CASE REPORT

Troia Med J 2024;5(1):47-49
DOI: 10.55665/troiamedj.1305000

Isolated trigeminal neuralgia as the presenting symptom of cerebellopontine infarction: a case report

Aslı Sena KAYA [1], Bekir Burak KILBOZ [1], Canan BOLCU EMİR [1]

[1] *Prof Dr Cemil Taşcıoğlu City Hospital, Neurology Department, İstanbul*

ABSTRACT

Trigeminal Neuralgia (TN) characteristically presents as unilateral recurring shock-like pain in the trajectory of the trigeminal nerve and its branches. Secondary TN is rare due to an ischemic lesion of the trigeminal root entry zone in pons. Here we report a patient with a cerebellopontine infarction transecting the central trigeminal pathways, resulting in isolated trigeminal neuralgia. A 72-year-old male patient presented to our emergency department with numbness and recurrent shock-like pain attacks on the left side of his face lasting 3 to 5 seconds starting abruptly 20 days ago and increasing in frequency in the last two days.

Neurologic examination revealed slight hypoesthesia to touch on the left maxillary trigeminal nerve dermatome. There was no other abnormality in the neurological examination. In the initial work-up, Diffusion-Weighted Magnetic Resonance Imaging (MRI) showed hyperintensity on the junction of the pons and left inferior cerebellar peduncle, the root-entry-zone of the left trigeminal nerve, without hypointensity in Apparent Diffusion Coefficient (ADC) sequence. Noncontrasted Computer Tomography (CT) revealed hypodensity in the same region. After the admission, an MRI showed the lesion was T1-hypointense, T2-hyperintense, minimally heterogeneously IV gadolinium-contrast enhancing, consistent with subacute infarction.

Secondary TN without any other abnormal neurologic signs attributed to an ischemic lesion of the trigeminal root entry zone in pons is unusual, but not impossible, and responds well to carbamazepine treatment. Patients who present with TN should undergo a comprehensive work-up to identify probable secondary matters.

Keywords: Trigeminal neuralgia, Cerebellopontine angle, Cerebral infarction, Secondary

ÖZET

Serebellopontin iskeminin semptomu olarak izole trigeminal nevralji: bir olgu sunumu

Trigeminal Nevralji (TN) karakteristik olarak trigeminal sinir ve dallarının bölgesinde tek taraflı tekrarlayan şok benzeri ağrı olarak kendini gösterir. Ponsta trigeminal kök giriş bölgesinin iskemik lezyonuna bağlı sekonder TN çok nadirdir. Burada merkezi trigeminal yolları kesen ve izole trigeminal nevralji ile sonuçlanan serebellopontin enfarktı olan bir olguyu sunuyoruz. Yetmiş iki yaşında erkek hasta, 20 gün önce aniden başlayan ve son iki gündür sıklığı artan yüzünün sol tarafında 3-5 saniye süren uyuşma ve tekrarlayan şok benzeri ağrı atakları ile acil servisimize başvurdu. Nörolojik muayenede sol maksiller trigeminal sinir dermatomunda dokunmaya karşı hafif hipoestezi saptandı. İlk incelemede, Difüzyon Ağırlıklı Manyetik Rezonans Görüntüleme (MRG), Görünür Difüzyon Katsayısı'nda (ADC) hipointensite olmadan, sol trigeminal sinirin kök giriş bölgesi olan pons ve sol inferior serebellar pedinkül bileşkesinde hiperintensite gösterdi. Kontrastsız Bilgisayarlı Tomografide (BT) aynı bölgede hipodensite saptandı. Başvurudan sonra MRG, lezyonun subakut enfarkt ile uyumlu, T1-hipointintens, T2-hiperintens, minimal heterojen IV gadolinyum-kontrast tutulumu gösterdiği belirlendi. Ponstaki trigeminal kök giriş bölgesinin iskemik lezyonuna atfedilen başka herhangi bir anormal nörolojik belirti olmaksızın sekonder TN olağann dışıdır, ancak imkansız değildir ve karbamazepin tedavisine iyi yanıt verir. TN ile başvuran hastalar, olası ikincil sorunları belirlemek için kapsamlı bir incelemeden geçmelidir.

