

## Upper digestive hemorrhage secondary to major duodenal papilla Dieulafoy's lesion:

### Case report

Hemorragia digestiva alta secundária à lesão de Dieulafoy na papila duodenal maior: Relato de caso

Hemorragia digestiva alta secundária a lesión de Dieulafoy en la papila duodenal mayor: Reporte de caso

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### Abstract

**Introduction:** Dieulafoy's lesion (DL) is occasioned by a tortuous, persistent and large caliber artery that emerges the mucosa from the submucosa of an organ, eventually triggering gastrointestinal bleeding in the presence of eroding factors of the mucosa and arterial wall. The presence of DL has been described in many anatomic topographies and although it predominates in the upper digestive tract, the presence of this lesion exactly in the major duodenal papilla is a rare event. **Objective:** to report a case of upper gastrointestinal bleeding secondary to a major duodenal papilla DL. **Case report:** a 72 year-old female, admitted to hospital care with a clinical history of two months continuous, painless melena, multiple previous blood transfusions and symptomatic anemia. She was referred by another health service with the diagnostic hypothesis of hemobilia, suggested by two previous esophagogastroduodenoscopies. Her abdominal ultrasound and arteriography were normal. A third esophagogastroduodenoscopy evidenced active bleeding in the duodenal major papilla, and after a carefully analysis a papillar DL was diagnosed. It was treated by endoscopy with adrenaline 1:10000 injection and thermocoagulation. Following this procedure she evolved with severe acute pancreatitis due to papillitis and need of intensive care unit admission. No rebleeding was detected and hospitalar discharge occurred twenty days after hospitalization. **Conclusion:** The localization of a DL at the major papilla is a rare event and acute pancreatitis is a complication related to its endoscopic treatment.

**Keywords:** Gastrointestinal hemorrhage; Endoscopy; Ampulla of Vater.

### Resumo

**Introdução:** A lesão de Dieulafoy (DL) é ocasionada pela existência de uma artéria tortuosa, persistente e de grande calibre que emerge da mucosa para a submucosa de um órgão, e pode desencadear hemorragias gastrointestinais na presença de fatores erosivos da mucosa e da parede arterial. A DL já foi descrita em diversas topografias anatômicas e embora predomine no trato digestivo superior sua presença na papila duodenal maior é considerada um evento raro. **Objetivo:** relatar um caso de hemorragia digestiva alta secundária a uma DL localizada na papila duodenal maior. **Relato de caso:** uma mulher de 72 anos foi internada com história clínica de dois meses contínuos de melena indolor, múltiplas transfusões sanguíneas e anemia sintomática. A paciente foi encaminhada de outro serviço de saúde para o hospital de referência com hipótese diagnóstica de hemobilia, sugerida por duas esofagogastroduodenoscopias prévias. Sua ultrassonografia abdominal e arteriografia foram normais. Uma terceira endoscopia digestiva alta evidenciou sangramento ativo na papila duodenal maior e após uma análise cuidadosa evidenciou-se uma DL na papila duodenal

maior que foi tratada endoscópicamente através de injeção de adrenalina 1:10000 e termocoagulação. Após esse procedimento a paciente evoluiu com pancreatite aguda grave secundária a papilite e necessidade de cuidados em unidade de terapia intensiva. Não foram detectados novos sangramentos e a paciente recebeu alta hospitalar vinte dias após a hospitalização. Conclusão: a presença de uma DL na papila duodenal maior é um evento raro e a pancreatite aguda pode ser uma das complicações relacionadas ao seu tratamento endoscópico.

