

Concurrent Intracranial and Extracranial Arterial Aneurysms : Report of Three Cases

Seung-Ryeol Shin, M.D., Tae-Sun Kim, M.D.,
Jung-Kil Lee, M.D., Je-Hyuk Lee, M.D.

Department of Neurosurgery, Chonnam National University, Medical School, Gwangju, Korea

Systemic multiple aneurysms are rare, and an association between intracranial and visceral arterial or abdominal aortic aneurysm in the same patient is a very rare occurrence. We report herein three such cases. In one case, aneurysms of the right internal carotid artery(ICA) and the right middle cerebral arterial bifurcation(MCAB) coexisted with the inferior pancreaticoduodenal arterial pseudoaneurysm and two ileal arterial aneurysms. In another case, the patient had the A-com arterial aneurysm and the right renal arterial aneurysm. And in the other patient, he had the right vertebral artery dissecting aneurysm with the abdominal aortic aneurysm. Initially, all patients were referred to our hospital with subarachnoid hemorrhage(SAH), and thereafter first two patients developed visceral arterial aneurysm rupture in the course of hospital stay and in the last patient, the abdominal aortic aneurysm was detected incidentally during carotid angiogram for Guglielmi detachable coil(GDC) embolization of vertebral dissecting aneurysm. After thorough review of our cases together with pertinent literatures, we emphasize the possibility of underlying extracranial aneurysms in ruptured intracranial arterial aneurysm patient and it's uncommon but fatal complication.

KEY WORDS : Intracranial artery aneurysm · Extracranial arterial aneurysm.

Introduction

The association of intracranial and extracranial arterial aneurysms is very rare. Until 1983, only sixteen times reported⁶⁾ and thereafter additional a few cases were reported^{7,9,14)}. We report herein three cases of concurrent existence of intracranial and abdominal arterial aneurysms and review the pertinent literature on this subject.

Case Report

Case 1

A 75-year-old woman was taken to a local hospital complaining of suddenly developed severe headache followed by a short period of unresponsiveness on the evening of November 12th, 2002. Emergency brain computed tomography(CT) was checked, and she was transferred to our university hospital to undergo surgical treatment with presumed diagnosis of subarachnoid hemorrhage. She had a stiff

neck but was neurologically normal Hunt and Hess grade²⁾. She had been diagnosed to have hypertension for 10 years and had taken antihypertensive medication, but she was mildly hypertensive(blood pressure was 150/100mmHg.) at the time of admission. She didn't have any history of systemic disease nor habit of smoking and alcohol drinking nor familial diseases. Physical examination revealed no particular abnormalities. A CT angiogram(CTA) was carried out and revealed evidence of a subarachnoid hemorrhage(SAH), small amount of intraventricular hemorrhage(IVH) with hydrocephalus and two aneurysms at right distal ICA and right MCAB(Fig. 1A, B). Cerebral angiograms were obtained on November 14th. The right carotid angiogram showed a 1.3cm multilobulated aneurysm at right ICA and a 0.5cm aneurysmal sac at right MCAB(Fig. 1C).

At that time, she had no significant neurological changes and had no specific problem on physical examination and also blood pressure was normotensive and other vital signs were stable. So, operation was planned on the next week.

On November 16th, 2002, she suddenly complained severe abdominal and back pain with cold sweating. On physical examination, she had a markedly distended abdomen with tenderness in the right hypochondrium and weak bowel sounds. Blood pressure was 80/50mmHg. Abdominal CT disclosed bleeding in the abdominal cavity(Fig. 2A). When

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• Address for reprints : Tae-Sun Kim, M.D., Department of Neurosurgery, Chonnam National University, Medical School, 8 Hak-dong, Dong-gu, Gwangju 501-757, Korea
Tel : 062) 220-6607, 6600, Fax : 062) 224-9865
E-mail : taesun@cnuh.com

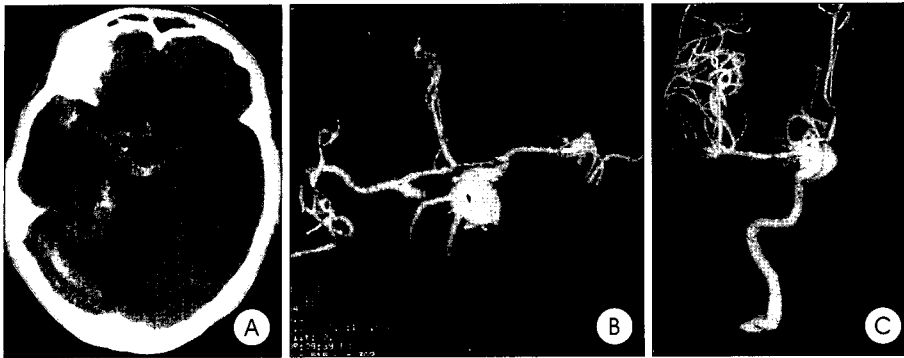


Fig. 1. A brain computed tomography(CT)(A) shows right sylvian, basal and ambient cistern dominant subarachnoid hemorrhage and CT angiogram(B) discloses aneurysms at right internal carotid artery and right middle cerebral arterial bifurcation. A right carotid angiogram shows a 1.3cm-sized multilobulated aneurysm at the right internal carotid artery and a 0.5cm-sized aneurysmal sac at the right middle cerebral arterial bifurcation(C).

