

Gastric antral vascular ectasia and solitary rectal ulcer syndrome – two rare diagnoses as the cause of anemia in a single patient: case report

Lumir Kunovsky^{1,2}, Milan Dastych¹, Radek Kroupa¹, Beata Hemmelova², Katarina Muckova³, Miroslava Chovancova³, Lenka Kuceroval¹, Jiri Dolina¹

¹Department of Gastroenterology, Faculty of Medicine, Masaryk University, University Hospital Brno Bohunice, Czech Republic

²Department of Surgery, Faculty of Medicine, Masaryk University, University Hospital Brno Bohunice, Czech Republic

³Department of Pathology, Faculty of Medicine, Masaryk University, University Hospital Brno Bohunice, Czech Republic

Summary

Gastric antral vascular ectasia (GAVE) and solitary rectal ulcer syndrome (SRUS) are both mentioned in the literature as rare causes of iron deficiency anemia and gastrointestinal (GI) bleeding. GAVE accounts for up to 4 % of upper non-variceal GI bleeding; SRUS is a rare benign disorder that presents with rectal bleeding. We present the case of a 75-year-old patient who was admitted to our facility with anemia. In the same patient, we encountered chronic bleeding from GAVE and SRUS. Both diagnoses were treated endoscopically: GAVE by argon plasma coagulation and a subsequent treatment with proton pump inhibitors and SRUS by adrenaline injection and clipping, consecutively treated with mesalazine enemas. The patient was successfully cured, resulting in a stable level of hemoglobin and no recurrent GI bleeding. We report a unique case of chronic GI bleeding caused by two uncommon diagnoses. The co-occurrence of GAVE and SRUS has not been previously described or published.

Key words: anemia – endoscopy – gastric antral vascular ectasia (GAVE) – gastrointestinal bleeding – solitary rectal ulcer syndrome (SRUS)

Cévní ektázie žaludečního antra a syndrom solitárního rektálního vředu – dvě vzácné diagnózy jako příčina anémie u téhož pacienta: kazuistika

Souhrn

Cévní ektázie žaludečního antra (gastric antral vascular ectasia – GAVE) a syndrom solitárního rektálního vředu (solitary rectal ulcer syndrome – SRUS) jsou uváděny v literatuře jako vzácné příčiny způsobující anémii z nedostatku železa a krvácení do gastrointestinálního traktu (GIT). GAVE může způsobovat nevarikózní krvácení z horního GIT do 4 %. V případě SRUS se jedná o vzácné benigní onemocnění, které se nejčastěji projevuje krvácením z konečníku. Prezentujeme případ 75letého pacienta, který byl přijat na naši kliniku pro anémii. U stejného pacienta jsme diagnostikovali chronické krvácení z GAVE a SRUS. Oba nálezy byly ošetřeny endoskopicky: GAVE pomocí argon plazma koagulace s následnou léčbou inhibitory protonových pump a SRUS opichem adrenalinovou injekcí a naložením klipu, poté následovala lokální léčba mesalazinovými klyzmaty. Pacient byl takto úspěšně vyléčen, s výsledným stabilním hemoglobinem a bez dalších známek krvácení do GIT. Prezentujeme ojedinělý případ pacienta s chronickou anémií, jež byla způsobena koincencí dvou vzácných onemocnění. Koincidence GAVE a SRUS nebyla zatím v literatuře publikována.

Klíčová slova: anémie – cévní ektázie žaludečního antra – endoskopie – gastrointestinální krvácení – syndrom solitárního rektálního vředu

Introduction

Gastric antral vascular ectasia (GAVE) is a rare cause of upper gastrointestinal (GI) bleeding. The etiology is still unclear, although there are some theories, including mechanical stress and excess of vasoactive instances [1–3].

GAVE is reported to be the cause up to 4 % of upper non-variceal bleeding [2,3]. This disorder is often linked with other chronic diseases such as autoimmune connective tissue diseases or in 30 % of the patients with liver cirrhosis [3,4]. GAVE has a characteristic endoscopic appear-

ance of linear, red stripes radiating toward the pylorus [5]. A punctate form has also been described, which appears more commonly in patients with liver cirrhosis [6].

