

Angiolipofibroma: Case report and literature re-review

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World Journal of Advanced Research and Reviews, 2023, 17(03), 437–442

Publication history: Received on 24 January 2023; revised on 07 March 2023; accepted on 10 March 2023

Article DOI: <https://doi.org/10.30574/wjarr.2023.17.3.0377>

Abstract

Angiolipofibroma is an extremely rare triphasic mesenchymal lesion. It consists of mature adipose tissue, fibrous tissue, and vessels. Very few cases reported in the current literature. This report presents a case of a polyp-like mass in detected in routine colonoscopy screening. We also present comprehensive data of previously reported cases found throughout the world.

Keywords: Angiolipofibroma; Mesenchymal lesions; Colorectum; Polyps

1. Introduction

With the introduction of the colonoscopy screening for bowel carcinoma early detection, the frequency of 4.6% in 2001 increased to 14.2% after 2001 [1]. Moreover, in 2018, The American Cancer Society updated its screening guidelines and lowered the starting age for colonoscopy screening to 45 years [2]. With the increasing number of screenings, new definitions of rare lesions have also appeared.

Angiolipofibroma (ALF) is a triphasic lesion consisting of characteristic histology of fibrous tissue, mature adipose tissue and proliferated vascular component including blood vessels and lymph vessels of different sizes [3]. This triphasic pattern differentiates itself from the most common types of the other mesenchymal lesions such as lipoma, leiomyoma, and gastrointestinal stromal tumor that is composed of only one type of tissue [4].

Here we presented clinicopathologic and endoscopic features of this unusual submucosal mesenchymal lesion located in the cecum.

2. Case presentation

A 70-year-old female admitted to the hospital for routine colonoscopy screening. She had no complaint such as constipation, diarrhea, rectal bleeding, abdominal pain, or weight loss. Physical examination revealed no findings. Her laboratory findings were within normal limits. In colonoscopy examination a sessile polyp, 0,9 cm in diameter was seen in the cecum. It had round and smooth surface covered by hyperemic mucosa. There were no other lesions seen in the rest of the colon, ileum, and upper gastrointestinal system. Endoscopic polypectomy was performed with snare and no complication occurred afterwards.

The polypectomy specimen admitted to the pathology laboratory was a circumscribed round polyp with a peduncle. It was 0.9 cm in diameter, of soft consistency covered by normal-looking grayish mucosa. The peduncle was measured 0.1 cm. The mucosal surface looked normal. No hemorrhage or necrosis was seen. The tissue was submitted to the routine pathology processing. On histological examination, overlying mucosa was seen normal. Beneath the muscularis mucosa a well demarcated but not encapsulated submucosal proliferation composed of mature adipose tissue, fibrous tissue

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and proliferated large and small vessels of varying nature (lymphatic, capillaries, small veins, venules, arterioles, and small arteries with thick wall) were seen (Figure-1,2).

