

Case Report

Ruptured Intrahepatic Arterial Aneurysm due to Segmental Arterial Mediolysis Treated with Coil Embolization

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A 67-year-old man presented with sudden-onset abdominal pain and hemorrhagic shock. Contrast-enhanced CT revealed a large hematoma in hepatic segments VI-VII with capsular rupture and hemoperitoneum. CT angiography demonstrated a saccular pseudoaneurysm in a posterior segmental branch of the right hepatic artery and a beaded appearance with irregular dilatation and segmental narrowing of the right hepatic artery branches, without inflammatory or atherosclerotic findings, supporting a diagnosis of segmental arterial mediolysis (SAM). Emergency angiography followed by coil embolization achieved hemostasis. Follow-up CT at 5 months showed complete exclusion of the aneurysmal lesions, with no evidence of recurrence or new vascular lesions. Coil embolization may be an effective, minimally invasive option for ruptured intrahepatic SAM lesions.

Key words: segmental arterial mediolysis, hepatic artery, intra-abdominal hemorrhage

Segmental arterial mediolysis (SAM) is a rare, non-atherosclerotic and non-inflammatory arteriopathy characterized by degeneration and lysis of the arterial media in medium- to large-sized arteries [1, 2]. Rupture of SAM-related visceral artery aneurysms can cause life-threatening intra-abdominal hemorrhage, with acute-phase mortality reported to be as high as 50% [2, 4]. SAM most commonly involves visceral arteries in the celiac and mesenteric territories [3-5]. Although a variety of visceral arteries may be affected, rupture of intrahepatic arterial lesions is uncommon [3-5]. Management depends on rupture status, vascular anatomy, and hemodynamic stability, ranging from conservative therapy to endovascular procedures or surgical intervention [2-4]. Because hepatic hemorrhage often presents with profound hemodynamic

instability, rapid diagnosis and timely hemostasis are critical.

Here, we report a rare case of hemorrhagic shock due to rupture of an intrahepatic arterial aneurysm with multiple aneurysmal lesions associated with SAM; hemostasis was successfully achieved by transcatheter arterial embolization (TAE) using coils.

Case Presentation

A 67-year-old man was transferred from a nursing home to our hospital because of sudden-onset abdominal pain. He had been bedridden since a previous brainstem hemorrhage but had retained the ability to communicate. On arrival, he was drowsy and hypotensive (65/30 mmHg) with tachycardia (115 beats/min), and hemorrhagic (hypovolemic) shock was suspected.

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He was afebrile on arrival. Initial laboratory tests showed a white blood cell count of 21,610/ μ l, a hemoglobin level of 9.7 g/dl, and a platelet count of 274,000/ μ l. The C-reactive protein level was 8.08 mg/dl. Liver functional reserve was Child–Pugh class B (7 points) (albumin 2.7 g/dl, total bilirubin 0.4 mg/dl, PT-INR 1.29, no ascites, and no hepatic encephalopathy). Coagulation-related parameters were as follows: PT-INR 1.29, fibrinogen 441 mg/dl, and lactate 1.3 mmol/l. After initial resuscitation with 1,000 ml of lactated Ringer's solution and transfusion of 2 units of red blood cells and 4 units of fresh frozen plasma, his systolic blood pressure transiently improved to 135 mmHg. Contrast-enhanced computed tomography (CT) was then performed. Blood cultures were obtained on arrival and on hospital days 3 and 5; all were negative.

CT demonstrated a large intrahepatic hematoma in segments VI and VII with capsular rupture and hemoperitoneum (Fig. 1A). CT angiography demonstrated a saccular pseudoaneurysm in a posterior segmental branch of the right hepatic artery and beaded caliber irregularity with aneurysmal dilatation in branches of the right hepatic artery (Fig. 1B). Definite active contrast extravasation was not identified on CT angiography, suggesting temporary hemostasis with pseudoaneurysm formation at the time of imaging. Contrast-enhanced CT of the whole abdomen revealed no aneurysms, dissections, or other vascular abnormalities in visceral arteries other than the hepatic artery. Based on the characteristic vascular morphology and

the absence of findings suggestive of inflammatory or atherosclerotic disease, the imaging findings were considered consistent with segmental arterial mediolysis (SAM). Because temporary hemodynamic stabilization had been achieved after resuscitation, emergency angiography was performed.

