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Clinical and CT Angiographic Follow-Up Outcome of Spontaneous Isolated Intramural Hematoma of the Superior Mesenteric Artery

Z. Xiaoq[1] · M. Hao[1] · L. Lin[1] · Y. Jiao[1] · J. Zou[1] · X. Zhang[1] · Y. Y. Hong[1]

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Abstract
Objectives The aim of this study was to evaluate the clinical course and vascular remodeling of spontaneous isolated intramural hematoma of the superior mesenteric artery (SIHSMA) after treatment.
Methods In this retrospective study, 24 consecutive patients with SIHSMA admitted from January 2009 through December 2016 were included in this study. The clinical characteristics, type and location of the dissection, clinical outcome and vascular remodeling were analyzed retrospectively.
Results The subjects included 21 men and three women, with a mean age of 50.58 years. The chief complaint was abdominal pain in all patients. The mean follow-up was 10.08 months. Among the 24 patients examined, two patients (8.33%) showed no obvious changes in the CT. Twenty patients were treated by conservative strategy. One case underwent exploratory laparotomy along with embolectomy and arteriotomy. Another three cases received angioplasty and stent implanting. Complete and partial remodeling was observed in 15 (62.5%) and four patients (16.67%). Three (12.5%) of the lesions had dissection remodeling and aneurysm change.
Conclusions SIHSMA represented variable vascular remodeling, while most of the patient got a complete resolution during follow-up, and its clinical course was benign in this study. Vast majority of patients can be managed conservatively when there are no signs indicating organ ischemia.

Keywords Superior mesenteric artery · Intramural hematoma · Remodeling pattern · Clinical course

Introduction

Spontaneous isolated dissection of the superior mesenteric artery (SISMAD) is very rare, with a reported incidence of 0.06% [1]. Superior mesenteric artery (SMA) is the most frequent site of isolated dissection among visceral arteries [2]. Given the infrequency of diagnosis in SISMAD, no related guidelines have been established.

With widespread application of computed tomography angiography (CTA) improved the ability to diagnose spontaneous isolated dissection of the superior mesenteric artery (SISMAD) at initial admission, an increasing number of studies concerning the management and clinical outcomes of SISMAD were published [3, 4]. However, no quantitative evidence is available regarding the clinical and angiography follow-up of the type of intramural hematoma of SISMAD.

The clinical presentation of SISMAD varies from incidental discovery from CTA without symptoms to acute

Z. Xiaoq and M. Hao contributed to the work equally and was regarded as co-first authors.

✉ Y. Y. Hong
rodgeryang1997@163.com

X. Zhang
xiweil092@163.com

[1] Division of Vascular Surgery, Department of General Surgery, First Affiliated Hospital of Nanjing Medical University, 300 Guangzhou Road, Nanjing 210029, Jiangsu, People's Republic of China

abdominal pain with peritonitis or bowel ischemia. It creates difficulties for surgeons in making adjustments to the treatment plan. Despite that, recent systematic reviews investigated the treatment strategies of SISMAD and advocated conservative management as the most common initial treatment [5, 6].

Several classification of SISMAD were put forward to the guidance treatment, such as Sakamoto [7], Yun [8], Luan [9], Li [10] and Yoo [11] classification. Sakamoto et al. simply categorized SISMAD into four types based on imaging appearances, but did not include total thrombotic occlusion of SMA trunk in their classification. Yun et al categorized SISMAD into three types according to the presence of false luminal flow and true lumen patency at the dissected segment. The Luan classification was based on the location of dissection, but does not include patency and re-entry of false lumen. On the other hand, the Li classification complements the Luan classification, but is too complicated and does not provide proper dissection length information. Yoo suggested a new classification that divided SISMAD into four types according to the morphologic features and emphasized disease severity by adding subtypes, providing a supplementary explanation for other theories. The differences of five classifications of SIHSMA are shown in Table 1.

Intramural hematoma (IH), known as a variant form of dissection, is characterized by the absence of intimal tear and direct flow communication between true and false lumen. To date, most published studies have been case series for SISMAD. The clinical course of and optimal treatment strategy for spontaneous isolated intramural hematoma of SMA (SIHSMA) have not been fully investigated. This study was conducted to investigate the clinical course and the vascular remodeling of SIHSMA.