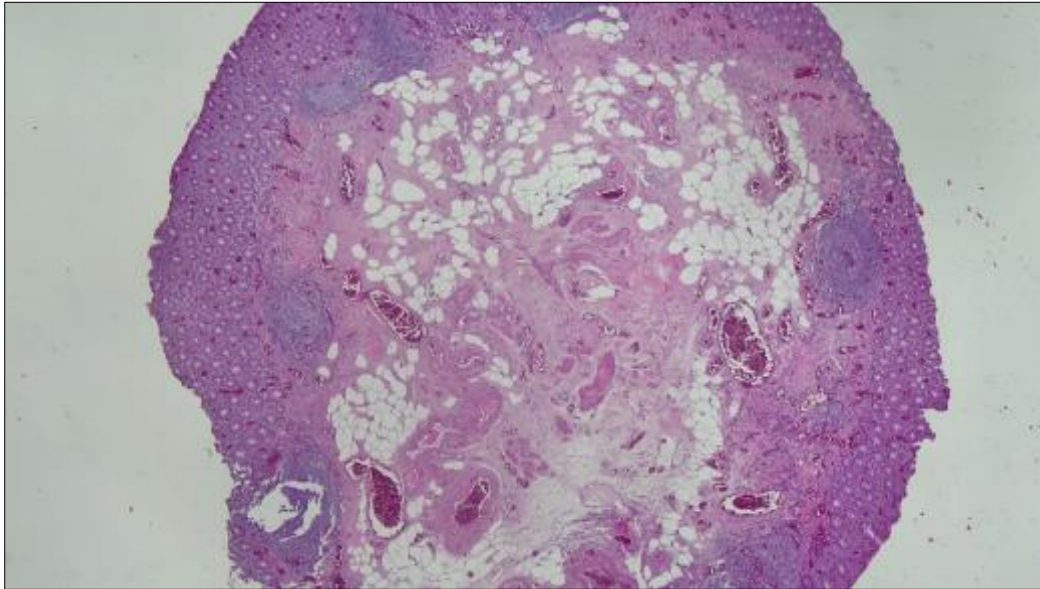


Figure 1 Polypoid tissue with a smooth surface covered with colonic mucosa is seen. There are round empty spaces corresponding to adipose tissue and vessels filled with erythrocyte in the center. Also note pink area that is fibrous tissue (H&Ex2)

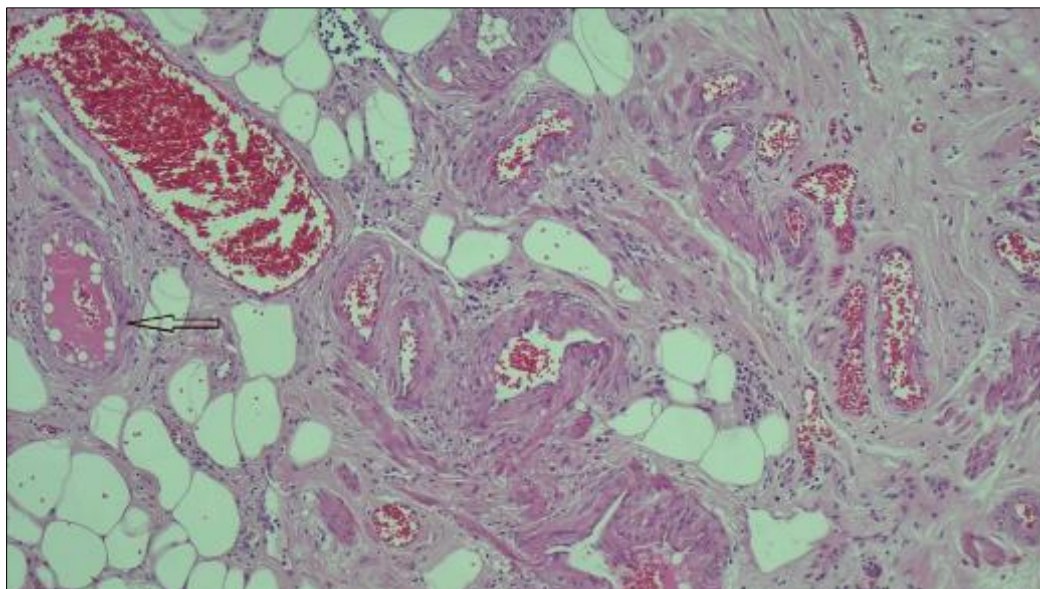


Figure 2 Closer examination of the stroma. There is a lymphatic vessel in which the lumen is filled with lymphatic fluid, not erythrocyte (arrow). Fibrous tissue is better seen at upper right (H&Ex10)

Thrombosis or vasculitis were not seen in the polyp stroma. Also, there was no nuclear atypia. Trichrome stain highlighted fibrous tissue (Figure-3).

