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<td>小山, 和行; 林, 三進; 木暮, 喬; 平川, 賢; 赤池, 陽</td>
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Abnormal Arteriovenous Communications of the Mesenteric Vessels
—Report of a Case of Angiodysplasia and Review of the Literature—

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Key Words: Angiodysplasia, Abnormal arteriovenous communication, Mesenteric vessels, Angiography, Diagnosis

腸間膜動脈における異常動静脉交通について

—Angiodysplasia の 1 例報告と文献的考察—

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小山 和行 林 三進 木暮 雅
平川 贅 赤池 陽

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腸間膜動脈における異常動静脉交通には angiodysplasia （いわゆる arteriovenous malformation, 動静脉奇形）と動静脉奇形がある。

我々は下血を主訴とする59歳の男性で血管造影を行なったところ、上腸間膜動脈の空腸枝、回腸枝および右結腸枝に多発する限局性の小さなangiodysplasia を認めた。文献上、消化管における angiodysplasia は血管造影による消化管出血部位の検索がなされると膣報告例が増え、すでに30例以上の報告がなされている。しかし本邦では4例の報告がなされているにすぎず、2例は Type 1 に属する限局性の単発性の小病変で、他の2例は Type 2 および Type 3 に属するびまん性の大病変であり、Type 1 に属する多発性の小病変を認めたのは本症例が本邦第1例目である。

Angiodysplasia は主として次の3型に分類される。Type 1 は限局する小病変で、潰血性、高齢者に多く、約80%は盲腸から右結腸曲に認められ、文献上307例が報告されている。このうち多発性のものは我々の症例を含む6例である。Type 2 はびまん性の大病変で、先天性、若年者に多く7例（うち空腸5例）が報告されている。Type 3 は消化管以外の臓器にも多発性に認められるもので56例が報告されている。

成因はType 1 では絶年変化による腸管壁の変化と血行動態の変化が挙げられている。

臨床症状は種々の腸管出血または貧血であり、罹患期間は大量下血例では短かく、貧血例では20～30年と長いとされている。

診断法としては、血管造影法が第1であり、その診断率は35%以上であり、特徴的所見として主に4項目が挙げられている。
Abnormal arteriovenous communications of the mesenteric vessels are angiodysplasia (so-called arteriovenous malformation) and arteriovenous fistula.

Angiodysplasia of the gastrointestinal tract is increasingly recognized as one of the major causes of massive or occult rectal bleeding.1)–37) The investigation of bleeding from the gastrointestinal tract has been considerably helped in recent years by selective mesenteric and celiac angiography.

More than three hundred cases have been reported in the literature since 1956 when Rutter38) reported a case of small submucosal vascular lesion in the ascending colon and without involving another organ system. Since 1960 when Margulis et al.39) introduced operative angiography to show the bleeding angiodysplasia of the cecum, small vascular lesions have been increasingly shown angiographically in evaluating the cause of rectal bleeding.

However, only four cases of angiodysplasia have been reported in Japan; two cases are Type 1 lesion,40)48)49) one case is Type 2 lesion49) and the other is Type 3 lesion.50) In our case there were separately three small lesions in the jejunum, ileum and ascending colon, which is the first case of multiply separated small angiodysplasia in Japan.

CASE REPORT

A 59-year-old man admitted to the hospital with melena. Three years before his admission, he had had esophageal transection with splenectomy for liver cirrhosis. He had been admitted with melena in this April and it disappeared after blood infusions in two days. No other investigation had been performed. On this occasion, in October, he noticed black stool in several days before his admission. Physical examination on current admission showed no other signs of disease. No lesions in cutaneous and buccal mucosa were revealed. Hemolysis showed normocytic, normochromic anemia and clinical laboratory findings were within normal limits.

Barium swallowing and barium enema also failed to show any lesion in the gastrointestinal tract. Upper gastrointestinal endoscopic examination to duodenal loop and colonoscopic examination to the mid-portion of the transverse colon failed to show any lesion.

