

## CASE REPORT

# Rectal angiolipoma: A case report and review of literature

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## Abstract

Angiolipoma is a rare vascular variant of the benign lipomatous tumors and is generally seen in subcutaneous tissues. We report a 70-year-old female with abdominal distension not related to rectal small polypoid mass with peduncule described as angiolipoma by histologically, and review the literature.

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## INTRODUCTION

Lipomas, lipomatosis, lipoblastoma, lipoblastomatosis, angiolipomas, myolipomas, chondroid lipomas, spindle cell lipomas, and pleomorphic lipomas are the benign adipose tissue tumors<sup>[1]</sup>. Angiolipoma is one of the rare subcutaneous tissue tumors with its characteristic histology consisting of mature adipose tissue and interspersed proliferated vascular component<sup>[1-3]</sup>. It accounts for 5%-17% of lipomas<sup>[2,4,5]</sup>, and is rarely seen in the gastrointestinal system (GIS). The locations of gastrointestinal angiolipoma according to the current literature are oropharyngeal region<sup>[4,6-8]</sup>, esophagus<sup>[9]</sup>, stomach<sup>[10-13]</sup>, duodenum<sup>[14,15]</sup>, small intestine<sup>[5,16-18]</sup>, ileocecal valve<sup>[19-21]</sup>, colon<sup>[14,22-24]</sup> and liver<sup>[25-28]</sup>. Angiolipomas localized in the gastrointestinal tract, characterized as submucosal solitary sessile<sup>[10,11,18,20]</sup> or endoluminal polypoid

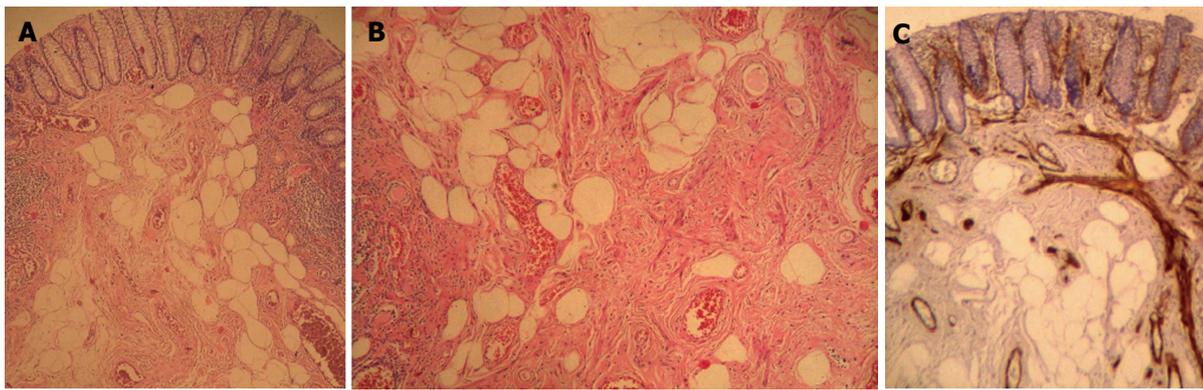
lesions<sup>[9,12,14,23,24]</sup>, are diagnosed generally by postoperative histopathologic evaluation or after polypectomy. Radiologic techniques such as barium radiographs, enteroclysis, abdominal ultrasound, computed tomography (CT), and magnetic resonance imaging (MRI) can demonstrate the lesion in the gastrointestinal tract before histopathologic diagnosis<sup>[24]</sup>.

## CASE REPORT

A 70-year-old woman admitted to the hospital with complaints of abdominal distension and constipation. She had no abdominal pain, rectal bleeding or weight loss. Her total blood count, serum biochemistry, sedimentation rate and abdominal ultrasound were normal. A polyp (1 cm in diameter) with lobulated and degenerated hyperemic mucosa and normal peduncle was seen at proximal rectum. Polypectomy was performed by endoscopic polypectomy snare and no complication occurred. Angiolipoma was diagnosed after histopathologic evaluation in mature adipose tissue and proliferous blood vessels (Figure 1). There was no fibrin thrombus and no muscle structure in histologic examination with HMB45 (monoclonal mouse anti-human melanosome, Dako, USA) and anti-smooth muscle antibody (mouse anti-human primary alpha-smooth muscle actin, Dako, USA) tissue stainings. There were no other lesions in total colonoscopy and esophagogastroduodenoscopy. No photograph of the polyp was taken before resection because externally there was no special difference in adenomatous polyps with degenerated surface mucosa.