Selective hepatic angiography demonstrated a pseudoaneurysm in the posterior segmental branch, which was considered the culprit lesion, and beaded caliber irregularity with aneurysmal changes in both the posterior and anterior segmental branches (Fig. 2A–C). No definite extravasation was identified on the initial angiography. Treatment was initiated in the posterior branch. After distal embolization with absorbable gelatin sponge particles as a temporary embolic agent, selective coil embolization of the pseudoaneurysm was performed using detachable platinum coils (1.5 mm $\times$ 4 cm, two coils; 2.5 mm $\times$ 8 cm, one coil). The anterior branch was treated similarly: following distal embolization with absorbable gelatin sponge particles, aneurysmal lesions were selectively embolized using detachable platinum coils (4 mm $\times$ 15 cm, two coils; 4 mm $\times$ 12 cm, one coil). Final angiography confirmed complete exclusion of the pseudoaneurysm and aneurysmal lesions without residual abnormal pooling (Fig. 2D). Hemodynamics remained stable after embolization (systolic blood pressure, approximately 140 mmHg), and the patient was admitted to the intensive care unit for close monitoring. No clinical evidence of liver failure, hepatic ischemia, or biliary complications was observed. Serum transaminase levels peaked on day 2

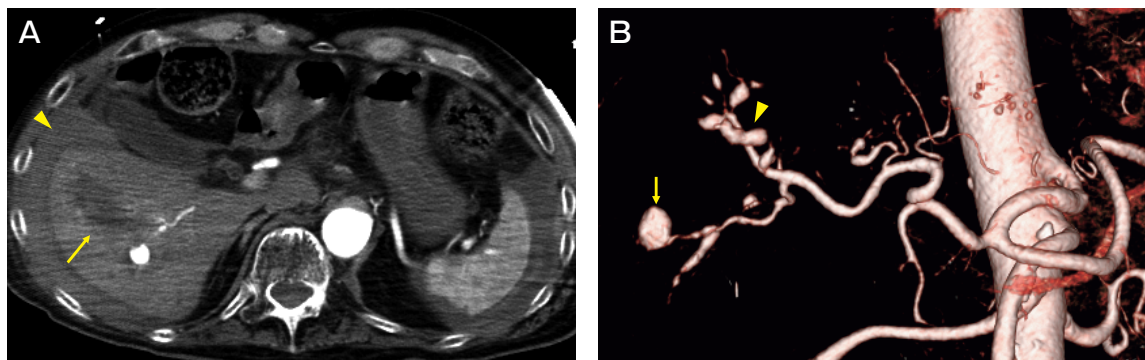


Fig. 1 Contrast-enhanced abdominal CT and CT angiography. (A) Contrast-enhanced abdominal computed tomography (CT) shows a large intrahepatic hematoma mainly in segments VI and VII (arrow) with capsular rupture and hemoperitoneum (arrowhead). (B) CT angiography demonstrates a saccular pseudoaneurysm in the posterior segmental branch of the right hepatic artery (arrow) and beaded caliber irregularity with aneurysmal dilatation in branches of the right hepatic artery (arrowhead), consistent with segmental arterial mediolysis. Definite contrast extravasation was not clearly identified at the time of imaging.

