

Duodenal Lipomatosis as a Curious Cause of Upper Gastrointestinal Bleed: A Report with Review of Literature

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ABSTRACT

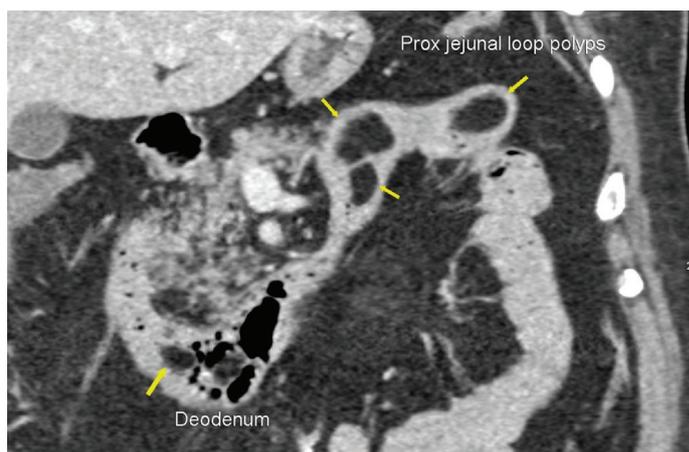
Lipomas of the gastrointestinal tract are rare. Duodenal lipomas are incidental and mostly asymptomatic. Tumours may produce symptoms of abdominal pain and discomfort or cause bleeding due to ulceration or intestinal obstruction due to intussusception. We describe a 45-year-old man presenting in emergency with 3 days history of melena with normal gastroduodenoscopy and contrast enhanced computed tomography revealing multiple polypoid lesion in duodenum and proximal jejunum suggestive of lipoma. Due to ongoing bleed, he underwent laparotomy with duodenectomy and uneventful postoperative recovery. Our review of cases published in last 67 years indicate that duodenal lipomas are rare to occur but commonly found in second part, they may be seen in third and fourth part of duodenum which may be missed on endoscopy. They can be multiple and may present as severe UGI bleeding which could be managed surgically. Though CT is diagnostic, histopathology confirms the diagnosis which shows lipomatous lesion composed of mature adipose arranged in lobules.

Keywords: Bleeding, Duodenectomy, Lipoma, Melena

CASE REPORT

A 45-year-old male presented with severe melena for 3 days. He was non alcoholic, non smoker and diabetic for 8 years with no significant medical or surgical illness in the past. On admission his pulse was 104/min with blood pressure of 100/70 mmHg and no significant per abdominal finding except melena was confirmed on digital rectal examination. His Hb (haemoglobin) was 8g/dl, blood glucose was 200mg/dl with dyslipidemia and other haematological and biochemical parameters were normal. Chest x-ray, ECG (electrocardiogram), echocardiogram was normal. Upper Gastrointestinal (UGI) endoscopy was normal and could be visualized till second part of duodenum. Ultrasound scan showed only fatty liver. CECT (Contrast Enhanced Computed Tomography) with angiography was done, revealed multiple lipomatous filling defects in 3rd and 4th part of duodenum including proximal jejunum [Table/Fig-1] and no active blush but pooling of the contrast in 3rd part of duodenum.

Patient was resuscitated with 2 units of blood transfusion and blood sugar level was controlled. Post transfusion Hb on day 2 of admission was 9.0g/dl which again dropped to 7 g/dl on day 3. In view of drop in haemoglobin and with doubtful therapeutic use of enteroscopy in multiple and large polypoidal lesion located in distal duodenum, it was decided to go ahead with surgery. Intraoperatively, multiple masses could be palpated in 2nd, 3rd and 4th part of duodenum and proximal jejunum and rest of the bowel appeared healthy. After complete Kocherisation, duodenum 2 cm distal to ampulla was excised along with 10 cm of proximal jejunum. A single polyp close to ampulla was removed after transfixation of the base. Intestinal continuity was maintained by doing side to side duodenojejunostomy. Operative time was 150 minutes and blood loss was 50 ml. Patient was started on liquid diet on postoperative day 4 and was discharged from the hospital on day 7. Postoperative recovery was uneventful. Specimen was examined which showed four large lipomatous polyps with largest dimension of 4 cm arising from third and fourth part of duodenum with ulcerated lesion at base responsible for recent bleeding episode [Table/Fig-2]. Definite diagnosis was a duodenal lipoma based on histological examination [Table/Fig-3] which showed polypoidal lesion lined by small intestinal mucosa

