ANNALI ITALIANI DI CHIRURGIA

Digital Edition
e-publish on-line
ISSN 2239-253X
Direttore Nicola Picardi

Unusual cause of upper gastrointestinal bleeding.
Report of the case of hemorrhagic duodenal lipoma with review of the literature

Ann Ital Chir, 2022; 11 - Sept. 5
pii: S2239253X22038014
Online Epub

Branko Brankovic*/**, Goran Stanojevic*/**, Milica Nestorovic*/**, Tijana Dencic***, Nikola Milutinovic**

*Department of Surgery, Faculty of Medicine, University of Nis, Nis, Serbia
**Clinic for Digestive Surgery, University Clinical Centre Nis, Nis, Serbia
***Department of Pathology, Faculty of Medicine, University of Nis, Nis, Serbia

Unusual cause of upper gastrointestinal bleeding. Report of the case of hemorrhagic duodenal lipoma with review of the literature.

Duodenal lipomas are uncommon and rare causes of gastrointestinal bleeding. Here, we present the case of a 45-year-old male patient who was admitted to University Clinical Centre because of melaena. After initial diagnostics, including echosonography, esophagogastroduodenoscopy revealed bleeding from protruding blood vessel at the polypoid submucosal change in the posterior duodenal bulb. Upon two urgent unsuccessful endoscopic hemostasis, a duodenotomy was performed. Definitive diagnosis was based on histological findings, describing duodenal lipoma with Bruner's gland hyperplasia. Upper GI bleeding is a serious challenge that requires adequate diagnostics necessary for the right choice of therapeutic approach. Unsuccessful endoscopic hemostasis could be followed by serious complications in bleeding duodenal lipoma when surgery should be always considered as the treatment of choice in patients with this kind of bleeding tumor.

KEY WORDS: Bruner Glands Hyperplasia, Duodenal Lipoma, Upper Gastrointestinal Bleeding

Introduction

Upper gastrointestinal (GI) bleeding of different etiologies is common in gastrointestinal pathology. The blood loss is usually manifested as hematemesis or melena or both. The main cause of this medical condition is bleeding from esophagus, stomach, or duodenum. The literature data reported esophagogastric varices, erosive esophagitis, and peptic ulcer as the three most common causes of upper GI bleeding [1]. Also, although considered as very rare in duodenum (about 5% gastrointestinal stromal tumors - GISTs), these tumors make up 30% of primary duodenal tumors, so it must be considered in differential diagnosis of the cases of upper GI bleeding [2], while duodenal lipoma is rather an unexpected cause.

Lipomas in gastrointestinal (GI) tract are very rare and uncommon. They are benign and slow growing tumors. The most of them occur in colon (64%), which is the common site for this kind of GI tumors [3]. There are also some literature data about ileal lipomas [4,5]. Manna et al. reported a rare case of acute gastrointestinal hemorrhage and acute anemia due to distal ileum lipoma [5]. The incidence of lipomas in stomach and duodenum is extremely low. Duodenal lipomas represent only 4% of all GI lipomas present in GI tract and they are usually asymptomatic [6]. Also, bleeding is not the frequent symptom of duodenal lipomas. There are only few cases of bleeding duodenal lipoma worldwide reported in literature [7].

Pervenuto in Redazione Febbraio 2022. Accettato per la pubblicazione Aprile 2022
Correspondence to: Branko Brankovic, Financially supported by the Ministry of Education, Science, and Technological Development of the Republic of Serbia Grant No. 451-03-9/2021-14/200113). Svetozara, Markovica 1/9, 18000 Nis, Serbia (e-mail: stojanovicivana38 @gmail.com)

