**Case Report**

**Delayed Collet-Sicard syndrome after internal carotid dissection and Jefferson fracture. Case report and Review of Literature**

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**ABSTRACT**

**Background:** Lower cranial nerve palsies, or Collet-Sicard syndrome, can be caused by many different etiologies including head trauma, basilar occipital fractures, tumors, and interventions. Few reports describe different presentations of this condition, and we present here a case study to increase awareness of and add to the variable spectrum.

**Case Description:** A 56-year-old who had been hit while diving was admitted to our department. On examination, he was conscious without any signs of lateralization but presented with severe neck pain. CT brain and cervical spine revealed a C1 fracture with bilateral symmetrical fracture of the anterior and posterior arches (Jefferson's fracture) and slight bilateral joint dislocation C1-C2 discreetly predominant on the left. One week later, he presented with dysarthria, dysphonia, swallowing disorder, anisocoria, tongue deviation, and palate deviation (XII, IX, and X). CT Angiography showed dissection of the internal carotid artery immediately after the carotid bulb. He has been treated conservatively with curative anticoagulants with stable symptoms. No surgical intervention had been proposed.

**Conclusion:** Adding to the literature, delayed Collet-Sicard syndrome and lower cranial affection can be caused by missed carotid wall hematoma following severe craniocervical trauma associated with Jefferson's fracture.

**Keywords:** Collet-Sicard syndrome, Injury, Internal carotid dissection, Jefferson fracture, Lower cranial nerves

**INTRODUCTION**

In 1915, Frederic Collet[2] first reported the clinical presentation of the first case of the last four cranial nerves paralysis (IX-XII) following a bullet injury in the mastoid region in a World War I soldier. Later, a new report was added by Jean Sicard.[21] Other possible causes could be closed head trauma or occipital condylar rare fracture. Subsequent possible mechanisms of cranial nerves affection could be explained by transmitting the trauma energy through the skull base, plugging jugular foramen by a displaced bony fragment.[8,19]

Very few reports in the literature are describing the situation with different presentations. Usually, the symptoms and signs are dramatic and presented soon after trauma. Consequences of intracranial or even extracranial events can contribute to nerve affection mechanisms and
subsequent manifestations such as bulbar palsy, motor, sensory deficits, dysphagia, or dysphonia.\textsuperscript{[5]}

Each single case study could add to the spectrum of this rare presentation. Some patients were treated conservatively with very good outcome and recovery or stable clinical course.\textsuperscript{[13]} While others were treated surgically, especially in the presence of bony fragments compression on the nerves.\textsuperscript{[20]}

We present here a unique case of Jefferson fracture C1 with internal carotid dissection (ICD) causing delayed lower cranial nerve affection to add to the spectrum in the literature.

**CASE DESCRIPTION**

A 56-year-old male had been hit while diving in a swimming pool and presented with severe post-traumatic cervical pain, with active and passive movements. On clinical examination, he was conscious and oriented without any signs of lateralization. In addition, there was no motor or sensory deficit.

X-ray cervical spine did not show any abnormalities. Computed tomography (CT) of the brain and cervical spine study showed C1 fracture; bilateral symmetrical fracture of the anterior and posterior arches of C1 (Jefferson's fracture) with slight bilateral joint dislocation C1-C2 discreetly predominant on the left. Spine magnetic resonance imaging (MRI) showed straightness of the cervical spine, and no disturbance of the coronal statics, and no constitutional cervical ductal stenosis as a secondary stenosis. No abnormality of the medullary signal as well as paraspinal soft tissue. Cervical angioscan was not done in the first admission.

On admission, the patient was managed conservatively in the form of anxiolytics, calcium channel inhibitors, and strong opioid analgesics. No surgical intervention was required, and the clinical situation remained stable with pain improvement.

Patient was discharged 2 days after admission on conservative treatment and neck collar.

One week later, he presented with mild complaints of dysarthria, dysphagia, and dysphonia. Clinical examination found no deficit except deviated tongue and palate, without ptosis or anhydrosis. Normal blood pressure and oxygen saturation, but sinus bradycardia were observed and required intensive care unit admission.

CT angiography showed right internal carotid artery occlusive dissection and carotid wall hematoma just above the carotid bulb [Figure 1] and showed ophthalmic artery opacification. Brain MRI excludes cerebral strokes.

The patient was observed in the ICU for a few days before he was discharged to the service. Introduction of curative anticoagulant was conducted. No surgical intervention had been proposed. The patient remains stable.

