

Intra-abdominal hemorrhage due to segmental arterial mediolysis of an ovarian artery pseudoaneurysm and concomitant aneurysmal subarachnoid hemorrhage: illustrative case

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BACKGROUND Aneurysmal subarachnoid hemorrhage (SAH) is one of the most severe neurosurgical diseases in which systemic management is important from the acute phase to the chronic phase. The authors reported a case of aneurysmal SAH associated with intra-abdominal hemorrhage possibly caused by segmental arterial mediolysis (SAM).

OBSERVATIONS A 60-year-old woman collapsed suddenly at home. On arrival at our hospital, she was comatose and her head computed tomography (CT) showed SAH, probably from an anterior cerebral artery aneurysm. Simultaneous body CT to screen for pneumonia associated with COVID-19 incidentally detected an intra-abdominal hematoma and the bleeding point. Emergent ventriculostomy was conducted first. Because abdominal angiography detected a ruptured pseudoaneurysm of an ovarian artery, emergency embolization was subsequently performed for hemostasis. However, she deteriorated again, and her pupils became fully dilated. The patient died on day 3 of hospitalization.

LESSONS Patients with aneurysmal SAH rarely have intra-abdominal hemorrhage in the acute stage and may have a fatal outcome. Intra-abdominal hemorrhage should be suspected in the setting of unstable vital signs, and prompt treatment is necessary.

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KEYWORDS subarachnoid hemorrhage; intra-abdominal hemorrhage; segmental arterial mediolysis; ovarian artery pseudo-aneurysm

Subarachnoid hemorrhage (SAH) is one of the most severe neurosurgical diseases in which systemic management is important from the acute phase to the chronic phase. To date, several reports have documented SAH with intra-abdominal hemorrhage. As a cause of intra-abdominal hemorrhage, segmental arterial mediolysis (SAM) was first described in 1976.¹ It is also gaining attention as a cause of intra-abdominal hemorrhage associated with SAH. The clinical course of such a patient is presented along with a review of the relevant literature.

Illustrative Case

A 60-year-old woman collapsed suddenly at home and was brought to our institution by ambulance. She had a past history of unilateral severe renal atrophy and hypertension. On arrival, her consciousness was E1V1M3, and her pupils were isocoric, normal

in size. There were no meaningful findings, including no anemia, on laboratory examination. Electrocardiography showed no findings suggestive of Takotsubo cardiomyopathy. Head computed tomography (CT) showed SAH, intraventricular hemorrhage, and intracerebral hematoma in the right frontal lobe with acute hydrocephalus, suggesting the presence of an anterior cerebral artery aneurysm on the right side (Fig. 1). After the head CT, whole-body CT was performed to screen for pneumonia due to COVID-19. However, immediately after these scans, the patient's blood pressure decreased suddenly, and her breathing deteriorated. Endotracheal intubation was subsequently performed, and continuous administration of noradrenaline was started. Whole-body CT showed hematoma from the perirenal space to the intrapelvic space on the right side. Additional contrast-enhanced CT showed an abnormal bright-

ABBREVIATIONS CT = computed tomography; SAH = subarachnoid hemorrhage; SAM = segmental arterial mediolysis.

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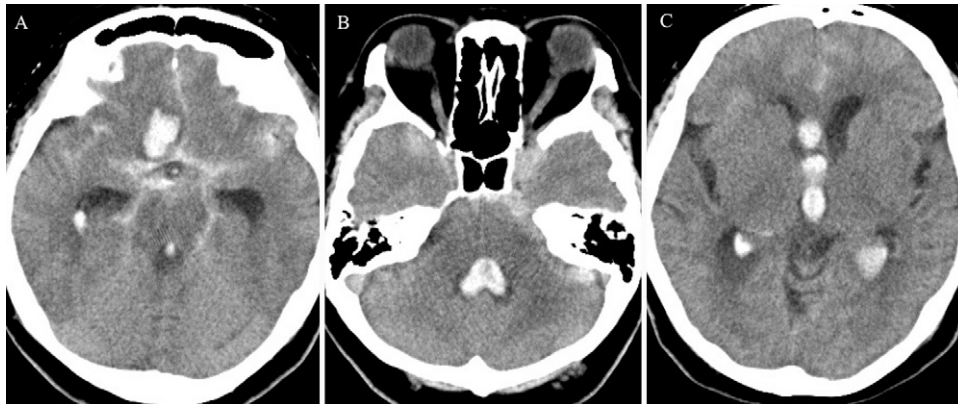


