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International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Primary aortogastric fistula following Nissen fundoplication: A case report

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ARTICLE INFO

Article history:

Received 12 October 2020
 Received in revised form
 29 November 2020
 Accepted 30 November 2020
 Available online 3 December 2020

Keywords:

Nissen fundoplication
 Aortogastric fistula
 Gastrointestinal bleeding
 Endovascular
 Case report

ABSTRACT

INTRODUCTION: Aortoenteric fistula (AEF) is a rare condition and consists of an abnormal communication between the aorta and the gastrointestinal (GI) tract. The duodenum is the most common location. Fistulas involving the stomach are very uncommon and account for only 2% of the cases. AEF typically results in rapid and fatal exsanguination as diagnosis is frequently missed or made too late (Bixby et al., 2018; Koungias et al., 2003; Lookman, 1959; Genc et al., 2000; Ong et al., 2019; Li et al., 2020).

PRESENTATION OF CASE: A 59 years old female with a history of Nissen fundoplication presented with lower gastrointestinal bleeding. Esophagogastroduodenoscopy (EGD) showed a large blood clot in the gastric fundus with no visible source of active bleeding. A mesenteric angiogram, performed for persistent gastro-intestinal bleeding and following two episodes of cardiac arrest, showed no evidence of active bleeding. The left gastric artery was prophylactically embolized. Persistent hemorrhage prompted an exploratory laparotomy followed by a left thoracotomy and confirmed the diagnosis of an aortogastric fistula (AGF). The patient expired intra-operatively.

DISCUSSION: AGF is a very rare but often fatal condition (Busuttill and Goldstone, 2001). Computerized tomography angiography (CTA) can be a key to the diagnosis (Raman et al., 2012). EGD and catheter angiography have low sensitivity (Kuhara et al., 2015; Manduch et al., 2008). Definitive diagnosis is usually made during surgical exploration or autopsy (Wasvary et al., 1997). While open surgical repair is considered the gold standard therapy, endovascular therapy is becoming the preferred initial treatment option (Bixby et al., 2018).

CONCLUSION: AGF should be considered in the differential diagnosis of GI bleeding, especially in patients with massive hemorrhage where EGD and mesenteric angiography are not diagnostic.

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1. Introduction

Aortoenteric fistula (AEF) is an abnormal communication between the aorta and the gastrointestinal (GI) tract and may result from aortic or GI pathology [1–6]. It is a rare condition with an incidence of 0.04–0.07% [7]. Most AEFs are secondary to procedures involving the aorta, i.e. as a complication of aortic repair (surgical or endovascular) [1–9]. Primary AEF is much less common and refers to a communication between the native aorta and the GI tract [1]. In both primary and secondary AEF, the duodenum is the most common location, accounting for 63–81% of the cases. Fistulas involving the stomach are very uncommon, with aortogastric fistulas (AGF) accounting for only 2% of AEF cases [2,6]. It typically results in a rapid and fatal exsanguination. Delayed intervention is associated with a mortality rate of nearly 100%. We describe a case of a primary AGF that occurred 4 years after Nissen fundoplication.

The purpose of this article is to raise awareness of this rare but rapidly fatal condition and the need to include it in the differential diagnosis of massive GI bleeding [1,2,4,10]. Treatment strategies are also postulated. This work has been reported in line with the SCARE criteria [28].

2. Presentation of case

A 59 years-old female with a history of alcohol abuse, presented to our community hospital emergency department with generalized weakness, recurrent syncope, and melena. Her past surgical history includes a laparoscopic Nissen fundoplication performed four years prior with an uneventful perioperative course. She was afebrile, tachycardic (heart rate at 120 beats per minute), hypotensive (blood pressure 77/43 mm Hg) and her physical exam revealed tremors and rigors. Pertinent abnormal laboratory values included a low hemoglobin (6.7 g/dL), elevated lactate (6.3 mmol/L), and elevated anion gap (22 mEq/L). Her creatinine and bilirubin levels were normal. She was admitted to the intensive care unit (ICU) with a diagnosis of GI bleeding and alcohol withdrawal syndrome and

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was managed medically with fluid resuscitation and blood transfusion. An esophagogastroduodenoscopy (EGD) demonstrated a large blood clot in the gastric fundus, with no evidence of esophageal or gastric varices. No source of active hemorrhage was identified. Upon return to the ICU the patient had a large black bowel movement but remained hemodynamically stable. Two hours later she developed a large bright red bowel movement, became unresponsive and experienced pulseless electrical activity (PEA). She was successfully resuscitated after two minutes but remained hemodynamically unstable requiring ongoing intravenous vasopressor support. A second code occurred during preparation for angiography which lasted five minutes. Three vessel mesenteric and abdominal aortic angiographies (anteroposterior views) showed no evidence of active bleeding. The left gastric artery was prophylactically embolized using Gelfoam slurry and coils. Clinical evidence of hemorrhage persisted however and prompted a repeat EGD in the interventional radiology suite. This demonstrated fresh blood in the gastric lumen but once again no clearly identifiable source of bleeding.

