CASE REPORT

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Hematochezia with Colonic Polypoid Angiodysplasia in a Young Female Patient

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A 18-year-old girl visited the hospital due to hematochezia. Colonoscopy revealed a 6-mm Yamada type II polyp with stigmata of bleeding, and a shallow ulcer on top was found at the cecum base. The polyp was removed by snare polypectomy, and hematochezia stopped thereafter. Angiodysplasia was diagnosed histopathologically. Generally, angiodysplasia appears as a flat or elevated, bright-red lesion on endoscopy, with a polypoid shape being extremely rare. This case is significant because the lesion occurred at the youngest reported age and was the smallest that has been reported, and is the only polypoid arteriovenous malformation to be discovered in the cecum. **(Gut and Liver 2008;2:126-129)**

Key Words: Hematochezia; Angiodysplasia; Polyp; Cecum; Young

INTRODUCTION

Arteriovenous malformation (AVM) of gastrointestinal tract is one of the significant causes of lower gastrointestinal bleeding, along with diverticulum, neoplasm, and internal hemorrhoid. AVM is characterized by indolent massive bleeding and chronic anemia that recur without any specific medical history or family history.[1] It is extremely rare in people under 50 years old.[2] Generally, angiodysplasia appears as a flat or an elevated, bright red lesion on endoscopy.[3] In addition, polypoid shape is extremely rare in colonic angiodysplasia. Recently, we experienced an extremely rare case of polypoid angiodysplasia of colon.

CASE REPORT

A 18-year-old girl came to the hospital after three times of hematochezia. She had no history of medical or surgical illnesses. Her vital signs were: blood pressure 113/80 mmHg, pulse rate 90 beats/min, and body temperature 36.5°C. On arrival, the level of hemoglobin was 8.7 g/dL and hematocrit was 26%. Other laboratory tests and physical findings were not remarkable. Upper gastrointestinal endoscopy showed chronic superficial gastritis without evidence of bleeding. As a consequence, colonoscopy was performed with an aid of Olympus video colonoscope (H260AL, Tokyo, Japan) to find the focus of gas-

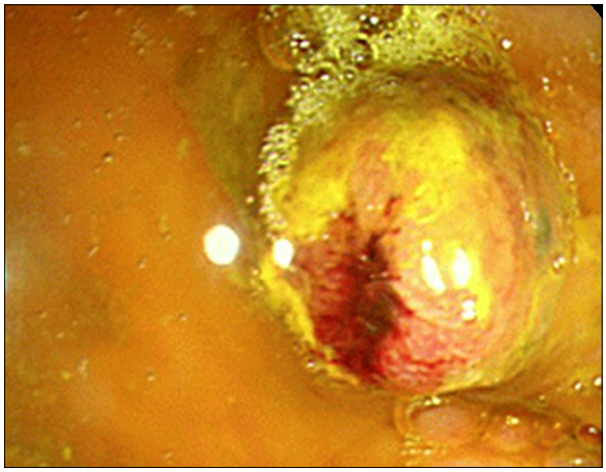

Fig. 1. Colonoscopic view of the polypoid lesion with stigmata of bleeding (6 mm, Yamada type II), with a shallow ulcer on top at the cecum base.

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trointestinal bleeding. A polypoid lesion with stigmata of bleeding and shallow ulcer on top was noted at the cecal base (size: 6mm, Yamada type: II) (Fig. 1). Snare polypectomy was performed (Fig. 2). Histopathologic findings revealed erosion and subacute nonspecific inflammation with ectatic blood vessels in mucosa and thick-walled blood vessels in submucosa (Fig. 3). A possibility of angiodysplasia or inflammatory fibroid polyp was considered. CD34 marker specific for inflammatory fibroid polyp was negative (Fig. 4), and thus polypoid angiodysplasia was diagnosed. Hematochezia ceased shortly after colonoscopic polypectomy. During the follow-up, there was no evidence of rebleeding.