**DISCUSSION**

Lower cranial nerves affection could be caused directly or indirectly by several mechanisms following different causes as shown in [Table 1].\textsuperscript{[1-4,6-9,11-18,20-22]} Primary causes could be open, closed cranio-cervical junction traumas, bullet injuries are part of the main causes and rarely reported.\textsuperscript{[5]} Following possible mechanisms and contributing factors causing nerve palsy could be transmitting energy through the skull base, bony fragments in the jugular foramen, and displacement of the cranium in relation to the cervical spine during trauma itself, brainstem edema, and ischemia.\textsuperscript{[6]}

Internal carotid artery dissection and occlusion are reported following different ways of cervical and head trauma.\textsuperscript{[10,19]} We report a unique case study presentation of carotid wall hematoma and dissection associated with Jefferson fracture and causing lower cranial nerve affection in the context of Collet-Sicard syndrome.\textsuperscript{[2,21]}

The wide spectrum of etiologies, mechanism, clinical presentation, and outcome has been reported through few case reports. High-resolution imaging studies are required to precisely define the mechanism of the conditions and degree of affection. Craniocervical CT and MRI are routinely used in the reported cases, but angiography is not routinely required.\textsuperscript{[13]}

Different management strategies can be applied including conservative and surgical intervention depending on the primary etiology. Moreover, with conservative treatments, outcome varies from good recovery\textsuperscript{[6]} to stable clinical condition.\textsuperscript{[9]} Surgical techniques also vary depending on the etiology to be either major surgery or removing the bony fragments compressing the cranial nerves in the jugular foramen.\textsuperscript{[20]}

![Figure 1: CT angiography axial section image at cervical spine (C1) shows occluded dissected internal carotid artery on the right side (black arrow) in comparison to the patent internal carotid artery on the left (white arrow).](image_url)
Spontaneous internal carotid artery dissection has been reported with headache, neck pain, and Horner syndrome. Several internal carotid artery pathologies can cause Collet-Sicard syndrome and lower cranial nerve affections such as aneurysm, dissection, and coiling. Mild cervical trauma has been reported to cause Collet-Sicard syndrome but without Jefferson fracture. We report here the first case combining Jefferson fracture and ICD causing delayed Collet-Sicard syndrome. Rereading of the first MRI showed a suspicion of right ICD. We believe that an early detection of carotid dissection will allow preventing lower nerves injury by the introduction of curative anticoagulants.

CONCLUSION

Adding to the literature, delayed Collet-Sicard syndrome and lower cranial affection can be caused by carotid wall hematoma following severe craniocervical trauma associated with Jefferson's fracture.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

Table 1: Possible etiologies of Collet-Sicard syndrome in the literature.

| Author          | Year  | No.  | Mechanism/Association                                  | Management             | Outcome               |
|-----------------|-------|------|--------------------------------------------------------|------------------------|-----------------------|
| Collet[1]       | 1915  | 1    | Gunshot injury                                        | Surgery                | Improved              |
| Sicard[2]       | 1917  | 1    | Head trauma                                            | Conservative           | Stable                |
| Lapresle et al.[3] | 1980  | 1 (28 y) | Angiography                                           | Conservative           | Improved              |
| Hashimoto et al.[4] | 1988  | 1(71 y) | Severe head and cervical spine injury, OC fracture     | Conservative           | Good recovery         |
| Silvestrini et al.[5] | 1991  | 1 (51 y) | Coiling of the left internal carotid artery just below the skull base | Conservative           | Improved              |
| Prick and Verhagen[6] | 1992  | 1 (61 y) | Cardiac surgery complication (Patent Ductus Arteriosus) | Conservative           | Normal after 1 year   |
| Sharma et al.[7] | 1994  | 1 (35 y) | OC fracture                                            | Surgery                | Partial neurological recovery |
| Rees et al.[8]  | 1997  | 1     | Spontaneous ICD                                       | Gastrostomy            | Deteriorated          |
| Larson et al.[9] | 1997  | 1 (67 y) | Jugular foramen tumor                                  | Conservative           | Rapidly progressive   |
| Miyazaki et al.[10] | 2000  | 1 (52 y) | OC fracture involving the jugular foramen              | Conservative           | Stable                |
| Heckmann et al.[11] | 2000  | 1 (44 y) | ICD                                                   | Conservative/anticoagulants | Stable               |
| Mohr et al.[12] | 2005  | 1 (35 y) | ICD                                                   | Conservative           | Improved              |
| Erol et al.[13] | 2007  | 1 (31) | Closed HI, OC fracture + EDH                           | Conservatively          | Partial neurological recovery |
| Battaglia et al.[14] | 2009  | 1 (57 y) | ICD post HI                                           | Conservative/anticoagulants | Persistent symptoms   |
| Handleyet al.[15] | 2010  | 1 (30 y) | IJV thrombosis                                         | Conservative           | Dramatically improved |
| Petrović et al.[16] | 2011  | 1 (57 y) | Neck fibrosarcoma                                      | Surgical Resection     | Not reported           |
| Rios et al.[17] | 2015  | 1 (81 y) | Jugular foramen tumors                                 | Stereotactic radiosurgery | Only dysphagia has improved slightly |
| Neo et al.[18]  | 2017  | 1 (71 y) | IJV thrombosis induced by central venous catheter     | Anticoagulants         | Recanalization and improved clinically except wasted tongue |
| De Oliveira et al.[19] | 2019  | 1 (61 y) | Jugular Foramen Schwannaoma                           | Radiotherapy           | Stable                |