Anahtar kelimeler: Trigeminal nevralji, Serebellopontin açı, Serebral enfarktüs, Sekonder

Cite as: Kaya AS, Kılboz BB, Bolcu Emir C.Isolated trigeminal neuralgia as the presenting symptom of cerebellopontine infarction: a case report.Troia Med J 2024;5(1):47-49. DOI: 10.55665/troiamedj.1305000

Corresponding author: Canan Bolcu EMİR
Address: Darulaceze caddesi, 34384, Şişli/İstanbul/ Turkey
E-mail: emir.canan@yahoo.com.tr
Phone: +905324047500

Date of arrival: 28.05.2023, *Date of acceptance*: 23.12.2023

INTRODUCTION

Historically called "tic douloureux", trigeminal neuralgia (TN) characteristically presents as unilateral recurring shock-like pain in the trajectory of the trigeminal nerve and its branches. [1]. Although classical TN can occur by demyelination and alterations in the microvasculature of sensory trigeminal afferents, secondary TN can arise due to other events such as multiple sclerosis and compression by a space-occupying lesion in the brainstem [2]. Secondary TN due to an ischemic lesion of the trigeminal root-entry-zone in pons is very rare, let alone an isolated TN without any other abnormal neurologic signs, and classified in ICHD-3 as "Trigeminal neuralgia attributed to other cause" [1]. Here we report a patient with a cerebellopontine infarction transecting the central trigeminal pathways, resulting in isolated trigeminal neuralgia.

CASE REPORT

A 72-year-old male patient presented to our emergency department with numbness and recurrent shock-like pain attacks on the left side of his face lasting 3 to 5 seconds starting abruptly 20 days ago and increasing in frequency in the last two days. His past medical history was remarkable for hypertension, hyperlipidemia, coronary artery disease, nephrolithiasis, benign prostate hyperplasia, umbilical hernia, and cataracts. He was using aspirin inconsistently, his other previous medications were statin, nebivolol, and perindopril with indapamide.

Neurologic examination revealed slight hypoesthesia to touch on the left maxillary trigeminal nerve dermatome; without any pathological signs in other cranial nerves, motor or sensory disturbances on extremities, dysmetria, dysdiadochokinesia or ataxia.

In the initial work-up, Diffusion-Weighted Magnetic Resonance Imaging (MRI) showed hyperintensity on the junction of the pons and left inferior cerebellar peduncle, the root-entry-zone of the left trigeminal nerve, without hypointensity in Apparent Diffusion Coefficient (ADC) sequence. Noncontrasted Computer Tomography (CT) revealed hypodensity in the same region. After the admission, an MRI showed the lesion was T1-hypointense, T2-hyperintense, minimally heterogeneously IV gadolinium-contrast enhancing, consistent with subacute infarction. (Figure 1)

His complete blood count, coagulation panel, biochemistry tests, and serologic studies were within normal limits. Echocardiography showed left atrium enlargement. Doppler ultrasonography revealed proximal left internal carotid artery (ICA) calcific plaque, narrowing the lumen by 40%, without any stenosis in the right ICA and vertebral arteries.

We suggested the patient use aspirin 300 mg/daily for the treatment of stroke and we prescribed carbamazepine 100 mg, twice daily, for TN. He expressed that the pain did not repeat after the initialization of carbamazepine treatment. Informed consent has been obtained and this report was processed according to the principles expressed in the Declaration of Helsinki.

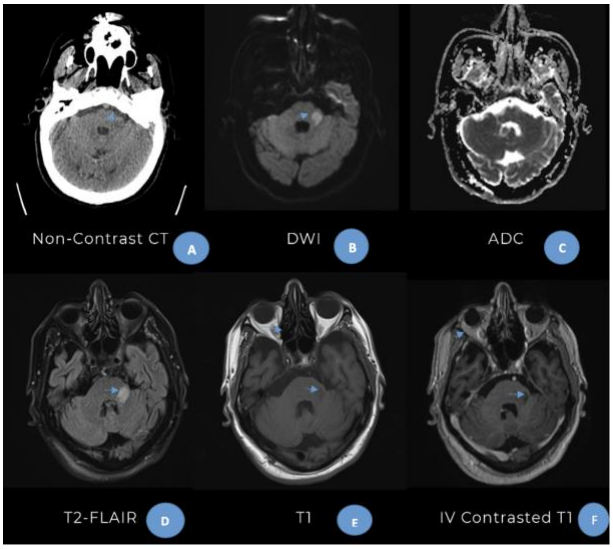

Figure 1: A) Noncontrasted Computer Tomography revealed hypodensity in the junction of the pons and left inferior cerebellar peduncle, **B)** Diffusion-Weighted Magnetic Resonance Imaging (MRI) showed hyperintensity on the junction of the pons and left inferior cerebellar peduncle, the root entry zone of the left trigeminal nerve, **C)** No hypointensity in Apparent Diffusion Coefficient sequence, **D-E-F)** MRI showed the lesion was T1-hypointense, T2-hyperintense, minimally heterogeneously IV gadolinium-contrast enhancing, consistent with subacute infarction.