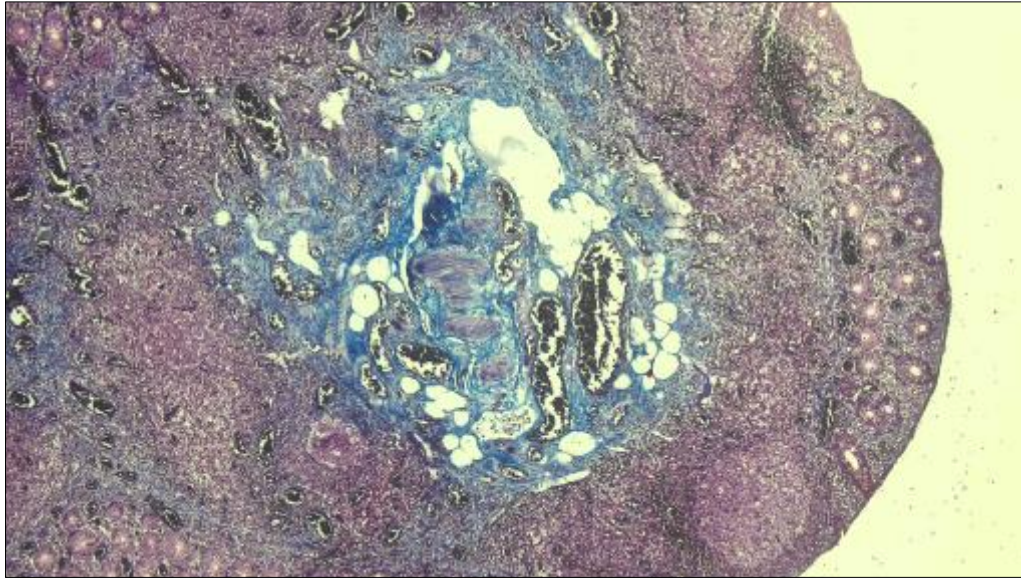


Figure 3 Fibrous tissue is highlighted by Trichrome stain, which colors it green (H&Ex4)

Desmin, D2-40 (Podoplanin), CD31, and HMB45 were applied immunohistochemically. While Desmin demonstrated arterial walls, D2-40 and CD31 demonstrated lymphatic and vascular endothelium respectively (Figure-4). HMB45 was negative.

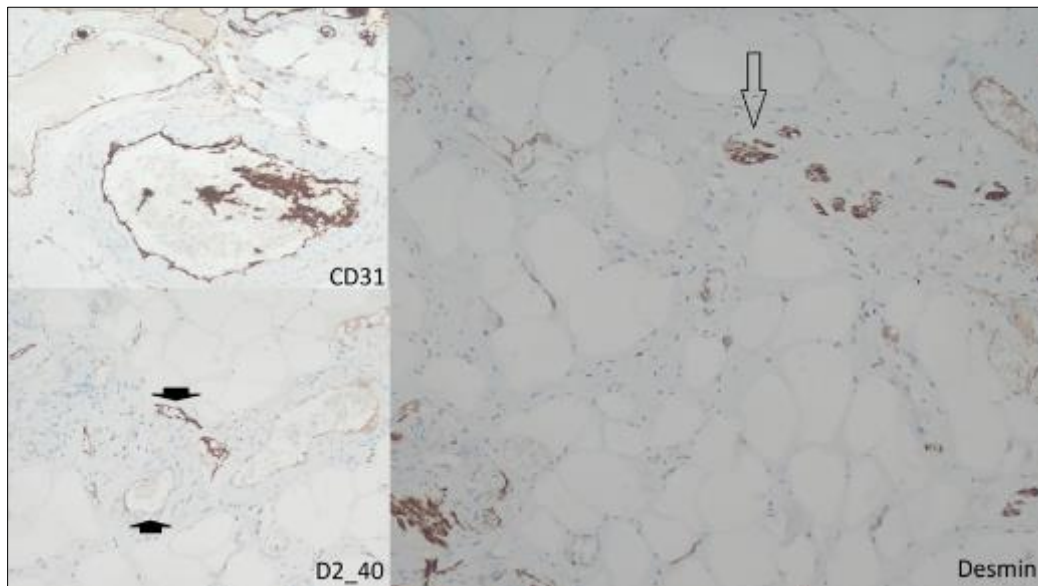


Figure 4 Fibrous tissue is negative around adipose tissue with Desmin. Desmin, which is seen as brown color here demonstrates muscle around arterial walls (arrow). Upper left: Vascular endothelium is seen brown color with CD31; and bottom left: While lymphatic endothelium is brown (arrowhead), vascular endothelium is negative with D2-40 (arrowhead) (H&Ex10)

3. Discussion

While most colonic polyps are originated from the mucosa, some of them are of mesenchymal type. Majority of these mesenchymal type polyps are composed of one single tissue type, i.e., adipocyte, neuron, and endothelium and called as lipoma, neuroma, and hemangioma. On the other hand, a small subset of them is composed of two or more tissues such as angiolipoma, fibrolipoma, angiomyolipoma. These mixed mesenchymal lesions are extremely rare.

