

# Locked-In Syndrome Following Cervical Manipulation by a Chiropractor: A Case Report

Guillaume Giordano Orsini<sup>1</sup>, Giorgios-Emmanouil Metaxas<sup>2</sup>, Vincent Legros<sup>3\*</sup>

<sup>1</sup> Emergency Department, University Hospital of Reims, Reims, France

<sup>2</sup> Radiology Department, University Hospital of Reims, Reims, France

<sup>3</sup> Intensive Care Unit, University Hospital of Reims, Reims, France

## ABSTRACT

**Introduction:** Vertebrobasilar occlusion poses difficult diagnostic issues and even when properly diagnosed has a poor prognosis. Newer studies highlight a better outcome when thrombectomy was carried out between six and twenty-four hours after an initial diagnosis of stroke. This paper reports a case where a patient suffered a vertebrobasilar stroke secondary to a traumatic bilateral vertebral arteries dissection was treated with late thrombectomy.

**Case presentation:** A 34-year-old woman was manipulated on the cervical spinal column by a chiropractor. Following three weeks of cervical pain, she presented with severe aphasia and quadriplegia (NIHSS = 28). An MRI scan indicated ischemia of the vertebrobasilar system. Thirty-one hours after the onset of these symptoms, a thrombectomy was performed. After one month, the patient could move her head and the proximal part of her limbs but remained confined to bed (NIHSS = 13).

**Conclusion:** The current case illustrates the benefit of late mechanical thrombectomy for a posterior cerebral circulation infarct. Although there was a delay in treatment, partial recovery ensued.

**Keywords:** locked-in syndrome, stroke, vertebral artery dissection, thrombectomy

Received: 25 August 2018 / Accepted: 22 May 2019

## INTRODUCTION

Vertebrobasilar occlusion poses difficult diagnostic issues and even when properly diagnosed has a poor prognosis and attempts at recanalization can be frustrating with a mortality rate as high as 44% [1]. Newer studies highlight a better outcome when thrombectomy is carried out between six and twenty-four hours of the initial diagnosis of cerebral circulation infarct.

This paper reports a case where a patient suffered a vertebrobasilar stroke secondary to a traumatic bilateral vertebral artery dissection. The advantage of aggressive therapy by late mechanical thrombectomy is discussed.

## CASE PRESENTATION

A 34-year-old woman suffered from low cervicalgia. She consulted her chiropractor who manipulated the cervical vertebrae. Following this procedure, the pa-

tient experienced pain for several weeks without any other symptoms.

Three weeks later, at 4 a.m., she awoke with an intense headache and paraesthesia of the right side of her face and right arm.

She attended the nearest emergency department where a cerebral CT and blood tests were performed, and all interpreted as normal. She was discharged from the hospital at the end of the day. On her way back home, she became aphasic and quadriplegic. The French out-of-hospital emergency medical service responded quickly, and when they arrived at her house found her to be conscious but aphasic and with spasticity of all four limbs. She was transferred to a nearby community hospital and was prescribed clorazepate (20mg, intravenous (IV) for 48h) for hysterical conversion disorder. The following day, physical examination recorded a temperature of 40°C, a Babinski sign on the left side, a right hemi-spasticity, persistent aphasia and impaired consciousness. She was transferred to the in-

\* Correspondence to: Vincent Legros, Intensive Care Unit, University Hospital of Reims, 45 rue Cognacq Jay 51100 Reims, France. E-mail: vlegros@chu-reims.fr

tensive care unit of the University Hospital of Reims, France.

