]

Since it is a rare clinical condition, there are no protocols to systematize care, which could compromise patients. As such, we believe that the greater the number of cases described in the literature and the more the subject is discussed, the greater the likelihood that future patients will have favorable prognosis. Therefore, the objective of this study is to describe a case of aortoesophageal fistula treated successfully. The patient signed a free and informed consent form and the study protocol was approved by the institutional Ethics Committee (decision number 5.927.474).

CASE REPORT

The patient was an 81-year-old female, with hypertension and diabetes who was admitted to the emergency department complaining of persistent coughing with onset 1 month previously, associated with hematemesis and weakness. His condition had worsened the previous day, with massive hematemesis, with the appearance of fresh blood. On physical examination, she had good color, was breathing well, with attenuated auscultation at the base of the right lung but normal cardiac auscultation, and her abdomen was painful on deep palpation in the epigastric area.

Initially, laboratory tests were unremarkable, but a chest X-ray showed consolidation at the base of the right lung. In view of this, the initial approach was empirical antibiotic therapy with intravenous Levofloxacin and the patient was admitted for etiological investigation of the hematemesis.

A UDE showed a lesion with fibrin-covered central erosion in the esophagus, 35 cm from the upper dental arch, with no signs of active bleeding (Figure 1). To enhance the investigation, a chest CT with contrast was ordered, showing signs of rupture of the descending aorta, with hematoma contained in the retrocardiac and retrocrural space, measuring 97 mm x 64 mm x 43 mm (Figure 2). There were no significant findings involving the heart or pericardium and no sign of enlarged lymph glands in the mediastinum or pulmonary region.

The patient underwent thoracic aortography, which confirmed presence of a descending thoracic aorta aneurysm at the level of the diaphragm (Figure 3). The decision was taken to conduct endovascular repair, deploying a straight endoprosthesis measuring 33 mm

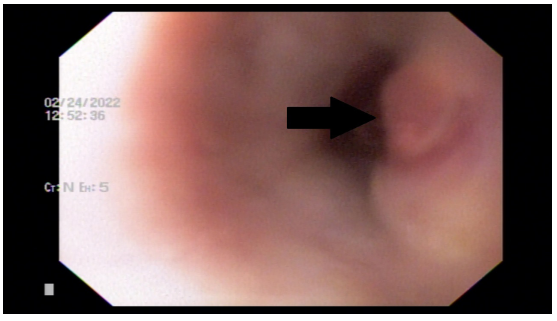

Figure 1. Central erosion of the esophagus (arrow).

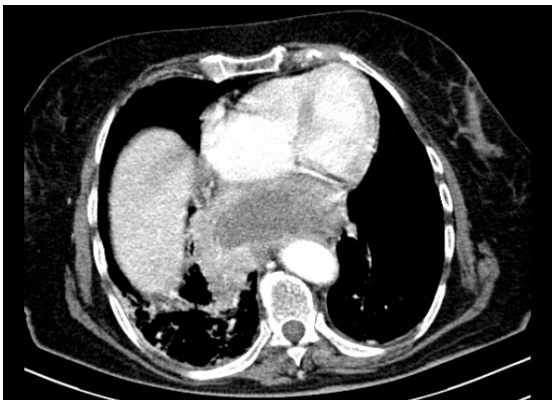

Figure 2. Computed tomography of the thorax (initial work-up) showing retrocardiac hematoma.

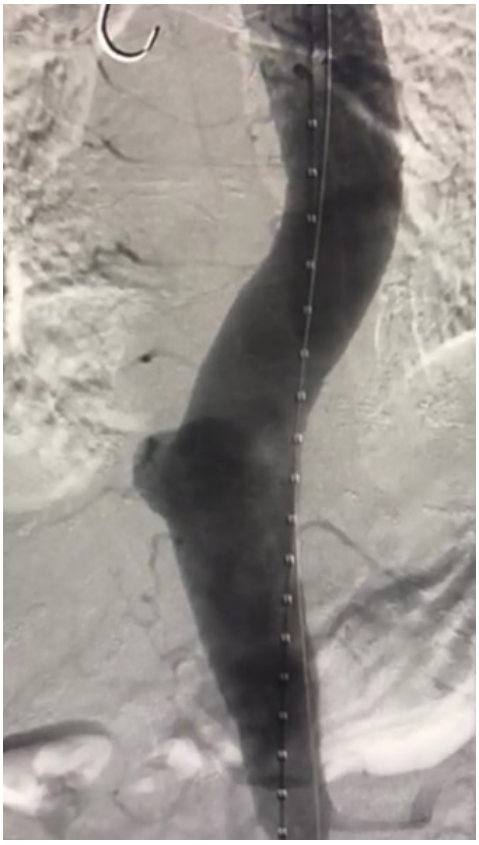

Figure 3. Arteriography showing aneurysm of the thoracic aorta.