Exploratory laparotomy was then undertaken which revealed a large amount of free intraperitoneal blood as well as a tense, distended stomach. An anterior gastrotomy was created upon which massive hemorrhage was encountered originating from the proximal posterior stomach near the level of the diaphragm. The patient lost her pulse immediately upon entering the stomach but was recovered after achieving a measure of control with manual pressure applied directly over the fistula. Extensive scarring and adhesions at the aortic hiatus prevented supra-celiac aortic clamping. Similar chronic findings were noted within the lesser sac near the aorta. A left thoracotomy was performed in an attempt to gain proximal control of the aorta. Dense fibrosis and scarring between the medial aspect of the left lung and the aorta was encountered. During the thoracic dissection, PEA occurred once again and resuscitation efforts were unsuccessful, at which time the patient expired. The area of hemorrhage was re-inspected and the diagnosis of AGF confirmed. The fistula was substantial measuring 10 mm in diameter. No evidence of active infection or tumor was identified, and the aorta was soft and pliable and free of atherosclerotic or aneurysmal disease. Biopsies were obtained and showed no malignancy or specific underlying etiology.

3. Discussion

AGF is a very rare, often fatal condition and requires rapid recognition and treatment [7]. The diagnosis is frequently missed or made too late for surgical therapy to be effective, and mortality remains high despite intervention [1,2,4,8]. In many cases, the initial hemorrhage will cease spontaneously, potentially allowing sufficient time for diagnosis and surgical intervention. This “herald bleed” has been attributed to a spasm of the intestinal musculature that closes the fistula temporarily [1,2,4,9]. The interval between initial hemorrhage and death from exsanguination was found to vary from several hours to 8 months [11].

AGF following Nissen fundoplication has been reported [1–3,13–16], with presentation up to 5 years following surgery. Factors promoting gastric ulceration into the aorta after Nissen fundoplication include local ischemia from ligation of short gastric arteries, local suture irritation, direct surgical trauma, delayed gastric emptying due to vagal nerve injury, hypergastrinemia, and perioperative subclinical contained perforation. These changes may result in gastric ischemia and ulceration which, given the proximity of the aorta to the stomach, can result in a fistula [1,12]. Post-surgical inflammation also results in adhesions surrounding the fundal segment of the fundoplication wrap and fixation of the stomach close to the aortic wall [1,2].

A triad of massive upper GI bleeding in the absence of an obvious source, pulsatile abdominal mass, and recurrent cardiovascular collapse disproportionate to the degree of hypovolemia was described by Lewis and Allan [2,12]. This complete triad is present only in 11% of patients. A history of gastro-esophageal surgery or distortion of the GI anatomy should however raise the index of suspicion [12].

Computerized Tomography Angiography (CTA) of the abdomen with intravenous contrast is considered the most effective examination in the evaluation of AEF. It has a sensitivity of 40–90% and a specificity of 33–100% [21]. The acquisition protocol consists of a phase without contrast injection, an arterial phase and a delayed portal venous phase [21]. Given the disease rarity, there are very few systematic descriptions of key CT Findings [22] and there are no large series to validate their accuracies [23]. Direct extravasation of contrast from the aorta into the bowel lumen is the most specific CT finding, however, it is rarely seen and is influenced by the timing of examination in relation to the bleeding [6]. The leakage of enteric contrast directly into the periaortic space is also a very specific finding, but extremely rare [22]. Diagnosis of AEF depends on a number of non-specific CT Findings, the most important of which is ectopic gas within or directly adjacent to the aortic lumen. Other findings include effacement of the periaortic or perigraft fat plane and the fat plane between the aorta and bowel, perigraft soft tissue thickening, perigraft fluid, perigraft hematoma, focal bowel wall thickening adjacent to the graft, intramural hematoma, disruption of the aortic wrap if present, pseudoaneurysm or aneurysm bulge, increased soft tissue between the graft and aortic wrap, and dystrophic vascular graft calcifications. These non-specific findings are also found in myriad other conditions including perigraft infection, immediate post-operative period, aortitis, mycotic aneurysms, and perianeurysmal fibrosis [17,23]. A CTA was not performed following the mesenteric angiogram due to the patient’s hemodynamic instability. We believe that an opportunity to have completed this scan was present following the first EGD while the patient was hemodynamically stable.

