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Abnormal Arteriovenous Communications of The Mesenteric Vessels:

Report of a case of Type 2 lesion of angiodysplasia

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腸間膜動脈における異常動静脈交通について

—Angiodysplasia Type 2 の1例報告と文献的考察—

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腸間膜動脈支配域における異常動静脈交通には Angiodysplasia (いわゆる arteriovenous malformation, 動静脈奇形) と動静脈瘻とがある。

我々は食道静脈瘤に対する食道離断術を目的として入院した58歳の男性で、精査のために腹部血管撮影を行なったところ、上腸間膜動脈の空腸枝支配域に、動脈相で腸管壁の濃染と、毛細管相で腸管壁に拡張した異常血管と還流静脈の早期出現、および静脈相で還流静脈の濃染と還流遅延を示した Angiodysplasia の Type 2 を認めた。

文献上十二指腸以下の消化管における Angiod-

ysplasia は血管撮影による消化管出血部位の検索がなされるにつれ報告例が増え、Type 1 から Type 3 まででみるとすでに400例に達している。

しかし、Type 2 に属するものは、8例が報告され(本邦では1例のみ)ているにすぎず、本症例が9例目(本邦2例目)である。

Type 2 はびまん性の大病変で、先天性と考えられ、若年者に多く、9例中8例が小腸に認められている。

臨床症状は、吐血、下血、タール便など種々の消化管出血または貧血である。

診断法としては血管撮影法が最も有効であるが、最近は術中に病変部位の同定のために Dop-

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pler 法や Indigo Carmin 注入法が行なわれつつある。治療としては、部分切除または広汎切除が行な

われる。

我々は、空腸での Angiodysplasia の Type 2 の 1 例について文献的考察を加え報告した。

With the advent of angiography angiodysplasia (arteriovenous malformation) of the gastrointestinal tract is increasingly recognized as one of the major causes of massive or occult rectal bleeding¹⁾⁻¹⁴⁾.

About four hundred cases have been reported in the literature since 1960 when Margulis et al. introduced operative angiography to show the bleeding angiodysplasia of the cecum¹⁾. It has been known that there are three types of angiodysplasia due to incidence and largeness⁷⁾¹⁵⁾.

After we reported a case of multiple Type 1 lesion and discussed angiodysplasia¹⁵⁾, we had a case of Type 2 lesion.

In English and Japanese literature there have been reported only eight cases of Type 2 lesion, which shows diffuse and large lesion, and commonly in the small bowel and probably congenital in origin.

This is the second case of Type 2 lesion in Japan, in which abnormal arteriovenous communications were shown in angiography.

Case report

A 58-year-old man was admitted to the hospital for esophageal varices surgery. Past history revealed no history of blunt trauma to his abdomen. The patient had no abdominal surgery and denied previous episodes of hematemesis and rectal bleeding. Blood chemistry showed slight liver dysfunction and laparoscopy revealed liver atrophy and splenomegaly. Liver biopsy showed liver cirrhosis. Angiography was performed for evaluating liver cirrhosis and collateral ways after an overnight fast. Selective superior mesenteric arteriography showed early, diffusely and intensely opacified jejunal wall, and abnormally dilated vessels in the jejunal wall, and dilated, intensely opacified and slowly emptying jejunal vein, namely Type 2 lesion of angiodysplasia. There were not found such abnormal vessels as intensely opacified and draining veins in other intestinal vessels.

Esophageal transection with splenectomy was performed and surgical treatment for angiodysplasia was not performed, because rectal bleeding or melena has not been revealed and it was difficult to evaluate the bowel lesion during surgery. Postoperative course was uneventful in these two years.

Table. Summary of mesenteric angiodysplasia in cases of Type 2 lesion

Author	Case		Symptoms	Location	Treatment
	Age	Sex			
Pierce	48	M	Tarry stool	Jejunum and proximal ileum*	Wide resection in three times, totaling 330 cm.
Moore	17	M	Melena	Jejunum	Segmental resection
	37	M	Melena, anemia	Jejunum	Segmental resection
	34	M		Transverse colon	Right colectomy
	48	F		Jejunum	Segmental resection
Matsumoto	43	F		(Ileum)**	Segmental resection
	40	M	Hematemesis	Jejunum and ileum	Wide resection of small bowel (270 cm)
Crawford	66	F	Melena, anemia	Jejunum and ileum*	Segmental resection
Present case	58	M	No bleeding episode	Jejunum	No surgical treatment

M=male, F=female, *=angiography was not performed, **=angiography showed normal.

