

Case Report

A Four-Vessel Cervical Artery Dissection Associated with a Tonic Seizure

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Abstract

The four-vessel cervical artery dissection is extremely rare. To our knowledge, its association with epilepsy has never been reported. A Computerized Tomography (CT) and CT Angiography (CTA) were performed in a 60-year-old patient who experienced a tonic seizure followed by right hemiplegia, right supranuclear facial weakness, dysphasia, and consciousness disorder. CTA revealed a dissection and occlusion of the left Internal Carotid Artery (ICA), a segmental occlusion of the left Vertebral Artery (VA), a hypoplasia, irregular luminal narrowing, and an occlusion of the right VA, as well as a dissection of the right ICA. Serial CT scans showed an infarction of the right occipital lobe and a larger ischemic region occupying most of the left occipital and temporal lobes, and some deep structures. The patient died, in spite of an intense anticoagulant and antiedema therapy. This could be the first patient reported with association of the four-vessel cervical artery dissection and a tonic seizure.

Keywords: Arterial dissection; Cervical arteries; Occlusion; Stenosis; Cerebral ischemia; Brain edema

Introduction

The human brain is supplied by two paired vessels: the Internal Carotid (ICA) and the Vertebral Arteries (VA) (Figure 1). Dissection of their cervical or cranial segments is usually related to the intimal tear, which enables a penetration of blood under pressure into the vessel wall with a consecutive intramural hematoma formation [1,2]. The latter leads to a lumen narrowing or occlusion and a subsequent hypoperfusion or ischemia of the cervical spinal cord, brain, retina, or certain peripheral nerves [3-5]. Dissection in general is a rare event commonly occurring in between 1.5 and 3 individuals per 100,000 population annually [1,4]. Usually 1 or 2 cervical vessels are affected, infrequently 3, and extremely rarely both ICAs and VAs [6-26]. Among the 190 patients with cervical and cranial arteries dissection, enrolled at our Hospital during the last 10 years, a single patient with bilateral carotid and vertebral artery dissection was revealed, which is an exceptional event, particularly when associated with seizures.

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Case Presentation

The 60-year-old patient was first admitted to a provincial Hospital for a tonic seizure manifested by abrupt contractions of the body musculature with an extension of the arms, legs and head, and consciousness disorder. This event was followed by dysphasia, hemiparesis, supranuclear facial weakness, and confusion. A CT examination was performed, which showed an ischemic area within the left occipital lobe. Since the consciousness disorder worsened two days later, he was transferred to our Hospital. The patient was admitted in a comatose state. The neurological examination revealed a right gaze deviation, a slower pupillary reaction bilaterally, right supranuclear facial weakness, and right hemiplegia. Laboratory analyses showed hyperglycemia and hyperlipidemia. The patient had a history of diabetes, dyslipidemia, hypertension, and cigarette smoking, but there were no signs of any hereditary disease, including a vascular collagen disorder. Since the patient had three grafts of aorto-coronary bypass placed two years ago, the echocardiography was performed. It showed blood pressure of 150/90 mm Hg, sinus rhythm, a heart rate of 82/min, a normal appearance of heart valves, atria and ventriculi, and a slight coronary insufficiency. The consciousness disorder was evaluated as 7 on the Glasgow Coma Scale, his stroke score as 27 on the NIHSS scale, and score 5 on the Rankin scale, respectively. A Computerized Tomographic (CT) examination was performed presenting ischemic areas within the right and left cerebral hemispheres. Since a vascular dissection was suspected, CT Angiography (CTA) was immediately undertaken, which showed a flame sign of the left ICA, i.e. a tapered occlusion at 18 mm from its origin (Figure 2). A similar occlusion was noticed in the left VA (Figure 2) which occupied its intradural (V3) portion, The right VA presented a hypoplasia, as well as multiple