FIG. 1. Initial CT on admission (A–C) shows SAH and intracerebral hematoma in the frontal lobe with acute hydrocephalus and intraventricular hemorrhage, suggesting a ruptured anterior cerebral artery aneurysm.

signal spot suggesting extravasation in the hematoma cavity, and one of the abdominal arteries that ran forward was considered the responsible artery although head CT angiography was postponed because of her poor condition (Fig. 2). Emergency ventriculostomy was conducted first to decrease intracranial pressure when her vital signs recovered slightly. However, she was still in need of catecholamine, and vital signs remained unstable. In this case, it was determined that endovascular embolization of the intra-abdominal hemorrhage was necessary first to stabilize the vital sign. Abdominal angiography showed a pseudoaneurysm of the right ovarian artery (Fig. 3A–C). This aneurysm was consistent with a diagnosis of SAM related to a ruptured cerebral aneurysm. The responsible aneurysm causing intra-abdominal bleeding was obliterated using gelatin sponge and pushable coil embolization although extravasation occurred during the procedure (Fig. 3D and E). Immediately after treatment, the patient's vital signs worsened again.

Because she then became comatose and had dilated pupils, intracranial vascular evaluation and the treatment of SAH were cancelled. Although there was no radiographic proof regarding the presence of a cerebral aneurysm, the initial head CT showing thick SAH in the inter-hemispheric fissure with intracerebral hemorrhage in the right frontal base strongly suggested a ruptured anterior cerebral artery aneurysm. Cone-beam CT showed no increase of the intracranial hematoma, but the cortical sulci seemed to be very tight, indicating extremely high intracranial pressure. Unfortunately, the patient died on day 3 of hospitalization.

Discussion

Observations

The concept of SAM was first described by Slavin et al. in 1976.¹ The common sites were reported to be the superior mesenteric artery (53.1%), hepatic artery (44.8%), celiac artery (35.7%),