Angiolipofibroma, as a neoplasm is seen in different parts of the body [5-15]. In the gastrointestinal tract, it was reported in buccal mucosa [16], oral cavity [17], esophagus [18], distal duodenum [19], ileum [20], cecum [3,20], colon and rectum [21,22, Table-1].

Table 1 Clinicopathological characteristics of gastrointestinal Angiolipofibroma

case	Age	sex	complaint	localization	Diameter (cm)
1 ¹⁶	59	F	Feeling of polyp-like mass	buccal mucosa	1,2
2 ¹⁷	NA	NA	NA	oral cavity	NA
3 ¹⁸	62	NA	dysphagia	esophagus	20x5
4 ¹⁹	73	M	Recurrent GIS bleeding	distal duodenum	12x2
5 ²⁰	54	M	Anemia and melena	Ileocecal valve	3
6 ²⁰	45	F	diarrhea	cecum	NA
7 ²⁰	53	F	NA	descending colon	NA
8 ²⁰	53	M	NA	sigmoid colon	0,7
9 ²⁰	57	M	Crohn's disease	cecum	0,8
10 ²⁰	51	F	NA	descending colon	1,2
11 ²⁰	31	M	Crohn's disease	transverse colon	0,7
12 ²⁰	34	F	diarrhea	rectum	0,6
13 ²⁰	23	F	Crohn's disease	cecum and ascending colon	0,8 multiple
14 ²⁰	73	F	Herniorrhaphy complication	Small intestine	2,3
15 ²⁰	72	M	Crohn's disease	Small intestine	1,5
16 ³	66	M	abdominal pain	cecum	1
17 ²¹	58	F	mass protruded from anus	sigmoid colon	8x3
18 ²²	NA	NA	NA	transverse colon	NA
Present case	70	F	unremarkable	cecum	0,9

The etiology is not clear, but it is believed that it is not a true neoplasm but disorganized overgrowth of mature cells and tissues normally present in the colonic submucosa. Agostini-Vulaj et al suggested that it may be reactive, as 4 out of 11 of their cases had Crohn's disease, previously damaged mucosa [20]. Our patient has no Crohn's disease.

Differential diagnosis must include lipomatous and vascular tumors of gastrointestinal tract. These are vascular lipoma, fibrolipoma, angiomyolipoma. There are currently no defined criteria for the quantity of each component necessary to make the diagnosis. Barron et al divided their 404 benign colonic lipomatous lesions as to whether it contained more than 30% vascular or fibrous tissue [23]. They did not define triphasic lesion. In our case fibrous tissue occupied less than 30% and we believe that some of their vascular lipoma and fibrolipoma cases are actually ALF, since scarce fibrous tissue can be overlooked while naming it.

Other entities one should keep in mind in the differential diagnosis include angiolipoma, which has fibrin thrombi in capillary-sized vessels instead of arterial and venous vessels; CD31 positive inflammatory fibroid polyp; and HMB45 positive angiomyolipoma. The case presented by Aye KS et al contained proliferated smooth muscle spindle cells with eosinophilic cytoplasm embedded in loose myxoid stroma [21]. This could be angiolipoma as they didn't apply HMB45 stain.

Like all lipomas, ALF is treated by surgery. Recurrence was not reported [5,16,22].

4. Conclusion

While gastrointestinal mesenchymal lesions are very rare, Angiolipofibroma is the rarest one among these groups. Its characteristic triphasic histology helps to aid its differential. Cases in the literature are approximately 1 cm in diameter, preferably located in the colon and seen in all age groups.

Compliance with ethical standards

Acknowledgments

Authors thank Dr Ayhan Hilmi Cekin for colonoscopic procedure.

Disclosure of conflict of interest

The authors declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Statement of ethical approval

The present case report and literature re-review does not contain any studies performed on animals/humans subjects by any of the authors.

Statement of informed consent

Informed consent was obtained from the case presented here.

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