DISCUSSION

Angiodysplasia is known as AVM, angioma, vascular ectasia, and this is a degenerative lesion, which increases the proportion of aging. It results from intermittent low grade obstruction of submucosal veins since it penetrates the muscular layers of the colon and cause small AVM.[4] Histologically, it is noted as a hypertrophy of submucosal layer, infiltration of inflammatory cell, and irregularly thickened vessel.[3] Cavett *et al.* reported that 80% of angiodysplasia was investigated in distal ileum, ascending colon, hepatic flexure, and especially in cecum (45%).[5]

According to the Medline search, only nine cases[3,6-12] of

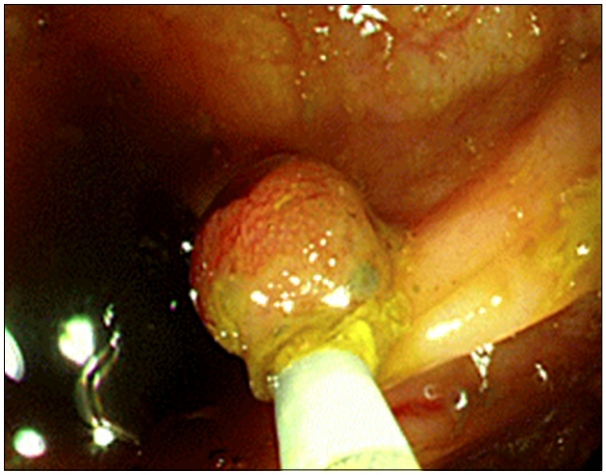

Fig. 2. Colonoscopic view of the polyp removed by snare polypectomy.

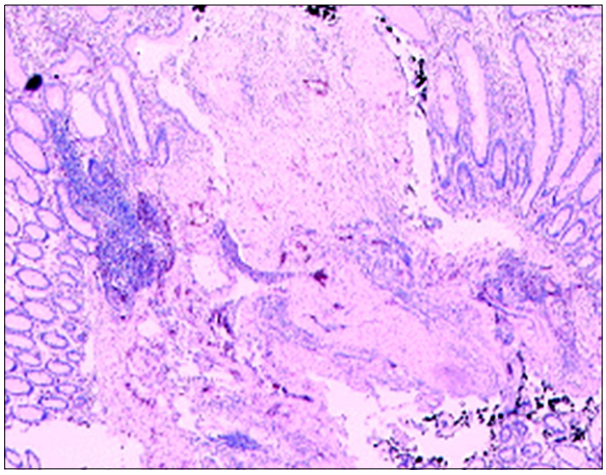

Fig. 4. Staining was negative for CD34 marker, which is specific for inflammatory fibroid polyps (×40).

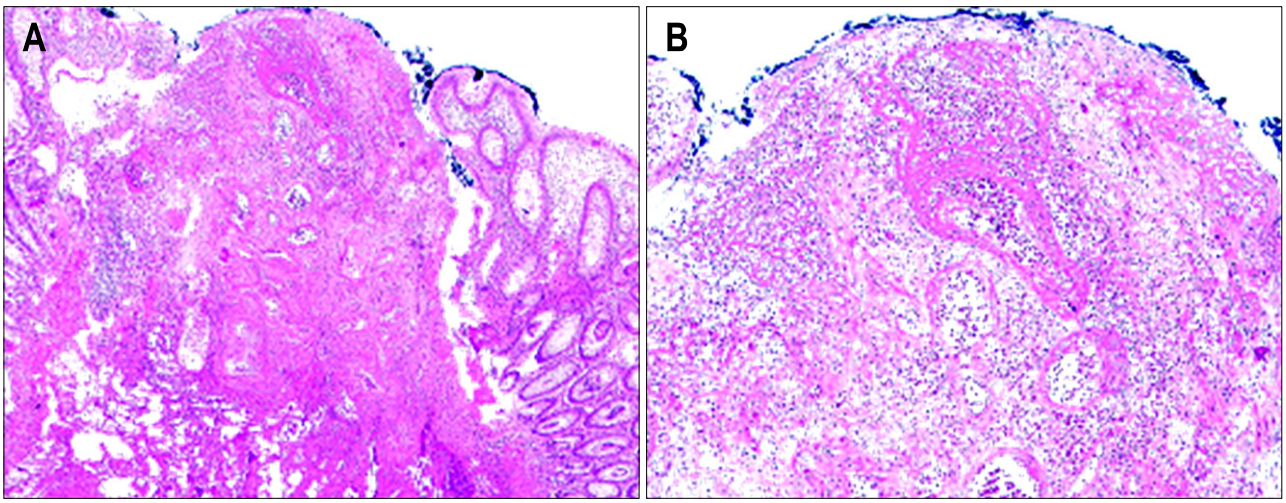

Fig. 3. (A) The submucosa layer was thickened and had an eroded surface (elastic stain, original magnification (×40)). (B) The thickened submucosa is due to proliferation of both large and small vessels and cell infiltrates (Elastic stain, (×100)).