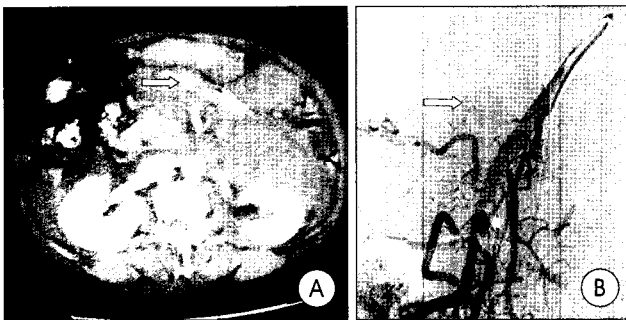


Fig. 2. An abdominal computed tomography shows aneurysm (arrow) and hematoma in abdominal cavity(A). A Celiac arteriogram shows a 1.5cm pseudoaneurysm arising from the inferior pancreaticoduodenal artery of superior mesenteric artery(large arrow) and small pseudoaneurysm arising from ileal artery(small arrow)(B).

vital signs were stabilized, we carried out a celiac arteriogram on November 20th, 2002, that revealed several aneurysms in the inferior pancreaticoduodenal artery(PDA) and ileal artery (Fig. 2B). We tried transarterial embolization(TAE)^{3,4,5,16} because of patient's high operative risks such as old age and pre-operative state of ruptured intracranial aneurysm, but failed to cannulate the feeding artery and to embolize the aneurysm. So, 6 days later, we tried TAE again, but also failed. Finally we suggested exploratory laparotomy to her family but they refused any further more treatments and evaluations. Three days later, mental state was more deepened to comatose state which was speculated to be a result of the rebleeding of ICA aneurysm but we could not check brain CT due to her family's refusal, and on January 14th, 2003, she died.

Case 2

A 52-year-old woman was admitted to our hospital complaining of suddenly developed severe headache on the morning of January 28th, 2003. She had a stiff neck and vomited once at ER. Blood pressure was elevated to 200/140

mmHg. On neurological examination, she was in drowsy mental state (Hunt and Hess grade 3, GCS 13; (E;3, V;4, M;6)) with isocoria and there was no weakness in extremities. She didn't have any history of systemic disease nor familial disease. Physical examination revealed no particular abnormalities. Laboratory examinations were normal except slightly elevated AST(41U/L) and positive result of HBV viral markers(HBs Ag, Anti-HBc and Anti-HBe). A CT angiogram was

carried out and revealed evidence of a subarachnoid hemorrhage, IVH with hydrocephalus, intracerebral hemorrhage(ICH) in left frontal lobe(Fig. 3A) and a 0.6cm-sized anterior communicating(A-com) artery aneurysm with adjacent vasospasm (Fig. 3B). Immediately, emergency operation was carried out under general anesthesia. A right pterional approach revealed an aneurysm of the A-com artery and the aneurysm was clipped. Thereafter her mental status showed no significant change with gradual stabilization of vital signs until the 8th hospital day.

On February 5th, 2003, her mental status was deepened from GCS 14 to GCS 12. A brain CT revealed increased SAH which was speculated to be the leakage of aneurysmal clipping, so we performed revision and clip repositioning with SAH removal on February 6th, 2003. But, on the day night, her mental state more aggravated to stuporous state and blood pressure decreased to 80/60mmHg with severe abdominal distension. On laboratory examinations, blood hemoglobin

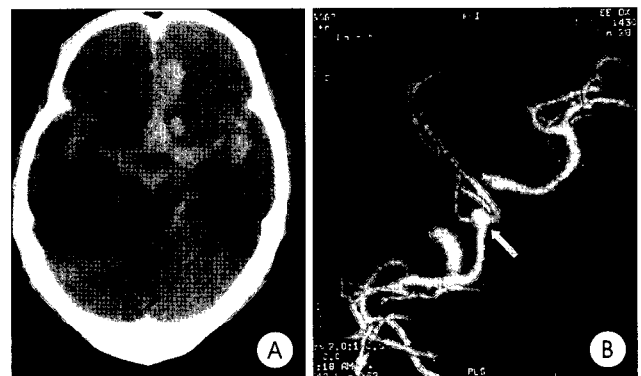


Fig. 3. A brain computed tomography(CT) shows thick subarachnoid hemorrhage with intracerebral hemorrhage(ICH) in the left frontal lobe intra cerebral hemorrhage(A) and CT angiogram shows 0.6cm A-com artery aneurysm(arrow)(B).