## DISCUSSION

Angiolipomas are benign lipomatous lesions, which were first defined by Bowen<sup>[29]</sup> in 1912 and differentiated from lipomas histopathologically by Howard in 1960<sup>[30]</sup>. The lipomatous benign lesions are seen as musculoskeletal masses especially in soft tissues and bones<sup>[1,2]</sup>. They are generally subcutaneous and capsulated, about 50% patients feel painful on palpation, and seen in young adult males in their 2nd or 3rd decades of life<sup>[1,2,24]</sup>. They are also localized in neck<sup>[4,31]</sup>, maxillofacial region<sup>[32-35]</sup> and non-subcutaneous tissues such as epidural<sup>[36]</sup>, spinal<sup>[37-39]</sup>, and suprasellar tissues<sup>[40]</sup>. Lipomas are seen more frequently, but angiolipomas are rare lesions<sup>[1,5]</sup>. Gastrointestinal angiolipoma is also a rare pathological condition<sup>[1,24]</sup>. The other uncommon localizations of angiolipomas are the renal tissue<sup>[41-44]</sup>, adrenal tissue<sup>[45,46]</sup>, breast<sup>[47]</sup>, thyroid glands<sup>[48]</sup>, parotid gland<sup>[49]</sup>, mediastinum<sup>[50]</sup>, orbits<sup>[51]</sup>, myocardium<sup>[52]</sup> and scrotum<sup>[53]</sup>.



**Figure 1** Histopathologic appearance of small rectal pedunculated angiolipoma (macroscopically 7 mm x 5 mm x 4 mm in diameter with normal mucosal appearance). **A:** Proliferated blood vessels and adipose cells in mature adipose tissue in submucosa of colon at small magnification (light microscope, HE); **B:** Proliferated vascular tissues and mature adipose tissue in submucosa. There was no fibrin thrombus in blood vessels (light microscope, HE); **C:** SMA staining of angiolipoma.

**Table 1** Gastrointestinal angiolipomas defined in literature

Literature	Localization	Case (age/sex)	Polyp size	Polyp peduncle	Symptom	Preoperative diagnostic imaging methods (except endoscopic procedure)	Therapy	Follow-up (F/U)/ recurrence
Jensen EH <i>et al</i> <sup>[9]</sup> 2006	Esophagus	85 yr/M	39 mm x 25 mm	Pedunculated large polyp	A 2-mo dysphagia to solids foods, complete esophageal obstruction; had prolapsed to the level of the GE junction	CT, EUS	Anterior esophagotomy, transection of polyp using an endoscopic stapling device (open surgical excision)	No F/U
DeRidder Ph <i>et al</i> <sup>[10]</sup> (1989)	Stomach	59 yr/M	6 mm	Submucosal well demarcated mass	Chronic occult GI bleeding		Laparotomy-excision	12 mo/No recurrence
McGregor DH <i>et al</i> <sup>[11]</sup> (1993)	Stomach	69 yr/M	50 mm x 40 mm x 20 mm	Gastric submucosal mass on great curvature, without peduncle	Chronic hemorrhage and severe anemia		Exploratory laparotomy, mass resection	30 mo/No recurrence
Hunt J <i>et al</i> <sup>[12]</sup> (1996)	Stomach	27 yr/F	80 mm x 55 mm x 45 mm	Large polypoid mass with large peduncle	Suggestive of intussusception through the pylorus, acute GI hemorrhage		Exploratory laparotomy, distal partial gastrectomy, Billroth I gastric reconstruction, resection of polyp	not given/No recurrence
Mohl W <i>et al</i> <sup>[14]</sup> (2004) <sup>2</sup>	Duodenum, Colon	The first was 66 yr/M, the second was 75 yr/F (2 pts with 1 duodenal and colonic A-L other duodenal A-Ls)	1st-10 mm located near papilla of Vater, and 2nd-23 mm at the upper duodenal knee	Both with peduncle	1st acute, 2nd chronic GI bleeding Upper GI bleeding due to duodenal A-L		Endoscopic snare polypectomy for 2 duodenal and for 1 colonic A-Ls	not given/No recurrence
Jung IS <i>et al</i> <sup>[15]</sup> (2004)	Duodenum	60 yr/F	35 mm x 4 mm	With peduncle	Dyspepsia for 6 mo	EUS	Endoscopic polypectomy	No F/U
Kaneko T <i>et al</i> <sup>[16]</sup> (1996) <sup>1</sup>	Meckel's diverticulum accompanied A-L				Intussusception			
Ferrozzi F <i>et al</i> <sup>[3]</sup> (1998)	Ileal				With tuberous sclerosis	CT		