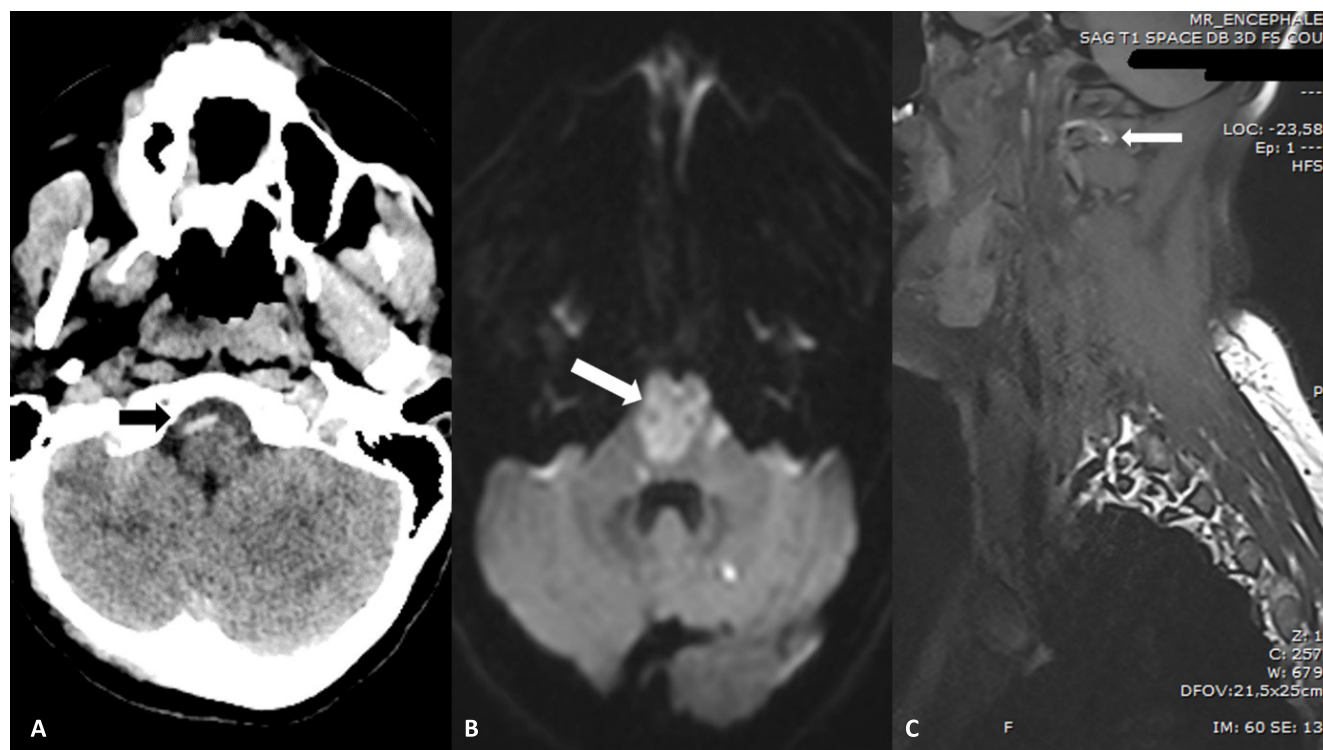
On arrival at the hospital, a cerebral CT was performed and interpreted as normal. At that time her Glasgow Coma Scale was recorded as 6/15 (E2, V2, M2), her pupils were reactive in myosis, she has bilateral pyramidal irritation signs with a National Institute of Health Stroke Scale (NIHSS) of 28. There were no meningeal signs or cranial nerve impairment. A lumbar puncture was immediately undertaken along with blood tests. She was intubated and sedated with propofol 120mg/h and remifentanyl 0.1µg/kg/mn, and probabilistic bi-antibiotic therapy with antiviral drugs was commenced (ceftriaxone 6g per day (100mg/kg/24h) in IV, amoxicillin 12g per day (200mg/kg/24h) in continuous perfusion and acyclovir 600mg/8h (10mg/kg/8h) in IV).

A review of the CT revealed a spontaneous hyperdensity of the right vertebral and basilar arteries and a systematised hypo-density of the pons. At the same time, the results of the lumbar puncture were received by phone call and reported as 0/mm<sup>3</sup> white cells, 1000/mm<sup>3</sup> red cells. A cerebral MRI was performed as an emergency and a diffusion hyper-signal with ADC restriction of the pons, the right and left cerebellum, a

minor FLAIR hyper-signal of the pons was found to exist. The T2-weighted gradient echo (Tw GE) reveals a thrombus in the basilar artery. The cervical T2 Fat Sat shows a hyper-signal of the left and right vertebral arteries wall, predominant on the second and third parts. This was suggestive of vertebrobasilar system ischemia secondary to a bilateral vertebral arteries dissection following the chiropractor's cervical manipulation (Figure 1).

An hour after her arrival in the ICU, and therefore about thirty-one hours after the beginning of the deficit, a manual thrombectomy was performed by an interventional neuro-radiologist. It highlighted the bilateral vertebral arteries dissection and an occlusive thrombus in the basilar artery. Amoxicillin, ceftriaxone and acyclovir were stopped, as was the sedation an hour after the recanalization was complete (TICI 2b-3) (Figures 2-4).

Seven days later, the patient was tracheotomised. A follow-up tomodensitometry showed an infarct of the pons. After a month, the patient was able to breathe unaided and attempted to talk with a phonatory cannula. She was able to move her head and the proximal part of her limbs but remained confined to bed.



**Fig. 1.** A- Cerebral CT showing spontaneous hyperdensity of the right vertebral artery; B- Cerebral MRI with diffusion hypersignal of the pons and in the left cerebellum; C- Cervical T2 FAT-SAT MRI showing a hypersignal in the wall of the third part of the left vertebral artery.



**Fig. 2.** Arteriography of the left vertebral and basilar arteries. The upper → shows the lack of basilar artery opacification. The lower → shows the occlusion of the last part of the right vertebral artery. The star shows the tight and irregular aspect of the right vertebral artery.



**Fig. 3.** Arteriography post thrombectomy with complete recanalization of the basilar artery.

## DISCUSSION

This case highlighted a clinical improvement following mechanical thrombectomy and demonstrated the efficacy of this procedure carried out at a late date following the onset of a severe stroke of the posterior cerebral circulation. The entire vertebrobasilar system was obstructed by a massive clot running from the ending of the two vertebral arteries to the basilar artery. A multidisciplinary dialogue between junior and senior intensivists and neuro-radiologist discussed the legitimacy and use of an aggressive recanalization therapy despite the late deadline. The treatment resulted in an improvement of consciousness and the onset of neurological recovery after a month of follow-up despite the discouraging initial MRI.