Selective inferior mesenteric, superior mesenteric and celiac angiography were performed. Selective superior mesenteric angiography demonstrated three angioclypsias; one was in the jejunal branch with early opacified, dilated, dense and slowly emptying vein, one was in the ileal branch of the ileocolic artery with abnormal clusters of the artery, early opacified, dilated, dense and slowly emptying vein, the other was in the middle colic artery in the hepatic flexure of the ascending colon with a vascular tuft, early opacified, dilated, dense and slowly emptying vein (Fig. 1. A-C). The feeding arteries were slighlty dilated in all lesions.

Surgical treatment was not done because of multiple lesions and improved melena with blood infusion therapy.

DISCUSSION

For vascular malformations of the gastrointestinal tract, the classification of vascular malformations
Fig. 1 Superior mesenteric angiogram showing three angiodyplasia in the jejunal branch, ileal branch and right colic branch.

A. Arterial phase showing slightly dilated feeding arteries; jejunal branch with abnormal clusters (arrow), ileal branch with abnormal clusters (open arrow) and right colic branch with a small vascular tuft (arrow head).

B. Late arterial phase showing early opacified and dilated veins; jejunal branch (arrow), ileal branch (open arrow) and right colic branch (arrow head).
C. Early venous phase showing dilated, densely opacified and slowly emptying veins; jejunal branch (arrow), ileal branch (open arrow) and right colic branch (arrow head).

According to Kajser\textsuperscript{49} or Gentry et al.\textsuperscript{86} had been used (Table 1). All kinds of lesions of vascular origin had been reported as vascular malformation and had been discussed mainly in reference to phlebectasia and hemangioma.\textsuperscript{46-48} These vascular lesions had been diagnosed by surgery and pathology. The roentgenologic diagnosis has depended on the calcifications or phlebolith in the plain abdominal films and on the filling defect, stenosis and intussusception in barium contrast study.\textsuperscript{51-60}

Small vascular lesions of the gastrointestinal tract have been shown with the advent of angiography.

Table 1 Classification of vascular malformations of gastrointestinal tract by Gentry et al. (1949)

<table>
<thead>
<tr>
<th>Class</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>Benign vascular lesions</td>
<td></td>
</tr>
<tr>
<td>I. Capillary hemangioma</td>
<td>(simplex, most single)</td>
</tr>
<tr>
<td>II. Mixed hemangioma</td>
<td></td>
</tr>
<tr>
<td>III. Cavernous hemangioma</td>
<td>Multiple phlebectasia (small cavernous)</td>
</tr>
<tr>
<td></td>
<td>Simple polypoid (single cavernous)</td>
</tr>
<tr>
<td></td>
<td>Diffuse expansive (single, contiguous)</td>
</tr>
<tr>
<td></td>
<td>Diffuse expansive (multiple, noncontiguous)</td>
</tr>
<tr>
<td>Malignant vascular lesions</td>
<td></td>
</tr>
<tr>
<td>A. Hemangioendothelioma</td>
<td></td>
</tr>
<tr>
<td>B. &quot;Benign metastatic hemangioma&quot;</td>
<td></td>
</tr>
<tr>
<td>C. Kaposi's sarcoma</td>
<td></td>
</tr>
<tr>
<td>D. Angiosarcoma</td>
<td></td>
</tr>
<tr>
<td>Location</td>
<td>No. of cases</td>
</tr>
<tr>
<td>------------------</td>
<td>--------------</td>
</tr>
<tr>
<td>Duodenum</td>
<td>4</td>
</tr>
<tr>
<td>Jejunum</td>
<td>23</td>
</tr>
<tr>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
</tr>
<tr>
<td>Ileum</td>
<td>14</td>
</tr>
<tr>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
</tr>
<tr>
<td>Cecum</td>
<td>93</td>
</tr>
<tr>
<td></td>
<td></td>
</tr>
<tr>
<td>Ascending colon</td>
<td>32</td>
</tr>
<tr>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
</tr>
<tr>
<td>Right colon</td>
<td>123</td>
</tr>
<tr>
<td>Hepatic flexure</td>
<td>2</td>
</tr>
<tr>
<td>Transverse colon</td>
<td>2</td>
</tr>
<tr>
<td></td>
<td></td>
</tr>
<tr>
<td>Splenic flexure</td>
<td>2</td>
</tr>
<tr>
<td>Descending colon</td>
<td>1</td>
</tr>
<tr>
<td>Sigmoid colon</td>
<td>3</td>
</tr>
<tr>
<td>Rectum</td>
<td>1</td>
</tr>
<tr>
<td>Omentum</td>
<td>1</td>
</tr>
<tr>
<td>Total</td>
<td>301</td>
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</table>