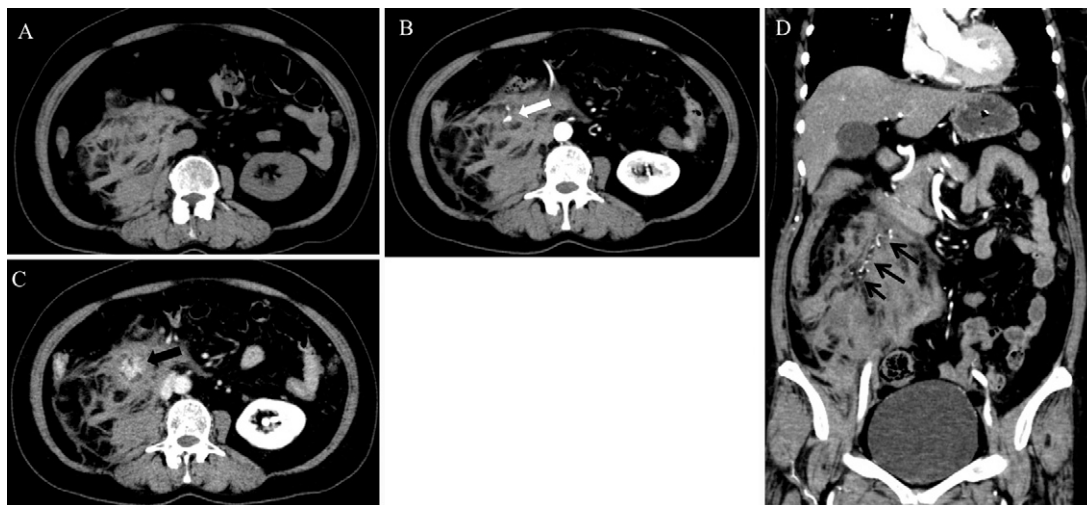


FIG. 2. A: Abdominal plain CT on admission shows massive intra-abdominal hematoma on the right side. B: Abdominal contrast-enhanced CT in the arterial phase demonstrates a spot sign in the center of the hematoma (*white arrow*). C: Abdominal contrast-enhanced CT in the venous phase demonstrates a high-density area that has increased in size, suggesting the source of the bleed (*black arrow*). D: Abdominal contrast-enhanced coronal CT shows hematoma from the perirenal area to the intrapelvic space, and the elongated right abdominal artery descends through the hematoma (*black arrows*).

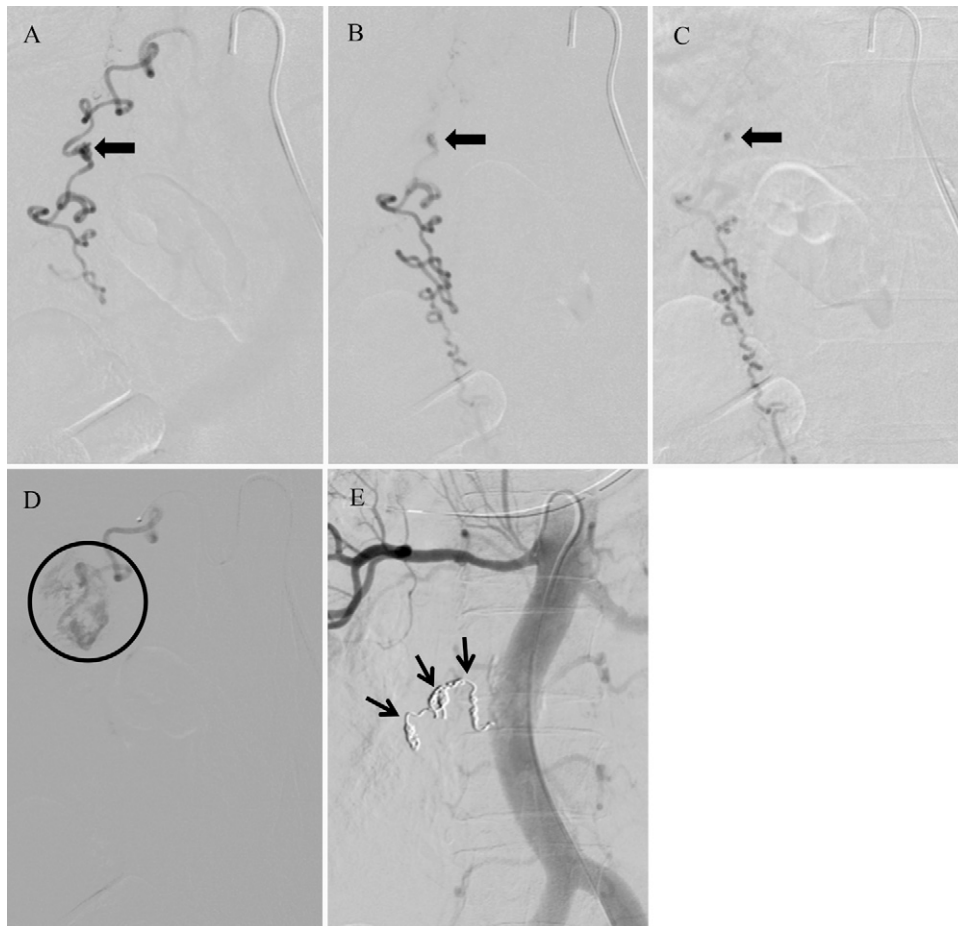


FIG. 3. Digital subtraction angiography, right ovarian artery injection (**A–C**), demonstrates the pseudoaneurysm with contrast retention in the midportion of the ovarian artery (*black arrows*). **D**: Microcatheter injection during the embolization procedure shows extravasation from the pseudoaneurysm (*black circle*). **E**: The ovarian artery is successfully obliterated using gelatin sponge and pushable coil embolization (*black arrows*).

