

## **Angiolipofibroma of the Right Colon: A Rare Type of Submucosal Polyp**

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**Colorectal hamartomatous polyps originating from the submucosa represent a very small percentage of all colonic polyps. Mesenchymal hamartomatous polyps are formed by more than one type of mesenchymal tissue. We report the pathological findings of an unusual presentation of a polypoid proliferation of vascular, mature fatty and fibrous connective tissue. The histological findings support the diagnosis of a hamartoma rather than a true neoplasm.**

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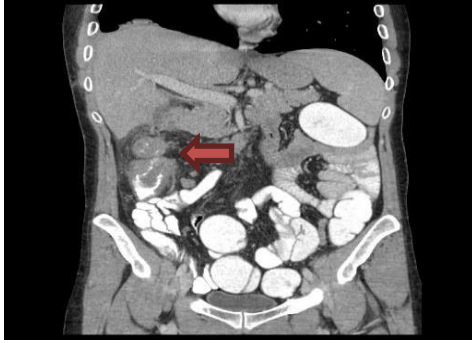
It is well-known that most colonic polyps are of epithelial origin and usually originate in the mucosa; however, a very small subset of polyps originates from the abnormal proliferation of submucosal mesenchyma. Common types include lipomas, leiomyomas, vascular, and gastrointestinal stromal tumors (GISTs)<sup>1</sup>. The majority of reported submucosal polyps are composed of a single type of tissue, such as lipoma, hemangioma, and leiomyomas. Therefore, hamartomatous submucosal polyps formed by more than one type of tissue are extremely rare.

The aim of this report is to present an unusual case of polypoid proliferation of vascular, mature fatty and fibrous connective tissue.

### **THE CASE**

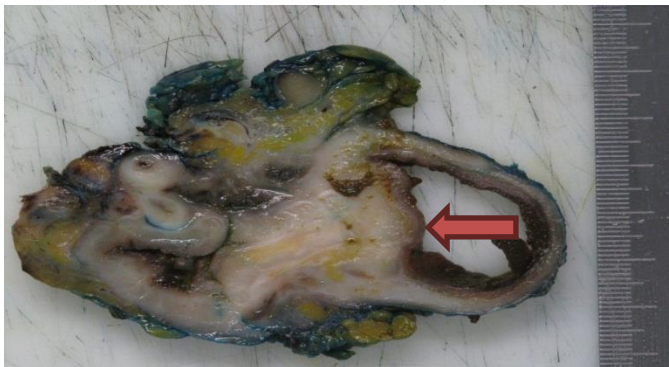
A thirty-year-old Bahraini male complained of right-sided abdominal pain radiating centrally for three years. The pain was progressive in nature, 8 on a scale of 10 in severity and was aggravated by spicy food. The patient had a history of weight loss (from 114 kg to 79 kg) in the last two months. He suffered from vomiting and diarrhea 4 to 5 times a day but no history of loss of appetite or rectal bleeding.

The patient had a history of long-standing Crohn's disease and was on medications. CT abdomen and pelvis showed an irregular polypoid wall thickening of the right side colon involving the ileocecal junction and extending to the hepatic flexure and is associated with peri-colic mesenteric fat stranding and mesenteric adenopathies, see figure 1.

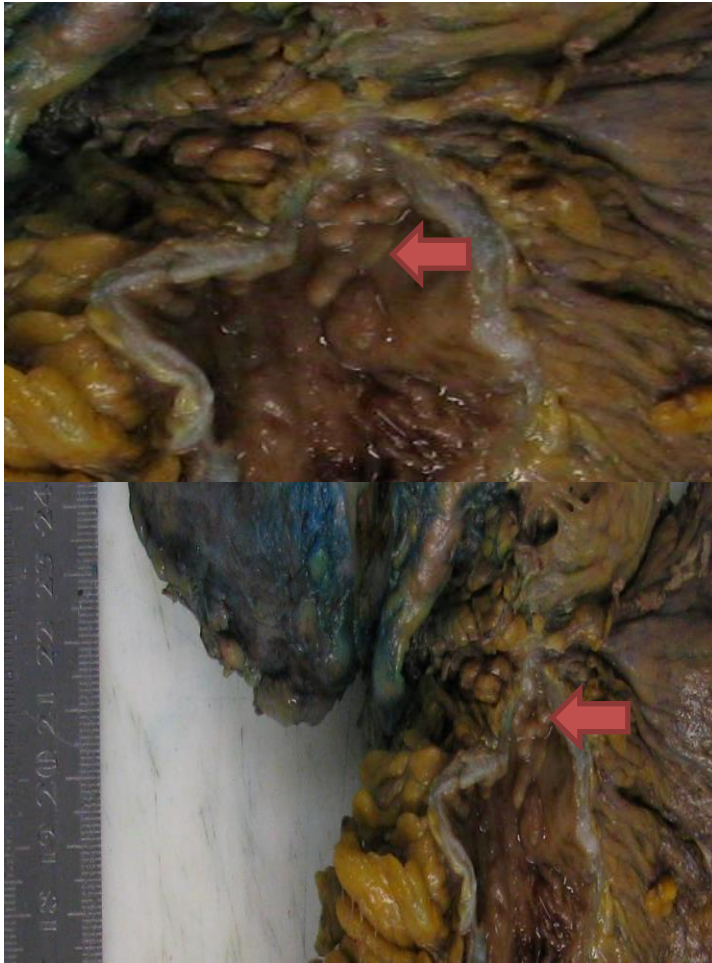


**Figure 1: CT Abdomen Shows Wall Thickening of the Right Colon**

The specimen was a right hemicolectomy consisting of 70 mm terminal ileum, 190 mm of cecum and ascending colon and 50x7 mm appendix. The terminal ileum segment had cobblestone-like mucosal appearance, severely congested, scarred and variably thickened. The serosal surface of the cecum showed marked fat puckering; mainly at the posterior part and on serial sectioning, 60 mm (length) x 120 mm (diameter) cecal mass was found. The cecal mass is formed by marked mural fibromuscular hypertrophy of the ileocecal valve and cecum in which scattered inflamed, possibly perforated diverticulae were noted, see figure 2. The ascending colon revealed several sessile and pedunculated yellowish-gray mucosal polyps, up to 30 x 15 mm, see figure 3.