Manner M <i>et al</i> <sup>[17]</sup> (2001)	Proximal ileum	71 yr/F	38 mm	With peduncle	Occult bleeding, ileoileal intussusception	US, CT	Small bowel resection	
Kwak HS <i>et al</i> <sup>[18]</sup> (2003)	Small bowel/proximal ileum	75 yr/M	30 mm	Intraluminal lobulated polypoid mass with peduncle	Epigastric discomfort, loss of appetite, weight loss	Enteroclysis, MRI	Surgery	No F/U
Aouad K <i>et al</i> <sup>[19]</sup> (2000)	(Bauhin valve) ileocecal valve				Gastrointestinal hemorrhage			
Kato K <i>et al</i> <sup>[20]</sup> (1999) <sup>1</sup>	Ileocecal valve	69 yr/M	52 mm x 50 mm x 40 mm	Without peduncle, a submucosal smooth surface mass A-L	3-d right lower quadrant abdominal pain	Contrast enhanced abdominal CT	Laparoscopy-assisted ileocecostomy, and a side-to-side anastomosis extracorporeally (a minimally invasive laparoscopic technique)	5 yr/No recurrence
Saroglia G <i>et al</i> <sup>[21]</sup> (1996)	Ileocecal valve	55 yr/M	55 mm	Submucosal mass	Invagination	Barium contrast graphy	Surgery	No F/U
Vandamme J <i>et al</i> <sup>[22]</sup> (1964)	Descending colon	43 yr/M	150 mm x 40 mm	With peduncle	GI bleeding and non painful sub-obstruction by invagination	Barium contrast graphy	Surgery, colon resection	not given/No recurrence
Okuyama T <i>et al</i> <sup>[23]</sup> (2002)	Sigmoid colon	49 yr/M	65 mm x 23 mm	Pedunculated polyp with smooth surface	Asymptomatic, during routine exam positive fecal blood	Double contrast enema, enhanced CT	Hemostatic clip and endoscopic electro-surgical polypectomy	No F/U
Chen YY <i>et al</i> <sup>[24]</sup> (2005)	Transvers colon	70 yr/M	50 mm	With peduncle	Colonic obstruction	US, Abdominal CT, Colonic barium enema, colonoscopic examination	Surgical segmental resection	2 yr/No recurrence
Kacar S <i>et al</i>	Rectum	70 yr/F	10 mm	With peduncle	Asymptomatic	-	Polypectomy performed by endoscopic polypectomy snare	6 mo/No recurrence Under F/U

<sup>1</sup>Abstract of reference (full text not available); <sup>2</sup>Three lesions of angiolipoma in two patients, were defined. GE: Gastroesophageal; A-L: Angiolipoma.

In the musculoskeletal system, angiolipomas are 70% multiple and 10% familial<sup>[5]</sup>. It can have an autosomal dominant penetrance<sup>[1,22]</sup>. The mode of inheritance in GIS is still not clear<sup>[54]</sup>.

Histologically, it is comprised of mature adipose tissue and proliferated vascular tissues. It can be classified as predominantly lipomatous or angiomatous type, based on the ratio of adipose tissue and vascular tissue composition<sup>[1,2,5]</sup>. Fibrin thrombus due to microtraumas is generally seen in angiomatous lesions<sup>[5,11,20,23,48]</sup>. Angiolipomas localized in muscle and cutaneous tissue can have two different behavior patterns: infiltrative or non-infiltrative<sup>[5]</sup>. But the gastrointestinal cases defined in the literature have been non-infiltrating and showed no recurrence during follow-up periods (Table 1).

