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A Rare Case of Transverse Colonic Cavernous Hemangioma: An Ambiguous Preoperative Diagnosis, An Inappropriate Biopsy, A Fortunate Outcome, One Good Lesson and Some Serious Reflections

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Abstract

Background: Cavernous hemangioma of colon is an uncommon vascular malformation, usually presenting with painless rectal bleeding and anemia. Examinations of computerized tomography, magnetic resonance image and colonoscopy are helpful to establish a definitive diagnosis.

But sometimes however, it is difficult to recognize this kind of tumor due to lack of specific findings. Surgical treatment is often the first choice. Other treatments like sclerotherapy, electrocautery, radium implantation or cryosurgery have also been described.

Case presentation: We reported one case of cavernous hemangioma located in transverse colon treated by laparoscopic segmental colonic resection. The diagnosis we suspected before the operation was gastrointestinal stroma tumor. But the paraffin pathology proved it a hemangioma. Fortunately, protractor biopsy did not lead to hemorrhage and the hemangioma was resected successfully.

Conclusion: Colonic hemangiomas are extremely rare in clinical practice. Some of them have untypical radiological and endoscopic manifestations, which make preoperative diagnosis ambiguous. Endoscopic biopsy should be cautious. Consultation with experienced radiologists, endoscopic physicians and multidisciplinary team is beneficial.

Choice of the surgical method depends on intraoperative judgement and the result of frozen pathology. And paraffin pathology decides whether carry out an extended or radical colectomy afterwards.

Keywords: Transverse colon; Cavernous hemangioma; Laparoscopic surgery; Intraepithelial neoplasia

Abbreviations

CT: Computed Tomography; GI: Gastrointestinal; HE: Hematoxylin and Eosin; IHC: Immunohistochemistry; MRI: Magnetic Resonance Image

Introduction

Colonic cavernous hemangioma is a rare benign vascular tumor. This disease was first described in 1839 by doctor Philips and only less than 200 cases have been reported since then [1,2]. In most cases, it happens in rectosigmoid and transverse colonic localization is extremely uncommon.

This special type of vascular tumor in the gastrointestinal tract is considered as a progressive intestinal hamartoma [2-6], on the border between malformations and tumors.

Recurrent, painless rectal bleeding and anemia are the common symptom. Accurate diagnosis is important because reckless biopsy can cause severe hemorrhage [7].

Case Study

A 56-year-old man presented with a two-month history of rectal bleeding. The endoscopic report described a hemispherical mass located at the transverse colon. Since endoscopic physicians suspected it as GIST by its shape **(Figure 1),** they take the biopsy. The pathologic result demonstrated tubular adenoma and low-grade intraepithelial neoplasia. Abdominal and pelvic computed tomography (CT) showed irregular thickening of colonic hepatic flexure wall (**Figure 2**).

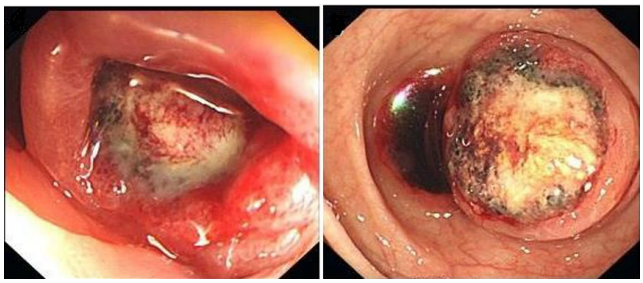

Figure 1 The hemispherical tumor locates at colonic hepatic flexure with ulceration on its surface. Distance between the lesion and anal margin is 75 cm.

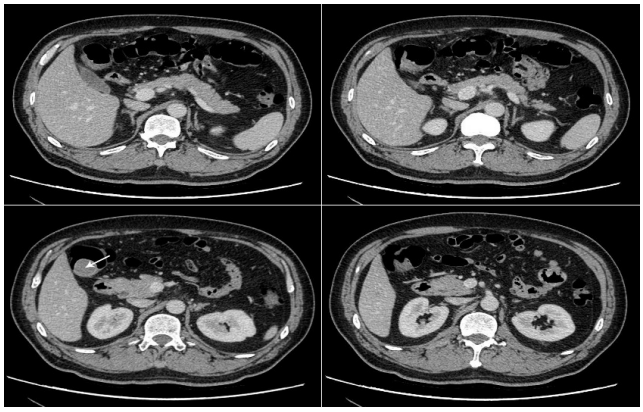

Figure 2 The colonic wall at hepatic flexure becomes irregular thickening and forms a nodule (white arrow) jutting out into the lumen.

