

Endoscopic diagnosis of primary aortoenteric fistula: case report

Diagnóstico endoscópico de fistula aortoentérica primária: relato de caso

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ABSTRACT: *Study type:* Case report. *Importance of the problem:* The incidence of the primary aortoenteric fistula is from 0.04% to 0.07% in the population. Its mortality is almost 100% if left untreated or when associated with a gastrointestinal hemorrhage. The diagnosis from an upper gastrointestinal endoscopy makes up only 25% of the cases, as computerized tomography is the gold standard for confirming this pathology. *Comments:* We reported a rare condition found in the third segment of the duodenum in this study. Intensive active bleeding; diagnosed by upper digestive endoscopy and aneurysm hemostasis, was performed with an endoscopic “clip” to control bleeding. The patient was stabilized from the hemodynamic point of view; however, he was referred to the Intensive Care Center (ICC) for better care due to the severity of the case. Computerized tomography was performed the following day and possible surgical intervention. However, the patient evolved hypertensive symptoms. With rebleeding (11 hours after diagnosis), clinical treatment was unsuccessful. Thus, it was impossible to perform a new endoscopic hemostasis, evolving into hypovolemic shock followed by cardiorespiratory arrest and death.

Keywords: Vascular fistula; Aortic aneurysm; Aorta, abdominal; Endoscopy, gastrointestinal; Fistula; Endoscopy.

RESUMO: *Modelo do estudo:* Relato de Caso. *Importância do problema:* A fistula aortoentérica primária tem incidência de 0,04% a 0,07% na população, tendo mortalidade de quase 100% se não tratada ou quando associada à hemorragia gastrointestinal. O diagnóstico realizado por endoscopia digestiva alta representa apenas 25% dos casos, sendo a tomografia computadorizada o padrão-ouro para confirmar essa afecção. *Comentários:* Nesse estudo, relatamos uma afecção rara encontrada na terceira porção do duodeno, com intenso sangramento ativo, sendo diagnosticada por endoscopia digestiva alta e realizada a hemostasia do aneurisma com “clip” endoscópico para controle do sangramento. Em seguida o paciente se estabilizou do ponto de vista hemodinâmico, porém, devido a gravidade do caso, foi encaminhado ao Centro de Terapia Intensiva (CTI) com o propósito de melhores cuidados e realização de Tomografia Computadorizada no dia seguinte, e possível intervenção cirúrgica. Entretanto, o paciente evoluiu com quadro hipertensivo, com ressangramento (11 horas após o diagnóstico), sem sucesso com o tratamento clínico e sem condição para realizar nova hemostasia endoscópica, evoluindo para choque hipovolêmico, seguido de parada cardiorrespiratória e óbito.

Descritores: Fístula vascular; Aneurisma aórtico; Aorta abdominal; Endoscopia gastrointestinal; Fístula; Endoscopia.

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INTRODUCTION

The aortoenteric fistula (AEF) is an abnormal communication between the aorta and the digestive viscera. It is rare and potentially fatal. This affliction is classified as primary and secondary. The secondary is a result of previous surgical intervention, usually by an aortic prosthesis¹. The primary aortoenteric fistula (PAEF) is a direct communication between an abdominal aorta aneurysm and the duodenum or another digestive viscera, with an incidence ranging from 0.04% to 0.07%. The mortality rate of PAEF is almost 100% if untreated or when associated with a gastrointestinal hemorrhage². The clinical presentation of PAEF is quite often insidious unless the doctors suspect gastrointestinal bleeding (80%) included episodes of hematemesis and melena (64%), abdominal pain (32%), and palpation of a pulsatile abdominal mass (25%). Other symptoms include dorsal pain, fever, sepsis, and shock³.

The upper gastrointestinal endoscopy (GIE) is the main exam performed for detecting gastrointestinal bleeding from an unknown source. However, the sensitivity of the PAEF is low (25%), and it must only be performed if the patient is hemodynamically stable. Many authors agree that this exam requires experienced operators due to the need to evaluate the full extension of the duodenum and avoid imprudent handling that may provoke the removal of a clot. The diagnostics of PAEF are generally obtained through a computerized tomography (CT) using intravenous contrast. Its specificity ranges from 85% to 100% and sensitivity from 50% to 94%, showing air bubbles on the wall of the aorta and the presence of the contrast in the duodenal lumen⁴.

The gold standard is conventional surgery for PAEF treatment in hemodynamically stable patients that can be performed using the following technique: aorto-aortic bypass with a Dacron Silver prosthesis or a polytetrafluoroethylene with an omental covering; replacing the aortic tract by a venous prosthesis; cryopreserved arterial homografts; replacing the in situ AEF by a prosthetic graft; a silver-covered prosthesis; and duodenal repair⁵. We recommend performing a CT, in this way, as a highly effective diagnostic modality for PAEF and the conventional surgical treatment (laparotomy) as the preferred treatment due to lower mortality rates⁶.