**Palavras-chave:** Hemorragia gastrointestinal; Endoscopia; Papila duodenal maior.

### Resumen

Introducción: La lesión de Dieulafoy (DL) es ocasionada por una arteria tortuosa, de gran calibre y persistente que emerge de la mucosa hasta la submucosa de un órgano, desencadenando finalmente una hemorragia gastrointestinal en presencia de factores erosivos de la mucosa y de la pared arterial. La presencia de DL se ha descrito en muchas topografías anatómicas y aunque predomina en el tracto digestivo superior, la presencia de esta lesión exactamente en la papila duodenal mayor es un evento raro. Reporte de Caso: una mujer de 72 años, ingresada en atención hospitalaria con historia clínica de dos meses continuos de melena indolora, múltiples transfusiones sanguíneas previas y anemia sintomática. Fue remitida por otro servicio de salud con la hipótesis diagnóstica de hemobilia, sugerida por dos esofagogastroduodenoscopias previas. Su ecografía y arteriografía abdominal fueron normales. Una tercera esofagogastroduodenoscopia evidenció sangrado activo en la papila duodenal mayor, y después de un análisis cuidadoso se diagnosticó DL papilar. Se trató por endoscopia con inyección de adrenalina 1:10000 y termocoagulación. Después de este procedimiento evolucionó con pancreatitis aguda grave por papilitis y necesidad de ingreso en unidad de cuidados intensivos. No se detectaron nuevas hemorragias y el alta hospitalaria se produjo veinte días después de la hospitalización. Conclusión: la localización de una DL en la papila duodenal mayor es un evento raro y la pancreatitis aguda es una complicación relacionada con su tratamiento endoscópico.

**Palabras clave:** Hemorragia gastrointestinal; Endoscopia; Papila duodenal mayor.

## 1. Introduction

Dieulafoy's lesion (DL) is occasioned by a tortuous, large caliber and persistent artery that emerges the mucosa from the submucosa, eventually triggering sudden massive gastrointestinal bleeding in the presence of eroding factors of the mucosa and arterial wall. Although it represents a diagnostic challenge, the presence of DL has been described in many anatomic topographies such as esophagus, stomach, duodenum, jejunum, ileum, colon, rectum and bronchus (Lai et al., 2020; Oladunjoye et al., 2020; Wang et al., 2017; Malliaras et al., 2016; Nguyen et al., 2015; Baxter & Aly 2010). DL predominates in the upper digestive tract, especially in stomach and duodenum, but the presence of this lesion exactly in the major duodenal papilla is a rare event and is associated with a high risk of complications associated with the therapeutic resources. This case report refers of a patient with major duodenal papilla DL and the impacts of this event on the diagnoses, clinical management and patient recovery.

## 2. Methodology

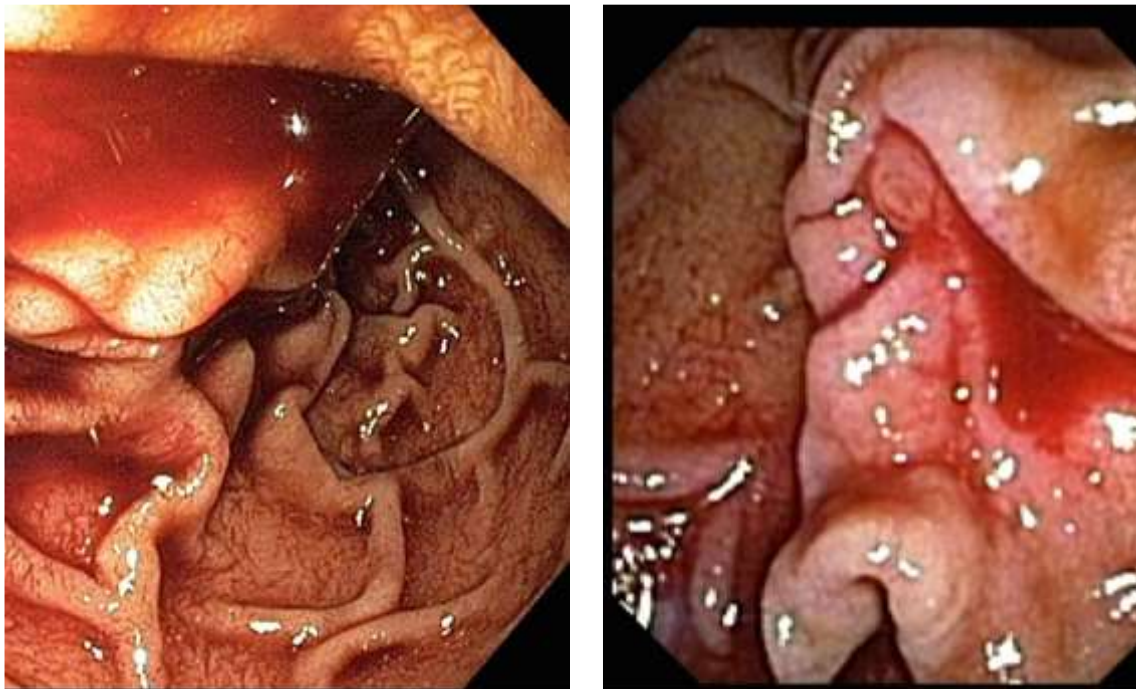
This is a descriptive case study elaborated by an interview with the patient and review of the medical record (Pereira et al., 2018). This study was completed in accordance with the Helsinki Declaration. Written informed consent was obtained from the patient for publication of this case and any accompanying images.