**Figure 2: Marked Mural Fibromuscular Hypertrophy**

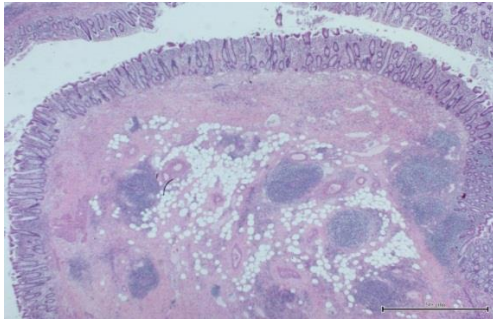


**Figure 3: Ascending Colon Revealed Several Sessile and Pedunculated Yellowish Grey Mucosal Polyps**

The appendix was unremarkable, and the mesenteric fat was relatively unaffected. Several mesenteric lymph nodes were retrieved, and the largest measures 14 mm. No gross evidence of malignancy was found.

A 40x12x9 mm piece of colonic wall doughnut (an anastomosis with a circular stapling device) was also included. Several sections were taken and fixed in 10% formalin, embedded in paraffin and stained with hematoxylin and eosin.

Microscopy revealed the following: the ascending colon polyps were benign angiolipofibromas, composed of mixed mature mesenchymal tissue elements, including fibrovascular tissues, strands of smooth muscle, scattered islands of fat and reactive lymphoid follicles, see figure 4.



**Figure 4: Polyp Section Showing Mesenchymal Tissue Elements, Including Fibrovascular Tissues, Strands of Smooth Muscle, Scattered Islands of Fat and Reactive Lymphoid Follicles**

The cecal mass is formed by a well-established diverticulae with evidence of diverticulitis. There was an abscess formation possibly due to rupture of a diverticulum. There is also marked transmural inflammation including apparent eosinophils, leukocytosis, and scattered reactive lymphoid follicles. There was a prominent neuromuscular hypertrophy with substantial subserosal involvement.

The appendix is relatively unaffected. All mesenteric lymph nodes were reactive, and no granulomas were identified. Neither dysplasia nor neoplasia was found. The terminal ileum showed features of non-specific chronic active ileitis with secondary mucosal edema and variable ischemia.

Scattered superficial fissures, crypt architectural distortion and occasional crypt abscesses are also noted in the background and despite the absence of granulomas; the overall appearance is of chronic inflammatory bowel disease (Crohn's disease).

## **DISCUSSION**

It is well-known that the gold standard procedure for colonic screening is colonoscopy to detect colorectal cancer or polyps<sup>2</sup>. In this case, we found submucosal polyps composed of structures normally present in the submucosa supporting the diagnosis of a hamartomatous reaction rather than a true neoplasm.

Angiolipofibromas are rare in the gastrointestinal tract. Few cases were previously reported; the first reported case was a large pedunculated esophageal polyp from a 62-year-old patient who presented with dysphagia and treated by surgical excision<sup>3</sup>. The second case was an excision of a 120 mm finger-like polypoid lesion extending from the distal duodenum to the proximal jejunum in a 73-year-old male who presented with recurrent gastrointestinal bleeding<sup>4</sup>. Unlike the previous cases, the lesion in our case was much bigger and located in the colon. Demir et al described a similar lesion originating in the mesocolon of the sigmoid, composed of vascular proliferation in a fibro-fatty tissue background; it was published in the name of "angiolipomatous mesenchymal hamartoma". Groisman et al described a cecal submucosal polyp formed by adipose tissue, fibrous tissue, and vascular proliferation and lymph vessels in variable sizes under the name "angiolipofibroma of the cecum"<sup>5,6</sup>.

Other differential diagnoses considered in such cases are neoplasms of adipose or vascular origin arising in the gastrointestinal tract. Nineteen angiolipomas were documented, one in

the esophagus, three in the stomach, eight in the small intestine, and seven in the colon<sup>7-25</sup>. However, the usual histological appearance of angiolipoma is the branching networks of capillary-type proliferating vessels of focal fibrin thrombi; our case included vascular proliferation without fibrin thrombi formation. Seven angiolipomas were documented in the colon<sup>26-33</sup>. It could be differentiated from our tumor by lack of fibrous proliferation.

Another common differential diagnosis is lipomatous neoplasms formed by adipose tissue and commonly found in the right colon<sup>34</sup>.

Vascular lesions seen in the gastrointestinal tract formed solely by vascular proliferation include lymphangiomas, hemangiomas, hemangiolymphangiomas, and congenital vascular malformations<sup>35</sup>. All the above mentioned vascular polyps which lack lipomatous tissue are not considered in our differential diagnosis. Our case is different because it showed prominent fibrous tissue proliferation.

## CONCLUSION

**We report a rare case of submucosal mesenchymal hamartomatous polyposis, which we believe to be the first reported case in the colorectum in Bahrain. This lesion is of a benign hamartomatous origin which could be treated successfully by surgical excision.**

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