DISCUSSION

Secondary trigeminal neuralgia due to ischemic processes are classified under "Trigeminal neuralgia attributed to other cause" in ICHD3 and have diagnostic criteria of (a) recurrent pain fulfilling the criteria for TN, (b) etiological disorder not described in the other titles of classification but known to be able to cause TN, and (c.) TN either led to the disorder's discovery or developed after the onset of the disorder [1]. In our case, ischemic infarction was discovered during the investigation of the etiology of facial pain and hypoesthesia.

TN frequently affects V2 and V3 dermatomes, slightly more often on the right side. Any additional abnormal neurological examination findings should be investigated in the case of secondary TN, although mild hypoesthesia is commonly reported in people with classical TN [3]. Our patient also had hypoesthesia, leading to his TN's etiological discovery.

The pathophysiological mechanism is thought to be identical in secondary TN with classical TN, which is demyelination and microvascular changes, but secondary TN is attributed to a distinct lesion affecting trigeminal root entry zone such as

demyelinating plaque, tumor, arteriovenous malformation [4] or stroke, like our case. Identification of secondary events with neuroimaging is important because they may require additional treatment or preventative measures in multiple sclerosis, tumors, or stroke.

There are few cases of TN with ischemic lesions discussed in the literature. Cabral et al. reported a 67-year-old female with left TN, chronic left pontine ischemic lesion, ipsilateral Babinski sign, and brisk reflexes on the right side; without decreased sensation in V2-V3 dermatomes [5]. A 68-year-old male with a wedge-shaped ischemic lesion right trigeminal nerve root entry zone causing right TN and hypoesthesia on the right V2 presented by Katsuno et al., notes only 9 cases previously reported when the case was published in 2010 [6] Peker et al. described a 72-year-old female with left TN with V2-3 hypoesthesia, trigger point at the left upper lip, and a lesion that is T1-hypointense, T2-hyperintense, not contrast enhancing, not diffusion restricting, decreased N-acetyl aspartate without a significant increase in choline or a lactate peak on MR spectroscopy, which is consonant with ischemic lesion [7].

Treatment for TN does not differ between classical and secondary forms. The first line of treatment is sodium channel blockers such as carbamazepine or oxcarbazepine [8] which swiftly relieved our patient's symptoms. Adding pregabalin, gabapentin, lamotrigine, or baclofen to the therapy and a combination of carbamazepine and oxcarbazepine can be considered if the monotherapy is not efficacious or the side effects prevent reaching the maximum dose of the monotherapy [8].

Conclusion

Secondary TN with mild hypoesthesia to touch on the maxillary division of trigeminal nerve dermatome attributed to an ischemic lesion of the trigeminal root entry zone in pons is unusual, but not impossible, and responds well to carbamazepine treatment. Sometimes spontaneous pain relief is possible, but it can last up to three months and in this condition, neuropathic pain treatment is essential for increasing the quality of life of the patient. Patients without a history of facial pain and at high risk of cerebrovascular disease, presenting with pain mimicking TN should undergo a comprehensive work-up to identify probable secondary matters and be treated with caution. Pain mimicking TN induced by ischemic stroke in the brainstem has a good prognosis, and most of the symptoms can be relieved spontaneously or by drug treatment.

Conflict of interest: None
Funding: None

REFERENCES

1. Headache Classification Committee of the International Headache Society (IHS) The International Classification of Headache Disorders, 3rd edition. Cephalalgia 2018;38: 1–211
2. Maarbjerg, S., Di Stefano, G., Bendtsen, L. & Cruccu, G. Trigeminal neuralgia – diagnosis and treatment. ;2017:37: 648–657.
3. Maarbjerg, S., Gozalov, A., Olesen, J. & Bendtsen, L. Trigeminal Neuralgia - A Prospective Systematic Study of Clinical Characteristics in 158 Patients. Headache J. Head Face Pain 2014; 54: 1574–1582
4. Nomura, T., Ikezaki, K., Matsushima, T. & Fukui, M. Trigeminal neuralgia: Differentiation between intracranial mass lesions and ordinary vascular compression as causative lesions. Neurosurg. Rev;1994:17: 51–57
5. Cabral, G., Lorga, T., Caetano, A., Baptista, T. & Baptista, M. V. Secondary Trigeminal Neuralgia: Do Not Forget the Pontine Ischemic Infarction. Eur. Neurol. 2021;84: 391–392
6. Katsuno, M. & Teramoto, A. Secondary Trigeminal Neuropathy and Neuralgia Resulting from Pontine Infarction. J. Stroke Cerebrovasc. Dis.2010;19: 251–252
7. Peker, S., Akansel, G., Sun, I. & Pamir, N. M. Trigeminal Neuralgia Due to Pontine Infarction. Headache 2004; 44: 1043–1055
8. Cruccu, G. et al. AAN-EFNS guidelines on trigeminal neuralgia management. Eur. J. Neurol.2008;15: 1013–1028.