It is pathophysiologically believed that one of the reasons for the formation of PAEF is caused by the presence of an abdominal aorta aneurysm that, when quickly pulsating against the duodenal wall and necrosis occurs, thereby causing the formation of the fistula⁷.

The objective of this study is to present a rare affection regarding this, with high mortality, and difficult to diagnose through endoscopy, whereas endoscopic hemostasis was performed using a clip on the aneurysm during the procedure to stabilize the patient

hemodynamically and subsequently perform the defined treatment as recommended in the medical literature.

The study was approved by the "Comitê de Ética em Pesquisa" (Research Ethics Committee) (CEP) at the University of José Rosário Vellano/UNIFENAS, and certified by "Certificado de Apresentação de Apreciação Ética" (the Ethics Appreciation Presentation Certificate) (CAAE) number 23638619.9.0000.5143.

CASE REPORT

The patient was male, 50 years old, hypertensive, diabetic, a smoker, and cirrhotic due to Child-Pugh C alcoholism. He had undergone several hospitalizations due to esophageal varicose veins and hepatic encephalopathy. He was diagnosed with slight erosive enanthematous antral gastritis three years previously. There is not any record of previous surgeries. He was taking Metformin and Propranolol. He was admitted to *Hospital Universitário Alzira Velano*, in southern Minas Gerais State, due to complaints of vomiting during the three previous days, and there was no mucus, pus, or blood in the regurgitated food-related to pyrosis and a moderately diarrhetic condition. Melena and hematochezia were observed. The physical exam displayed: paleness 2+/4+, hydrated, anicteric, acyanotic, afebrile, preserved peripheral pressure, absence of edemas, and jugular turgency. Glasgow 15, isochoric, photoreagent pupils, and no motor deficit. Arterial Blood Pressure (BP): 100X60mmHg. The physical exam was unchanged, and only the liver was palpable at 1cm from the right costal border. The following laboratory exams were requested that showed the following: Red blood cells: 2.65 million/mm³; Hemoglobin 8.70 g/dL; Hematocrits: 24.70%; Platelets: 65,000/mm³; Leucocytes: 9,900/mm³. Afterward, he was admitted to the emergency room for hemodynamic stabilization and followed by an Upper Digestive Endoscopy. At that time, he was administered red blood cell concentrate, two plasma concentrates, volemic expansion was performed with crystalloids, and vasoactive drugs were administered. The patient could only perform the EGD 9 hours after being admitted to the hospital due to hemodynamic instability. During the procedure, a severe pulsatile bulging was noticed in the third duodenal segment with a spraying bleeding orifice similar to an aneurysm, thereby confirming the diagnosis of an aortoenteric fistula. At that moment, hemostasis of the aneurysm was performed using two endoclips to stop the bleeding. After the procedure, having stopped the bleeding momentarily, the patient was transferred to the Intensive Therapy Unit (ICU) for greater care and monitoring. The patient needed to be stabilized again, and red blood cell concentrates, plasma, and vasoactive drugs had to be administered again. Around three hours after the EGD, new laboratory exams showed: Hemoglobin: 3.8g/dL; Hematocrit: 11.1%; Platelets: 60,000/mm³; Prothrombin

Time: 29.6 seconds; Prothrombin Activity: 38.8%; INR: 2.48; Activated Partial Thromboplastin Time: 40.6 seconds. After 11 hours of monitoring in the ICU, he developed hemodynamic instability again, displaying enterorrhagia and following cardiopulmonary arrest (CPA). It was necessary to insert an endotracheal tube for mechanical ventilation. The patient underwent four CPAs, and in the last one, after ten resuscitation cycles, he maintained in an asystole condition until death.