Table 1 The differences of five classifications of SIHSMA

Classification	Definition
Type Iva, IVb Li thrombosed false lumen without an ULP (subdivided into IVa, patent true lumen; IVb, severe stenosis of the true lumen; and IVc, occlusion of the true lumen)	Completely
Type III Yoo thrombosed false lumen	Completely or partially
Type IIb Yun	Visible false lumen but not visible re-entry site
Type IV Sakamoto thrombosed false lumen without ulcer like projection (ULP)	Completely
Type Luan	Not described

Methods

A retrospective study was performed on 24 consecutive patients (21 males, three females, age: range 42–74 years, mean 50.58 years, all Han) who were diagnosed with SIHSMA at Jiangsu Province Hospital between January 2009 and December 2016. The mean follow-up was 10.08 months (range 1–45 months). Patients with SIHSMA were identified by searching the medical records for SISMAD, acute or chronic vascular disorder of intestines and aortic dissection at our Department of General Surgery. Their medical and imaging records were reviewed retrospectively. The changes in the patients' images on CT scans and their responses to different treatment modalities were analyzed. If a patient had another artery involvement, such as an aortic dissection, that patient was excluded from the study. Other types of SISMAD with entry, re-entry sites or progression to dissecting aneurysm were excluded.

Twenty-one study patients were admitted from the emergency department because of the acute onset of abdominal pain, and another three subsets without symptoms were inadvertently found by characteristic contrast-enhanced CT findings. We evaluated the demographics and clinical manifestations of the patients on admission retrospectively. Patients' in-hospital and outpatient clinical follow-up outcomes and vascular remodeling were recorded by vascular surgeons. All patients were identified by CTA in our study. All arteries were measured and classified according to anatomic descriptors. Peritonitis signs of bowel wall necrosis and perforation (e.g., abdominal wall rebound tenderness, muscle rigidity or shock) were considered an indication for exploratory laparotomy.

Initially, all patients underwent conservative management based on our protocol consisting of bowel rest with fasting, control of blood pressure, vasodilator drug, antiplatelet agent and short-term anticoagulation. Symptomatic patients underwent close clinical and hemodynamic monitoring, and blood pressure was controlled in patients with hypertension. Fasting was continued until the abatement of pain and oral intake was restarted upon the patient's toleration. Anticoagulation therapy consisted of low molecular weight heparin (enoxaparin) and oral anticoagulants (e.g., warfarin, rivaroxaban). Antiplatelet drugs were simultaneously administered for 3 to 6 months. In case symptoms persisted around 1 week, conservative management with fasting was continued if there were no signs of clinical and hemodynamic deterioration suggesting peritonitis. All people were given medical treatment during the

first 3 to 5 days. If the abdominal pain did not subside, the endovascular stent placement was then performed in patients.

A CT scan was performed 7 days after the initial diagnosis, 1 month, and 6 months after admission, to re-evaluate the dissected lesion and check the status of the intestine. Types of vascular remodeling were described according to the mean diameter and length of intramural hematoma. No remodeling was described as no change or increase in the mean diameter and length of intramural hematoma, even formation of pseudoaneurysm and dissection. Partial remodeling was described as the mean diameter, and length of intramural hematoma decreased. Complete remodeling was described as intramural hematoma disappeared.

Due to no related guidelines have been established of SISMAD, we treated the patients according to clinical symptoms. All study participants, or their legal guardian, provided informed written consent prior to study enrollment.

The continuous variables were recorded as means standard deviation (SD). Numerical values are expressed as mean ± standard deviation (SD). Statistical analysis of the difference between statistics was assessed by the Student unpaired t test. All statistical analyses were performed using statistical software SPSS (version 23.0). A value of $P < 0.05$ was considered to indicate variance homogeneity.

Results

We evaluated the demographics and clinical manifestations of the included patients on admission (Table 2). The median duration of abdominal pain before admission was 99.96 h (range, 6–504 h). Periumbilical tenderness was found in nine patients, and no patients showed rebound tenderness. Atherosclerotic risk factors included hypertension in 8 patients, hyperlipidemia in none patients, smoking in 3 patients, cardiac disease in one patient.

Patients' in-hospital and discharged clinical follow-up outcomes and vascular remodeling were collected retrospectively. Table 3 depicted the degree of stenosis, distance from ostium, and length of IH.

The mean distance from the SMA ostium to the beginning of the IH was 17.19 mm (range 0–80 mm). The mean length of dissection was 77.89 mm (range 7.7–140 mm), and the mean degree of stenosis was 64.83% (range 25–98%) (Table 3). The aim of treatment was symptom relief and vascular remodeling. All patients were relieved of symptoms after treatment. Twenty patients were treated by conservative strategy. One case underwent exploratory laparotomy along with embolectomy and arteriotomy. Another 3 cases received angioplasty and stent implanting.