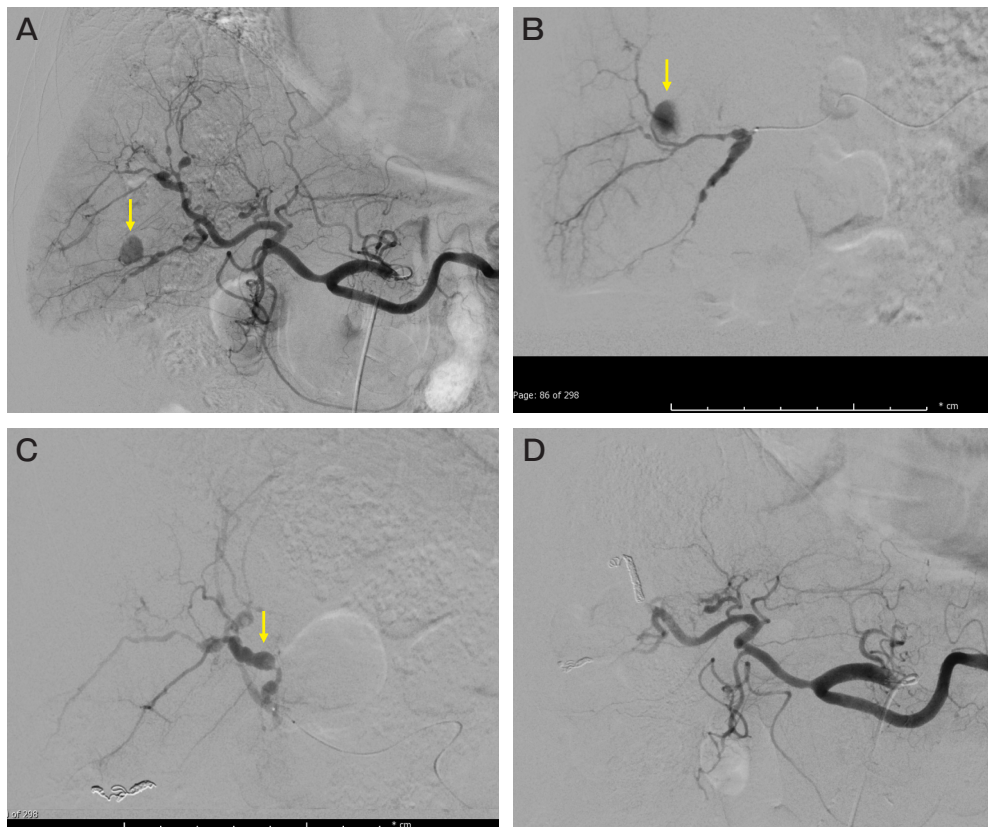


Fig. 2 Angiography and transcatheter arterial embolization. (A) Selective hepatic angiography shows beaded caliber irregularity with alternating dilatation and narrowing of the right hepatic artery branches. The arrow indicates the pseudoaneurysm in the posterior segmental branch. (B) Superselective angiography of the posterior segmental branch demonstrates the pseudoaneurysm (arrow), which was considered the culprit lesion. (C) Superselective angiography of the anterior segmental branch demonstrates aneurysmal dilatation and caliber irregularity (arrow). (D) Completion angiography after transcatheter arterial embolization (TAE) confirms complete exclusion of the pseudoaneurysm and aneurysmal lesions with no residual abnormal pooling.

after embolization (AST 368 u/l; ALT 422 u/l) and subsequently improved. Follow-up CT on day 8 showed a marked decrease in hemoperitoneum and reduction of the intrahepatic hematoma, with complete aneurysm occlusion and no new vascular lesions. The patient was discharged on day 14 after embolization without complications. Follow-up outpatient CT at 5 months showed no recurrence of aneurysms (Fig. 3A, B).

Ethics approval: This study was approved by the Ethics Committee of Kochi Health Sciences Center (approval No. 251109).

Discussion

We report a case of hemorrhagic shock due to rupture of an intrahepatic arterial aneurysm with multiple

aneurysmal lesions associated with segmental arterial mediolysis (SAM); the case was successfully managed with transcatheter arterial embolization (TAE) using coils.

SAM is a rare, non-atherosclerotic and non-inflammatory arteriopathy characterized by mediolysis of the arterial media, which can lead to arterial dissection, aneurysm formation, stenosis, and occlusion [1,2]. A characteristic feature of SAM is its biphasic course: an initial injurious phase with acute weakening of the arterial wall, resulting in dissection and/or hemorrhage, followed by a reparative phase in which granulation tissue and fibrosis contribute to arterial wall healing [7]. With increasing awareness and widespread use of contrast-enhanced CT and CT angiography, SAM is being recognized more frequently; however, uncertainties