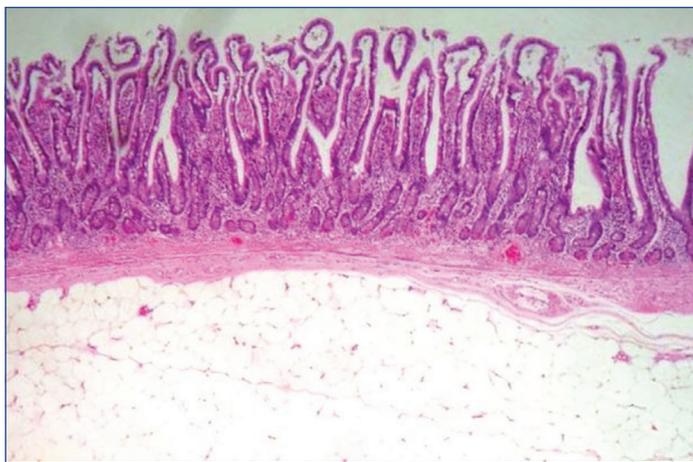


[Table/Fig-1]: 320 slice CECT abdomen coronal section showing polyps in distal duodenum and jejunum.



[Table/Fig-2]: Grossly, multiple lipomatous polyp involving D3, D4.

with mild mucosal inflammation. The submucosa was expanded and showed lipomatous lesion composed of mature adipose tissue and arranged in lobules interspersed with rare thin delicate fibrovascular septa possessing bland features.



[Table/Fig-3]: Histopathological examination confirming duodenal lipoma (H&E 4x).

DISCUSSION

Gastrointestinal (GI) lipomas are benign and slow growing tumours of submucosal origin. They are often found incidentally but may be symptomatic and can present with mild to severe gastrointestinal bleeding, intussusceptions, abdominal pain, constipation and diarrhoea. Bleeding is an uncommon presentation for duodenal lipoma. Lipomas have been found throughout the GI tract but occur most commonly in the colon, ileum, and jejunum [1-3]. Lipomas of the duodenum are relatively rare and are generally located in the second portion of the duodenum [2]. In a review of 1200 consecutive duodenoscopies, lipomas were found in only 2 patients [4]. In one series of 115,251 routine autopsies reported by Suire and Gousse [5], there were only 26 lipomas of the duodenum. Gastrointestinal lipomas account for 4% of all

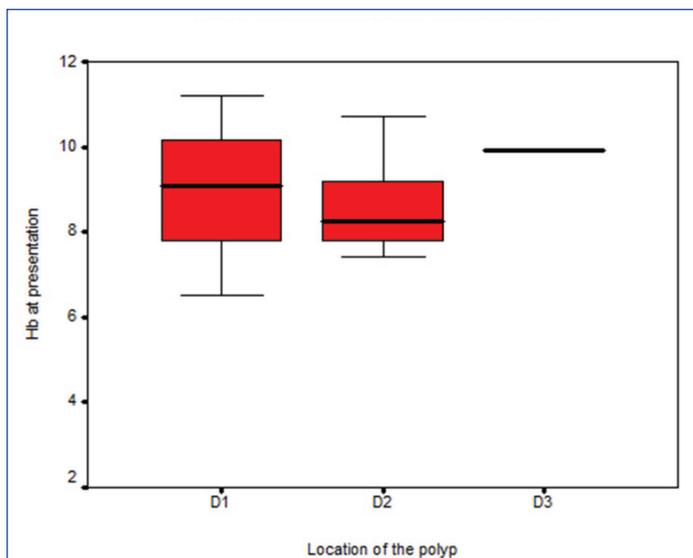
benign gastrointestinal tumours. Of gastrointestinal lipomas 64% are seen in the colon, but only 4% occur in the duodenum [6]. The peak incidence is around the 5th and 7th decade of life, with a slight female preponderance.