Case Report

A 45-year-old male patient was admitted from Clinic for Gastroenterology to our Department for gastrointestinal proximal segment bleeding and melena. After hospitalization in a regional hospital, where he was unsuccessfully conservatively treated due to upper GI bleeding, he was admitted at the Clinic for Gastroenterology of University Clinical Centre Nis with anemia and low hemoglobin. On admission at the Clinic for Gastroenterology, the patient was conscious, oriented, afebrile, pale, with anemic mucoses. His blood pressure was 130/75 mmHg, heart rate rhythmic.
Echosonography found bleeding pseudopolypoid change. The patient underwent two urgent upper GI endoscopies which revealed protruding bleeding vessel at the polypoid submucosal change in the posterior duodenal bulb. Since, due to active bleeding, endoscopic hemostasis was not successful two times, the patient was transferred to the Clinic for Surgery. At admission, the patient's laboratory finding revealed anemia (RBC 3.11 X 1012/L, hemoglobin 85 g/L, hematocrit 0.27, MCH 27.3, RDW 15.3)
According to algorithm for patients with refractory bleeding despite a second therapeutic endoscopy, and threatening hemorrhagic shock, surgery was required. After adequate preoperative preparation, the patient underwent surgery. Using upper medial laparotomy, duodenotomy

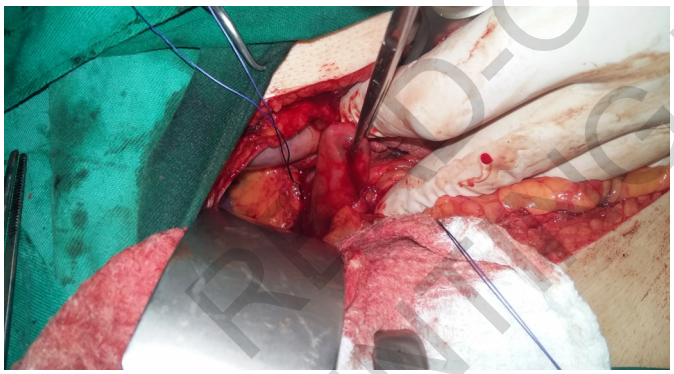

Fig. 1: Surgical excision of the tumor.

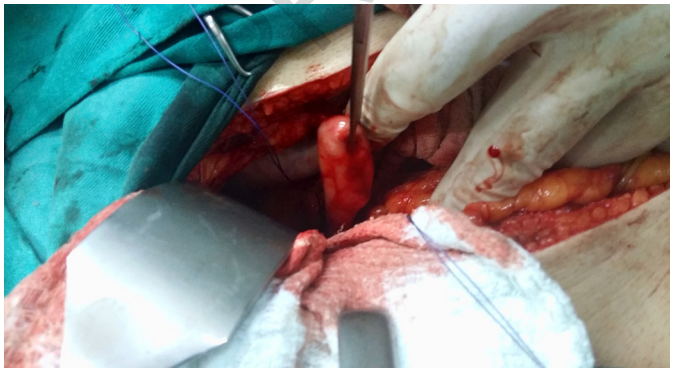

Fig. 2: Surgical excision of the tumor.

Fig. 3: Excised haemorrhagic duodenal lipoma.

and the tumor excision has been performed. It was relatively well-limited smooth bleeding tumor formation. Then mucose and submucose reconstruction has been done, followed by duodenal suture, lavage and drainage, while the excised change was sent for histopathological examination (Figs. 1, 2).
The excised tumor dimensions were 35x20x15 mm (Fig. 3). The differential diagnosis was gastrointestinal stromal tumor (GIST) and lipoma and final diagnosis was confirmed histologically.
After the procedure, the patient was observed for bleeding and treated conservatively. Ten days after he was discharged without any signs of bleeding. His laboratory results at the dismission were as follows: RBC 3.5 X 10^{12}/L, WBC 10.6 X 10^{9}/L, PLT 449 X 10^{9}/L, hemoglobin 99 g/L, hematocrit 0.30, CRP 66.5 mg/L, blood urea nitrogen (BUN) 2.2 mmol/L, total protein level 55.7 g/L, albumin 33 g/L, amylase 106 U/L. The patient was recommended a diet.

Histopathological Examination

The tissue was fixed in 10% formalin solution for histological examination and routinely processed and embedded in paraffin blocks. Serial sections of 4 microns were prepared from these blocks. Histopathological sections from paraffin blocks after deparaffinization were stained with hematoxylin-eosin (HE) method. Microscopic images of pathohistological specimens were recorded with a digital camera (Eclipse 50i; Nikon; Tokyo).

