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DOI: 10.17235/reed.2020.6899/2020
Link: PubMed (Epub ahead of print)

Please cite this article as:
Pérez Fernández Alejandro, Rubio Mateos José María, Sánchez Fernández María Josefa, Molina Muñoz José Manuel, Chacón Martínez Silvia, Rodrigo Agudo José Luís, Esteban Delgado Pilar, Pérez-Cuadrado Martínez Enrique, Pérez-Cuadrado Robles Enrique. Recurrent small-bowel bleeding from a Dieulafoy's lesion after combined endoscopic treatment. Rev Esp Enferm Dig 2020. doi: 10.17235/reed.2020.6899/2020.

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OR 6899 inglés

Recurrent small-bowel bleeding from a Dieulafoy's lesion after combined endoscopic treatment

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Received: 28/01/2020

Accepted: 11/02/2020

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ABSTRACT

Introduction: Dieulafoy's lesion of the small bowel is an uncommon cause of gastrointestinal (GI) bleeding that often recurs after endoscopic treatment.

Material and methods: we report an observational, descriptive, retrospective, single-center study in 15 patients with small bowel bleeding who were diagnosed with a Dieulafoy's lesion by capsule endoscopy or double-balloon enteroscopy.

Results and conclusions: all patients underwent combined endoscopic treatment. During a median follow-up of 33.5 months (range, 2-145), three of the 12 cases that stayed in follow-up (25 %) recurred, all within 48 hours after treatment. Two were successfully re-treated with a repeat endoscopic procedure.

Keywords: Gastrointestinal hemorrhage. Dieulafoy's lesion. Capsule endoscopy. Double-balloon enteroscopy. Treatment failure.

INTRODUCTION

Dieulafoy's lesion (DL) is a rare cause of small-bowel (SB) bleeding and the most common location is the stomach (1). It is endoscopically defined when one or more of the following characteristics is observed: arterial bleeding, blood vessel with/without bleeding or an adhered clot with a narrow point of attachment to the mucosa. All are associated with a small mucosal defect or surrounded by normal mucosa (1). Furthermore, it may be accompanied by an underlying condition such as diverticula (2). Diagnosis is challenging since bleeding is intermittent and endoscopic evidence is absent, except for bleeding. The recurrence risk is around 12.5-20 % and the ideal management of these lesions is poorly defined (3,4). The goal of the study was to assess the clinical characteristics of patients with DL in the SB and recurrent bleeding after combined treatment with double-balloon enteroscopy (DBE) in a reference center.

MATERIAL AND METHODS

Design

An observational, descriptive, single-center study was performed.

Patients

All patients with SB bleeding from a DL who were diagnosed by capsule endoscopy (CE) or DBE from December 2004 to December 2019, and underwent combined endoscopic treatment by DBE were included in the study. Those who received simple endoscopic treatment were excluded.

Variables

Diagnosis with DL was defined as jet bleeding without an underlying mucosal lesion. Follow-up ensued from therapeutic DBE to study completion (December 2019); to this end, electronic medical records were reviewed and patients were contacted by telephone. Recurrence was considered when overt GI bleeding occurred after DBE, or when a

hemoglobin decrease of at least 2 g/dl was found after covering transfusional needs with prior hemoglobin stabilization.

Procedures

1. Preanesthetic study. Peri-procedural management of anti-platelet drugs and/or anticoagulants, thromboembolic risk and assessment of the indication for orotracheal intubation.
2. CE. A Pillcam™ SB2 (Given Imaging Ltd., Yoqneam, Israel) or SB3 (Given Imaging Ltd., Mansfield, USA) capsule was used.
3. DBE. The EN-450p5, EN-450t5 or EN-580T (Fujifilm®, Japan) enteroscope was used. The patient fasted for the previous eight hours when the oral route (OR) was used. With regard to the anal route (AR), a standard preparation was used, similar to that of colonoscopy. Combined endoscopic treatment was performed first using a 1:10000 diluted adrenaline injection and/or low-flow (0.5 l/min, 40 W) argon plasma coagulation (APC) (Erbe Elektromedizin, Tübingen, Germany), which was followed by clips (Resolution™ Clip; Boston Scientific, Natick, Mass., USA). DL sites were tattooed with India ink.

Statistical analysis

A descriptive analysis was performed using the SPSS® Statistics v.19 software (IBM, SPSS, IL, USA). Qualitative variables were expressed as numbers and percentages, and quantitative non-normal variables as the median and range.