Table 2 Demographics and clinical manifestations of the included patients on admission

Study population	
Features ($N = 24$)	
Age, years, mean ± SD	50.58 ± 7.84
Male gender	21 (78.5%)
Coexisting medical conditions	
Hypertension	8 (32%)
Hyperlipidemia	0
Diabetes mellitus	0
Chronic kidney disease	0
Coronary artery disease	1 (4%)
Atrial fibrillation	0
Chronic obstructive pulmonary disease	0
History of abdominal surgery	3 (12%)
Current smoker	3 (12%)
Presentation	
Asymptomatic	0
Symptomatic	24 (100%)
Periumblical tenderness	9
Rebound tenderness	0

A stent (6 * 60 mm * 2; 7 * 80 mm * 1; Luminex stent, Bard Peripheral Vascular, Angiomed GmbH & Co, Medizintechnik KG, Karlsruhe, Germany.) was placed in the affected SMA in each case (Fig. 1). There have been various morphologic changes detected by follow-up CT angiography, and the results are summarized in Table 4. Among the 24 patients examined, two patients (8.33%) showed no obvious changes in the CT. However, 22 patients (91.6%) showed angiographic improvement to complete remodeling (Fig. 2). Complete and partial remodeling was observed in 15 (62.5%) and four patients (16.67%) (Fig. 3). Partial remodeling is defined as: The length and extent of intramural hematoma decreased, and the degree of stenosis did not increase. About three(12.5%) of the lesions had progressed and had aneurysm changes.

Discussion

An increase in number of SISMAD has been reported in recent years with the development of advanced imaging technology. The etiology, risk factors, natural course, and the optimal treatment for SISMAD have not been firmly established due to the complexity and rarity of the condition. Factors likely influencing the nature of SISMAD have been complicated as causative, and include a history of connective tissue disorder, fibromuscular dysplasia (FMD), cystic medial degeneration, atherosclerosis, segmental arterial mediolysis (SAM), and elastic tissue disorders

Table 3 Lesion characteristics of superior mesenteric artery dissection

Patient	The median distance from the SMA ostium to the beginning of the IH (mm)	IH length (mm)	Degree of stenosis (%)
1	0	29	36
2	0	95	76
3	0	129	90
4	19.8	112	90
5	27.7	135	36
6	23	44	73
7	36.3	38.5	66
8	18.6	42.1	37
9	31.9	41.2	74
10	30	52	50
11	16	46	98
12	0	110	90
13	0	92	90
14	35	66	75
15	13	85	88
16	4.7	110	60
17	0	140	47
18	43	67	25
19	0	110	62
20	24	53	42
21	0	7.7	66
22	0	112	90
23	80	55	48
24	9.7	98	47
Mean ± SD	17.19 ± 19.52	77.89 ± 37.2	64.83 ± 21.77
P	< 0.05	< 0.05	< 0.05

IH intramural hematoma

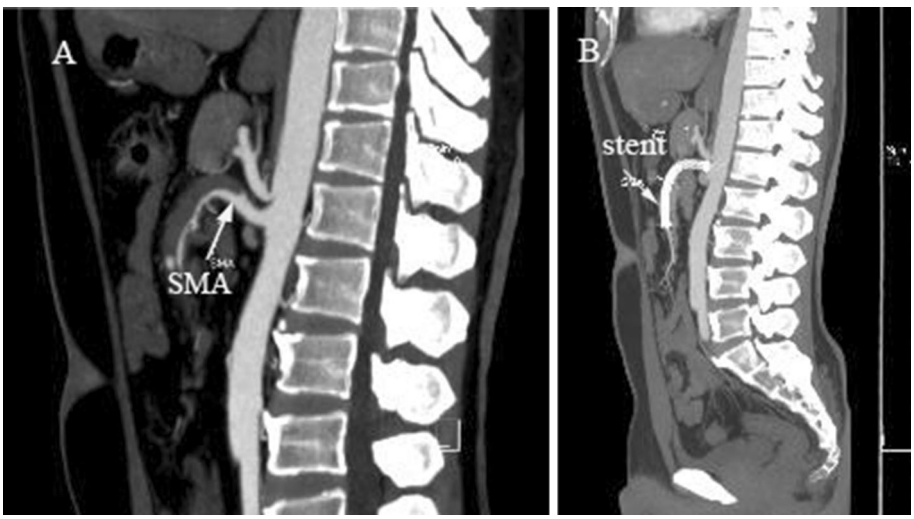

Fig. 1 A 50-year-old man with epigastric pain. **A** Enhanced CT showed an intramural hematoma (arrow). **B** A stent (7 * 80 mm * 1; Luminex stent, Bard Peripheral Vascular, Angiomed GmbH & Co, Medizintechnik KG, Karlsruhe, Germany.) was placed in the affected SMA. After 7 months, the intramural hematoma was completely remodeled and the stent was normally patent (arrowhead)