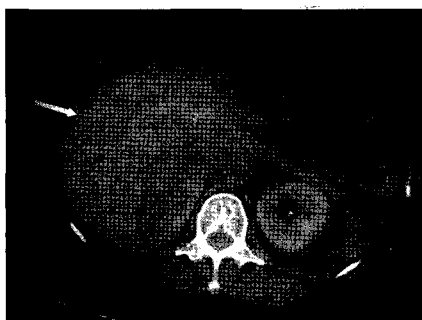


Fig. 4. An abdomen computed tomography reveals huge acute hematoma (arrow) in the right perirenal space.

decreased from 11.2g/dl to 7.0g/dl. Abdominal CT (Fig. 4) was carried out and huge acute hematoma in right perirenal space with active extravasation of contrast media was disclosed. An urological consultant recommended a right nephrectomy with presumed diagnosis of spontaneous right kidney rupture. We carried out emergency exploratory laparotomy under general anesthesia. A large-volume hematoma in the right perirenal space and blood in the peritoneal cavity was explored, and an evacuation of the clot from the surface of the kidney revealed renal arterial rupture. Hemostasis was obtained by resection of right kidney and ligation of right renal artery and vein. The etiology of intraperitoneal hemorrhage, presumed by the operator (urologist), was rupture of the renal artery aneurysm. The operation was successful but she died about 2 months later because of combined multiple poor medical conditions such as hepatic failure, renal failure and septic condition.

Case 3

A 77-year-old man with essential hypertension developed sudden onset severe headache and visited local hospital, 20 days before he was transferred to the department of neurology of our hospital. He had been treated medically, but there was no improvement of symptom. So he visited our neurology department. At admission, he had no specific symptom except

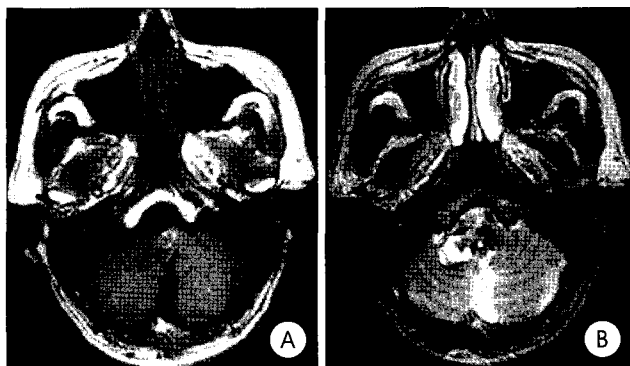


Fig. 5. A brain magnetic resonance image shows a mass lesion in the right cerebellomedullary cistern, which is iso-signal intensity in T1-weighted axial image (A) and high and iso-signal intensity in T2-weighted axial image (B). Medulla and cerebellum are compressed by this mass lesion.

headache. His neurologic examinations revealed no abnormalities and he only had essential hypertension for 30 years as a past medical history. Also his familial history was clean. A Brain magnetic resonance imaging (MRI) was checked on July 1st, 2003. This brain MRI showed 2.7 × 1.8 cm sized lobulated mass in right cerebellomedullary cistern, which was high and iso-signal intensity in T1 WI (Fig. 5A) and iso-signal intensity in T2WI (Fig. 5B). Medulla and cerebellum were compressed by this mass lesion.

This lesion was thought to be the thrombosed giant aneurysm and so he was transferred to our department. At first, we checked a brain CTA for the purpose of detecting aneurysm on July 3rd, 2003. The brain CTA disclosed about 2.7 × 1.8 cm sized mass in right cerebellomedullary cistern, consistent with thrombosed right vertebral arterial dissecting aneurysm with wall calcification (Fig. 6). We performed GDC embolization successfully on July 5th, 2003. During this procedure, at lower abdominal aortogram, an abdominal aortic aneurysm involving supra- and infrarenal abdominal aorta was incidentally detected (Fig. 7). Because the patient had no abdominal aortic aneurysmal symptom, he was discharged on July 15th, 2003, and consulted to out-patient department of general surgery. An abdominal CT was obtained on July 25th, 2003. At this abdominal CT, 5 × 4.6 cm abdominal aortic aneurysm of distal infrarenal aorta with intraluminal thrombus and severe atherosclerotic change in abdominal aorta and both common iliac arteries



Fig. 6. A brain computed tomography angiogram discloses a 2.7 × 1.8 cm sized mass in the right cerebellomedullary cistern, consistent with thrombosed right vertebral arterial dissecting aneurysm with wall calcification.