RESULTS

Patients

In total, 1,816 CEs and 470 DBEs were performed with an indication for SB bleeding and DL was identified as the cause in 14 (2.98 %) DBEs and three (0.16 %) CEs. Two patients were excluded as they only received endoscopic treatment with APC. Hence, 15 subjects were included, of which three were lost to follow-up. The median patient age was 71 years (range: 55-86) and the most common presentation was overt GI bleeding (Table 1). All patients included in the study required transfusions.

Diagnostic tests and therapeutics

The median number of diagnostic tests (radiography and endoscopy) performed for all 15 patients until their diagnosis and treatment by DBE was 4 (range, 2-6) (Table 2). Thirteen underwent CE (86.7 %) before DBE with the following findings: a diagnosis was reached in two (13.3 %), abundant fresh blood remnants were found in ten (Fig. 1) (76.92 %), other vascular lesions were seen in five (38.46 %) and the examination was normal in one case (6.7 %).

The median number of DBEs until diagnosis was 1 (range, 1-3) (Fig. 2) and ten patients only required one DBE procedure for diagnosis and treatment (66.7 %). The median DBE duration in the nine patients where the time was recorded was 60 minutes (range: 35-90). The OR was used for 13 patients (86.7 %) and the AR for two (13.3 %). Endoscopic treatment ensued with APC + adrenaline in nine subjects (60 %), APC + adrenaline + clip in four (26.7 %); APC + clip in one (6.7 %) and adrenaline + clip in one (6.7 %). No endoscopic treatment-derived complications were reported in our series.

Follow-up

During a median follow-up of 33.5 months (range, 2-145), three of 12 patients (25 %) had a recurrence, all within 48 hours after treatment. Rescue therapy with a second DBE procedure using similar hemostatic measures was performed for two cases, which managed to control the bleeding. One patient required emergency surgery for hemorrhagic shock after the second DBE. Of the three patients with recurrent disease, two had been treated with APC and adrenaline and one with APC, adrenaline and clips.

DISCUSSION

The present study was performed in a series of 15 patients with SB DL and a recurrence rate of 25 % was found after combined endoscopic treatment amongst the 12 subjects who were followed up. Bleeding was controlled with a rescue DBE procedure in two of three patients, with no recurrences observed in the long term. However, a low number of patients is the primary limitation of our study.

In our series, we observed that the factors most commonly associated with bleeding from DL included advanced age, high blood pressure and heart valvular disease. This accounts for the

fact that treatment with antiplatelet agents and anticoagulants are commonplace. The most common presentation was overt bleeding in the form of melena, which is consistent with other series of subjects with proximal SB (jejunum) disease (1,3,5). The diagnostic yields and agreement between CE and DBE in SB bleeding is variable according to lesion type and procedure timing (6,7). In our series, CE had a low diagnostic yield for DL, while it was very useful to orient towards an OR or AR approach for DBE.

With regard to therapy modality, while discrepancies exist, at least two hemostatic techniques are recommended for DL. In our experience, we prefer to initially use diluted adrenaline injections, since applying a second endoscopic treatment such as APC or clip placement is easier once the bleeding has been reduced or quelled (4). The recurrence rate was similar to that reported in other series (1,3), although three patients (all three referred from other institutions) were lost to follow-up and this aspect could not be verified. In contrast to other series where recurrence developed after days or months (4), recurrence in our study was always early, within 48 hours after the procedure. Tattooing during DBE was useful to facilitate the identification of the bleeding origin in recurrent cases, whether using the same technique or in a refractory subject during surgery.

To conclude, DL is an uncommon but serious cause of GI bleeding in the SB, usually located in the jejunum (3). It may be successfully managed with combined endoscopic hemostatic therapy via DBE, nearly always via the OR. Recurrent bleeding is common, hence we recommend that treated lesions be tattooed to facilitate their subsequent localization.