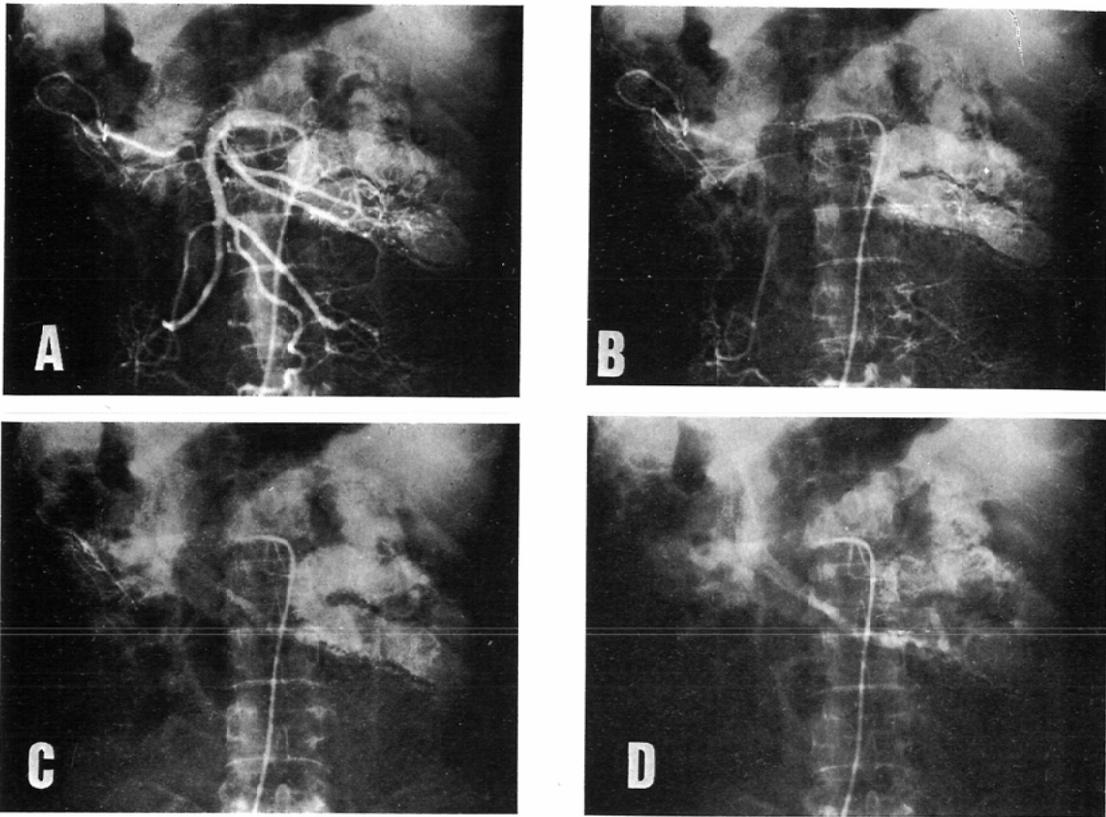


Fig. Superior mesenteric arteriogram showing Type 2 lesion of angiodyplasia in jejunal branch.

- A. Arterial phase showing early opacified jejunal wall.
- B. Late arterial phase showing abnormally dilated vessels in jejunal wall and intensely opacified jejunal wall and showing no abnormal vessels in other branches of superior mesenteric artery.
- C. Capillary phase showing abnormally dilated vessels in jejunal wall and intensely opacified jejunal wall, and intensely opacified jejunal vein.
- D. Venous phase showing slowly emptying and intensely opacified jejunal vein and portal vein and showing no definite draining veins of other branches of superior mesenteric vein.

Discussion

Vascular lesions of the gastrointestinal tract have been reported as angioma, telangiectasia, arteriovenous malformation, angiodyplasia and colonic vascular ectasia.