(Marfan syndrome and EhlerseDanlos disease) [2, 12, 13]. In the present study, eight patients (33.3%) presented with hypertension an three patients (12.5%) smoked; however, there was no atherosclerotic change in the orifice and the proximal segment of SMA. Strong male preponderance is another characteristic of SISMAD [8]. A systematic review

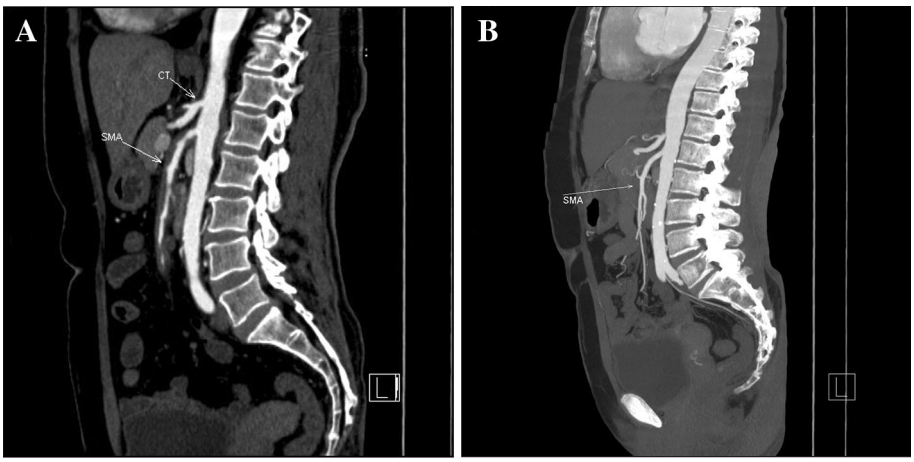

Fig. 2 A 51-year-old woman with acute abdominal pain and an spontaneous isolated intramural hematoma of the superior mesenteric artery (SIHSMA). **A** Enhanced CT showed an intramural hematoma (arrow). Then, the patient was conservatively followed up because there was no sign of bowel ischemia. **B** After 19 months, the intramural hematoma was complete resolution (arrowhead)

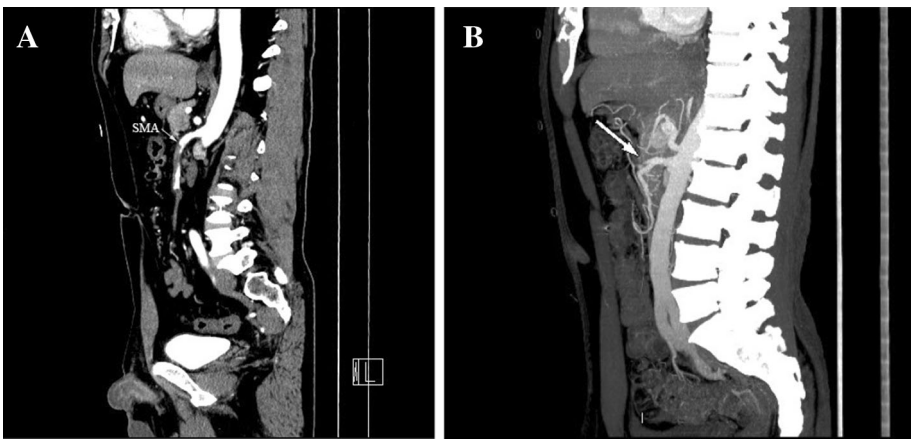

Fig. 3 A 42-year-old man with epigastric pain. **A** Enhanced CT showed an intramural hematoma (arrow). **B** After 26 months, the intramural hematoma was partial remodeling with a dissection change (arrowhead)

Table 4 Morphologic changes detected by follow-up CT angiography

Follow-up result	*N* (%)
Complete remodeling	15 (62.5)
Partial remodeling	4 (16.67)
No change	2 (8.33)
Dissection remodeling or aneurysm change	3 (12.5)

included fifty-one articles of SISMAD indicating ethnic differences that Asia (China, Korea, and Japan) are areas with the highest incidence of disease [5]. On the whole, SISMAD is very uncommon.