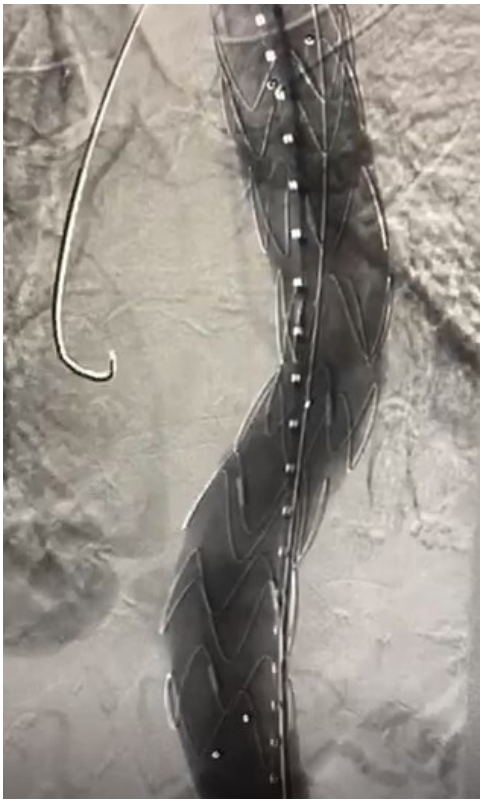

Figure 4. Endovascular repair of the thoracic aortic aneurysm with a straight endoprosthesis, showing absence of endoleaks.

x 33 mm x 170 mm into the thoracic descending aorta to the level of the celiac trunk (Figure 4). There were no complications during the procedure.

After endovascular treatment, a second UDE was performed, finding an orifice approximately 10 mm in size at the site of the erosion observed previously (Figure 5). In view of this, a nasogastric probe was fitted to feed the patient.

After 9 days in hospital, with no further episodes of hematemesis and with improving clinical status, the patient was discharged from hospital, feeding via the nasogastric probe and with a prescription for Levofloxacin to complete the 14-day antibiotic course.

Sixty days after hospital discharge, a control chest CT showed the endoprosthesis in the descending aorta at an appropriate position to treat the aortic rupture, with hematoma contained in the retrocardiac and retrocrural space, measuring 31 mm x 9 mm (Figure 6). Around 70 days after discharge, UDE was ordered again which revealed healing at the site of the previous erosion (Figure 7). The biopsy only showed mild chronic esophagitis and so the nasogastric probe was removed and the patient was instructed to start oral feeding. At outpatient follow-up, 3 months after discharge, the patient reported good tolerance of oral feeding and no complaints of any type.

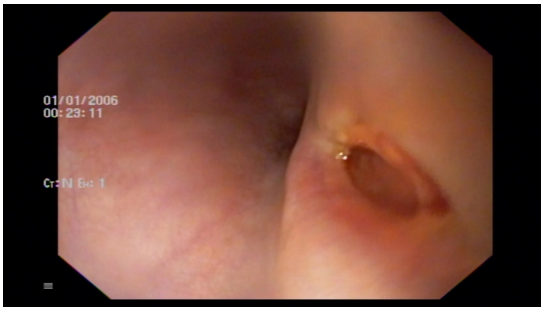

Figure 5. Orifice in the posterior wall of the esophagus.

DISCUSSION

Of the several possible causes of a primary aortoesophageal fistula, primary aortic aneurysm, foreign body ingestion, and thoracic cancer are the most important.[9] The etiology of the fistula in the case described here was probably multifactorial, since the aortic aneurysm was relatively small and unlikely to be the only cause.