Malan and Puglionisi used the term "angiodyplasia" in the wide spectrum of congenital vascular lesions as abnormally developed or residual embryonal vascular network in the extremities¹⁵⁾. In these vascular lesions abnormal arteriovenous communications are shown in "trunkular arteriovenous fistula" and "arteriovenous angioma".

Szilagy et al. used "arteriovenous fistulas" as abnormal arteriovenous communications and classified into four groups¹⁶⁾.

Although some authors classify angiodyplasia into three groups in angiography; 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 tract¹⁷⁾¹⁸⁾.

However, we consider that these classifications are not reasonable for evaluating vascular lesions with abnormal arteriovenous communications. We consider that vascular lesions with abnormal arteriovenous communications in angiographic evaluation should be classified into two main categories, namely "arteriovenous fistula" and "arteriovenous malformation (angiodysplasia)"¹³⁾¹⁹⁾²⁰⁾. Arteriovenous fistula is a lesion that a direct communication is shown between trunks or branches of artery and vein, and arteriovenous malformation is a lesion that an indirect communication is shown as arterio-capillary-venous angiectasia.

The term "angiodysplasia" will be used such lesions which vascular components of abnormal vessels are unidentified its origin or which includes wide spectrum of vascular lesions, and the term "arteriovenous malformation" will be used such lesions which vascular components are identified in consisting of artery, capillary and vein. However, it is quite difficult to distinguish capillary and vein from arterialized vessels in histology. Now, we use the term "angiodysplasia" and "arteriovenous malformation" in the same meanings¹⁵⁾.

There has been reported only 9 cases of Type 2 lesion including our case; 4 cases in the jejunum and mean age is 40.0⁷⁾, 3 cases in the jejunum and ileum and mean age is 51.3⁴⁾¹²⁾¹⁴⁾, 1 case in the ileum and age is 43⁷⁾, and 1 case in the transverse colon and age is 34⁷⁾. The mean age in 9 cases is 43.4 (range 17-66) consisting of 3 women and 6 men. Of these cases, the lesion was shown in angiography in 7, the lesion was not shown in one⁷⁾ and angiography was not performed in one⁴⁾.

Although little has been discussed regarding the pathogenesis and causative factors of Type 2 lesion, it has been suggested that it is congenital in origin because of its largeness and diffusion and prevalence in younger patients and similar pathogenesis has been considered in angiodysplasia of the extremities¹⁵⁾.

It is possible to speculate that hemodynamic change, namely portal hypertension due to liver cirrhosis, would be accelerate vascular dilatation and hemostasis, in where there would be some factors as the nature of weakness of vascular wall and surrounding tissues and as the residual embryonal vascular network.

Clinical symptoms of angiodysplasia, Type 2 lesion, are hematemesis, rectal bleeding, melena, and anemia with occult rectal bleeding⁴⁾⁷⁾¹²⁾. The duration is from two months to 6.5 years.

We have found angiographic findings to be helpful in diagnosing Type 2 lesion of angiodysplasia, as follows;

1. Normal or slightly dilated feeding artery,
2. Diffuse and intense opacification of the bowel wall,
3. Abnormally dilated vessels in the bowel wall, and
4. Early and densely opacified, slowly emptying vein (s).

Other diagnostic methods are exploration⁴⁾, direct visualization such as the measurement of the venous pressure, transillumination⁴⁾²¹⁾, intraoperative doppler technique²²⁾, indigo carmin solution injection¹⁴⁾ and endoscopy²³⁾⁻²⁵⁾.

The usual treatment is segmental or wide resection of the involved bowel.

Conclusion

The case of a 58-year-old man with mesenteric angiodysplasia, Type 2 lesion is reported.

Angiography revealed a large and diffuse lesion in the jejunum, with diffuse and intense opacification of jejunal wall, abnormally dilated jejunal vessels and intensely opacified and slowly emptying jejunal vein.

This is the second case of Type 2 lesion of angiodysplasia in Japan. The pathogenesis of Type 2 lesion is unclear and has been considered congenital in origin.

In our case portal hypertension in liver cirrhosis has been shown and this would be suggested that hemodynamic change is one of the causative factors.

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