Pathological Findings

The resected specimen demonstrated the bright yellow pedunculated polypoid mass with greasy cut surface without any erosion or ulcer, measuring 35x20x15 mm.
Pathological report showed an encapsulated well circumscribed collection of mature adipose tissue without atypia in the submucosa, but a lobular proliferation of

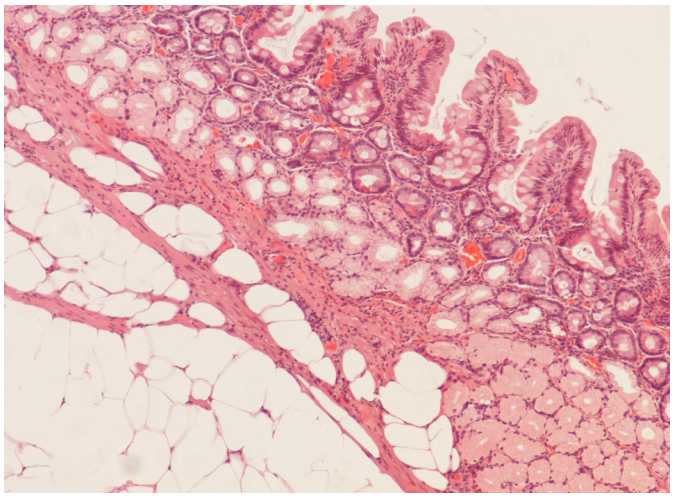

Fig. 4: Microscopic view of the lobulated submucosal duodenal lipoma with Brunner gland hyperplasia (HEX200).

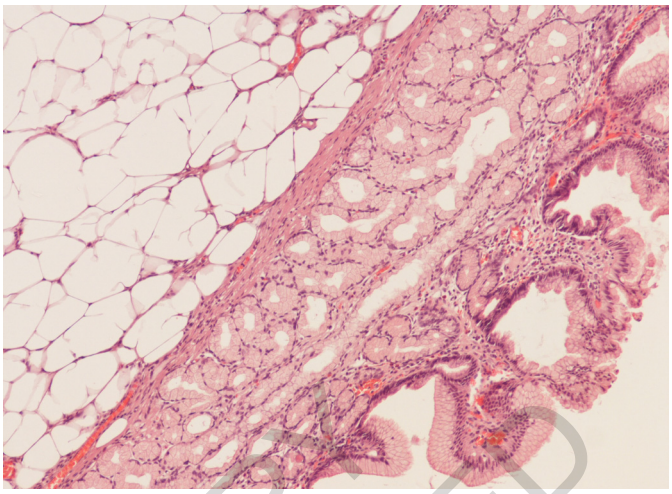

Fig. 5: Pathological image of the duodenal lipoma in the submucosa under a normal duodenal mucosal layer (HEX200).

Table I - *Literature data about hemorrhagic duodenal lipoma cases.*

Authors	Year	Sex	Age	Dimension in cm	Location
Tung et al.[20]	2001	Male	73	2	D2
Sou et al.[21]	2006	Female	81	5	D3
Tsukamoto et al.[22]	2008	Female	75	12	D1
Murata et al.[23]	2008	Male	67	4	D2
Long et al.[24]	2008	/	/	4	D3
Mohamed et al.[25]	2008	Female	70	5,5	D2
Mendez et al.[26]	2008	Male	70	6	D3
Chang et al.[27]	2010	Female	59	4	D2
Ouwerkerk et al.[28]	2010	Female	52	1.7	D1
Kadaba et al.[29]	2011	Female	60	6	D1
Efe et al.[30]	2012	Male	76	4	D2
Thorlacius et al.[31]	2013	Male	66	3.5	D2
Yaman et al.[32]	2014	Female	59	4	D2
Dinesh et al.[33]	2016	Male	45	4	D234
Ødorf and Knuthsen[34]	2017	Male	73	3	D2
Lan et al.[35]	2018	Female	65	2.7	D3
Gwak et al.[7]	2019	Female	85	2	D2
Yoshi et al.[19]	2020	Male	72	4	D1
Baiss et al.[36]	2021	Male	58	10	D2
Our case	2022	Male	45	3.5	D2

Although bleeding is the rare symptom of these kind of tumors, it is necessary to consider duodenal lipomas as the cause of GI bleeding in differential diagnosis.