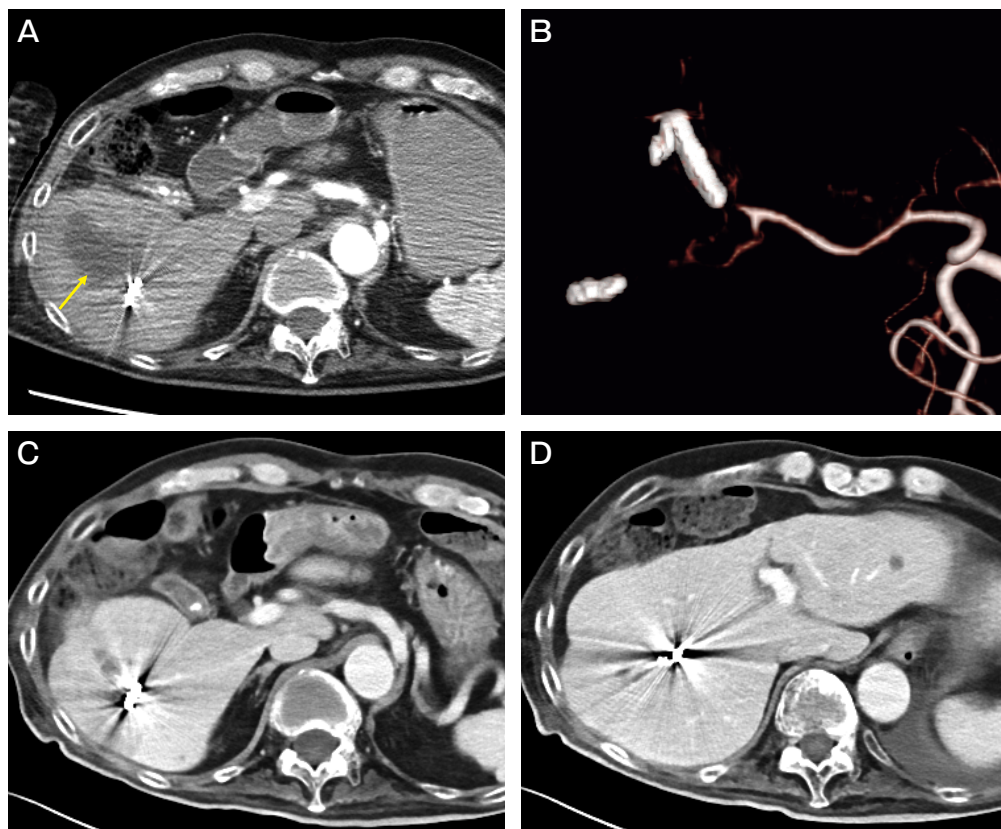


Fig. 3 Follow-up CT after transcatheter arterial embolization (TAE). (A) Contrast-enhanced CT on day 8 after TAE shows a marked decrease in hemoperitoneum and reduction of the intrahepatic hematoma (arrow). (B) CT angiography (volume-rendered image) at 5 months after TAE shows no recurrence of the pseudoaneurysm or other intrahepatic aneurysmal lesions. (C, D) Contrast-enhanced axial CT images at 5 months after TAE demonstrate coil artifacts in the posterior (C) and anterior (D) segmental branches of the right hepatic artery, with no evidence of residual or recurrent aneurysmal lesions.

remain regarding pathologic–radiologic correlation, disease progression, and prognosis [7].

Typical radiological findings of SAM include aneurysms, arterial dilatation, stenosis, and occlusion, which may persist as sequelae after the initial event [5,7]. Although SAM is considered an acute and segmental process, clinical manifestations can overlap with other vasculopathies and vascular disorders, which may complicate diagnosis [7]. SAM most commonly involves visceral arteries in the celiac and mesenteric territories, including the gastroduodenal and colonic arteries [3–5]. Rupture can be fatal, whereas spontaneous stabilization or regression has also been described during follow-up [3,4].

In a multi-institutional retrospective observational study by Shimohira [3], most unruptured SAM lesions remained stable or regressed without rupture. Notably,

a high rupture rate was reported for lesions involving the middle colic artery, suggesting substantial heterogeneity in rupture risk depending on the involved vessel [3]. In contrast, intrahepatic arterial lesions accounted for 7.3% of SAM cases and remained unruptured during the observation period [3].

Rupture of intrahepatic aneurysms due to SAM is rare. Including the present case, seven cases of intra-abdominal hemorrhage caused by rupture of intrahepatic arterial lesions associated with SAM have been documented (Table 1) [8–12]. Hemostasis was achieved by TAE in three cases, including ours [8,9]. One case was successfully managed conservatively, with subsequent regression of the lesion [10]. In the remaining cases, endovascular control was difficult, and surgical intervention was required. One patient developed abdominal compartment syndrome due to massive

Table 1 Reported cases of intra-abdominal hemorrhage due to intrahepatic arterial lesions of segmental arterial mediolysis (SAM) (including the present case)