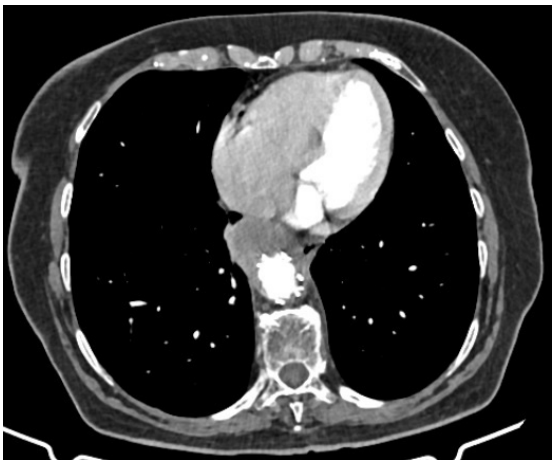

Figure 6. Computed tomography of the thorax (control) showing retrocardiac hematoma retracting and the endoprosthesis in the aorta.

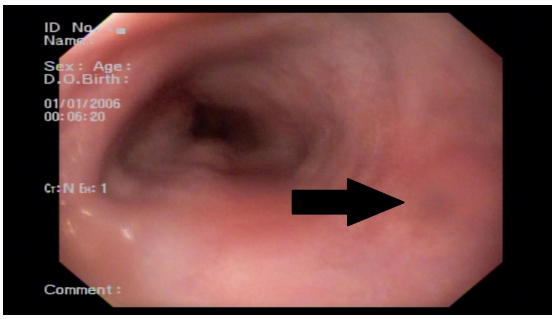

Figure 7. Erosion healed (arrow).

Diagnosis of upper digestive hemorrhages starts with UDE, in which the findings most characteristic of aortoesophageal fistulas are as follows: a submucosal protrusion similar to a tumor, an ulcerative lesion, or a pulsatile protrusion with a central fistula.[8] The patient described here had erosion of the esophagus, with no neoplastic characteristics and with a completed healing process, which could indicate a foreign body injury, corroborating the hypothesis of multifactorial etiology.

If UDE is inconclusive, CT with contrast should be ordered, since it can confirm diagnosis in 30 to 61% of these cases.[5] In the present case, CT was extremely important because it showed the source of the patient's bleeding.

If available, endovascular treatment is the first choice for diseases of the thoracic aorta, since it has lower rates of morbidity and mortality than open surgical treatment, with an 87.3% success rate in cases of aortoesophageal fistula.[10] In cases in which endovascular treatment is not feasible, open repair is chosen, with insertion of a prosthesis via a left thoracotomy, possibly requiring extracorporeal circulation to minimize complications, such as ischemia of the viscera and spinal marrow.[11]

Management of the esophageal injury will depend on the characteristics of the injury and the availability of resources at the institution. Primary closure is one possible option, but the success rate is higher after partial or total esophagectomy, with primary anastomosis, combined with a jejunostomy or gastrostomy, to avert dehiscence of the anastomosis and facilitate patient feeding.[12] If there is an infectious process with purulent secretion via the fistula orifice, one option for treatment is to insert a pigtail esophageal stent via the fistula, combined with jejunostomy.[13]

Conventional treatment of aortoesophageal fistulas involves repair of both the aortic and the esophageal injuries, which can be performed concomitantly or separately from the surgical process. Recent reports also describe the treatment option of only treating the aortic injury, using endovascular techniques to repair the aortoesophageal fistula.[14] In the current case, it was decided to employ endovascular treatment in the aorta and manage the esophageal erosion conservatively, since it was relatively small, with viable margins, and free from infection. In combination, a nasogastric probe was fitted, to improve patient nutrition.

CONCLUSIONS

Aortoesophageal fistula is a rare pathology with high mortality. Diagnosis is difficult and in order to arrive at a diagnosis in time to treat the patient, the care team must know of and suspect this disease. As such, all and any cases should be reported in order to provoke debate on the subject, accumulate knowledge, and benefit affected patients. This report demonstrates that when such injuries are of small diameter, it is possible to repair the aorta only using endovascular techniques, with conservative follow-up of the esophageal injury.

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Critical revision of the article: LKM, HHG
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Statistical analysis: N/A.
Overall responsibility: LKM

*All authors have read and approved of the final version of the article submitted to J Vasc Bras.