Solitary rectal ulcer syndrome (SRUS) is also described as an infrequent benign entity that can cause iron deficiency anemia and whose symptoms usually include rectal bleeding. SRUS is often linked with a defecation disorder [7,8]. Its etiology is unknown; it is mostly reported with chronic mucosal and hypoperfusion induced ischemic injury to the rectal mucosa. SRUS is associated with a paradoxical contraction of the pelvic floor, leading to a mucosal prolapse and a pressure necrosis of the rectal mucosa [7]. Endoscopic findings of SRUS can imitate diseases such as inflammatory bowel diseases and neoplasms [7]. Symptoms of SRUS include rectal bleeding, mucus discharge from the rectum, straining during defecation, constipation, rectal prolapse, and lower abdominal pain [7–9].

Fig. 1A. Characteristic endoscopic view of GAVE stripes in the antrum creating a watermelon stomach



Fig. 1B. Detail of one of the GAVE stripes with dilated blood vessels



Case report

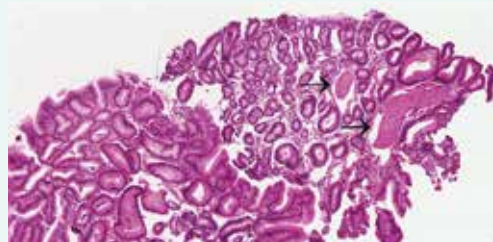
Our patient is a 75-year-old polymorbid man whose case history includes hypertension, diabetes mellitus, dyslipidemia, hyperuricemia, atrial fibrillation for which he was taking anticoagulants, chronic ischemic heart disease, hemorrhoids, diverticulosis of the sigmoid colon, and gastroduodenal ulcer disease. The patient had a history of constipation and occasional episodes of rectal bleeding.

The patient was admitted to our ward for severe sideropenic anemia, with a hemoglobin level of 64 g/L. Dysphagia, vomiting, hematemesis, and melena were not present at the time of admission. The patient was hemodynamically stable. Two blood transfusions were given with no suitable hemoglobin elevation. Esophagogastroduodenoscopy (EGD) and colonoscopy were planned to exclude GI bleeding. EGD showed characteristic findings of GAVE (fig. 1A, 1B), with no signs of acute bleeding. Treatment with argon plasma coagulation (APC) was chosen (fig. 1C) and followed with proton pump inhibitor medication. Diagnosis of GAVE was proved histologically (fig. 2). There was a sudden occurrence of enterorrhagia after a few days, and a colonoscopy was performed; the colonoscopy detected a rectal ulcer 13 cm from the

Fig. 1C. Endoscopic view after APC treatment



Fig 2. Gastric antral biopsy: biopsy showed dilated capillaries (arrows) in the superficial lamina propria with fibrin thrombi and mild foveolar hyperplasia



anal verge, covered by a clot (fig. 3A). Endoscopic treatment of an adrenaline injection and a lining of the clip was performed. Local rectal enema therapy with mesalazine was started. Ten days later, the control colonoscopy showed healing of the rectal ulcer (fig. 3B). The patient seemed to be stable, and his hemoglobin level normal. Liver ultrasound elastography excluded cirrhosis, and EGD excluded esophageal or gastric varices. Transrectal ultrasonography (TRUS) was added, with no signs of infiltration of deeper layers. Finally, histology confirmed the diagnosis of SRUS (fig. 4).

Discussion

Our patient had several possible causes of GI bleeding in his case history: hemorrhoids, diverticulosis of the sigmoid colon, and gastroduodenal ulcer disease. However, none of them was the cause of the patient's chronic anemia. Two rare entities were shown by endoscopy: GAVE and SRUS.

Fig. 3A. Endoscopic view of solitary rectal ulcer with a clot 13 cm from the anal verge



Fig. 3B. Healing rectal ulcer after 10 days of treatment



GAVE can be associated with other chronic diseases such as liver cirrhosis, systemic sclerosis, diabetes mellitus, and cardiovascular disease [3,6]. Our patient underwent ultrasound elastography, which excluded liver cirrhosis. Patients with liver cirrhosis and GAVE usually have a punctate form of GAVE instead of the more common linear striped appearance on the endoscope [3,6]. The etiology of GAVE in our patient's case seems to be cardiovascular.

