

CASE REPORT

Cortex-sparing infarction in triple cervical artery dissection following chiropractic neck manipulation

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ABSTRACT

Background: Multivessel cervical dissection with cortical sparing is exceptional in clinical practice. **Case presentation:** A 55-year-old man presented with acute-onset neck pain with associated sudden onset right-sided hemiparesis and dysphasia after chiropractic manipulation for chronic neck pain. **Results and Discussion:** Magnetic resonance imaging revealed bilateral internal carotid artery dissection and left extracranial vertebral artery dissection with bilateral anterior cerebral artery territory infarctions and large cortical-sparing left middle cerebral artery infarction. This suggests the presence of functionally patent and interconnecting leptomeningeal anastomoses between cerebral arteries, which may provide sufficient blood flow to salvage penumbral regions when a supplying artery is occluded. **Conclusion:** Chiropractic cervical manipulation can result in catastrophic vascular lesions preventable if these practices are limited to highly specialized personnel under very specific situations.

Keywords: trivessel dissection, cortical sparing, magnetic resonance imaging, chiropractics, stroke

INTRODUCTION

Cervical artery dissection (CeAD) is a common cause of ischemic stroke under the age of 45 and accounts for about 13–22% of ischemic strokes in this population.^{1,2} 15–20% of these patients present with multivessel CeAD.³ Simultaneous dissection of three or more cervical arteries is a rare phenomenon occurring in 1.5–1.6% of spontaneous and 0.1% of traumatic extra- and intracranial artery dissections.⁴ Cervical manipulation is still widely practiced in massage parlors and barbers in the Middle East. Unaware of potential underlying spinal column or

vascular disease, patients have a neck manipulation at barber shops even though they are unsure of its therapeutic benefit, let alone the potential risk involved. It is generally agreed that the complications associated with cervical manipulation are under-reported, most probably since dissections can remain silent for days or weeks.^{5,6}

We report a case of bilateral internal carotid artery (ICA) dissection, associated with vertebral artery (VA) dissection following neck manipulation which resulted in bilateral anterior cerebral artery territory infarctions and large left middle cerebral artery infarction with cortical sparing, suggesting pre-existing collateral arteriolar connections and its influence in the different cortical involvement among stroke subtypes and on prognosis.⁷

CASE REPORT

A 55-year-old man, smoker, with history of diabetes mellitus, hypertension and dyslipidemia, presented to the emergency department with acute-onset neck pain with associated sudden onset right-sided hemiparesis and dysphasia. One week before the onset of his symptoms he had cervical manipulation for chronic neck pain. There was no family history of stroke or heritable connective tissue disorder. There were no features of neurocutaneous disorders, Marfan's syndrome or Ehlers-Danlos syndrome. Neurological exam of the patient revealed a Glasgow Coma Scale (GCS) of 14/15, right facial palsy of 1/3 and a normal gag reflex on admission. Brain CT scan showed ill-defined hypodensity in the left centrum semiovale, and bilateral old lacunar strokes.

The patient was started on aspirin and enoxaparin for deep venous thrombosis prophylaxis. After 24 hours he developed total hemiplegia and global aphasia with deterioration of his consciousness (GCS 8/15).

At that time, neurological examination showed a stuporous patient with global aphasia, right-sided gaze palsy, right facial weakness, tongue palsy, absent gag reflex, and total right flaccid hemiplegia with absent deep tendon reflexes and right Babinski sign. Sedimentation rate, electrolytes and complete blood count were all normal. The following abnormal values were found: HbA1 8.6%, CRP 51 mg/L. Specific investigations for a hereditary or acquired collagen disorder were all normal as well as values of serum reagin, total cholesterol, plasma triglycerides, homocysteine, anticardiolipin antibodies IgG and IgM, anti-Ro antibody, antinuclear antigen, anti-smooth muscle antibody, anti-Jo-1 antibody, anti-La antibody, anti-RNP antibody, anti-Sc170 antibody, rheumatoid factor and echocardiogram. Within 24 hours of the onset of symptoms, magnetic resonance imaging and magnetic resonance angiography (MRI/MRA) showed bilateral anterior cerebral artery territory infarctions and large left middle cerebral artery infarction with cortical sparing (Fig. 1A). Evaluation of the neck and head showed bilateral internal carotid and left vertebral artery dissection (Fig. 1B, C). After a thorough intensive care reassessment, where a controlled blood pressure target of 130/90 mm Hg was a treatment goal, the patient started to improve. Within 48 hours his GCS was 12/15 and the patient was able to communicate and to perform limited movements of the right extremities.



Figure 1. (A). Left fronto-temporo-parietal large infarction as well right frontal cerebral infarction with cortical sparing in MCA and bilateral ACA territory (FLAIR image). (B–C). Absence of flow along the whole course of the intracranial internal carotid artery on both sides as well as the left vertebral artery with the dominant right vertebral artery, likely secondary to dissection. (MR Angiography).

Three weeks after admission he was discharged to rehabilitation. Currently his neurological examination reveals GCS 14/15 (following simple commands), motor aphasia, and residual right hemiparesis (2/5).