renal artery (25.9%), splenic artery (24.5%), cerebral vessels (13.3%), and inferior mesenteric artery (10.5%), and multiple lesions were reported in 62.2%.² Although the definitive diagnosis is supposed to be made pathologically, it is difficult to harvest the specimen routinely in clinical practice. Therefore, clinical diagnostic criteria have been proposed based on imaging, laboratory data, and clinical symptoms.³ Imaging features include dissection, a spindle-shaped aneurysm, and occlusion. Other differential diagnoses to consider for SAM include fibromuscular dysplasia, polyarteritis nodosa, antineutrophil cytoplasmic antibody-associated vasculitis, giant cell arteritis, Takayasu disease, Behçet disease, mycotic aneurysm, type IV Ehlers-Danlos syndrome, Marfan syndrome, neurofibromatosis, pseudoxanthoma elasticum, atherosclerosis, and Kawasaki disease.³ The histopathological features of SAM include (1) a residual medial island with aneurysmal segmentation and fibrinous gaps due to vacuolization and lysis of the tunica media and (2) a medial gap with rupture of the intima while the dilated adventitia is preserved and no inflammation in the vessel wall or atherosclerosis of the vessel wall.⁴ Our patient also had an intra-abdominal hemorrhage due to the ruptured ovarian artery aneurysm. She was already semicomatose on arrival, and it was not possible to confirm her clinical symptoms, detailed neurological findings, or

laboratory data necessary for differential diagnosis. The possibility that a preexisting ovarian aneurysm caused intra-abdominal bleeding due to blood pressure fluctuation on arrival cannot be ruled out; however, ovarian aneurysm rupture is extremely rare. Only 25 cases were reported from 1963 to 2014, 18 (72%) of which were associated with pregnancy.⁵ In the present case, abdominal angiography showed a pseudoaneurysm of the ovarian artery. The patient was not pregnant, and abdominal bleeding occurred after SAH. Therefore, a clinical diagnosis of intra-abdominal hemorrhage associated with SAM was made. To the best of our knowledge, a total of 14 similar cases have been reported, including the present patient (Table 1).^{6–18} Multiple aneurysms were found both intracranially and extracranially. The location of ruptured cerebral aneurysms was the anterior circulation in 58.8% of cases and the posterior circulation in 41.2% of cases. The time from onset of SAH to onset of intra-abdominal hemorrhage varied from 0 to 30 days. Interestingly, 3 (21.4%) of 14 patients had both intracranial and intra-abdominal hemorrhage on the same day. The clinical manifestations at the time of onset included hypovolemic shock in 6 patients, abdominal pain in 3 patients, and impaired consciousness in 2 patients. Intra-abdominal hemorrhage due to SAM was most frequently observed in the splenic artery and superior

TABLE 1. Literature regarding intra-abdominal hemorrhage caused by SAM associated with aneurysmal SAH