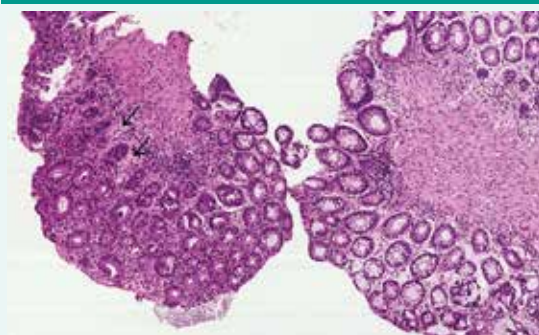
In a differential diagnosis, it is important to distinguish between GAVE and portal hypertensive gastropathy (PHG) or antral gastritis (AG). The treatment of these entities differs from that of GAVE. The treatment of GAVE should be performed endoscopically, whereas PHG treatment is focused on the reduction of portal pressure [1] and AG can be treated by medication.

APC is safe and effective in the treatment of GAVE, but recurrent bleeding occurs and may require more endoscopic sessions [5]. Some authors claim that endoscopic band ligation (EBL) is feasible and effective and should be performed as a method of first choice in GAVE treatment [2,6].

If severe recurrent and refractory bleeding occurs after APC or EBL treatment or endoscopic treatment is not effective, surgery is reserved as an efficacious method. It can, however, be associated with an increased risk of morbidity and mortality [3]. Jin et al published a case of a successfully surgically treated patient with refractory bleeding. In their case, a distal gastrectomy was performed in a hemodynamically unstable patient [4].

Clinical symptoms, endoscopic view, and histological findings constitute the criteria for the diagnosis of SRUS [8,9]. SRUS has been described in three most common endoscopic views. Endoscopic appearances

Fig. 4. Histopathological section of rectal lesion: examination of the rectal mucosa adjacent to the ulcer showed non-specific changes: superficial erosion, mixed inflammatory infiltration of the lamina propria mucosa. The following characteristic (traumatic-type) changes were found: starting fibromuscular obliteration of the lamina propria mucosae (arrows) and slight distortion of the crypts



range from erythematous lesions to ulcerative or polypoidal/nodular ones. The most common appearance reported is the ulcerative type [7] that our patient presented with.

Rectal bleeding was the reason to perform colonoscopy in our case. This symptom is reported as most common in SRUS, according to Abid et al [7] in 82 % and Abbasi et al [8] in 56 % of the cases. Constipation, which our patient also presented with, is reported as a less frequent symptom, 23 % in Abid et al [7] but 73 % in Abbasi et al [8]. Giving blood transfusions because of anemia caused by SRUS is uncommon [9]; however, severe anemia and chronic ischemic heart disease were indications for a blood transfusion in our case.

In our sample, the histopathological section confirmed a benign diagnosis. Some authors also recommend TRUS as helpful in ruling out an associated malignancy and recommend performing it routinely as a part of an evaluation in cases of suspected SRUS [10]. Nevertheless, performing a biopsy is mandatory to confirm SRUS and exclude potentially malignant diagnoses.

Hemorrhoids are often the reason for rectal bleeding, but a possible co-occurrence with another disease should not be excluded. Some authors published associated underlying conditions with SRUS. Abid et al [7] reported a slightly increased co-occurrence with hemorrhoids in 6 %. Co-occurrence with ulcerative colitis was present in 2.5 %, hyperplastic polyps in 3.5 %, adenomatous polyps in 2 %, and adenocarcinoma of the colon was observed in 2 % [7].

The treatment of SRUS ranges from a conservative treatment such as topical enemas (5-aminosalicylate or steroid), oral 5-aminosalicylate or sucralfate, biofeedback, followed by endoscopic steroid injections, to surgery (rectopexy, excision of ulcer) [9,11]. In a non-healing SRUS, conservative treatment management, laparoscopic resection rectopexy, and transanal endoscopic microsurgery were reported as safe, feasible, and effective treatment methods [12]. Endoscopy can also be used for the treatment of bleeding SRUS, which we successfully managed endoscopically by an adrenaline injection and clip ligation.

Conclusion

We describe a patient presenting with chronic iron deficiency anemia caused by a combination of two rare diagnoses. Endoscopy was chosen as a first-line treatment, followed by a drug treatment.