DISCUSSION

CeAD is a highly variable clinical presentation which is a challenge to diagnose. A single, unilateral headache or cervical pain in oligosymptomatic cases can be underestimated and only thorough interrogation can expose circumstances related to traumatic etiologies such as chiropractic manipulations that prompt immediate MRI-MRA. The exertion of forceful neck-rotation as part of a routine procedure by chiropractors is associated with bilateral CeAD. In a large multivariate analysis, patients with dissection-related cervical artery occlusion had a significantly increased risk of suffering a severe and incapacitating stroke.⁸

Our patient presented with a triple CeAD, two carotid arteries with one vertebral artery, which is about as common as bilateral vertebral artery dissection with unilateral internal carotid artery dissection.⁴ In the study of Arnold et al., dissection of both internal carotid arteries and a vertebral artery has been reported in 4 out of 740 (0.54%) cases of spontaneous CeAD. In that study, middle cerebral artery (MCA) territory stroke was observed as the most common presentation and found in 3 out of the 4 patients. Like in most cases reported, an underlying intrinsic vasculopathy could not be demonstrated in our patient.

Different to most cases with triple CeAD, our patient had ischemic stroke in the anterior cerebral artery (ACA) and MCA territory, but largely sparing the cortex in the MCA vascular territory. This suggests the presence of functionally patent and interconnecting leptomeningeal anastomoses (LMA) between cerebral arteries which may provide sufficient blood flow to salvage penumbral regions when a supplying artery is occluded.^{9,10,11} Another study examined the degree of cortical surface involvement in patients with complete occlusion of the MCA and more than 70% of those patients had cortex-sparing infarctions in contrast to classic full-territory infarctions.¹² This can be explained by LMA among cerebral arteries which salvaged the cortical areas in the vicinity of the MCA. The main sources of origin of the leptomeningeal arteries are from the ipsilateral ACA and posterior cerebral artery (PCA).⁷

Pre-existing collateral circulation also can be a mechanism to preserve cortical tissue mainly along the MCA territory through accessory MCA branches arising from both ACAs with more clinical relevance than generally thought.¹³

In our patient, the infarction in the ipsilateral ACA territory was not associated with an extensive cortical involvement in the MCA, which can be linked to a better prognosis.

Studies have revealed that 28% of strokes following CeAD were preceded by chiropractic neck manipulation.² Furthermore, patients most commonly affected are young healthy individuals with no or few vascular risk factors.^{1,2} CeAD after chiropractic neck manipulation occurs more often in the vertebral than in the carotid arteries.^{12,14} Unlike spontaneous vertebral dissections which do not show a preferred site of dissection, by far, the most vulnerable site during chiropractic cervical spine manipulation is at the atlanto-axial joint. Ipsilateral headache is the most common symptom of dissection and may be of gradual (hours to days) or sudden onset. Neck pain, facial pain, cerebral ischemia, cranial nerve palsies, and/or pulsatile tinnitus may also occur.⁴ Horner's syndrome accompanied by pain should raise the suspicion of internal carotid artery dissection. In general, the prognosis of extracranial CeAD is favorable, with most patients experiencing resolution of symptoms and minimal deficits. However, significant neurologic and cognitive deficits, or death from a massive stroke occur in 2 – 5% of patients.¹⁵

The treatment options in CeAD cases are limited to antiaggregants or anticoagulants depending on the extent of the dissection and its location and size of the involved brain tissue. There is no evidence of therapeutic superiority of anticoagulation over antiplatelet agents and randomized controlled prospective trials are required to further investigate this.¹⁶

MRI and MR angiography were the appropriate diagnostic procedures to identify and localize the site of the dissections and to assess the extent of ischemic brain area.¹⁷ At the moment of sudden deterioration of the patient, conventional angiogram was not opted for in view of the procedure being an added risk to the patient's delicate condition regardless of its usefulness for providing more precise anatomic details.

The sudden decline of our patient after a period of apparent stabilization confirms that CeAD is highly

variable both in presentation and course which is of poorer prognosis if a previous chiropractic maneuver results in bilateral cervical arterial occlusion and stroke, the only independent factors associated with a poor outcome in large studies.^{8,18}

Manipulation of the cervical spine is a potentially life-threatening procedure performed not only by qualified physiotherapists but also by empiric personnel in barbershops without any formal training. The risks clearly outweigh the benefits as there is evidence that cervical manipulation has not shown a clear effectiveness when compared with other less dangerous procedures to relieve neck pain.¹⁹

In this scenario, it would be worthy to restrict cervical manipulation only to those qualified professionals with medical training, aware of its potential risks. Nevertheless, in a more pragmatic perspective, educational programs could encourage and help other practitioners who use cervical manipulation to undertake

this training as a mandatory prerequisite to perform their jobs, preventing those rare, but life-threatening accidents.

CONCLUSION

Multivessel CeAD with cortical sparing is rare. It is important to consider the underlying mechanisms that permit the prompt reaction of leptomeningeal anastomotic vessels which remodelate in a rapid manner, blood recirculation into the penumbra area in spite of the artery occlusion, sparing cortical tissue along MCA and ACA territories with a more favorable prognosis. Cervical arteries dissections have to be carefully ruled out in the appropriate clinical settings since complete strokes may occur within 1 to 5 days of the onset of symptoms. Potentially dangerous chiropractic maneuvers must be avoided, and should be restricted to experts in order to minimize the risks of further cervical vessel lesions.

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