Table 1. Summary of Reported Polypoid Arteriovenous Malformations

Authors	Age/Sex	Chief complaint	Location	Maximal diameter (cm)	Treatment
Koziara et al[3]	84/F	Hematochezia	Sigmoid colon	3.5	Snare polypectomy
	58/M	Iron deficiency anemia	Transverse colon	1.5	Snare polypectomy
Ji et al[6]	81/M	Hematochezia	Transverse colon	3.5	Polypectomy with detachable snare
Park et al[7]	41/M	Hematochezia	Descending colon	1.0	Snare polypectomy
D'Arienzo et al[8]	53/M	Hematochezia	Sigmoid colon	2.0	Snare polypectomy
Maeng et al[9]	59/F	Hematochezia	Transverse colon	6.0	Surgery
Nasseri et al[10]	26/M	Hematochezia	Sigmoid colon	3.0	Loop diathermy & Epinephrine injection
Mckevitt et al[11]	24/M	Hematochezia	Rectum	0.7	Epinephrine injection&Snared and cauterized
Jung et al[12]	69/M	Hematochezia	Ascending colon	0.8	Polypectomy with detachable snare
Present case	18/F	Hematochezia	Cecum	0.6	Snare polypectomy

polypoid AVM have been reported up to date in colon after the first report of polypoid AVM of sigmoid colon and transverse colon (Table 1). Patient reported in this case is the youngest subject among polypoid colonic angiodysplasia reported so far. Moreover, the case was noticed in cecum. According to Table 1, 7 of 10 patients were over 40 years-old and mainly male in gender. Hematochezia was the most common chief complaint. Sigmoid colon and transverse colon were the most frequent site of the lesion. Maximal diameter was between 0.6 cm and 6 cm with our case being the smallest.

Moore *et al.*[13] classified intestinal AVM according to the angiographic characteristics, localization, age of the patient, and family history. Type 1 AVMs are solitary, localized lesions within the right side of colon and usually occur in older patients. Type 2 AVMs are larger, occasionally visible, and most common in the small intestine. Type 3 AVMs are punctate angiomas causing gastrointestinal hemorrhage. In our case report, angiodysplasia is small, polypoid and solitary in the cecum of an adolescent girl. According to this classification, our case cannot be classified based on Moore *et al.*[13]

In our opinion, this classification should be limited to flat or elevated general angiodysplasia. Besides, polypoid shaped AVM should be divided into a separate classification since polypoid shape is significantly different from other angiodysplasia in diagnosis and treatment. Unlike other angiodysplasia, angiodysplasia of polypoid shape can be easily diagnosed by endoscopy. In most of the cases, snare polypectomy or polypectomy with detachable snare can be applied rather than the surgical approach such as excision or colectomy. Accordingly, interval between the beginning of symptom to diagnosis and treatment would be shortened. Moreover, as shown in Table 1, sigmoid colon and transverse colon are the most frequent involved sites.

Massive bleeding after the removal of polypoid AVM is not rare. Dobrowolski S *et al.* concluded large pedunculated polyps with stalk are at high risk of hemorrhage.[14] Hachisu *et al.* reported detachable snare polypectomy diminished bleeding risk in >20 mm, pedunculated or semipedunculated polyp.[15] In addition, Ji *et al.*[6] proposed to utilize detachable snare to remove polypoid angiodysplasia, otherwise it must be removed after epinephrine or other sclerosing agent injection. Endoscopic ultrasonography (EUS) prior to polypectomy of polypoid angiodysplasia with stalk may be useful, because EUS is beneficial to investigate the internal structure and venous flow of large lesions prior to polypectomy.[16] In this case, we could resect it only by using snare polypectomy since the polyp was small (6 mm) and was Yamada type II shaped pedunculated one. There was no bleeding after the polypectomy. If large pedunculated or semipedunculated, EUS and detachable snare polypectomy must have been considered actively. In conclusion, when a colonic polypoid angiodysplasia is found, we must recognize the risk of massive bleeding after polypectomy. EUS might be useful, and the lesion can be removed safely by attaching detachable snare or injecting other sclerosing agents.

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