Brunner glands. The overlying duodenal mucosal layer was entirely normal (Figs. 4, 5). On these histopathological findings, a final pathological diagnosis of submucosal duodenal lipoma was made.

Discussion

The literature data about the cases of duodenal lipomas are very rare. The duodenal lipomas are benign and slow growing tumors of unknown etiology, usually occurring between fifth and seventh decade of life. Lipomas arise from adipocytes and there are hypotheses that these tumors could be associated with certain metabolic disturbances present in degenerative disease [8] or dyslipidemia [9], but mostly they might be the consequence of embryonic displacement of adipose tissue.

In the context of the structure, it is also important to discuss the unusual appearance of Bruner glands hyperplasia at the substrate of duodenal lipoma in our patient. Pironi et al [10] has reported an unusual case of massive GI bleeding due to Brunner's gland adenoma, which was successfully removed by open surgery, but we haven't find in the literature the hyperplasia of Bruner glands

in duodenal lipoma. Gastric hyperacidity was originally thought to induce Bruner's glands hyperplasia by gland stimulation [11], but only 45% of these patients had hyperacidity. Kovacevic et al [12] have reported that the concurrent Helicobacter pylori infection is very common in patients with Bruner gland tumors, but its pathogenic role in the development of Brunner's gland hyperplasia remains unclear. It is not clear what is the cause and the impact of Brunner's gland hyperplasia in this case of bleeding duodenal lipoma.

The differential diagnosis of duodenal lipomas is rather difficult because the symptoms of these tumors are not specific. The symptoms depend on their size and location and vary from epigastric fullness, abdominal pain, obstructive jaundice, ulcerations and bleeding to intussusception and bowel obstruction [13-15].

Duodenal lipomas are mostly asymptomatic, but symptomatic duodenal lipomas need either endoscopic or operative treatment. Depending on the size, the adequate therapeutic method should be used. There are reports that surgical resection is indicated for tumors with a size of 2 cm or more[16]. Beside size, the important factors for adequate therapy approach are the tumor shape and if it is bleeding or not. Upper gastrointestinal bleeding represents today a serious medical state, which diagnosis and surgical treatment modalities are studiously discussed by Cortese et al[17]. Acute massive upper GI bleeding often requires emergency surgery due to hemodynamic instability of the patient when endoscopic hemostasis has failed or when the tumor is very large[10]. Bleeding duodenal lipomas are extremely rare. It has been reported that they represent only 5.5% of all duodenal lipomas[18]. Large bleeding duodenal lipomas can be technically difficult for endoscopic excision, due to increased risk of bleeding, when a classic surgery is indicated [5]. But it is important to mention that, upon searching at PubMed and Google Scholar, Youshii et al[19] found only 16 cases of hemorrhagic duodenal lipoma published in the period between 2000 and 2019. Searching at available literature, up to now, we have found only 20 cases, including our one.

Conclusion

Upper GI bleeding is a serious challenge that requires adequate diagnostics necessary for the right choice of therapeutic approach. Duodenal lipomas, as very rare tumors, are usually not considered as the cause of GI bleeding, but they could cause severe GI bleeding unresponsive to endoscopic hemostasis. That is why, although endoscopic treatment of GI bleeding have increased an attention as minimally invasive therapeutic method, unsuccessful endoscopic hemostasis could be followed by serious complications in bleeding duodenal lipoma when surgery should be always considered as the treatment of choice in patients with this kind of bleeding tumor.

Riassunto

I lipomi duodenali rappresentano cause rare di sanguinamento gastrointestinale. Presentiamo qui il caso di un paziente maschio di 45 anni che è stato ricoverato al Centro Clinico Universitario per melena. Dopo la diagnostica iniziale, comprendente l'ecosonografia, l'esofagogastroduodenoscopia ha rivelato sanguinamen-to da un vaso sanguigno sporgente in corrispondenza della sottomucosa polipoide nel bulbo duodenale posteriore. Dopo due tentativi di emostasi per via endoscopica, eseguiti d'urgenza ma senza successo, è stata eseguita una duodenotomia. La diagnosi definitiva si basa sui reperti istologici, che descrivevano il lipoma duodenale con iperplasia della ghiandola di Bruner.