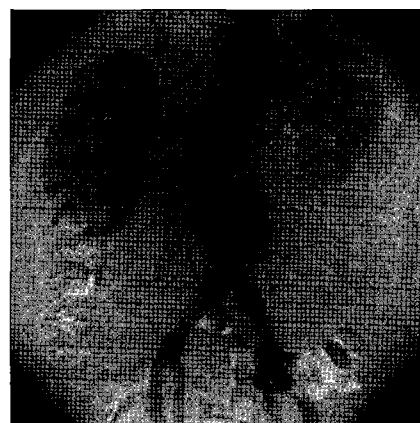


Fig. 7. A lower abdominal aortogram taken during 4 vessel cerebral angiogram reveals an abdominal aortic aneurysm involving the supra- and infrarenal abdominal aorta.

were confirmed. He was scheduled to undergo operation on August 13th, 2003.

Discussion

The infrequent association of intracranial and extracranial aneurysm has only been described sixteen times⁶⁾ until 1983, and thereafter additional a few cases were reported^{7,9,14)}. The incidence of visceral artery aneurysm is very rare. The splenic artery is the most common site of visceral artery aneurysm, accounting for 60 percent of such aneurysms. The hepatic artery is the second most common site (20 percent), and the remaining 20 percent are aneurysms of the superior mesenteric, celiac, gastroduodenal, left gastric, pancreaticoduodenal, jejunoileal, and inferior mesenteric arteries¹⁹⁾. Although splenic artery aneurysms are most common, its incidence lies within the suggested range of 0.04 percent to 0.16 percent¹²⁾ and that of intracranial saccular aneurysms between 4 to 8 percent, the two could be expected to be found together about once in every 10000. But this calculated frequency is not correct as we know.

PDA aneurysms are rare, constituting only 2 percent of all visceral artery aneurysms²⁰⁾, and the incidence of renal artery aneurysm based on autopsy studies is 0.01 percent⁸⁾. To the best of our knowledge, coexistence of intracranial aneurysm with PDA aneurysm haven't been reported in the literature. The pathology of PDA aneurysm was not certain in our case, but it was speculated to be the vascular disease of visceral arteries because gastroduodenal angiogram shows irregular beaded appearance of branches of gastroduodenal artery which is a feature characteristic of vascular disease. Also intracranial and renal aneurysms in the same patient have rarely been reported in the literature. Vaughan and Barry²²⁾ reported 3 case associated with fibromuscular dysplasia¹⁾ in 1971, and William²⁴⁾ reported one case which appears not to be associated with disease of the vascular tree in 1973. In our case, we could not find specific abnormal vascular finding on operation.

Abdominal aortic aneurysm is the most common intraabdominal aneurysm. This aneurysm is found in 2 percent of the elderly population and the incidence is increasing¹⁸⁾. However, only a few cases combined with intracranial aneurysm, which were reported. Norgard et al.¹⁴⁾ (1987) reported 5 of the 574 patients had both clinically diagnosed abdominal aortic aneurysm and clinically diagnosed intracranial aneurysm. Hideki et al.⁹⁾ (2001) also reported one case of ruptured intracranial aneurysm associated with unruptured abdominal aortic aneurysm. The etiology of aneurysms is apparently multifactorial. Norgard et al. detected high familial incidence of intracranial aneurysms and abdominal aortic aneurysms and

suggested that intracranial aneurysms and abdominal aortic aneurysms have common arterial etiologic factor. One autopsy study¹⁷⁾ strongly suggests that the development of intracranial aneurysm and abdominal aortic aneurysm in the same patient may be related to common genetic factors in the arteries, especially in the tunica media. Nowadays, abnormality of genes encoding matrix metalloproteinases and tissue inhibitors of metalloproteinases is thought to be the one possible factor responsible for the development of intracranial aneurysms and abdominal aortic aneurysms^{21,23)}. However, in our patient, familial history of intracranial aneurysm or abdominal aortic aneurysm was uncertain, although it was not researched definitely, and biopsy is not performed yet. So, we think further evaluation for the etiology of aneurysms in this patient should be performed more later on.

Conclusion

Systemic multiple aneurysms are rare, and an association between intracranial and visceral arterial aneurysms or abdominal aortic aneurysm in the same patient is a very rare occurrence but it may happen^{10,22,24)}. Hypertensive, hypervolemic therapy and administration of antiplatelet agents, which are often employed in the treatment for vasospasm in patients with aneurysmal SAH, may precipitate the rupture of concurrent asymptomatic extracranial aneurysms^{7,15)}. These cases emphasize the possibility of underlying extracranial aneurysms in ruptured intracranial arterial aneurysm patient and it's uncommon but fatal complication.

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