

Fenestration and Duplication of the Vertebrobasilar Artery Detected by Conventional Angiography and Magnetic Resonance Angiography

Jae Heung Ahn, M.D.,¹ Myoung Soo Kim, M.D.,¹ Hyun Koo Lee, M.D.,²
Seung Jun Lee, M.D.,¹ Hyo Il Park, M.D.,¹ Chae Heuck Lee, M.D.¹

Department of Neurosurgery,¹ Seoul Paik Hospital, Inje University College of Medicine, Seoul, Korea

Department of Neurosurgery,² Cheongju Saint Mary's Hospital, Cheongju, Korea

Objective : The purpose of our study is to examine the clinical significance of vertebrobasilar artery(VBA) fenestration and duplication. In addition, we review its incidence and pathogenesis.

Methods : Cerebral angiography was performed in 803 patients and magnetic resonance angiography(MRA) in 880; the patients had or were suspected to have cerebrovascular disease. We retrospectively reviewed angiography and MRA.

Results : Fifteen patients (eight men, seven women, 3 to 77 years of age, median age = 58 years) had a VBA fenestration and duplication. Seven (7/803 = 0.87%) of the patients undergoing cerebral angiography revealed fenestrations and one duplication of VBA. Ten patients (10/880 = 1.14%) among 880 patients that underwent MRA demonstrated fenestration of basilar artery(BA). Two of 66 patients that underwent both conventional cerebral angiography and cranial MRA showed a fenestration of BA. Twelve fenestrations were located in the proximal portion of the BA and one was in the mid portion of the BA. One vertebral artery(VA) fenestration was located in the intracranial portion of the right VA, and one VA duplication was at the level of C1-2 in the left VA.

Conclusion : In addition to medial defects, flow phenomena at the proximal end of fenestrations, where hemodynamic stress and increased turbulence are present, may contribute to aneurysm formation. And arterial fenestration is a predisposing factor in vascular injury and cerebral ischemia.

KEY WORDS : Fenestration · Vertebrobasilar · Clinical significance.

Introduction

Fenestration of a cerebral arteries is a known incidental angiographic or autopsy finding. Although usually not related to any clinical symptoms, there is evidence reported in the literature suggesting a relationship between fenestration and intracerebral aneurysms and transient ischemic attack, among other clinical correlations^{12,14,18,23,32,34}. In addition, the importance of understanding the complex anatomy of fenestrations and aneurysms is increased when Guglielmi detachable coil(GDC) embolization is used as an alternative treatment modality for cerebral aneurysms. However, there are few reports of fenestration of the vertebrobasilar arteries(VBA) in Korea^{1,12,19,34}. The purpose of this study was to determine the clinical implications of fenestration of VBA.

Materials and Methods

Retrospective review of the 803 conventional cerebral angiograms and 880 cranial MRAs performed over an eight-year-period yielded 15 patients with vertebrobasilar arterial fenestration. Conventional cerebral angiography (Philips V-5000, Philips Medical System, Eindhoven, Netherlands) was performed in 494 patients between July 1997 and June 2004, and cranial MRA (1.5T, Signa MR/i, General Electric, Milwaukee, WI, USA) was performed in 880 patients between May 2001 and June 2004 in one hospital.

In addition, conventional cerebral angiography (Philips V-3000, Philips Medical System, Eindhoven, Netherlands) was performed in 309 patients between June 2002 and February 2005 in another hospital. Both cerebral angiography and MRA

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• Address for reprints : Myoung Soo Kim, M.D., Department of Neurosurgery, Seoul Paik Hospital, Inje University College of Medicine, 85 Jeo-dong 2-ga, Jung-gu, Seoul 100-032, Korea Tel : +82-2-2270-0032, Fax : +82-2-2270-0573, E-mail : hanibalkms@hanmail.net

were performed in 66 of the patients. The MRAs and angiographies were undertaken for a variety of clinical reasons, including symptoms of cerebral ischemia, cerebral infarction, hemorrhagic contusion, intracerebral hemorrhage, headache, and dizziness. For the MRA studies, a three-dimensional time-of-flight (TOF) technique with a neurovascular phased array coil (MRI devices, Milwaukee, Wis., USA.) and a multiple overlapping thin slab acquisition technique was used. No intravenous paramagnetic contrast agent was administered to any of the patients. In each patient, a total of 20 maximum intensity projection (MIP) images in the frontal view (both from left lateral to right lateral, 180° and craniocaudally 180°) were routinely displayed stereoscopically. We retrospectively evaluated all of the angiograms and MRAs for cerebral arterial anatomic variations. In patients with fenestration, special attention was given to defining the origin and size of the fenestrated vessels. We also recorded the presence of associated vascular lesions, including cerebral aneurysms, vascular stenosis or occlusion, other anomalies.