luminal narrowing of the distal V2 and V3 segments, whilst the V4 portion was occluded (Figure 2). The cervical part of the right ICA was normal (Figure 2), but several luminal narrowing of its cavernous segment were noticed, compatible with dissection, which measured from 1.5 mm to 3.0 mm in diameter (Figure 3). Bilateral hemispheric infarctions were noticed in serial axial CT scans (Figures 4a through f). A large ischemic region of the left hemisphere occupied most of the occipital and temporal lobes. This massive ischemic areas involved the white matter and some portions of the cerebral cortex. In addition, the posterior part of the left thalamus and putamen were also affected, as well as a small part of the posterior limb of the internal capsule ipsilaterally. The ischemic region within the right hemisphere mainly occupied the occipital lobe. Signs of global brain edema were seen on CT scans as well. Due to such radiologic findings, anticoagulant and antiedema therapy was immediately introduced, whilst vascular surgery or endovascular interventions were not indicated due to the serious clinical state of the patient. However, in spite of intense medication, the patient died three days later.

infrequent, whilst four-vessel involvement has been found extremely rarely [6-26]. For instance, two, three and four-vessel dissections were reported by Béjot et al. [7] in 15.2% of their 983 patients, and by Hassan et al. [6] in 22% of 76 patients. Arnold et al. [9] revealed a three-vessel dissection in 3 cases (1.5%), and a four-vessel in one (0.1%) within a group of 740 patients. All in all, a four-vessel dissection has been reported so far in less than three dozen patients (Table 1). We found a bilateral carotid and vertebral arteries dissection in one case (0.52%) among 190 patients. The cervical arteries were most often affected in these instances (Figure 2), and rarely the cervicocranial or intracranial vessels (Table 1), as noticed in the right ICA of our patient (Figure 2). In any case, typical radiologic signs were observed,

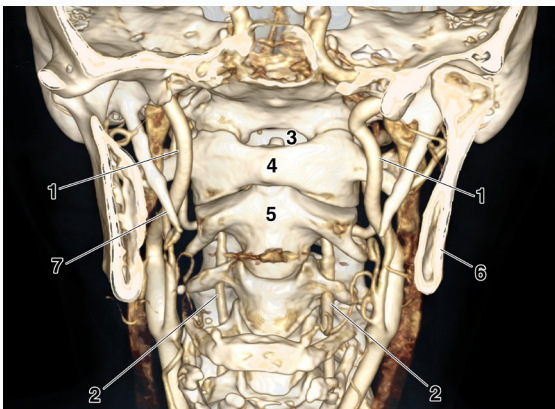


Figure 1: A coronal section of a 3D CTA image of the cervical arteries in a healthy individual. Note the right and left internal carotid arteries (1), and the right and left vertebral arteries (2). 3 – the foramen magnum; 4 – the atlas; 5 – the axis; 6 – the mandible; 7 – the styloid process.

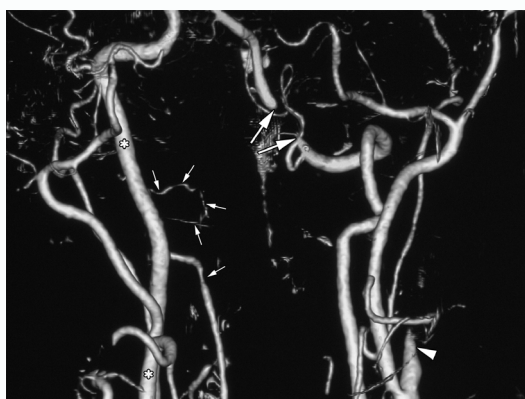


Figure 2: A 3D CTA image showing an occlusion of the initial part of the left internal carotid artery (arrowhead), an occlusion of the dural segment of the left vertebral artery (between the two larger arrows), and an irregular luminal narrowing of the hypoplastic right vertebral artery (smaller arrows). The cervical part of the right internal carotid artery is labeled by an asterisk (*).