After the multidisciplinary team (including surgeons, endoscopic physicians, pathologists and radiologists) consultation, the preoperative diagnosis was inclined to a benign lesion but hard to be excised by endoscopy. The final plan was resecting by surgery. Carbon nanoparticles were injected to the periphery of the lesion before the operation for the localization.

Laparoscopic exploration indicated the tumor was at the transverse colon close to hepatic flexure. We resected part of transverse and right colon, and then made an end to end anastomosis. The specimen extracted from the patients showed a 2.5 × 1.8 cm mass protruding into the lumen, with the ulceration on its surface **(Figure 3).**

Intraoperative frozen pathologic section report suggested significant inflammatory cells infiltration without cancer cell. The final paraffin pathologic diagnosis was polypoid cavernous hemangioma located at submucosa (**Figure 4**).

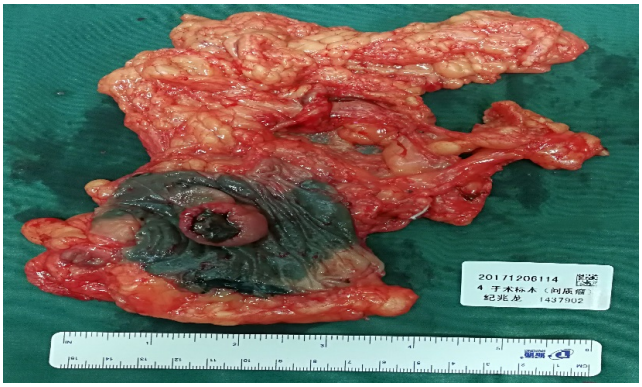

Figure 3 The resected specimen includes an 8.5 cm colonits mesentery and a part of omentum majus. Colonic wall is dyed black by carbon nanoparticles. The hemangioma, size of 2.5 × 1.8 × 1 cm, protrudes into the lumen with ulceration on its surface.

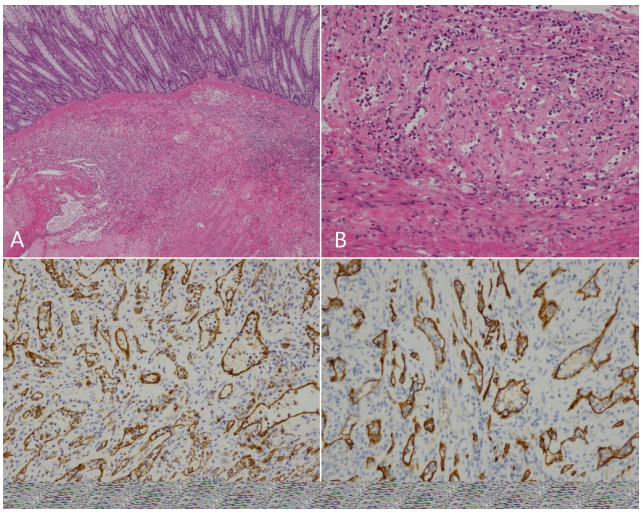

Figure 4 A: Hematoxylin and eosin (HE), 40x; B: HE, 200x; C: Immunohistochemistry (IHC) CD31 staining, 200x; D: IHC CD34 staining, 200x. The hemangioma, located at submucosa, is composed of dilated and varying in sized vessel lumens with large amounts of inflammatory cells infiltration. CD31 and CD34 staining are positive on the vascular endothelial cells.

Immunohistochemical staining showed: AE1/AE3 (-), CD34 (3+), Ki-67 (individual-), CD31 (3+), D2-40 (-), F8 (1+). The patient recovered with normal passage of gas and defecation on the 3rd and 6th day respectively. He discharged on the 7th day. The follow-up computed tomography after one month showed an intact and clear anastomotic stoma.