M: Male, F: Female, Parentheses show range and mean of age in where age is known cases of both sexes.

which may show the bleeding site if the rate of blood loss is more than 0.5 ml per minute. There has been confusion in terminology. For instances, “hemangioma”, “telangiectasia”, “arteriovenous malformation”,”angiodysplasia” and “colonc vascular ectasia” are often regarded as interchangeable. Halpern et al. first used the term angiodysplasia for vascular malformations such as aneurysm, discrete arteriovenous fistula, conglomerate masses of angiectasia, capillary angiodysplastic lesions, phlebectasia and angioma.

In recent years angiodysplasia has been used for such vascular lesions as arterio-capillary-venous angiectasia.

With the review of the literature we are able to agree with the classification by Moore et al.
except for one point, namely that Type 2 lesion includes the cases of multiple lesions involving other organ systems. We consider that Type 2 lesion includes the cases of large and diffuse lesions in the gastrointestinal tract and Type 3 lesion includes the cases of multiple lesions involving other organ systems simultaneously.

The three types of angiodysplasia are: Type 1 is small, localized lesion(s), most commonly in the right colon and probably acquired lesion, Type 2 is large, diffuse and occasionally visible lesion, commonly in the small bowel and probably congenital lesion, Type 3 is multiple lesions involving gastrointestinal tract and other organ systems. Sometimes this type is reported as Rendu-Osler-Weber syndrome, sometimes the lesion is one of a type which should be classified as Rendu-Osler-Weber syndrome.

There are 307 cases of Type 1 lesion. Of 301 cases of solitary lesion (Table 2); 4 cases in the duodenum, 1.3% in distribution; 23 cases in the jejunum and the mean age is 49.5 in where age is known cases (7.6%); 31 cases in the ileum and the mean age is 45.4 (4.7%); 21 cases in the cecum and the mean age is 60.2 (30.9%); 14 cases in the ascending colon and the mean age is 64.4 (10.6%); 123 cases reported in the right colon (40.9%); 2 cases in the hepatic flexure (0.7%); 2 cases in the transverse colon (0.7%); 2 cases in the splenic flexure (0.7%); 1 case in the descending colon (0.3%); 3 cases in the sigmoid colon (1.0%); 1 case in the rectum (0.3%); and 1 case in the omentum (0.3%).

<table>
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<th>Author</th>
<th>Case</th>
<th>Location</th>
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<tr>
<td>Calem</td>
<td>63</td>
<td>Duodenum, Jejunum</td>
</tr>
<tr>
<td>Marx</td>
<td>62</td>
<td>Jejunum, Cecum</td>
</tr>
<tr>
<td>Miller</td>
<td>Unknown</td>
<td>Jejunum, Sigmoid colon</td>
</tr>
<tr>
<td></td>
<td>Unknown</td>
<td>Ileum, Sigmoid colon</td>
</tr>
<tr>
<td></td>
<td>Unknown</td>
<td>Ileum, Cecum, Splenic flexure, Sigmoid colon</td>
</tr>
<tr>
<td>Present case</td>
<td>59</td>
<td>Jejunum, Ileum, Ascending colon</td>
</tr>
</tbody>
</table>

M: Male, F: Female

The lesions in the right colon, from cecum to hepatic flexure, account for over 80% and it is common in older patients over 50 years old.