Discussion

Most often a single cervical, cranial, or cervicocranial artery is affected by a dissection, and involvement of three arteries is very

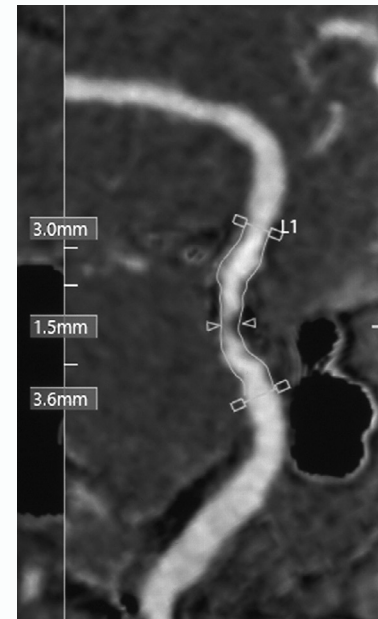


Figure 3: A CTA scan presenting a dissection of the cavernous segment of the right internal carotid artery. Note the luminal size measured in millimeters.

Table 1: Bilateral carotid and vertebral artery dissection in the literature.

No.	Authors [References]	Age/Sex	Location	Cause
1	Abe et al. [16]	52/M	Cervical	Spontaneous
2	Arnold et al. [9]	No data	Cervical	Spontaneous
3	Barroso & Demasles [18]	39/F	Cervical	Postpartum
4	Béjot et al. [7]	No data	Cervical	No data
5	Chakrapani et al. [10]	50/F	Cervical	Trauma
6	Chung et al. [19]	44/M	Cervical	Trauma
7	Coss & Jones [17]	23/F	Cervical	Trauma
8	Fukuda et al. [12]	No data	Cervical	Trauma
9	Golub et al. [11]	49/F	Cervical	Spontaneous
10	Goyal et al. [20]	28/F	Cervical	Postpartum
11	Hassan et al. [6]	50/F	Cervical	Spontaneous
12	Keilani et al. [15]	52/F	Cervical	Trauma
13	Koleilat et al. [23]	23/F	Cervical	Trauma
14	Kutlu et al. [21]	9/F	Cervical	Trauma
15	Leach & Malham [14]	No data	Cervical	Trauma
16	Marshman et al. [8]	29/F	Cervical	Spontaneous
17	Özkan Arat et al. [3]	35/F	Cervical	Trauma
18	Rees et al. [2]	43/M	Cervical	Spontaneous
19	Rudusky [22]	No data	Cervical	Trauma
20	Simon et al. [25]	31/F	Cervical	Postpartum
21	Surpur & Govindarayan [24]	36/F	Cervical	Drug
22	Yong & Heran [13]	45/F	Cerv./Cran.	Trauma