Results

Fifteen patients (eight men and seven women, 3-77 years of age, median age 58 years) had a fenestration or duplication of VBA. Two of the 66 patients who underwent both conventional cerebral angiography and cranial MRA showed fenestration of the basilar artery (BA).

Seven (7/803=0.87%) of the patients who underwent cerebral angiography had a fenestration or duplication of VBA. Ten patients (10/880=1.14%) among the 880 patients who underwent MRA had fenestration of the BA.

Table 1. Summary of fifteen patients with fenestration and duplication of vertebro-basilar artery

Case	Age/sex	Location	Fenestration or duplication	Associated vessel anomaly	Symptom	Study
1	F/70	BA, middle	fenestration	Basilar trunk aneurysm	SAH	A
2	F/53	BA, proximal	fenestration	left SCA aneurysm	SAH	A
3	F/31	BA, proximal	fenestration	no	headache, paresthesia	M, A
4	F/77	BA, proximal	fenestration	no	hemorrhage	M
5	M/58	BA, proximal	fenestration	no	infarction	A
6	F/63	BA, proximal	fenestration	no	dizziness	M
7	M/50	BA, proximal	fenestration	no	TIA	M
8	M/51	BA, proximal	fenestration	no	infarction	M
9	M/72	BA, proximal	fenestration	no	TIA	M
10	M/61	BA, proximal	fenestration	no	infarction	M
11	F/3	BA, proximal	fenestration	Moyamoya disease	infarction	M, A
12	M/65	BA, proximal	fenestration	AComA fenestration	ophthalmoplegia	M
13	F/56	BA, proximal	fenestration	no	headache	M
14	M/66	left VA	duplication	no	hemifacial spasm	A
15	M/48	right VA	fenestration	AComA aneurysm	SAH	A

BA : basilar artery, SCA : superior cerebellar artery, SAH : subarachnoid hemorrhage, TIA : transient ischemic attack
 AComA : anterior communicating artery, VA : vertebral artery, A : transfemoral cerebral angiography, M : magnetic resonance angiography

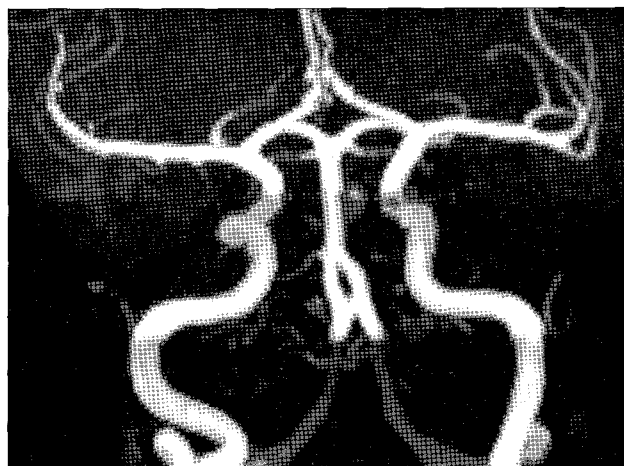


Fig. 1. Magnetic resonance angiography demonstrating fenestration at the vertebrobasilar junction.

Fenestration of the BA

Five fenestrated BAs were detected in conventional angiographic study group and 10 in MRA group : the incidences were 0.62% (5/803) and 1.14% (10/880), retrospectively. Two of the 66 patients who underwent both conventional cerebral angiography and cranial MRA demonstrated a fenestration of the BA.

Thirteen fenestrations of the BA were demonstrated by cerebral angiography or MRA (Table 1). Twelve fenestrations were located in the proximal portion of the BA and had a small slit-like shape (Fig. 1). One fenestration was located in the mid portion of the BA and had two associated aneurysms (Case 1, Fig. 2).

Fenestration and duplication of the vertebral artery

One fenestrated vertebral artery (VA) and one duplication of the VA were detected in the conventional angiographic study group. The incidences of fenestration and duplication of the VA were both 0.125% (1/803). One patient presented with a subarachnoid hemorrhage due to an anterior communicating artery aneurysm, and the other presented with contralateral hemifacial spasm (Table 1). One vertebral angiography showed an extracranial duplication of the VA at the level of C₁₋₂ (Case 14, Fig. 3). In addition, one vertebral angiography showed an intracranial fenestration of the right VA (Case 15, Fig. 4).

