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Case Report

Simultaneous Dissection of Intra- and Extracranial Vertebral Artery. Report of two Cases and Review of Literature

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Summary

Two patients who developed subarachnoid haemorrhage are presented. The first patient was a 41-year-old woman whose angiograms showed right extracranial vertebral artery (VA) dissection starting at the C2 level extending to the intracranial VA near the VA union. Proximal occlusion of the right VA by the endovascular approach was performed. The second patient was a 57-year-old man whose angiograms showed the left intracranial VA dissection distal to the posterior inferior cerebellar artery and an extracranial aneurysmal dilatation of the left VA at the C1 level and extracranial VA dissection in the V3 portion of the right VA. Left intracranial VA dissection was surgically trapped, and the remaining lesions were conservatively treated.

Simultaneous dissection of the intracranial and extracranial portions of the VA is rare. Such lesions usually cause brain ischaemia, but may cause intracranial subarachnoid haemorrhage.

Keywords: Arterial dissection; subarachnoid haemorrhage; treatment; vertebral artery.

Introduction

Arterial dissection of the intracranial and extracranial vertebral arteries (VAs) has been reported separately in the past. This is because their clinical pictures and management are different, i.e., the former causes ischaemia or subarachnoid haemorrhage that frequently requires surgical intervention while the latter causes solely ischaemia in the vertebrobasilar system and is treated conservatively [4, 10, 13]. Intracranial VA dissection causing subarachnoid haemorrhage is occasionally fatal [6], but extracranial VA dissection does not cause subarachnoid haemorrhage for apparent anatomical reasons. Extracranial VA dissection and intracranial VA dissection without basilar extension causing ischaemia have a favourable prognosis in the majority of cases [4, 5, 8, 10, 13]. Simultaneous dissection of the intracranial and extracranial portions of the VA usually causes brain ischaemia, and rarely results in subarachnoid haemorrhage [7, 23]. We report two such patients presenting with subarachnoid haemorrhage and discuss their clinical significance.

Case Presentation

Patient 1

A 41-year-old woman suddenly developed headache and nausea and lost consciousness. The patient was transferred to our hospital from the local hospital on the same day. Upon admission, she was drowsy, but no apparent focal neurological deficits were observed except for mild right cerebellar ataxia. Her past history was not contributory. There was no history of trauma. Computerized tomography (CT) showed moderate hydrocephalus and intraventricular haematoma, but subarachnoid haemorrhage was not apparent. Cerebral angiograms showed right VA dissection starting at the C2 level extending to the intracranial VA near the VA union with stagnation of the blood flow (Fig. 1a). We thought that right VA dissection caused subarachnoid haemorrhage, which resulted in ventricular haematoma although CT failed to show subarachnoid haemorrhage clearly. Early intervention was preferable to prevent rebleeding, but the neuro-interventionalist (neurosurgeon in our hospital) was not available at that time. Thus, proximal VA occlusion with detachable balloons and platinum coils was finally performed on day 11 without sequelae. Post-embolisation angiograms did not show the dissected segment of the right VA (Fig. 1b). The patient was discharged on Day 30 with mild trunk ataxia. No recurrence occurred during the 9-year follow-up period. At the last follow-up, the patient was neurologically normal.

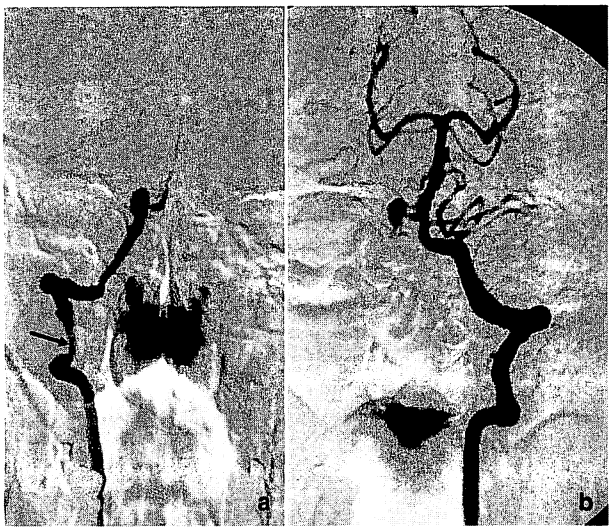

Fig. 1. Pre-embolisation right vertebral angiogram (a, frontal view) showing arterial dissection starting from the extracranial vertebral artery at the C2 level (arrow) extending to the intracranial vertebral artery near the union. Post-embolisation left vertebral angiogram (b, frontal view) fails to show the dissected portion of the right vertebral artery