No.	Author (Reference)	Year	Age (years)	Sex	Presenting symptom	Hemodynamic instability	Involved hepatic artery/branch	Treatment	Outcome
1	Tomonaga <i>et al.</i> [10]	2011	68	M	Abdominal pain	No	Multiple hepatic arteries	Conservative management	Survived
2	Marshall <i>et al.</i> [12]	2015	72	F	Abdominal pain	Yes	Right posterior segmental hepatic artery	TAE followed by right hemihepatectomy	Survived
3	Marshall <i>et al.</i> [12]	2015	65	F	Abdominal pain	Yes	Right posterior segmental hepatic artery	Right hemihepatectomy	Died
4	Beerle <i>et al.</i> [11]	2016	57	F	Abdominal pain	Yes	Right posterior segmental hepatic artery	Right hepatic artery ligation	Survived
5	Olivares <i>et al.</i> [8]	2018	73	F	Abdominal pain	Yes	Right hepatic artery	TAE	Survived
6	Kato <i>et al.</i> [9]	2023	68	F	Abdominal pain	No	Right posterior segmental hepatic artery	TAE	Survived
7	Present case		67	M	Abdominal pain	Yes	Right hepatic artery (anterior and posterior segmental branches)	TAE	Survived

TAE, transcatheter arterial embolization.

hemorrhage and underwent laparotomy with right hepatic artery ligation in addition to intraperitoneal decompression [11]. Two patients were reported by Marshall [12]: one presented in shock and required emergency laparotomy with hepatectomy, and the other underwent laparotomy and right hepatic lobectomy because adequate hemostasis could not be achieved after TAE. Importantly, our case includes mid-term follow-up CT at 5 months, demonstrating no recurrence of the pseudoaneurysm and no new vascular lesions suggestive of SAM recurrence. Moreover, by reviewing these seven reported cases (Table 1), we summarize their clinical presentations, management strategies, including endovascular therapy and surgery, and outcomes, thereby contextualizing the role and limitations of TAE in this rare entity.

Conditions that can mimic segmental arterial mediolysis (SAM) in the visceral arteries include inflammatory arteritides (*e.g.*, granulomatosis with polyangiitis, polyarteritis nodosa, eosinophilic granulomatosis with polyangiitis, Kawasaki disease, and other uncommon arteriopathies), mycotic aneurysms, cystic medial necrosis, and fibromuscular dysplasia [7]. Although inflammatory arteritides can overlap with SAM in clinical and laboratory findings, our case demonstrated characteristic imaging morphology—namely, a beaded appearance with alternating segmental dilatation and narrowing and multiple aneurysmal lesions—making

SAM more compatible than diffuse inflammatory arteritis [5, 7]. Mycotic aneurysm was considered less likely because blood cultures obtained on arrival and on hospital days 3 and 5 were all negative, and there were no imaging findings suggestive of marked perivascular inflammatory change [7]. Cystic medial necrosis rarely involves visceral arteries and is usually associated with stigmata of Marfan syndrome, which were not present in our patient. Fibromuscular dysplasia is uncommon outside the carotid and renal arteries, typically affects younger individuals, is often painless, and is rarely associated with hemorrhage, making it unlikely in the present case [7].

A limitation of this case is that vasculitis-specific testing, including ANCA titers, and histopathological confirmation were not performed; therefore, the diagnosis was based partly on exclusion, together with the clinical course and characteristic imaging findings. Nevertheless, the overall clinical presentation and vascular morphology were most consistent with SAM, which we considered the most plausible diagnosis.

Several technical considerations also bear mention. In our case, the culprit bleeding lesion involved a relatively broad area with multiple distal outflow branches; therefore, complete isolation of all distal vessels by coil embolization alone was technically difficult. A stepwise embolization strategy—distal embolization with absorbable gelatin sponge particles followed by selective

coil embolization—was used to reduce distal runoff and facilitate effective isolation of the bleeding segments. When selective catheterization was feasible, distal coiling was additionally performed, while proximal isolation was achievable with coils, enabling definitive exclusion of the culprit lesion.

In conclusion, SAM can cause life-threatening intra-abdominal hemorrhage when rupture occurs; however, when the patient's hemodynamics can be stabilized and the lesion is anatomically amenable, TAE with coil embolization can provide rapid hemostasis in a minimally invasive manner. Our case demonstrates that endovascular therapy can be a life-saving option even in rare cases of intrahepatic arterial involvement. Nevertheless, surgical intervention may be required when endovascular hemostasis is unsuccessful or when abdominal compartment syndrome develops.

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