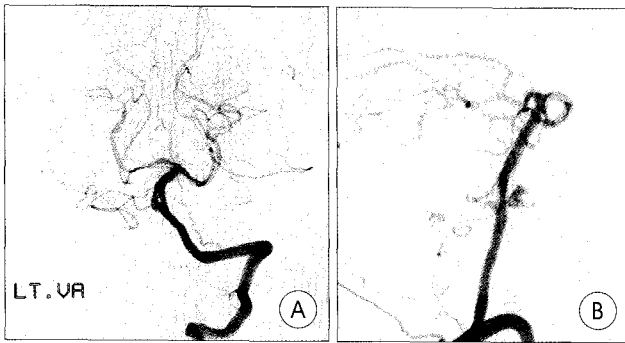


Fig. 2. Left vertebral angiogram showing fenestration at the basilar trunk and two aneurysms at the level of the fenestration (A : antero-posterior view, B : oblique view).

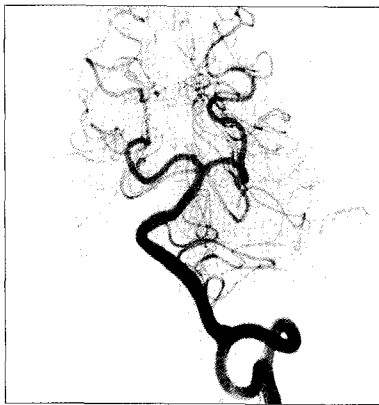


Fig. 3. Left vertebral angiography revealing that the left vertebral artery is duplicated after leaving the level of the C2 transverse foramen.

Discussion

Incidence of VBA fenestration

Fenestrations of the cerebral arteries are rare, and there is a significant discrepancy between their reported angiographic (0.03~1%) and post-mortem incidence (1.3~5.3%)^{2,24,28,30}.

Sanders et al., in their retrospective review

of 5,190 cerebral angiograms, reported 37 patients with 38 fenestrated arteries : 16 basilar (0.5%), 10 vertebral (0.19%), nine middle cerebral (0.17%), and three anterior cerebral (0.06%) arteries²⁴. Akira et al., reported a retrospective review of 600 MRAs. They stated that 10 BA fenestrations (1.7%) were detected by MRAs³¹. Only six fenestrations of the BA and two fenestrations of the VA have been previously reported in Korea^{1,12,19,34}. In this study, the retrospective incidence of BA fenestration was 0.62% (5/803) in the conventional cerebral angiography group and 1.14% (10/880) in the MRA study group. Moreover, in this study, the incidences of VA fenestration and duplication in the conventional cerebral angiography group were both 0.125% (1/803).

Pathogenesis of VBA fenestration

Fenestration of an intracranial artery refer to division of the lumen of an artery resulting in two distinct endothelium-lined channels, which may or may not share their adventitial layer²⁴.

Padgett elucidated the embryologic basis of fenestration of the basilar and vertebral arteries²². The basilar artery is formed during the second to fourth stages of embryonic development by the fusion of two primitive longitudinal neural arteries²².

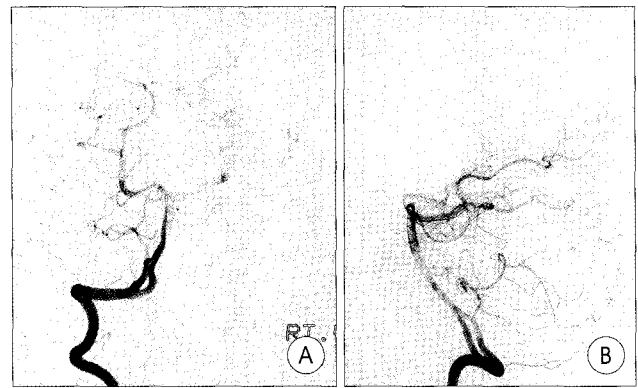


Fig. 4. Right vertebral angiogram showing fenestration of vertebral artery (A : antero-posterior view, B : lateral view).

In the early stages of fusion, these arteries are connected by several bridging areas; further fusion occurs to form the basilar artery. If these areas of irregularity persist, fenestration results; if fusion fails to occur, duplication, of the basilar artery occurs²⁹. Recently, a second embryological mechanism has been proposed for fenestration at the origin of the basilar artery, with intracranial basilar or vertebral artery fenestration consistent with persistent basivertebral anastomoses (temporary bridging arteries) beyond the fifth week of fetal life^{5,7,9,22,24}.

Fenestration of the vertebral artery is considered an anomaly occurring in the embryological developmental process. The vertebral artery forms in the embryo between the 32nd and 40th days of fetal age. Anastomoses developing between successive cervical segmental arteries lead to the formation of a primitive vertebral artery on each side in the neck. Primitive lateral basivertebral anastomoses appear transiently, while the basilar artery is formed by a pair of longitudinal neural arteries. When a portion of the primitive accessory basivertebral artery remains, this later forms an intracranial fenestration of the vertebral artery^{7,16}. Extracranial duplications or fenestrations are developed by different mechanisms. Apparently, extracranial duplication or fenestration results when a primitive segmental artery persists during the embryological development of the vertebral artery²³.