Several previous reports have highlighted the interest of early aggressive medical care of symptomatic vertebral artery dissections, particularly for the Locked-In Syndrome patient, and have indicated that a good functional outcome is possible [2]. Fields et al.(2012) showed a better recanalization rate and functional outcome with mechanical thrombectomy for stroke after dissection, with an improvement in the modified Rankin Score (mRS) for a patient with mechanical thrombectomy with or without stenting [3]. However, this study reported on a low number of patients with dissection and stroke. In another study, Lutsep et al. (2008) proved that patients with recanalization tended



**Fig. 4.** Total arteriography of the right vertebral and basilar arteries. → show the irregular aspect of the vertebral wall.

to have better outcomes than those without recanalization. Good outcomes (mRS score 0-3) were seen in 41%, and functional independence (mRS score 0-2) were seen in 33% of all patients. However, no study reported on the use of aggressive mechanical thrombectomy for vertebrobasilar occlusion in comparison with the natural history of the disease [1]. Epidemiologic studies showed that a complete recanalization was obtained for > 60% of patients after six months when medical treatment alone was employed [4].

Stenting of the vertebral artery is another therapeutic approach. Roth et al., ( 2010) reported an improve-

ment in the NIHSS of > 10 points, and 50% of patients showed an mRS < 2 at 90 days after stent-assisted mechanical recanalization [2,3,5–8].

More recently, Lindsay (2018) and Albers, et al.(2018) showed the benefit of thrombectomy, six to twenty-four hours after stroke [9,10]. These trials used the RAPID software to aid in decision-making, and it showed a thrombectomy effect up to 24 hours after symptom onset in selected patient with occlusion of proximal vessels [11]. However, as reported in an previous thrombectomy trial there was a correlation between early recanalization and clinical outcome (12), though the study was focused on the anterior cerebral circulation.

Further studies are necessary to confirm the efficacy of late thrombectomy on stroke with vertebrobasilar occlusion.

## ■ CONCLUSION

Neurological disabilities or conscience disorder in young people can be related to a stroke secondary to an artery dissection. Early diagnosis and endovascular repair condition the patient outcome. The present case shows the benefit of late (>24h) mechanical thrombectomy for a posterior cerebral circulation infarct.

## ■ CONFLICT OF INTEREST

None to declare.

## ■ BIBLIOGRAPHY

1. Lutsep HL, Rymer MM, Nesbit GM. Vertebrobasilar Revascularization Rates and Outcomes in the MERCI and Multi-MERCI Trials. *J Stroke Cerebrovasc Dis.* 2008;17:55-7.
2. Hauck EF, Natarajan SK, Horvathy DB, Hopkins LN, Siddiqui AH, Levy EI. Stent-assisted basilar reconstruction for a traumatic vertebral dissection with a large basilar artery thrombosis. *J Neurointerv Surg.* 2011;3:47-9.
3. Fields JD, Lutsep HL, Rymer MR, et al. Endovascular mechanical thrombectomy for the treatment of acute ischemic stroke due to arterial dissection. *Interv Neuroradiol.* 2012;18(1):74–9.
4. Arauz A, Márquez JM, Artigas C, Balderrama J, Orrego H. Recanalization of vertebral artery dissection. *Stroke.* 2010;41(4):717–21.
5. Alderazi YJ, Cruz GM, Kass-Hout T, Prestigiacomo CJ, Duffis EJ, Gandhi CD. Endovascular therapy for cerebrovascular injuries after head and neck trauma. *Trauma.* 2015;17(4):258–69.
6. Zaidat OO, Wolfe T, Hussain SI, et al. Interventional acute waschemic stroke therapy with intracranial self-expanding stent. *Stroke.* 2008;39(8):2392-5.
7. Levy EI, Ecker RD, Horowitz MB, Gupta R, Hanel RA, Sauvageau E, et al. Stent-assisted intracranial recanalisation for acute stroke: early results. *Neurosurgery.* 2006;58(3):458–63.
8. Roth C, Papanagiotou P, Behnke S, et al. Stent-assisted mechanical recanalisation for treatment of acute intracerebral artery occlusions. *Stroke.* 2010;41(11):2559–67.
9. Albers GW, Marks MP, Kemp S, et al. Thrombectomy for Stroke at 6 to 16 Hours with Selection by Perfusion Imaging. *N Engl J Med.* 2018;378(8):708-18.
10. Nogueira RG, Jadhav AP, Haussen DC, et al. Thrombectomy 6 to 24 Hours after Stroke with a Mismatch between Deficit and Infarct. *N Engl J Med.* 2018;378(1):11-21.
11. Ragoschke-Schumm A, Walter S. DAWN and DEFUSE-3 trials: was time still important? *Radiologe.* 2018;58(1):20-3.
12. Saver JL, Goyal M, Van der Lugt AAD, et al. Time to treatment with endovascular thrombectomy and outcomes from ischemic stroke: a meta-analysis. *JAMA.* 2016;316(12):1279–89.