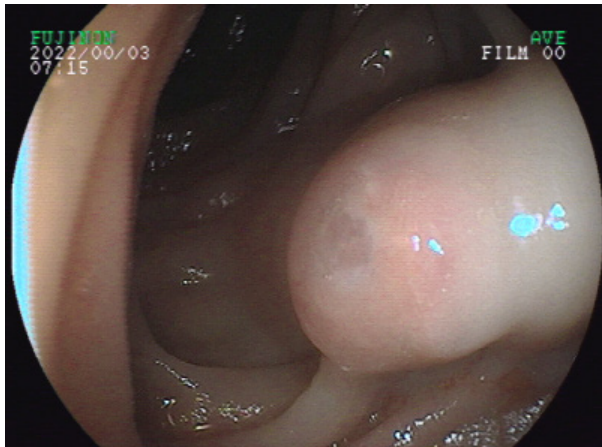


Figure 1: Aortoenteric fistula in the 3rd segment of the duodenum

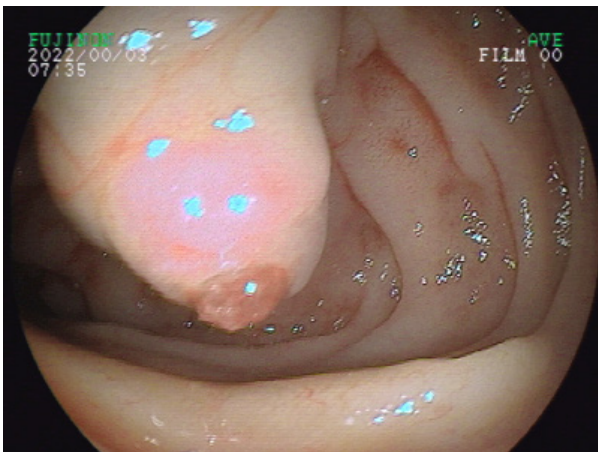


Figure 2: Aortoenteric fistula with a sentry clot

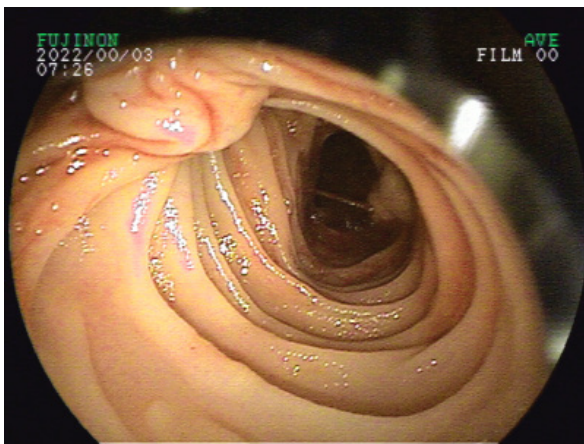


Figura 3: Fistulized aneurysm with active sprayed bleeding (as the arrow shows)

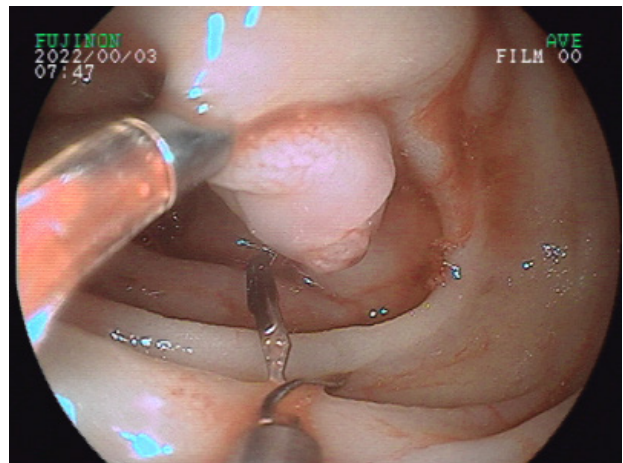


Figure 4: Clipped fistulized aneurysm

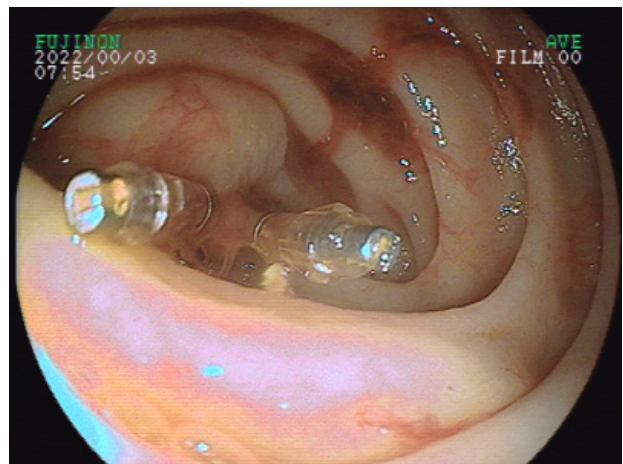


Figure 5: After clipping the fistulized aneurysm with two endoclips

DISCUSSION

The diagnosis of a PAEF is quite often delayed due to its rarity. Whereas 94% of the patients suffer from gastrointestinal bleeding, the classical triad of a palpable abdominal mass, abdominal pain, and gastrointestinal bleeding are present in 11% of the individuals⁴. It is important to emphasize our patient did not display abdominal pain when performing the palpitation during the physical exam. However, he only complained of pyrosis. He also was vomiting (without mucus, pus, or blood), melena, and enterorrhagia. Regarding this, the main diagnostic hypothesis would be a peptic ulcerous disease or rupture of esophageal varicose veins, since in his last EGD performed three years previously, it displayed slight erosive enanthematous antral gastritis and the presence of grade II esophageal varicose veins, treated with elastic ligatures. An EGD was requested again to search for the cause of the gastrointestinal bleeding based on his leading suspected clinical causes.