An online search in PubMed and Google scholar search engine for 'duodenal lipoma' and 'bleeding' generated 52 articles. Excluding 28 cases with presentation other than the UGI bleeding and concomitant small bowel or extra-intestinal lipomatosis, 24 case reports were identified to be published from 1948- 2015. Of which 23 cases of single and one case with two duodenal lipoma presenting as UGI bleeding has been reported in literature [1,2,6-26]. [Table/Fig-4] summarizes the review of literature for the duodenal lipoma presenting as UGI bleeding. Statistical analysis was done using the available data. Data entry was done in MS Excel spread sheet. Data analysis was done in SPSS (Version 16.0). Average age of presentation was 61 years (range 36 to 81) with female to male ratio of 11:10. Average maximum length of the polyp was 4.8cm (range 1.7 to 12 cm). Mean Haemoglobin at presentation was 8.5g/dl (range 4.3 to 11.2) with 60% occurring in second part of duodenum. On statistical analysis there was no significant association of size of the polyp with blood haemoglobin at presentation [Table/Fig-5]. Though majority of cases before 2000 were managed by transduodenal excision, recent reports suggest most of them are managed by endoscopic method. Management with duodenectomy was done with very few cases. [Table/Fig-6] indicates trend of significant change in management trending towards endoscopic interventions.

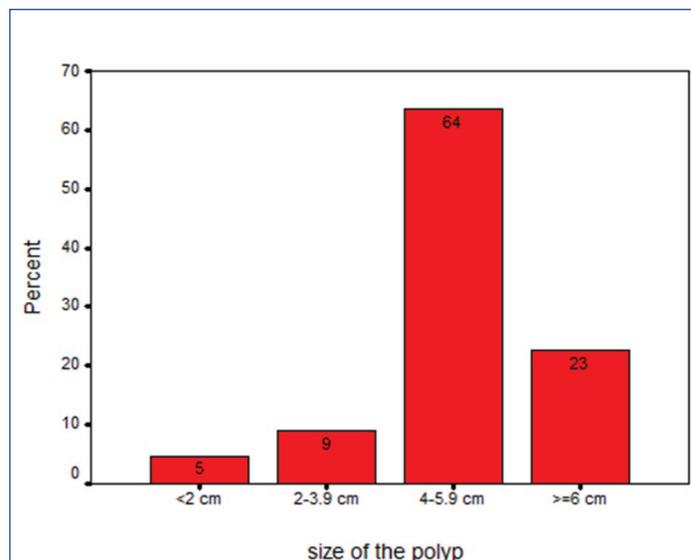
Duodenal lipomas are most often submucosal, but can also be subserosal. Duodenal lipoma presents as a round or ovoid, soft mass with regular or lobulated contours. They can be either sessile or pedunculated. The overlying mucosa of the duodenal lipoma is usually normal, but there may be areas of ulceration or

Sl No	Author	Year	Age in years	Sex	Location	Maximum Dimension in cm	Number	Hb at presentation in g%	Management
1	Yaman et al., [7]	2014	59	Female	D2	4	Single	9.7	Endoscopic Polypectomy
2	Thorlacius et al., [8]	2013	66	Male	D2	3.5	Single	9.2	Endoscopic Polypectomy
3	Efe et al., [9]	2012	76	Male	D2	4	Single	NA	Endoscopic Polypectomy
4	Kadaba et al., [10]	2011	60	Female	D1	6	Single	6.5	Transduodenal resection
5	Chang et al., [11]	2010	59	Female	D2	4	Single	8.2	Transduodenal resection
6	Ouwerkerk et al., [6]	2010	52	Female	D1	1.7	Single	9.1	Transduodenal resection
7	Mohamed et al., [12]	2008	70	Female	D2	5.5	Single	7.4	Endoscopic Polypectomy
8	Long et al., [13]	2008	NA	NA	D3	4	Single	NA	Endoscopic Polypectomy
9	Murata et al., [14]	2008	67	Male	D2	4	Single	10.7	Endoscopic Polypectomy
10	Tsakamoto et al., [15]	2008	75	Female	D1	12	Single	NA	Laparoscopic distal Gastrectomy
11	Menéndez et al., [16]	2008	70	Male	D3	6	Single	9.9	Duodenectomy(D3,D4)
12	Sou et al., [17]	2006	81	Female	D3	5	Single	NA	Endoscopic Polypectomy
13	Tung et al., [18]	2001	73	Male	D2	4.5	Single	7.8	Endoscopic Polypectomy
14	Krachman et al., [2]	1992	49	Male	D2	5.5	Single	NA	Transduodenal resection
15	Michel et al., [1]	1988	47	Female	D2	6	Single	9	Transduodenal resection
16	Michel et al., [1]	1988	54	Female	D2	6	Single	8	Transduodenal resection
17	Agha et al., [19]	1985	NA	NA	NA	NA	NA	NA	NA
18	Sarma et al., [20]	1984	63	Male	D2	3	Single	4.3	Transduodenal resection
19	Inoue et al., [21]	1983	47	Female	D1	5.5	Single	11.2	Resection
20	Makokha et al., [22]	1975	NA	NA	NA	NA	NA	NA	NA
21	Lemer et al., [23]	1971	63	Male	D2	4	Single	8.3	Transduodenal resection
22	Barr et al., [24]	1964	NA	NA	NA	NA	NA	NA	NA
23	Fawcett et al., [25]	1949	36	Male	D2	4	Two	NA	Partial duodenectomy
24	Allison et al., [26]	1948	70	Male	D3	5	Single	NA	Transduodenal resection
25	current case	2016	45	Male	D2,3,4	4	Four	NA	Duodenectomy(D3,D4)