Patient 2

A 57-year-old man suddenly developed loss of consciousness and respiratory disturbance and was transferred to our hospital immediately. Upon admission, the patient was comatose, but the light reflex was preserved bilaterally. The patient's past history was not contributory. There was no history of trauma. CT showed severe subarachnoid haemorrhage predominantly in the posterior fossa. Emergency angiograms revealed the left intracranial VA dissection distal to the posterior inferior cerebellar artery and an aneurysmal dilatation of the left extracranial VA at the C1 level (Fig. 2a) and extracranial VA dissection in the V3 portion of the right VA. Following the angiography, left intracranial VA dissection was surgically trapped and the remaining extracranial lesions were conservatively treated. Postoperatively, the clinical status improved drastically. The patient had mild left leg paresis and bilateral visual deterioration due to Terson's syndrome, but was otherwise normal on discharge 7 weeks later. Postoperative angiograms failed to show left V4 dissection distal to the posterior inferior cerebellar artery, but the remaining lesions were unchanged (Fig. 2b, c). Ophthalmic surgery was required for Terson's syndrome. At the 2-year follow-up from the ictus, the patient was neurologically normal.

Discussion

Extracranial VA Dissection and Spinal Subarachnoid Haemorrhage

Theoretically, extracranial VA dissection does not cause subarachnoid haemorrhage for anatomical reasons with the exception of lesions that cause spinal subarachnoid haemorrhage. Two cases with extracranial VA dissection in association with spinal subarachnoid haemorrhage have been reported in the literature [1, 9]. Kaplan *et al.* [9] reported a 41-year-old postpartum woman who had VA dissection from the C6–7 to C3 levels with a pseudo-aneurysm at the C5–6

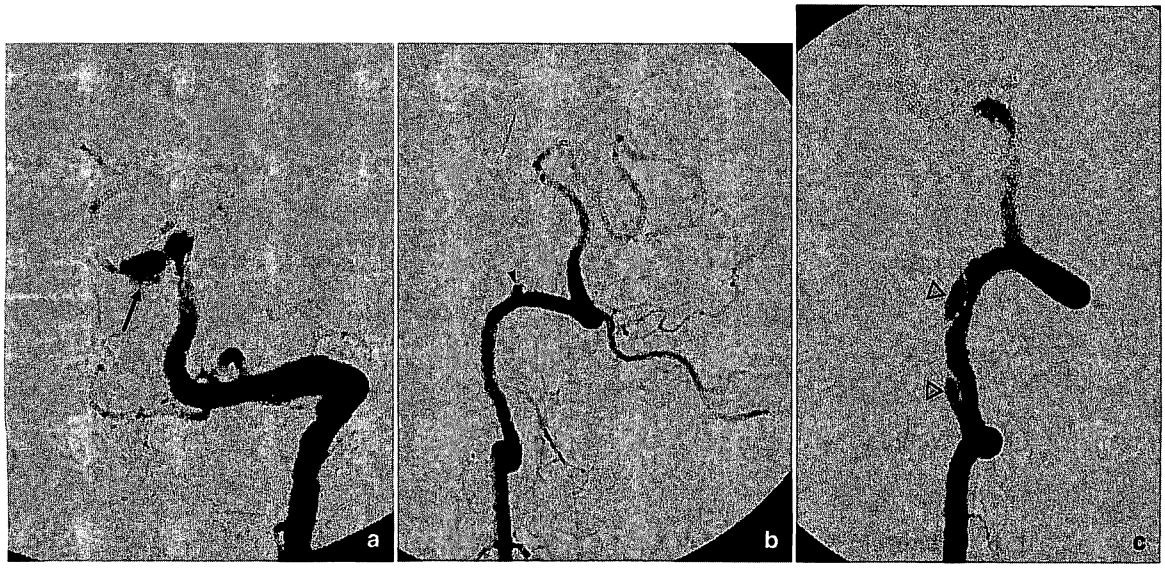

Fig. 2. Pre-operative left vertebral angiogram (a, frontal view) showing intracranial dissection (arrow) distal to the left posterior inferior cerebellar artery. Postoperative left vertebral angiogram (b, lateral view) shows disappearance of intracranial arterial dissection by trapping, but aneurysmal dilatation at the C1 level (arrowhead) remains unchanged. Postoperative right vertebral angiogram (c, lateral view) demonstrates extracranial vertebral dissection at the C1/2 (V3) level (open triangles)

level. Subarachnoid haemorrhage was presumed to be caused by the pseudo-aneurysm at the foramen of the C6 nerve root, which had a connection with the spinal subarachnoid space. The patient was treated by endovascular VA occlusion with detachable balloons. Biousse *et al.* [3] argued, however, that subarachnoid haemorrhage in Kaplan's case was caused by an undetected intracranial VA dissection even in the presence of a normal angiogram. Bahar *et al.* [1] reported a 40-year-old man with Behcet's disease who developed spinal subarachnoid haemorrhage. Angiography revealed right VA dissection in the V2 portion (C2–5 levels) with an intradural aneurysmal dilatation of the right radiculomedullary artery at the level of the C5. These two cases illustrate that extracranial VA dissection can be associated with spinal subarachnoid haemorrhage under special conditions.