An emergency laparotomy is recommended in patients who suffer from active gastrointestinal bleeding after concluding the diagnosis. However, the patient was found hemodynamically unstable. Therefore the surgical procedure was postponed, as in these cases, there is a high rate of mortality⁸. The gold-standard treatment for PAEF is conventional surgery, performing an anatomical in situ repair with an aortic graft or extra-anatomical graft in cases of infection². However, in unstable patients, without signs of sepsis and severe infection, the endovascular treatment with a bifurcated aortic endoprosthesis can be possible as an initial alternative for controlling bleeding and achieving acceptable results medium and short-term in secondary AEF⁸.

Even if there is no pathognomonic sign for the PAEF diagnosis, there is a defined occurrence as an Arterial Hemorrhage consisting of an episode of gastrointestinal bleeding that generally is self-constrained by forming a blood clot in the aortoduodenal fistula. However, after infusion therapy, increased arterial pressure, or endoscopic procedures, the blood clot can be removed and cause severe bleeding, with the tendency to develop into hemodynamic instability³. The patient in this reported case had undergone a volemic expansion with crystalloids as well as administering red blood concentrates, plasma, and vasoactive drugs, before and after the EGD for stabilizing him hemodynamically, which unfortunately may have contributed to causing, contrary to what was expected, a worsening of his condition by rebleeding.

Participation of the authors: *Vieira JPG*: Performed the bibliographic review on the researched topic and participated in writing the case report and discussion. *Silva IC*: Participated in preparing the clinical case by reviewing the patient's medical history. *Brandão JAA*: Participated in adapting the case report to the Medical Journal standards and participated in writing the discussion. *Andrade BBP*: Participated in writing the case report conclusion and correcting the article text, according to the evaluators' counsel from the Medical Journal. *Andrade LC*: Participated in preparing the initial project, article revision, coordination, and study guidance. *Nascimento AF*: Providing project guidance and submitted it to "Plataforma Brasil" (Brazilian Platform) and the Ethics Committee of the "Universidade José do Rosário Vellano" (UNIFENAS), Alfenas, MG.

There are no conflicts of interest.

REFERENCES

1. Ishimine T, Tengan T, Yasumoto H, Nakasu A, Mototake H, Miura Y, Kawasaki K, Kato T. Primary aortoduodenal fistula: a case report and review of literature. *Int J Surg Case Rep*. 2018;50:80-3. doi: 10.1016/j.ijscr.2018.07.019.
2. Varghese M, Jorgensen GT, Aune C, Bergan R, Norderval S, Moland J. Primary aortoduodenal fistula: a case report and a review of the literature. *Ann Vasc Surg*. 2016;34:271. doi: 10.1016/j.avsg.2015.12.013.
3. Varetto G, Gibello L, Trevisan A, Castagno C, Garneri P, Rispoli P. Primary aortoenteric fistula of a saccular aneurysm: case study and literature review. *Korean Circ J*. 2015;45(4):337-9. doi: 10.4070/kcj.2015.45.4.337.
4. Ho S, Liu B, Loya R, Koury I. Primary aortoenteric fistula: a rare case of a massive gastrointestinal bleed. *Cureus*. 2016;8:e766. doi: 10.7759/cureus.766.
5. Keunen B, Houthoofd S, Daenens K, Hendriks J, Fourneau I. A case of primary aortoenteric fistula: review of therapeutic challenges. *Ann Vasc Surg*. 2016;33:230.e5-230.e13. doi: 10.1016/j.avsg.2015.11.033.
6. Oizumi PSL, Silva LMA, Pereira JM, Oizumi RF, Filho SAF. Fístula aórtico-entérica primária relacionada a aneurisma de aorta abdominal. *Biosci J*. 2013;29(6). Available from: <http://www.seer.ufu.br/index.php/biosciencejournal/article/view/18275>.
7. Aguilar EM, Alonso SF, Fariña ES, Alonso LF, Pascual MA, Vallespuga RC. Estado actual sobre el diagnóstico y tratamiento de fistulas aortoentéricas. *Angiología*. 2014;66:246-53. doi: 10.1016/j.angio.2014.03.009.
8. Rossi FH, Izukawa NM, Prakashan AK. A case report of an endovascular treatment of secondary aortoenteric fistula. *J Vasc Bras*. 2012;11:236-9. doi: 10.1590/S1677-54492012000300012.

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