## 3. Case Report

A 72-year-old woman, with previous pathological history of diabetes, systemic arterial hypertension, dyslipidemia, hyperuricemia, chronic coronary artery disease, chronic degenerative changes in the lumbar spine, obesity and in previous treatment of deep venous thrombosis of the right lower limb for the last six months was referred by a less complex hospital service to the tertiary hospital "Santa Casa de Misericórdia de Passos" (SCMP) with symptomatic anemia and complaint of two months of continuous painless melena. During the past six months she was taking a daily dose of rivaroxaban (20mg) and related eventual use of self-administrated non-steroidal anti-inflammatory drugs, such as sodium diclofenac, which was promptly suspended. She did not have any past of gastrointestinal bleeding, smoking, use of alcohol, chronic liver or peptic ulcer diseases

or familiar history of gastrointestinal malignancy. She was admitted the origin hospital 14 days before, with initial hemoglobin of 6,4g/dL and in need of multiple blood transfusions though hospitalization to maintain clinical stability. During this period her abdominal ultrasound was normal and two esophagogastroduodenoscopy suggested that she had hemobilia. The patient was transferred to SCMP in order to perform arteriography and embolization of the bleeding site and subsequently investigate the cause of hemobilia. At SCMP she presented hemodynamically stable, her pulse rate was 80 beats/min, respiratory rate 20 breaths/min, blood pressure 120/70mmHg. She was pale and asthenic. Her laboratory values were: hemoglobin 9,2 g/dL (normal range: 12 to 16 g/dL); hematocrit 26,8% (normal range: 35 to 47%); white blood cell count 8500/mm<sup>3</sup> (normal range: 3600 to 11000/mm<sup>3</sup>); platelets 278000/mm<sup>3</sup> (normal range: 140000 to 450000/mm<sup>3</sup>); urea 55mg/dL (normal range: 15 to 39 mg/dL); creatinine 0,8mg/dL (normal range: 0,6-1,0mg/dL); seric amylase 31U/L (normal range: 25 to 115U/L); seric lipase 167U/L (normal range: 73 to 393 U/L);, prothrombin activity 86% (normal range: 70 to 100%); international normalized ratio 1,07; thromboplastin partially activated time relation 0,9 (normal range: 0,8 to 1,2) , Reactive protein C 13mg/dL (normal range: 0 to 3 mg/dL). Blood culture, urine culture and routine urinalysis did not present changes. Full dose endovenous pantoprazole was prescribed. Arteriography was performed and no bleeding was detected by the method, although new melena events and a decrease in hemoglobin levels was observed. A new esophagogastroduodenoscopy was performed and a carefully analysis evidenced a small vessel located at duodenal major papilla's entrance as the resource of the streaming hemorrhage (Figure 1), compatible with the diagnosis of a Dieulafoy's Lesion. The surrounding mucosa was normal.

**Figure 1.** Endoscopic views of an active bleeding coming from a Dieulafoy's lesion located at the major duodenal papilla.



Source: Authors.

Even with a high risk of papillitis, endoscopy therapy associating epinephrine 1:10000 injection and thermocoagulation was performed. In the subsequent days following this procedure, the patient evolved with pain in the upper abdomen, nausea and low diet acceptance. She was diagnosed with acute pancreatitis with systemic repercussions, like acute renal injury AKIN stage 2 and need of supplemental oxygen 2l/min. Laboratory data three days after endoscopic treatment : hemoglobin 7,4 g/dL (normal range: 12 to 16 g/dL); hematocrit 21,2% (normal range: 35 to 47%); white blood cell count 5600/mm<sup>3</sup> (normal range: 3600 to

11000/mm<sup>3</sup>); platelets 221000/mm<sup>3</sup> (normal range: 140000 to 450000/mm<sup>3</sup>); urea 102 mg/dL (normal range: 15 to 39 mg/dL); creatinine 3,6mg/dL (normal range: 0,6-1,0mg/dL); seric amylase 124U/L (normal range: 25 to 115U/L); seric lipase 677U/L (normal range: 73 to 393 U/L); Reactive protein C 441mg/dL (normal range: 0 to 3 mg/dL). After 48 hours she was classified with Atlanta's severe acute pancreatitis and transferred to intensive care unit. A computed tomography was performed to exclude perforative complications and a pancreatitis Balthazar's severity index grade B was identified. She was submitted to clinical management and also was diagnosed and treated for an urinary tract infection by *Candida non albicans*. New blood transfusion and parenteral iron administration was need to achieve clinical stabilization. She had no new bleedings during hospitalization, reversed clinical complications and received hospital discharge twenty days after admission. Control esophagogastroduodenoscopy was performed before discharge with normal results and the patient received orientation for outpatient follow-up.