EGD has a low sensitivity (20%) due to difficulty visualizing the culprit lesion in the setting of massive bleeding [8]. The fistula itself is rarely seen by endoscopy but the diagnosis may be suggested by the presence of blood and an extrinsic pulsatile mass. It is not uncommon for an incidental ulcer to be mistaken as the primary cause of bleeding. Catheter angiography has a low sensitivity (26%) and is influenced by the timing of contrast injection relative to the active hemorrhage [12]. Angiography is usually performed when the patient is hemodynamically stable; an obstructing thrombus plug within the fistula may prevent visualization of active contrast extravasation however [24]. In our case, evaluation of the supra-celiac aorta with angiography was not performed. It is possible that an aortogram performed in the lateral projection, with the diagnostic catheter placed in the distal thoracic aorta, could have identified the AGF.

Definitive diagnosis is often made during operative exploration or autopsy [15]. Surgical repair has included direct repair of the stomach and aorta, patching of the aortic segment, resection with in-situ synthetic aortic graft placement, and extra-anatomic reconstruction with aortic ligation. Given the small number of patients presenting with AEFs, different fistula locations, surgical histories and underlying etiologies are often included in the discussion. Our review yielded 5 patients with a presentation similar to ours: a history of Nissen fundoplication, no other noted foregut surgery or malignancy, no significant primary aortic pathology, and no identified foreign body or trauma [12–16]. None of these patients survived the perioperative period. As has been suggested in other reviews, consideration should be given to achieving vascular control via thoracotomy prior to entering the abdomen, or prior to opening the stomach if noted to be distended. The stomach may afford some element of tamponade at this juncture [18–20].

Fistula repair can be performed with endoscopic, endovascular, or open surgical approach. While open surgical repair has been considered the gold standard therapy, advances in endovascular techniques might make stenting, with or without repair of the fistula, the preferred initial treatment option [1]. Endovascular repair of the aorta combined with endoscopic injection of a fibrin sealant in the fistula tract has been described [25]. Transcatheter arterial embolization of the fistula using n-butyl cyanoacrylate mixed with iodized oil followed by aortic endograft placement was also reported [8]. In a systematic review including 41 patients with AEF who underwent endovascular repair, persistent/recurrent/new infection or recurrent hemorrhage occurred in 44% of patients after a mean follow up of 13 months [26]. In a multicenter study comparing open surgical repair and endovascular repair of AEF, endovascular repair had lower in-hospital mortality compared to open surgery (0% and 35% respectively) and lower morbidity (25% and 77% respectively). In the long term, however, the endovascular group had higher rates of recurrence, sepsis, reoperation, and AEF-related death. The early survival advantage in the endovascular group relative to the open surgical group was lost by the second year of follow up [27]. Endovascular repair leaves a gastric defect untreated [7,9], which exposes the graft to a contaminated environment leading to an increased risk of endograft infection and sepsis as well as recurrent bleeding. Additional radical aortic surgery should be performed once the patient is stable for increased long-term survival [7].

4. Conclusion

Aortogastric fistula is a rare but life-threatening complication following Nissen fundoplication. It poses a diagnostic challenge however given the low sensitivity of the available diagnostic examinations. Diagnosis requires a high index of suspicion. Early diagnosis and treatment are crucial as delay in treatment carries a very high mortality. We suggest the following recommendations:

1. Early use of CT scanning with an established GI bleed protocol might be of benefit in making the diagnosis.
2. In patients with massive GI bleeding and non-diagnostic EGD and mesenteric angiogram, a distal thoracic angiography to include a lateral projection should be performed.
3. Endovascular intervention should be considered as an initial therapy, to include stenting of the affected area with or without embolization of the fistula.
4. If open surgery is performed consider achieving proximal vascular control via thoracotomy prior to opening the abdomen or prior to gastrotomy if the stomach is distended.
5. Surgical aortic reconstruction should be considered following initial endovascular intervention once the patient is clinically stable.

Declaration of Competing Interest

The authors report no declarations of interest.

Funding

We did not receive funding from any source.

Ethical approval

Institutional Review Board (IRB) was waived. As per our institution policy, IRB review/approval is not required for case reports or series of less than 3 cases, provided that no patient identifying information is included.

Consent

Written informed consent was obtained from the patient's next of kin for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Abdel Aziz Jaffan, MD: Interventional Radiologist, performed the mesenteric angiogram and embolization procedure; first author; wrote, revised and edited the paper.

James Larson, MD, FACS: Surgeon, performed the open surgery; wrote, revised and edited the paper.

Sunil Kapur, MD: Gastroenterologist; performed the endoscopies; wrote, revised and edited the paper.

Registration of research studies

Not applicable.

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Provenance and peer review

Not commissioned, externally peer-reviewed.

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