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Table 1. Clinical characteristics of patients with a Dieulafoy's lesion in the small bowel

Variables	*n (%)*	*n (%)*	*n (%)*	*n (%)*
Gender	Male 6 (40)	Female 9 (60)		
Antiaggregants	Yes 6 (40)	No 9 (60)		
Anticoagulants	Yes 3 (20)	No 13 (80)		
NSAIDs	Yes 1 (6.67)	No 14 (93.33)		
Cardiovascular disease	Yes 11 (73.33)	No 4 (26.67)	AF: 2 (18.18)	Ischemic heart disease: 2 (18.18)
Type	HBP: 10 (90.91)	Valvulopathy: 5 (40.45)		
Valvulopathy location	Aortic: 4 (80)	Aortic and other: 1 (20)		
Valve type	Mechanical: 1 (20)	Biological: 4 (80)		
Overt GI bleeding	Yes 11 (73.33)	No 4 (26,67)		
Type of manifestation	Melena: 10 (90.9)	Rectorrhagia: 1 (9.1)		
Dieulafoy's lesion location	Jejunum: 13 (86.67)	Ileum: 2 (13.33)		

HBP: high blood pressure; AF: atrial fibrillation.

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Table 2. Characteristics of diagnostic tests, treatments and recurring Dieulafoy's lesions in the small bowel

No.	*Urgent DBE*	*DBE route*	*DBE time*	*Treatment*	*Recurrence*	*No. of diagnostic tests*	*No. of DBEs*	*CE findings*
1†	No	Oral		APC + adrenaline	No	4	1	Blood
2	No	Anal		APC + adrenaline	Yes	6	3	Blood
3	Yes	Oral	90	APC + clips	No	2	1	
4	No	Oral	90	APC + adrenaline	No	3	1	
5	No	Oral	35	APC + adrenaline + clip	Yes	4	1	Blood
6	No	Oral	50	APC + adrenaline	No	4	1	Blood
7	Yes	Oral	90	APC + adrenaline	No	6	3	Blood, vascular lesions
8	No	Oral		APC + adrenaline + clip	No	5	1	Normal
9†	Yes	Oral	60	APC + adrenaline	No	4	1	Blood, vascular lesions
10	No	Oral	45	APC + adrenaline + clip	No	4	1	Blood, vascular lesions
11	No	Oral	50	APC + adrenaline	No	4	1	Jet bleeding
12	Yes	Oral	75	APC + adrenaline	Yes*	6	2	Jet bleeding
13	Yes	Oral		APC + adrenaline + clip	No	5		Blood, vascular lesions
14	Yes	Anal		APC + clip	No	4	1	Blood
15†	No	Oral		APC + adrenaline	No	6	1	Blood, vascular lesions

DBE: double-balloon enteroscopy; CE: capsule endoscopy. *Emergency surgery for hemorrhagic shock. †Patients who were lost to follow-up.

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Fig. 1. Capsule endoscopy. Red blood in the jejunum with no apparent lesion.

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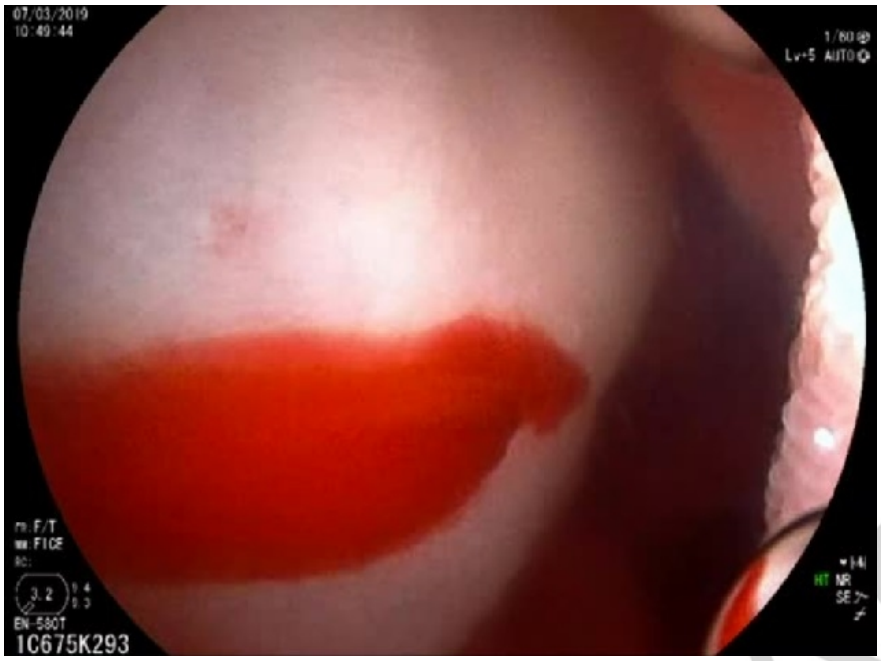

Fig. 2. Double-balloon enteroscopy. Jet arterial bleeding without an underlying lesion.