The incidence of SISMAD is likely underestimated because of no reliable clinical signs and laboratory features. In symptomatic patients, most common clinical manifestations were abdominal pain, back pain, and chest pain [14] which may be related to stenosis of the true lumen causing mesenteric ischemia [15], to rupture of the dissection causing a mesenteric hematoma [16], or to the dissection itself [17]. According to other researches, both the degree of true lumen stenosis and dissection length are relevant to symptoms among the CT features of SISMAD [10]. On the basis of Poiseuille's law of fluid dynamics, flow rate bears an inverse relation to distance. Thus, blood flow is decreased when the dissection is long adding the grade of intestinal ischemia. As a consequence, pain severity correlates with dissection length when viewed from the side. Management tends to depend on the presence or absence of symptoms, if there is no evidence of bowel infarction, bleeding or aneurysmal changes, conservative treatment with close follow-up is sufficient and could reduce unnecessary endovascular interventions [7, 18, 19].

There is no consensus on optimal treatment for SISMAD. Therapeutic strategy choice is based on patient symptoms and signs, morphologic characteristics of the dissection and the presence of edema/inflammatory/ischemic changes on CTA, and experience of the medical centers. Generally, an exploratory laparotomy is needed in cases with signs of peritonitis. In a systematic review study, conservative therapy was the treatment of choice in the majority of cases and the success rate of conservative treatment was 85% [20]. However, failure of conservative

treatment in SISMAD was not rare [21, 22]. It might be due to the insufficient observation periods and timing for surgery. Among conservative treatment cases from Dong et al. [23], the abdominal pain remarkably or completely resolved without peritoneal signs in five patients after the first three to 5 days medical treatment. After then, medical treatment continued instead of an immediate surgery, and ultimately achieved complete alleviation of abdominal pain within the following 1 week. Ahn et al. [24]. considered that endovascular procedures might have been unnecessary if the conservative treatment had not been terminated too early. Considering the self-limited course, symptom relief of SISMAD might be obtained with longer duration of conservative treatment [22]. In our study, 1 case underwent embolectomy and arteriotomy due to the lack awareness of the disease in 2010.

Patients with SIHSMA were included in this study. This disease was subscribed as TYPE IV of Li classification and TYPE III of Yoo classification with characteristics of a true lumen without occlusion or ulcerative change. We retrospectively studied 24 patients and observed the outcome after 10.08 months follow-up. The result was satisfactory that most patients caught complete remodeling without radiographic progression to an entry or occlusion. Of the patients included in this research, only three cases needed a stent implanting. Although one case was provided with conservative therapy initially, the abdominal pain still persisted and we chose endovascular intervention with a stent to dilate true lumen after 4 months. Postoperative symptoms were evidently relieved in endovascular group. We concluded that if people were diagnosed as spontaneous isolated intramural hematoma of SMA, hematoma absorption could be achieved in most cases which was a strong evidence for the efficacy of conservative treatment.

At early stage, SIHSMA could be misdiagnosed as superior mesenteric artery embolus according to the same CT manifestation, which was another acute onset disease and usually needed emergency surgery. Currently, the mainstay of diagnostic modalities in SMA embolus is the biphasic CT scan for vessel evaluation in the arterial phase and intestinal evaluation in the delayed phase [25]. Advanced bowel infarction can be strongly suspected in CT scan, in patients with specific intestinal findings, such as intestinal pneumatosis, portomesenteric venous gas, and nonenhancement of the bowel wall [26]. Effect of precontrast acquisition was limited for the detection of a SIHSMA. Patients with atheromatous changes were mostly accompanied by multiple sclerosis plaque in abdominal aorta.

The purpose of follow-up CT scanning was to examine the changes that occurred in the angiography and to determine the natural process of SIHSMA. The vascular remodeling of SISMAD can be classified into (1) no changes, (2) complete remodeling, (3) partial recanalization, (4) progression (total occlusion) or aneurysmal changes of the false lumen [18]. The majority of patients showed improvement or no change in both angiography and clinical symptoms after conservative management. However, Park et al. reported that complete remodeling was more likely to occur in patients with type II lesions by Yun's classification [27]. In our study, we identified that initial angiographic classification of intramural hematoma was more likely to be angiographic improvement to complete remodeling.

The present study was limited by the small sample size, single institution's data and its retrospective review nature which don't allow direct comparison with other treatment strategies. In addition, the observation time was too short to determine the development of progression and aneurysmal dilatation.

Conclusion

SIHSMA represented variable vascular remodeling while most of the patients got a complete resolution during follow-up, and its clinical course was benign in this study. Vast majority of patients can be managed conservatively when there are no signs indicating organ ischemia.

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Conflict of interest The authors declare no conflict of interest.

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