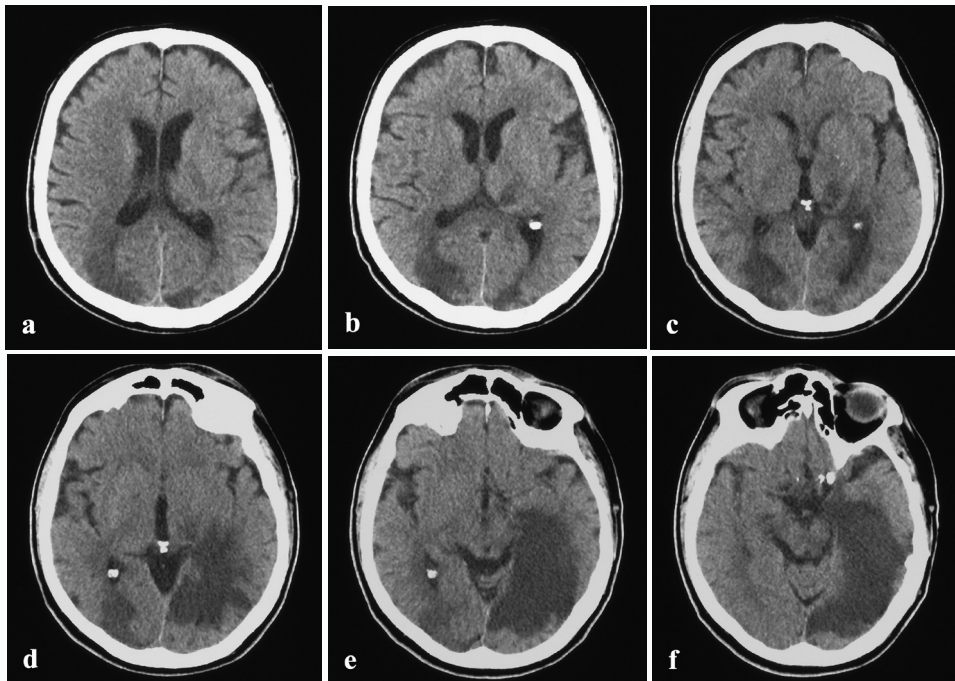


Figure 4a-f: Serial axial CT scans showing an ischemic area of the right occipital lobe and a large ischemic region of the left occipital and temporal lobes.

including an intimal tear, a flame type occlusion, or a string-and-pearl sign, i.e. irregular lumen narrowing of some arteries [1,4,5], as was the case with the right VA of our patient (Figure 2).

As regards causes and risk factors of dissection in general, usually blunt or mild trauma was noticed, as well as certain chromosomal or genetic anomalies, including connective tissue disorders, some infections, certain arteriopathies, migraine, hemodynamic changes, some drugs, iatrogenic injury, and pregnancy and the postpartum period [1,4,16,18,19,24,25]. Dissections without a certain cause are classified as spontaneous or idiopathic [1,2,4,6,8,9,11,16,20]. Similar factors were established in patients with a four-vessel dissection (Table 1). Most often a traumatic injury was noticed, i.e. in 50% (Table 1). Our male patient, aged 60 years, had no signs of connective tissue disease, or of hereditary disorders. However, he showed several important vascular risk factors, that is, hypertension, diabetes, hyperlipidemia, and smoking [1,4,19]. As regards the patient's epileptic seizure, tonic and tonic-clonic seizures, including status epilepticus, are extremely rarely associated with cervical arteries dissection and a subsequent brain infarction [26-30]. It is hard to determine whether a seizure was triggered the arterial dissection or it was a consequence of the four-vessel dissection in our patient without a history of epilepsy. If the seizure was a causal factor, muscle contractions are sufficiently strong to exert an abrupt compression and damage to the wall of the cervical arteries, and thus to lead to their dissection and brain hypoperfusion [29,30]. On the other hand, focal and global hypoxia or ischemia can result in extreme excitation and synchronization of the activity of some parts of the cerebral network and the spread of this abnormal electrical activity with a subsequent seizures appearance [30]. A three- or four-vessel dissection, like other situations with a brain hypoperfusion, may cause a global cerebral ischemia followed by brain edema [31]. This resulted in bilateral hemispheric infarctions in our patient. The left infarction (Figure 4) was larger than the right one due to a complete left ICA occlusion, and a segmental occlusion

of the left VA. The ischemic region mainly occupied the territory of the left posterior cerebral artery, due to the collateral blood flow from the opposite ICA through the circle of Willis and from the external carotid artery, which was sufficient to compensate for the supply in the middle and anterior cerebral arteries territory (Figure 4). Other authors also noticed such a collateral blood flow [11]. As regards the right occipital infarction, it was mainly a consequence of a hypoplastic, stenotic and occluded left VA, and probably of an intracerebral steal of blood from the right hemisphere by the opposite hemispheric circulation, as noticed in some other patients [32].

Conclusion

This is probably the first case of a four-vessel dissection associated with a tonic seizure. The extensive brain parenchyma injury and global cerebral ischemia were accompanied by global brain edema, which all caused deteriorating consequences in our patient. Delayed diagnosis and therapy of dissections also contributed to the lethal outcome.

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