Of 6 cases of multiple lesions (Table 3); 1 woman aged 62, 2 men aged 62 and 63, and 3 cases where age and sex are unknown. 5 cases have lesions both in the small and large bowel. There are 7 cases of Type 2 lesion; 5 cases in the jejunum and the mean age is 38.0; 1 case in the transverse colon and 1 unknown case. The mean age in 7 cases is 38.1 (range 17-48)
consisting of 2 women and 5 men.

There are 56 cases of Type 3 lesion; 9 women and the mean age is 53.1 (range 23–77, 6) and 15 men and the mean age is 54.3 (range 7–76) and 35 unknown cases. Little is known about the pathogenesis. Beley et al. showed the pathogenesis in the case of right colon as having begun with dilatation of the capillary ring and small arteriovenous communications occurring as the result of chronic, intermittent, low-grade obstruction of the submucosal veins. The prevalence of these degenerative lesions can be attributed to tension in the cap wall. However, it is noted that recurrent bleeding was shown after surgical treatment and in our case rectal bleeding was shown three years after esophageal transection with splenectomy. These findings suggest that hemodynamic changes, caused by previous surgery or vascular diseases such as aortic stenosis and atherosclerotic diseases, affect the pathogenesis.

Although little has been discussed regarding the pathogenesis and causal factors of multiple and of large and diffuse lesions, it has been suggested that it is congenital in origin because of its largeness and diffusion and prevalence in younger patients. Histological findings are ruptured mucosa at the bleeding site, dilated vessels in the submucosa, several large vascular channels in the mucosa or submucosa, and a tangle of dilated submucosal vessels shown by the specimen injection technique.

Clinical symptoms of angiodysplasia are massive rectal bleeding, melena and anemia with occult rectal bleeding. The duration is short in massive bleeding case, but it is longer than twenty years in anemic patients. There have been reported some cases accompanied with aortic stenosis, atherosclerosis and diverticulosis. The more effective diagnostic study is selective angiography. Total mesenteric angiography only fails to reveal angiodysplasia in a few cases. Diagnosis by this method can be accurately made in over 85% of the cases. Although some authors classify angiodysplasia angiographically into three groups; Type 1 is arteriovenous malformation; Type 2 is small vessel malformation and Type 3 is venous malformation, they use the classification in the lesions in the extremities and do not discuss the lesions in the gastrointestinal tract.

We have found angiographic findings by some authors to be helpful in diagnosing angiodysplasia. Angiographic findings with our modification are shown as follows:

1. normal or slightly dilated feeding artery,
2. small vascular tuft or lake in small solitary lesion, or abnormal clusters of small arteries in the bowel wall in large lesion,
3. accumulation of contrast material in the vascular spaces and intense opacification of the bowel wall,
4. early and densely opacified, slowly emptying and dilated draining vein.

The second diagnostic study is colonoscopy. Colonoscopic findings reveal normal to slightly reddish or ulcerated mucosa during bleeding. Richardson et al. revealed the lesions in 12 of 29 cases and there were 6 false negatives. Rogers et al. revealed the lesions in 5 cases and treated bleeding successfully in 4 cases by electrocoagulation method.

The third, and least reliable method of diagnosis is exploration and direct visualization such as the measurement of the venous pressure in the dilated veins and transillumination.
The usual treatment of patients is wide resection of the bowel such as subtotal colectomy. Occasionally, electrocoagulation and drug infusion therapy for bleeding are performed.

CONCLUSION

The case of a 59-year-old man with mesenteric angiodysplasia, Type 1 lesion, is presented. Angiography revealed these solitary, localized small lesions in the jejunum, ileum, and ascending colon.

Angiodysplasia of the gastrointestinal tract is one of the major causes of massive or occult rectal bleeding and is shown successfully by selective mesenteric and celiac angiography.

Until now only four cases were reported in the Japanese literature; two cases are solitary Type 1 lesion, one case is Type 2 lesion and the other is Type 3 lesion.

This is the first case of multiple Type 1 lesion.

The pathogenesis of angiodysplasia is unclear and is partially explained as degenerative changes in aging in Type 1 lesion.

In our case surgery had been performed previously, namely esophageal transection with splenectomy, and this suggests that hemodynamic change is one of the major causes of angiodysplasia.

REFERENCES


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