#### 4. Discussion

In this case report the patient had some classical factors associated to DL such as age, cardiovascular disease, hyperlipidemia, systemic arterial hypertension, diabetes mellitus, use of nonsteroidal anti-inflammatory drugs and anticoagulants (Inayat et al., 2018; Nguyen & Jackson 2015).

The presence of a DL located at duodenal major papilla simulated hemobilia on a first impression, but although it was mentioned as one of the initial hypotheses, the patient did not have any traumatic history or neoplastic lesion, no compatible image exam finding and that possibility was completely ruled out after arteriography study. Only on a third esophagogastroduodenoscopy DL was identified. The active bleeding during procedure was an important key to elucidate the lesion's nature. As a matter of fact, to diagnose a Dieulafoy's Lesion may be challenging even to an experienced endoscopist (Stojakov et al., 2007; Nojkov & Cappell, 2015). Previous studies demonstrated that even 49% of DL were identified on initial endoscopy and 33% of the patients required a repeated endoscopy (Reilly & Al-Kawas, 1991). In some cases the combination between endoscopic and radiological methods can be considered to achieve diagnoses (Inayat et al., 2018).

Technical literature records that advances in the endoscopy field have allowed an increased rate of detection and therapeutic efficacy, low recurrence and excellent long-term prognosis. Only patients under antiplatelet therapy are more likely to have an early relapse (Massinha et al., 2019). The most common endoscopic therapies of DL are injection with epinephrine and thermocoagulation (monopolar, bipolar or heater probe therapy). Sclerosant agents increase the risk of perforation and cyanoacrylate glue or fibrin glue can result in damage to the endoscope. Mechanical methods, such as hemoclips and endoscopic band ligation, have a theoretical benefit to cause less tissue damage than thermocoagulation, but they can be difficult to administer in some anatomic areas, such as duodenal major papilla. We should consider that epinephrine injection can work as an alternative to reduce active bleeding, allowing a more definitive method such thermocoagulation or mechanical means. Angiography may be used to embolise DL if there is active bleeding during the realization of the procedure, however the risk of organ ischemia is higher with this method. Surgical treatment is reserved only to a small portions cases refractory to endoscopic and angiographic methods (Baxter & Aly, 2010; Nguyen et al., 2015; Lim et al., 2009).

In this specific case, the use of combined therapy (epinephrine plus thermocoagulation) allowed success hemostasis of duodenal major papilla DL bleeding, but was followed by a transitory edema and initiated a severe acute pancreatitis episode. Although rare, duodenal DL lesions was previous described (Inayat et al., 2018; Ibrarullah and Waghlikkar, 2003; Murali et al., 2017) including one bleeding Dieulafoy's-Like lesion resembling the duodenal papilla (Bilal et al., 2015) and a DL inside a duodenal diverticulum (He et al., 2020). A Periampullary lesion has been previous described as cause of an obscure gastrointestinal bleeding and needed surgery treatment after endoscopic and arteriographic fail (Rana et al., 2010). Han and coworkers (2021) had recently described a Dieulafoy Lesion of the Major Papilla, which was clearly separated from the bile duct

and pancreatic duct orifices [13]. In our case the Dieulafoy's lesion was located exactly at the entrance major duodenal papilla and its treatment triggered transitory disfunction of that organ, causing severe acute pancreatitis.

## 5. Conclusion

DL lesion must be remembered as a cause of gastrointestinal bleeding and its diagnosis may be challenging. The localization of a DL at the major papilla is a really rare event and medical team must predict acute pancreatitis as a complication related to its endoscopic treatment.

Future observations may focus on the systematization of clinical protocols for the management of hemorrhages caused by Dieulafoy's lesion, seeking to emphasize the specificities of each anatomical topography in which it can be found and its impact on the patient's treatment.

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