Il sanguinamento del tratto gastrointestinale superiore è una sfida seria che richiede un'adeguata diagnostica necessaria per la giusta scelta dell'approccio terapeutico. L'emostasi endoscopica non riuscita potrebbe essere seguita da gravi complicazioni nel caso di lipoma duodenale sanguinante, laddove la chirurgia dovrebbe essere sempre considerata come il trattamento di scelta nei pazienti con questo tipo di tumore sanguinante.

References

1. Kamboj AK, Hoversten P, Leggett CL: *Upper Gastrointestinal Bleeding: Etiologies and Management*. Mayo Clin Proc, 2019; 94(4):697-703.

2. Giordano A, Moroni M, Di Filippo G, Cammelli F, Baraghini M, Giudicissi R, Vellei S, Feroci F, Cantafio S: *Emergency duodenal resection for giant GIST with acute gastrointestinal bleeding.* Ann Ital Chir, 2021; 10:S2239253X21036707.

3. O'Riordan BG, Vilor M, Herrera L: *Small bowel tumors: An overview*. Dig Dis, 1996; 14:245-57.

4. Feo CV, Zese M, Tralli G, Targa S, Galeotti R, Rizzati R: *A rare case of ileo-ileal intussusception due to a bleeding lipomatous mass treated by laparoscopic ileal resection*. Ann Ital Chir, 2019; 8:S2239253X19030391.

5. Manna E, Frattaroli FM, Polettini E, Nunziale A, Pappalardo G: *A rare cause of acute gastrointestinal hemorrhage: Ileal lipoma.* Ann Ital Chir, 2017; 88:73-75.

6. Cao D, Sun X, Du J, Sun Y: *Duodenal lipoma associated with ectopic duodenal glands*. Clin Pract, 2011; 1:e128.

7. Gwak SY, Lee MK, Lee YK: *A case of a bleeding duodenal lipoma successfully controlled by endoscopic resection*. Clin Endosc, 2020; 53:236-40.

8. Jeong IH, Maeng YH: *Gastric lipomatosis*. J Gastric Cancer, 2010; 10(4):254-58.

9. Bilgic Y, Altinsoy HB, Yildirim N, Alatas O, Kanat BH, Sahin A: *Familial abdominal and intestinal lipomatosis presenting with upper GI bleeding*. Case Rep Gastrointest Med, 2015; 2015:123723.

10. Pironi D, Vendettuoli M, La Gioia G, Arcieri S, Filippini A: *A large Brunner's gland adenoma: an unusual cause of gastrointestinal bleeding*. Ann Ital Chir, 2010; 81:147-51.

11. Kaplan EL, Dyson WL, Fitts WT Jr: *The relationship of gastric hyperacidity to hyperplasia of Brunner's glands*. Arch Surg, 1969; 98:636-39.

12. Kovacevic I, Ljubicic N, Cupic H: *Helicobacter pylori infection in patients with Brunner's gland adenoma*. Acta Med Croatica, 2001; 55:157-60.

13. Tjandra D, Knowles B, Simkin P, Kranz S, Metz A: *Duodenal lipoma causing intussusception and gastric outlet obstruction*. ACG Case Rep J, 2019; 6:e00157.

14. Pei MW, Hu MR, Chen WB, Qin C: *Diagnosis and treatment of duodenal lipoma: A systematic review and a case report*. J Clin Diagn Res, 2017: 11:PE01-PE05.

15. Chen HT, Xu GQ, Wang LJ, Chen YP, Li YM: *Sonographic features of duodenal lipomas in eight clinicopathologically diagnosed patients*. World J Gastroenterol, 2011; 17(23):2855-59.

16. Cortese F, Colozzi S, Marcello R, Muttillo IA, Giacovazzo F, Nardi M, Mero A: *Gastroduodenal major haemorrhages in critical patients*. Ann Ital Chir, 2013; 84:671-79.

17. Yu HG, Ding YM, Tan S, Luo HS, Yu JP: *A safe and efficient strategy for endoscopic resection of large, gastrointestinal lipoma*. Surg Endosc, 2007; 21:265-69.

18. Michel LA, Ballet T, Collard JM, Bradpiece HA, Haot J: *Severe bleeding from submucosal lipoma of the duodenum*. J Clin Gastroenterol, 1988; 10:541-45.