Simultaneous Dissection of the Extracranial and Intracranial VA

Although Mokri *et al.* [13] reported that six of 28 VA dissections (21%) involved both the extracranial (C1–2 segment) and intracranial portions, simultaneous arterial dissections in the intracranial and extracranial VA are rare, judging from the scarcity of reported cases in the literature. Of the 24 *intracranial* VA dissections in Yamaura's series [20], which included intracranial and extracranial VA dissection distal to the exit from the transverse process of the atlas, no VA dissection occurred in the segment proximal to the dural penetration. To our knowledge, there have been 11 cases in the literature including our two cases [2, 5, 7, 8, 10, 11, 12, 23] of simultaneous spontaneous dissection of the extracranial and intracranial portions of the VA (Table 1). Reported cases with VA dissections at the V3/4 junction described vaguely are excluded. Six cases reported by Mokri *et al.* [13] were also excluded due to lack of detailed information. Of the 11 cases mentioned above, there were 7 women, 3 men and one patient whose sex was not given, ranged in age from 21 to 57 (mean of 37.1 years). VA dissection caused ischaemia in the vertebrobasilar system in 7 cases and subarachnoid haemorrhage in the remaining 4 cases. VA dissection was on the right side in 3 cases, on the left in 6, and bilateral in 2. In one case [8], dissection was observed in the entire left VA from the V1 to the V4 segments and basilar artery. Treatment was conservative in 7 patients, and in the remaining four patients (subarachnoid haemorrhage: 3 patients and ischaemia: 1 patient), either endovascular proximal occlusion with balloons/coils or surgical trapping was performed. Outcome was no deficits in 3 patients, favourable in 6 patients, moderately disabled in 1 patient, and severely disabled in 1 patient.

There are histological differences between extracranial and intracranial VAs. The intracranial (intradural) VAs lack an external elastic lamina and have a thicker internal elastic lamina, thinner adventitia, and fewer elastic fibers in the media than the extracranial VAs [19]. Extracranial arterial dissection occurs within the media or between the media and adventitia [17]. However, intracranial arterial dissection occurs between the internal elastic lamina and the media that forms intramural haematoma [22], or involves subadventitia which may rupture into the subarachnoid space [6, 16, 22]. From angiographic study, however, it is impossible to define VA dissection as either true or false aneurysm because angiography shows only intraluminal structures. Since the cases with simultaneous dissection of the extracranial and intracranial portions of the VA presented with either ischaemia or subarachnoid haemorrhage, it is also impossible to attribute the dissection to any specific layer of the VA. It is not likely that dissection of the long segment of the VA remains in the same parietal plane. The rarity of haemorrhage in extracranial VA dissection can be attributed to the anatomical structures which surround the VA in the spinal column or to the presence of the external elastic lamina and thick adventitia. There is a possibility that in rupture of the extracranial VA, dissection results in a spontaneous arteriovenous fistula, which has a communication to the surrounding venous plexus.

Youl *et al.* [23] emphasized that extracranial vertebral angiograms should be obtained as well as intracranial angiograms in the diagnosis of intracranial subarachnoid haemorrhage especially when the presence of intracranial VA dissection is not convincing. Since extracranial VA dissection is frequently observed bilaterally [4, 10, 13], VAs should be examined carefully for possible bilateral VA dissection as well as for evaluation of the collateral circulation which is of importance in formulating treatment strategies. Since cervical carotid artery dissections are commonly associated with VA dissection, 4-vessel cerebral angiography of both intracranial and extracranial vessels is necessary [13].