Authors & Year	Age (yrs)/Sex	WFNS Grade	Cerebral Aneurysm	Tx for Cerebral Aneurysm	Symptoms of SAM	Location of SAM	Interval Btwn Ab Hemorrhage & SAH	Path Dx of SAM	Outcome
Isla et al., 1988 ¹⁶	68/F	II	A com a	Clipping	NA	NA	1 mo	No	Improved
Fuse et al., 1996 ¹⁸	56/F	II	Lt ICA, rt MCA	Clipping	Ab pain	Celliac a, gastric a	16 days	No	Improved
Sakata et al., 2002 ¹¹	48/M	V	Rt VA, lt ICA	No	Impaired consciousness	Superior mesenteric a, renal a, external iliac a	NA	Yes	Died
Soga et al., 2009 ¹³	73/F	V	NA	No	Impaired consciousness	Superior mesenteric a	NA	Yes	Died
Matsuda et al., 2012 ¹⁰	59/M	I	Rt proximal ACA, rt distal ACA, lt VA	Coating	NA	Splenic a, renal a, gastroduodenal a	NA	No	Improved
Stetler et al., 2012 ¹⁴	59/F	NA	Rt P com a	Coiling	Hypovolemic shock, ab pain	Hepatic a	Postop day 3	No	Improved
Cooke et al., 2013 ⁶	unknown (45–55/M)	V	Lt VA	Coiling	Incidental	Mammary a, renal a	NA	No	Improved
Shinoda et al., 2016 ¹²	47/M	V	Lt VA	Coiling	Hypovolemic shock, ab pain	Middle colic a	8 days	Yes	Improved
Welch et al., 2017 ¹⁷	61/M	NA	Posterior spinal a	Embolization	NA	Splenic a, posterior spinal a	Same day	No	Improved
Hellstern et al., 2018 ⁸	30/M	IV	BA, bilat ICA	Coiling & flow diverter	Hypovolemic shock	Splenic a, hepatic a	Same day	Yes	Improved
Hayashi et al., 2018 ⁷	49/F	NA	Lt ICA	Clipping	NA	Splenic a, hepatic a, superior mesenteric a, gastroduodenal a	4 days	No	Improved
Isaji et al., 2018 ⁹	45/M	II	Rt VA	Coiling	Hypovolemic shock	Middle colic a	8 days	No	Improved
Tanaka et al., 2020 ¹⁵	60/M	II	Lt ICA	High-flow bypass & trapping of C2 segment	Hypovolemic shock	Superior mesenteric a	12 days	Yes	Died
Present case	60/F	V	Rt ACA suspected	No	Hypovolemic shock	Ovarian a	Same day	No	Died

a = artery; ab = abdominal; ACA = anterior cerebral artery; A com a = anterior communicating artery; BA = basilar artery; Dx = diagnosis; NA = not applicable; ICA = internal carotid artery; MCA = middle cerebral artery; Path = pathological; P com a = posterior communicating artery; SAH = subarachnoid hemorrhage; SAM = segmental arterial mediolysis; Tx = treatment; VA = vertebral artery; WFNS = World Federation of Neurosurgical Societies.

mesenteric artery (16.7%), followed by the renal artery and hepatic artery (12.5%). Because there have been no reports describing SAM involving the ovarian artery, this may be the first case of bleeding from this vessel. Previous reports indicate that various therapeutic approaches for SAM have been adopted, including coil embolization (28%), abdominal organ surgery (24%), open arterial repair (21%), and medical management (20%). Case-specific treatment modalities yielded symptom relief in most (91%) patients, with a mortality rate of 7%.² In patients with SAH attributed to SAM, 4 of 14 patients (28.6%) died even after treatment for SAM (Table 1). These data indicate that patients with SAM associated with SAH often have a poor prognosis. Although the cause of SAM remains controversial, several authors suggested that a catecholamine surge may be relevant to intra-abdominal bleeding as well as Takotsubo cardiomyopathy.¹⁹ One article proposed that norepinephrine is the cause of SAM, reporting that SAM was found in the renal arteries, intrahepatic arteries, and coronary arteries in dogs injected with ractopamine (a β 2-adrenergic agonist). Ractopamine causes the release of norepinephrine from the peripheral nervous system.²⁰ Catecholamines are sometimes used to treat vasoconstriction, and some reports have suggested that this may also be a cause of the development of SAM. When a sudden worsening of the vital signs occurs, it is important to consider the possibility of not only Takotsubo cardiomyopathy but also intra-abdominal hemorrhage. In addition to routine examination for cerebrovascular diseases, systemic examination should be performed as soon as possible.

Lessons

Although it was possible to incidentally detect intra-abdominal hemorrhage in our patient, SAH and intra-abdominal hemorrhage can occur almost simultaneously. It is necessary to keep in mind the possibility of intra-abdominal hemorrhage when vital signs deteriorate during the acute care of a patient with SAH. Prompt diagnosis and appropriate treatment are of great importance.

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Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: Tao, Matsubara, Uno. Acquisition of data: Matsubara, Kinoshita, Fukunaga, Yamamoto. Analysis and interpretation of data: Tao, Yamamoto. Drafting the article: Tao, Matsubara. Critically revising the article: Tao, Yagi. Reviewed submitted version of manuscript: Tao, Matsubara. Administrative/technical/material support: Matsubara, Fukunaga, Yamamoto. Study supervision: Matsubara, Uno.

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