19. Yoshii H, Izumi H, Tajiri T, Mukai M, Nomura E, Makuuchi H: *Surgical resection for hemorrhagic duodenal lipoma: A case report*. Tokai J Exp Clin Med, 2020; 45(2):75-80.

20. Tung CF, Chow WK, Peng YC, Chen GH, Yang DY, Kwan PC: *Bleeding duodenal lipoma successfully treated with endoscopic polypectomy*. Gastrointest Endosc, 2001; 54(1):116-67.

21. Sou S, Nomura H, Takaki Y, Nagahama T, Matsubara F, Matsui T, Yao T: *Hemorrhagic duodenal lipoma managed by endoscopic resection*. J Gastroenterol Hepatol, 2006; 21:479-81.

22. Tsukamoto Y, Samizo M, Takahashi T, Miyashita M, Maekawa T: *A case of a giant lipoma of the duodenal bulb resected by laparoscopic assisted distal gastrectomy*. Jpn J Gastroenterol Surg, 2008; 41:510-15.

23. Murata A, Osoegawa T, Ijyu M, Kanayama K, Tanaka M, Nakamura K: *Pedunculated duodenal lipoma treated with endoscopic polypectomy with a detachable snare*. Fukuoka Igaku Zasshi, 2008; 99:131-35.

24. Long BW, Goodwin E, Halford P, Almas J: *Diagnosis and endoscopic resection of a bleeding duodenal lipoma*. J Miss State Med Assoc, 2008; 49:3-6.

25. Mohamed HK, Suresh CC, Alexander KG, Neena M: *Bleeding duodenal lipoma treated with endoscopic polypectomy*. Trop Gastroenterol, 2008; 29(3):167-68.

26. Menéndez P, Cubo T, Padilla D, Villarejo P, Gambí D, Alonso A, Martín J: *Gastrointestinal bleeding due to duodenal lipoma*. Rev Esp Enferm Dig, 2008; 100(7):442-43.

27. Chang CW, Chu CH, Shih SC, Chen MJ, Yang TL, Chang WH: *Duodenal polypoid lipoma with bleeding*. Am J Surg, 2010; 200:e49-e50.

28. Ouwerkerk HM, Raber MH, Freling G, Klaase JM: *Duodenal lipoma as a rare cause of upper gastrointestinal bleeding.* Gastroenterology Research, 2010; 3(6):290-92.

29. Kadaba R, Bowers KA, Wijesuriya N, Preston SL, Bray GB, Kocher HM: *An unusual cause of gastrointestinal bleeding: Duodenal lipoma*. Case Rep Gastroenterol, 2011; 5:183-88.

30. Efe C, Purnak T, Ozaslan E, Kücükciloglu Y: *Which is the best approach for the hemorrhagic duodenal lipoma: endoscopic resection or surgery?* Am J Surg, 2012; 203(40):558.

31. Thorlacius H, Weiber H, Ljungberg O, Nielsen J, Toth E: *Endoscopic diagnosis and treatment of a giant duodenal lipoma presenting with gastrointestinal bleeding*. Endoscopy, 2013; 45(Suppl 2 UCTN):E385-86.

32. Yaman, Derici H, Paksoy S: *Symptomatic duodenal lipoma with endoscopic snare polypectomy*. Ulus Cerrahi Derg, 2014; 30(2):103-05.

33. Dinesh Z, Majid W, Tiwari P: *Duodenal lipomatosis as a curious cause of upper gastrointestinal bleed: A report with review of literature*. J Clin Diagn Res, 2016; 10(5):PE01-PE04.

34. Ødorf K, Knuthsen S: *Duodenal lipoma is a rare cause of gastrointestinal bleeding*. Ugeskr Læger, 2017; 179:V10160730.

35. Lan HC, Chen TS*: A rare cause of occult gastrointestinal bleeding*. Dig Liver Dis, 2018; 50(6):620.

36. Baiss M, Rahali A, Elmajdoubi H, Mdaghri J, Lahlou K, Mssrouri R, Benmar S, Settaf A: *Giant duodenal lipoma: An unusual cause of gastrointestinal bleeding (a case report)*. Pan Afr Med J, 2021; 38:342.