Supported by Ministry of Health, Czech Republic – conceptual development of research organization (FNBr, 65269705).

References

1. Patwardhan VR, Cardenas A. The management of portal hypertensive gastropathy and gastric antral vascular ectasia in cirrhosis. *Aliment Pharmacol Ther* 2014; 40(4): 354–362. Available at DOI: <<http://dx.doi.org/10.1111/apt.12824>>.
2. Wells CD, Harrison ME, Gurudu SR et al. Treatment of gastric antral vascular ectasia (watermelon stomach) with endoscopic band ligation. *Gastrointest Endosc* 2008; 68(2): 231–236. Available at DOI: <<http://dx.doi.org/10.1016/j.gie.2008.02.021>>.
3. Kar P, Mitra S, Resnick JM et al. Gastric antral vascular ectasia: case report and review of the literature. *Clin Med Res* 2013; 11(2): 80–85. Available at DOI: <<http://dx.doi.org/10.3121/cmr.2012.1036>>.
4. Jin T, Fei BY, Zheng WH. Successful treatment of refractory gastric antral vascular ectasia by distal gastrectomy: A case report. *World J Gastroenterol* 2014; 20(38): 14073–14073. Available at DOI: <<http://dx.doi.org/10.3748/wjg.v20.i38.14073>>.
5. Chiu YC, Lu LS, Wu KL et al. Comparison of argon plasma coagulation in management of upper gastrointestinal angiodysplasia and gastric antral vascular ectasia hemorrhage. *BMC Gastroenterol* 2012; 12: 67. Available at DOI: <<http://dx.doi.org/10.1186/1471-230X-12-67>>.
6. Keohane J, Berro W, Harewood GC et al. Band ligation of gastric antral vascular ectasia is a safe and effective endoscopic treatment. *Dig Endosc* 2013; 25(4): 392–396. Available at DOI: <<http://dx.doi.org/10.1111/j.1443-1661.2012.01410.x>>.
7. Abid S, Khawaja A, Bhimani SA et al. The clinical, endoscopic and histological spectrum of the solitary rectal ulcer syndrome: a single-center experience of 116 cases. *BMC Gastroenterol* 2012; 12: 72. Available at DOI: <<http://dx.doi.org/10.1186/1471-230X-12-72>>.
8. Abbasi A, Bhutto AR, Taj A et al. Solitary Rectal Ulcer Syndrome: Demographic, Clinical, Endoscopic and Histological Panorama. *J Coll Physicians Surg Pak* 2015; 25(12): 867–869. Available at DOI: <<http://dx.doi.org/10.2015/JCPSP.867869>>.
9. Urganci N, Kalyoncu D, Eken KG. Solitary rectal ulcer syndrome in children: a report of six cases. *Gut Liver* 2013; 7(6): 752–755. Available at DOI: <<http://dx.doi.org/10.5009/gnl.2013.7.6.752>>.
10. Sharma M, Somani P, Patil A et al. Endoscopic ultrasonography of solitary rectal ulcer syndrome. *Endoscopy* 2016; 48(Suppl 1): E76–E77. Available at DOI: <<http://dx.doi.org/10.1055/s-0042-102449>>.
11. Smejkal M, Adamek S, Bitnar P et al. Rehabilitation and modern approaches to the treatment of solitary rectal ulcer syndrome. *Gastroent Hepatol* 2014; 68(5): 451–455. Available at DOI: <<http://dx.doi.org/10.14735/amgh2014451>>.
12. Ihnat P, Martinek L, Vavra P et al. Novel combined approach in the management of non-healing solitary rectal ulcer syndrome – laparoscopic resection rectopexy and transanal endoscopic microsurgery. *Wideochir Inne Tech Maloinwazyjne* 2015; 10(2): 295–298. Available at DOI: <<http://dx.doi.org/10.5114/wiitm.2015.52060>>.

Lumir Kunovsky, M.D.

✉ lumir.kunovsky@gmail.com

Department of Gastroenterology, Faculty of Medicine, Masaryk University, University Hospital Brno Bohunice, Czech Republic

www.med.muni.cz

www.fnbrno.cz

Received 25. 3. 2017

Accepted 28. 4. 2017