Eight maxillofacial angiolipomas had been reported in the literature until 1989<sup>[7]</sup> and 17 head and neck angiolipomas until 1999<sup>[4]</sup>. There are also cases in the literature of oropharyngeal angiolipomas including tongue, cheek mucosa and palate<sup>[4,6-8]</sup>.

Angiolipomas localized in the GIS or liver. Although hepatic localization of lipomatous tumors is seen more frequently than gastrointestinal tract, hepatic angiolipomas are also rare lesions in this group<sup>[27,28,55,56]</sup>. Most lipomatous

lesions of the liver<sup>[55,56]</sup> and hepatic angiolipomas are associated with tuberous sclerosis complex<sup>[26]</sup>.

Seven cases of angiolipoma had been reported in the gastrointestinal tract in the literature up to 2005<sup>[24]</sup>. To the best of our knowledge, 17 cases of gastrointestinal angiolipoma have been reported to date (1 esophageal<sup>[9]</sup>, 3 gastric<sup>[10-12]</sup>, 2 duodenal<sup>[14,15]</sup>, 4 small bowel<sup>[3,16-18]</sup>, 3 ileocecal valve<sup>[19-21]</sup>, and 4 colonic)<sup>[14,22-24]</sup>. Based on these reports, our case would be the first angiolipoma defined in the rectum.

There are syndromes accompanying angiolipomas, such as familial angiolipomatosis, which is rare and benign with mostly autosomal dominant inheritance and multiple angiolipomas of the extremities and trunk<sup>[57]</sup>. Birt-Hogg-Dube syndrome is an autosomal-dominant condition characterized by fibrofolliculomas, trichodiscomas, and acrochordons<sup>[58]</sup>. There is only one ileal angiolipoma-associated tuberous sclerosis in the literature<sup>[3]</sup>. Hunt *et al*<sup>[12]</sup> described a case with solitary gastric Peutz-Jeghers polyp and angiolipoma presenting as acute hemorrhage.

Angiolipomas are diagnosed generally as single lesions in the gastrointestinal tract, but Mohl *et al*<sup>[14]</sup> described a case of both colonic and duodenal angiolipoma. Angiolipomas in the gastrointestinal tract, as shown in Table 1, can be sessile<sup>[10,11,18,20]</sup> or pedunculated polypoid

Table 2 Histopathological findings of cases in literature

Literature	Histopathological findings under light microscope
Jensen EH <i>et al</i> <sup>[9]</sup>	A submucosal polypoid mass, consisting of benign lipomatous tissue mixed with vascular channels, consistent with benign angiolipoma
DeRidder <i>et al</i> <sup>[10]</sup>	Submucosa contained a well demarcated nodule composed of fat, extending from the under surface of the muscularis mucosa to the deep margin of biopsy. There were separate, well developed small arteries and veins
McGregor D <i>et al</i> <sup>[11]</sup>	A circumscribed, lobular, pinkish-tan soft ovoid mass showing proliferation of mature adipose tissue and vascular tissue surrounded by a thin to focally thick fibrous capsule in the submucosa, no definite fibrin thrombi
Hunt J <i>et al</i> <sup>[12]</sup>	A solitary Peutz-Jeghers-type polyp showing typical glandular epithelium, overriding a 5 cm gastric angiolipoma
Mohl W <i>et al</i> <sup>[14]</sup>	2 submucosal duodenal angiolipomas, one with large central vessel and one colonic angiolipoma
Yung IS <i>et al</i> <sup>[15]</sup>	A submucosal lesion composed of mature adipose tissue and small vessels with fibrin thrombi within the vascular channels
Manner M <i>et al</i> <sup>[17]</sup>	An angiolipoma with 20% vascular component and mucosal ulceration
Kwak HS <i>et al</i> <sup>[18]</sup>	Mature, highly vascular adipose tissue consistent with angiolipoma
Kato K <i>et al</i> <sup>[20]</sup>	A circumscribed proliferation of mature adipose and vascular tissue surrounded by a thin to focally thick fibrous capsule in the submucosa, with fibrin thrombi in small capillaries
Saroglia G <i>et al</i> <sup>[21]</sup>	Mature adipose and vascular tissue
Vandamme J <i>et al</i> <sup>[22]</sup>	Angiolipoma with multiple small vessels and adipose tissue
Okuyama T <i>et al</i> <sup>[23]</sup>	Encapsulated by a thin layer of connective tissue, arising in the submucosa and composed of mature adipose tissue and proliferating capillaries, many fibrin thrombi in capillary lumen
Chen YY <i>et al</i> <sup>[24]</sup>	Encapsulated by a thin layer of connective tissue arising from the submucosa, histologically was comprised of mature adipose tissue and proliferative blood vessels
Kacar S <i>et al</i>	Proliferated submucosal blood vessels and mature adipose tissue. There was no fibrin thrombus