The fenestration of the extracranial VA is divided into two types according to the embryological origin. In the segmental recanalisation type, a fenestration remains in the vertebral canal between the two consecutive transverse processes while in the remnant vessel type (duplication), one of the vessels leaves the vertebral canal, enters into the spinal canal and courses in the subarachnoid space between the two consecutive transverse foramina. The duplicated vessel is an embryological remnant vessel of the paramedian longitudinal system, i.e., the lateral spinal artery¹⁵. In this study, Case 14 had a remnant type duplication of extracranial VA, and Case 15 an intracranial fenestration of the VA.

Clinical consideration of VBA fenestration

The medial walls of fenestrations have focal defects at both ends of fenestration. Focal defects on the medial wall of fenestration may lead to aneurysm formation, similar to those seen at cerebral arterial bifurcations^{5,7,8}. In addition to medial defects, flow phenomena at the proximal end of fenestrations, where hemodynamic stress and increased turbulence are present, may contribute to aneurysm formation⁶. There is much literature suggesting a relationship of fenestration to intracerebral aneurysm²³. Campos et al. presented 21 cases of vertebralbasilar junction aneurysms associate with fenestration⁶. In Case 1, we found two aneurysms at the site of the fenestration. To the best of our knowledge, this is only the third time that this abnormality has been reported^{6,29}. Fenestrations of the intracranial vessels can mimic true vascular pathological features. Kalia et al. reported a unique case of a patient with an subarachnoid hemorrhage, in whom a partially thrombosed basilar artery fenestration simulated an aneurysm on preoperative angiography¹³.

In most cases, fenestration of the vertebral artery was an incidental finding on angiography and was asymptomatic. However, in one case, it was reported that an injury to a duplicated VA by the lateral cervical spinal puncture provoked an acute subdural hematoma that resulted in death, although the fenestration itself was asymptomatic²³. In most of the cases reported previously, however, duplication of the vertebral artery was an asymptomatic, incidental finding on angiography and was occasionally associated with other vascular abnormalities such as cerebral aneurysm, arteriovenous malformation, and duplication of the basilar artery¹¹. Some authors stated that duplication of the vertebral artery itself had no clinical significance²⁶. Wollschlaeger et al., however, based on an observation made at autopsy, stated that this anomaly might give rise to symptom as the result of a small ring of pulsating compression around a cranial nerve³³. Schmidt and Pfingst reported the case of a 39-year-old woman with a left extracranial duplication of the vertebral artery; the patient had suffered from occipital neuralgia since her youth. They observed the anomalous artery of the duplication compressing the left C1 and C2 sensory roots at operation and postulated that it might have had clinical effects²⁵. And Hasegawa et al., reported a case with extracranial duplication of the vertebral artery compressing the upper cervical cord intradurally at the level of the atlas¹¹.

There have been several reports of a saccular aneurysm occurring at the fenestration of the VA^{3,15,18}. Although the occurrence of aneurysms in the fenestration is a subject of controversy, there is evidence to suggest that the existence of defects in the tunica media of the vessel at each end of the fenestration and local hemodynamic forces at the proximal site may pre-

cipitate aneurysm formation⁶. A fenestration of the VA associated with an arteriovenous malformation is much less common^{20,30}. There are several reports of vertebralbasilar fenestrations occurring either alone or with associated conditions such as neuralgia^{14,17,21} and vertebralbasilar ischemia^{4,12,27,32}. Steven et al. postulated that transient left lateral medullary ischemia in a patient with a fenestrated left vertebral artery resulted from intimal injury to the vertebral artery, the most common site for such injury being at the level of the fenestration. They proposed that arterial fenestration is a predisposing factor in vascular injury and cerebral ischemia²⁷. Hong et al. reported a vertebral angiography-demonstrated anomaly of the left vertebral artery, and a large intraluminal thrombus was detected just proximal to the fenestration. The vertebral artery was occluded proximal with a balloon, and the patient became free from ischemic attacks¹².

Because of recently accumulated clinical experience and advances in GDC production technology, GDC provides an alternative treatment modality for aneurysms. In the case of the surgically difficult aneurysms such as basilar or vertebral artery aneurysm with fenestration, GDC embolization is a highly effective treatment modality. Vigil et al. suggested that vertebralbasilar junction aneurysms with associated fenestrations could be successfully treated with GDCs¹⁰.

Conclusion

In this study, the incidence of BA fenestration was 0.62% (5/803) in the conventional cerebral angiography group and 1.14% (10/880) in the MRA study group. And the incidences of VA fenestration and duplication in the conventional cerebral angiography group were both 0.125% (1/803). In addition to medial defects, flow phenomena at the proximal end of fenestrations, where hemodynamic stress and increased turbulence are present, may contribute to aneurysm formation. And arterial fenestration is a predisposing factor in vascular injury and cerebral ischemia.

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