ICD: Internal carotid dissection, HI: Head injury, IJV: Internal jugular vein, OC: Occipital condyle, EDH: Epidural hematoma

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Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Battaglia F, Martini L, Tannier C. Syndrome de Collet-Sicard après dissection carotidienne. Rev Neurol (Paris) 2009;165:588-90.
2. Collet F. Sur un nouveau syndrome paralytique pharyngo-larynge par blessure de guerre. Lyon Med 1915;124:121–9.
3. de Oliveira IA, Cordeiro BM, Coelho RD, de Sousa AF, Barros YC, Teixeira KI. Collet-Sicard syndrome: Case report. Curr Res Integr Med 2019;4:16-8. Available from: https://www.pulsus.com/abstract/colletsicard-syndrome-case-report-5036.html. [Last accessed on 2020 Dec 03].
4. Erol FS, Topsakal C, Kaplan M, Yıldırım H, Özveren MF. Collet-Sicard syndrome associated with occipital condyle fracture and epidural hematoma. Yonsei Med J 2007;48:120-3.
5. Grundy DJ, McSweeney T, Jones HW. Cranial nerve palsies in cervical injuries. Spine (Phila Pa 1976) 1984:9:339-43.
6. Gutierrez Rios R, Castrillo Sanz A, Gil Polo C, Zamora Garcia MI, Morollon Sanchez-Mateos N, Mendoza Rodriguez A. Collet-Sicard syndrome. Neurologia 2015;30:130-2.
7. Handley TP, Miah MS, Majumdar S, Hussain SS. Collet-sicard syndrome from thrombosis of the sigmoid-jugular complex: A case report and review of the literature. Int J Otalaryngol 2010;2010:203587.
8. Hashimoto T, Watanabe O, Takase M, Koniyama J, Kobota M. Collet-Sicard syndrome after minor head trauma. Neurosurgery 1988;23:367-70.
9. Heckmann JG, Tomandl B, Duhm C, Stefan H, Neundörfer B. Collet-Sicard syndrome due to coiling and dissection of the internal carotid artery. Cerebrovasc Dis 2000;10:487-8.
10. Holyst J. Internal carotid artery occlusion due to intraoral trauma. Acta Neurochir (Wien) 1976;33:325-9.
11. Lapresle J, Lasjaunias P, Thévenier D. Transitory paralysis of cranial nerves IX, X and XII as well as the left VII after angiography. Contribution to the ischemic pathology of the cranial nerves. Rev Neurol (Paris) 1980;136:787-91.
12. Larson WL, Beydoun A, Albers JW, Wald JJ. Collet-Sicard syndrome mimicking neuralgic amyotrophy. Muscle Nerve 1997;20:1173-7.
13. Miyazaki C, Katsume M, Yamazaki T, Aoki K, Kuroki T, Takasu N. Unusual occipital condyle fracture with multiple palsies and Wallenberg syndrome. Clin Neurol Neurosurg 2000;102:255-8.
14. Mohr A, Ebert S, Knauth M. Spontaneous dissection of the internal carotid artery with ipsilateral Collet-Sicard syndrome. Rofo 2006;178:444-6.
15. Neo S, Lee KE. Collet-Sicard syndrome: A rare but important presentation of internal jugular vein thrombosis. Pract Neurol 2017;17:63-5.
16. Petrović S, Grozdanović D, Kovačević P, Višnić M, Petrović D. Collet Sicard syndrome as atypical presentation of neck fibrosarcoma: A case report. Bosn J Basic Med Sci 2011;11:137-40.
17. Prick MJ, Verhagen WI. The Collet-Sicard syndrome as a complication of cardiovascular surgery. J Neurol Neurosurg Psychiatry 1992;55:741.
18. Rees JH, Valentine AR, Llewelyn JG. Spontaneous bilateral carotid and vertebral artery dissection presenting as a Collet-Sicard syndrome. Br J Radiol 1997;70:856-8.
19. Schneider RC, Lemmen LJ. Traumatic internal carotid artery thrombosis secondary to nonpenetrating injuries to the neck; a problem in the differential diagnosis of craniocerebral trauma. J Neurosurg 1952;9:495-507.
20. Sharma BS, Mahajan RK, Bhatia S, Khosla VK. Collet-Sicard syndrome after closed head injury. Clin Neurol Neurosurg 1994;96:197-8.
21. Sicard JA. Syndrome du carrefour condylechire posterieur (type pur de paralysie laryngee associee). Marseille Med 1917;53:383.
22. Silvestrini M, Floris R, Tagliati M, Stanziione P, Simonetti G. Collet-Sicard syndrome caused by a coiling of the internal carotid artery. Riv Neurol 1991;61:135-6.

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