lesions<sup>[9,12,14,23,24]</sup>, and the cases in the literature are generally diagnosed based on abdominal pain, obstruction, invagination, intussusception, and acute or chronic bleeding.

Diagnosis of GIS angiolipomas can be made radiologically via contrast barium enema, enteroclysis, abdominal ultrasound, endoscopic ultrasound, abdominal tomography, or MRI before the polypectomy or resection if the lesion is large<sup>[9,15,17,18,20,23,24]</sup>. Large lesion size and the symptom are the main factors in the radiologic diagnosis, but final diagnoses are done histologically. There is filling-defect in gastrointestinal lumen detected by barium enema<sup>[24]</sup> and enteroclysis<sup>[18]</sup>, hyperechoic lesion in transabdominal ultrasonography<sup>[17,24]</sup>, submucosal lesion in gastrointestinal wall in endosonography<sup>[9]</sup>, hyperechoic lesion in CT<sup>[3,9,14,17,20,23]</sup>, and central high signal intensity with peripheral iso-signal intensity on T1-weighted in-phase images in MRI<sup>[18]</sup>.

Surgical resection is preferred to endoscopic resection

in the case of broad-based or pedunculated large polyps due to risk of perforation and bleeding<sup>[9,10,11,12,17,18,20,24]</sup>. For pedunculated polyps, injection of epinephrine, or use nylon loop or metal hemostatic clip are suggested before polypectomy to decrease the risk of postoperative bleeding<sup>[14,23]</sup>. These patients can be treated by standard polypectomy. The level of urgency is also an important factor when choosing the most optimal treatment. In the case of intussusception<sup>[16,21]</sup>, obstruction<sup>[9,24]</sup> or acute bleeding<sup>[12,19,22]</sup>, emergent surgery is generally suggested. In some cases, minimally invasive laparoscopic techniques can be performed successfully. A cecal lesion was treated successfully with laparoscopy-assisted ileocecostomy and a five-year follow-up showed no recurrence in this patient<sup>[22]</sup>.

Our patient was diagnosed by histopathologic evaluation after snare polypectomy of a single, small polyp with thin peduncle in the rectum. There was no complication after polypectomy. The peduncle was histologically normal. There were proliferated blood vessels and mature adipose tissue in the submucosa and no fibrin thrombus was found. Mature adipose tissue and proliferated blood vessels are the diagnostic components as defined in the literature. Fibrin thrombus in the vascular component is a common finding<sup>[5,11,20,23]</sup>. The histopathological findings of the cases in the literature are given in Table 2.

The polyp was not responsible for the dyspeptic symptoms and constipation in our patient. Although the polyp could have caused rectal bleeding, no bleeding occurred in our patient. The lesion was thus considered as asymptomatic. The sigmoid angiolipoma in literature<sup>[23]</sup> was asymptomatic, but 65 mm in diameter. Our case is the first rectal, small, asymptomatic angiolipoma diagnosed incidentally in the gastrointestinal tract.

Although angiolipomas are benign lesions among adipose tissue neoplasms, additional staining techniques are required to distinguish from malignant lipomatous forms like angioliposarcoma<sup>[27,28]</sup>. The resection material of our patient was stained by HMB45 and anti-smooth muscle antibody, and found negative for muscle fibers. The differential diagnosis was made between angiolipoma and angioliposarcoma.

In conclusion, it is important to remember that submucosal, sessile, or polypoid lesions in the gastrointestinal tract with or without symptoms can be an angiolipoma, a lipomatous tumor, although it is quite rare.

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