[Table/Fig-4]: Review of literature for the duodenal lipoma presenting as UGI bleeding.



[Table/Fig-5]: Relation of blood hemoglobin at the time of presentation with location of the polyp.



[Table/Fig-7]: Size wise distribution of duodenal lipoma presenting as bleeding.

Period	Transduodenal resection	Endoscopic Polypectomy	Duodenectomy	Laparoscopic distal Gastrectomy	Total
Before 2000	7(87.5%)	0	1(12.5%)	0	8
After 2000	4(21.4%)	8(57.1%)	2(14.3%)	1(7.1%)	15
	11	8	3	1	23

[Table/Fig-6]: Change of trend in management of bleeding duodenal lipoma.

erosion [27]. The mechanisms of erosion or ulceration are probably mucosal pressure atrophy or peristalsis that leads to elongating and stretching, with necrosis of the overlying epithelial layers [1]. Though the predisposing factors responsible for bleeding are unknown sometimes ingested food material such as a chicken bone, may be a precipitating factor for bleeding in a large polyp as described by Michel et al [1].

Although endoscopic view supports the diagnosis, it is generally insufficient in making a definitive diagnosis and it may not visualize lipomas beyond second part of duodenum as in our case. However, CT scan of the upper gastrointestinal tract can fairly accurately facilitate the preoperative diagnosis of lipoma based on low attenuation signals of -50 to -100 Hounsfield units [28]. Though CT is helpful in diagnosis but is not precise to localize the bleeding point and origin of the lesion due to polypoid nature of lipoma. In our case CT showed the lesion in proximal jejunum but was arising from distal duodenum. EUS (Endoscopic Ultrasound) features of a homogenous, hyperechoic mass within the submucosal layer are highly characteristic of duodenal lipomas and in addition, EUS can also visualize the depth and invasion [29]. Malignant transformation of gastrointestinal lipomas has not been reported [30]. Capsule endoscopy may be helpful to diagnose the lesion in distal small bowel and more importantly to rule out any other concomitant cause of obscure GI bleed. In our case capsule endoscopy was not performed due to financial constraints of the patient which is also a limiting factor for capsule endoscopy in developing countries.

Symptomatic duodenal lipoma warrants treatment. Asymptomatic incidentally found lipoma may be observed but our review shows 61 % of duodenal lipoma presenting as UGI bleeding were 4 to 5.9cm of size as shown in [Table/Fig-7], raising the doubt, should asymptomatic giant lipoma (>4cm) be removed if amenable for endoscopic excision. Endoscopic techniques for removal of gastrointestinal lipomas include "snare" polypectomy, "endoloop", "unroofing", subtotal resection and submucosal dissection [18,31,32]. Limitations of endoscopic removal are location and multiplicity, as in present case. Though other methods of surgical removal by duodenotomy or duodenectomy have not reported

any major complication, endoscopic removal is preferred over surgical removal, if feasible. Our case is a very rare case of multiple duodenal lipoma presenting with melena involving distal duodenum which was managed in time with duodenal resection and had uneventful recovery.

CONCLUSION

In summary, though duodenal lipomas are rare to occur but commonly found in second part, they may be seen in third and fourth part of duodenum. Though rare but duodenal lipoma can be multiple and may be missed on UGI endoscopy. The trend of management is shifting from surgical approach to endoscopic approach but as in our case, surgery still remains an important tool in armamentarium.

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