It is rare but possible that a patient with intracranial VA dissection presenting initially with ischae-

Table 1. *Simultaneous Dissection of Intracranial and Extracranial Vertebral Artery*

No.	Authors	Year	Age/ gender	Onset	Symptoms and signs	Right VA	Left VA	Treatment	Follow-up angiogram	Follow-up period	Outcome	Remarks
1	Berger & Wilson	1984	27/F	ischaemia	brainstem symptoms	?	V3, V4 dissection (C2 level to VA union)	conservative treatment	...	4 years	independent, right hemiparesis	oral contraceptives
2	Caplan, *et al.*	1985	27/F	ischaemia	brainstem symptoms	hypoplastic	V3, V4 dissection	no anti-coagulation	...	5 years	significant residual deficit	oral contraceptives
3	Chiras, *et al.*	1985	43/F	ischaemia	left cervical pain, cerebellar ischaemia	VA dissection from C3 to basilar junction, aneurysmal dilatation at V4	V3, V4 dissection	conservative treatment	disappearance of stenosis, persistence of aneurysmal dilatation	3 months	favourable	
4	Heat & Easton	1985	25/?	ischaemia	cervical pain, lateral medullary syndrome	?	V1, V2, V3, V4, basilar artery dissection	conservative treatment	...	...	favourable	basilar artery occlusion
5	Mas, *et al.*	1987	36/F	ischaemia	left lateral medullary syndrome	normal	V3, V4 dissection, aneurysmal dilatation at C2	conservative treatment with aspirin	normal angiography	4 years 8 months	favourable	migraine, past user of oral contraceptives
6	Youl, *et al.*	1990	44/F	SAH	posterior cervical pain, headache, seizure	V4 dissection, aneurysmal dilatation at C1	normal	conservative treatment	resolved V4 dissection, disappearance of aneurysmal dilatation	6 weeks	no deficit	steroid treatment for systemic lupus erythematosus
7	McCormick & Halbach	1993	21/M	ischaemia	pontine, cerebellar, thalamic infarction	normal	VA dissection from C1 to proximal to PICA	proximal balloon occlusion	...	...	favourable	
8	Halbach, *et al.*	1993	43/F	SAH	?	?	extracranial to intracranial dissection	proximal balloon occlusion	collateral filling of the distal VA above balloons	17 months	favourable	
9	Mascalchi, *et al.*	1997	44/M	ischaemia	right lateral medullary syndrome	V3, V4 dissection with pseudoaneurysm	normal	conservative treatment	...	...	favourable	
10	Patient 1	2002	41/F	SAH	headache, disturbance of consciousness	V3, V4 dissection	normal	proximal balloon and coil occlusion	disappearance of VA dissection	9 years	no deficit	
11	Patient 2	2002	57/M	SAH	disturbance of consciousness	V3 dissection	V4 dissection, aneurysmal dilatation at C1	trapping of the left V4 dissection by surgery	...	2 years	no deficit	Terson's syndrome

F female; *M* male; *SAH* subarachnoid haemorrhage; *VA* vertebral artery.

mia will subsequently develop subarachnoid haemorrhage. There are four such patients in the literature [14, 15, 18, 21]. This indicates that anticoagulation or antiplatelet treatment for intracranial VA dissection with an initial ischaemic presentation has a potential risk of precipitating subsequent subarachnoid haemorrhage.

In conclusion, simultaneous dissection of the intracranial and extracranial portions of the VA is rare. Although such lesions usually cause brain ischaemia, they may cause intracranial subarachnoid haemorrhage.

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Comments

As the authors themselves admit, there have been previous reports on pathology, clinics and treatment of this kind of dissections; 9 concomitant intracranial and extracranial simultaneous dissections were found in the literature and included in table 1 (together with their own 2 cases). So this is some repeat performance; however, the cases are well documented, and a comprehensive and informative literature review is given.

M. Gaab

The authors should be commended for a very interesting report of two cases with VA dissection. The data in the literature dealing with VA dissection intradural and extradural are rare. The underlying cause(s) of the dissection is disease of the artery wall and minor trauma, or, in cases of normal artery wall, a forceful trauma to the VA. The ischaemic end result following dissection of the extracranial VA, and SAH end result following the dissection of intracranial VA, is no doubt dependent also on the differences in the structure of the artery wall in its extracranial and intracranial segment. The concomitant dissection of the artery, extra- and intradural leading to final ischaemia, is also well-explained. One is in agreement with the author's opinion that in SAH as well as ischaemia in the region – and in particular in cases without obvious cause (aneurysm) for SAH – both VAs should be visualized in their extradural and intradural segments.

Since the number of patients with of VA dissections published in the literature is small, the treatment is not yet well established, and will be modified in the future with better understanding of the underlying causes as well as with earlier diagnosis of the end results of the dissections.

Knowing the course of the VA, one has the impression that with vigorous abrupt movements (stretching and/or rotation) in the neck region – as they occur in extreme sports, mainly in teen-agers – the lesion of VA could occur in more cases than have been diagnosed so far.

V. Dolenc

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