Discussion

Hemangiomas are rare causes of gastrointestinal bleeding. Incidence of this kind of vascular malformation in gastrointestinal (GI) tract is only 0.06% [8]. And GI

hemangiomas account for 0.3% of all GI tumors. This corresponds to 3–4% of all benign intestinal tumors [7,9]. Rectosigmoid colon is the most commonly involved area while the transverse colonic hemangioma is exceedingly rare. Histologically, gastrointestinal hemangiomas are classified into four types: capillary, cavernous, arteriovenous and mixed types.

Capillary and cavernous subtypes are the most frequent. The proliferations of small capillaries, which are made of thin-walled spaces lined by endothelial cells, form the capillary hemangioma. The cavernous hemangiomas consist of large spaces lined by single or multiple layers of endothelial cells [10]. GI arteriovenous hemangiomas are extremely rare and have only been described in the small intestine [11].

They consist of arteries and veins with significant intimal thickening and irregular dilatation of the veins [12]. Gastrointestinal hemangiomas can appear as isolated mass (like this case) or clusters of polypoid lesions. In some cases, they can even extend into the mesentery and adjacent organs.

Patients with colonic hemangioma present with painless recurrent rectal bleeding, melena or anemia. Other symptoms like abdominal or pelvic pain, obstruction, intussusception or perforation are relatively uncommon. The diagnosis is usually established on the basis of characteristic endoscopic manifestation, as bluish or deep red submucosal lesions, with dilated veins in the colonic wall. But sometimes, these lesions are hard to recognize. Computed Tomography (CT) is an effective non-invasive diagnostic method. It can show the image of phlebolith and provide critical information about the extent, invasion and extra-intestinal involvements of hemangioma. Magnetic Resonance Image (MRI) is another valuable and specific diagnostic tool. The typical imaging findings on MRI is bright signal intensity on T2-weighted images and intermediate signal intensity on T1-weighted images, and blood vessels and calcifications appear with a signal voided on T1- and T2-weighted images.

In our case, phlebolith was vacant on CT images **(Figure 2)** and the ulceration on the mass covered up the features of hemangioma thus, made the endoscopic diagnosis difficult. Biopsy was thought to be the most effective diagnostic method for most colorectal tumors but happens to be the contradiction of hemangioma due to its risk of massive hemorrhage. We had to admit that our reckless action of biopsy was inappropriate.

We did not take the possibility of colonic hemangioma into account because our team has never seen it before. This case was an excellent lesson for us. Physicians must keep clear minds and be cautious in the face of this kind of situation. Consultation with experienced radiologists and endoscopic physicians is helpful. Multidisciplinary team consultation can provide a direction for treatment. The nature of tumor, which decides the surgical method, could be further judged by intraoperative observation and frozen pathology. Like in our case, we made a colonic segmental resection since the frozen pathology ruled out the carcinoma. But frozen pathology has limitations. If paraffin pathology found infiltrated cancer cells, we should make a radical right hemicolectomy afterwards.

It is clear that surgical resection is the first choice of large or diffuse tumors. When there are extracolonic organs involvements, a good exploration of the abdominal cavity should be performed [13]. Endoscopic resection has sometimes been performed for those pedunculated, small, solitary hemangiomas. Other non-invasive treatments like sclerotherapy, electrocautery, radium implantation or cryosurgery can only provide temporary good outcomes. Angiography and embolization have been described as options to control acute bleeding, but bleeding recurrence is a common issue [14].

Conclusion

In summary, hemangiomas located at transverse colon are extremely rare. They are more commonly appeared at rectosigmoid area and usually present with painless rectal bleeding and anemia. Computed tomography, magnetic resonance image and endoscopy are helpful for diagnosis and endoscopic biopsy should be avoided. Surgery is the most effective way to cure this disease. Given the rarity of this kind of disease, it is hard to take it into account before the surgery especially when the radiological and endoscopic manifestation is atypical. So, when the preoperative diagnosis is ambiguous, consultation with experienced radiologists, endoscopic physicians and multidisciplinary team is helpful. Surgeons can decide resected method by intraoperative observation and the frozen pathology results. Paraffin pathology determines if it need a subsequent treatment.

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