INTRODUCTION

Trigeminal neuralgia (TN) is classified as primary and secondary\(^5\). Primary TN is mainly due to a vessel compressing the root entry zone of the fifth nerve. Secondary or symptomatic TN can be due to a mass compressing the trigeminal nerve or due to multiple sclerosis. Brainstem lesions are known to cause facial pain. Yet, classical TN due to brainstem lesions is very rare. In this report two cases with brainstem vascular malformations were presented and the literature was reviewed.

CASE PRESENTATION

Case 1: A 52-year-old female patient presented to our clinic with 2 years history of typical TN on the right side. Her first complaint was begun after an attack of headache, dizziness and ataxia. These complaints were resolved in a few days without any medication.

After a few weeks she developed typical TN in the right side. She was using carbamazepine 800 mg/day. Her neurological examination was normal. Her recent cranial MRI showed nonenhancing, heterogenous signal intensity in the pons.
and hypointense area around the lesion in T2-weighted images (Fig. 1). The T1-weighted image demonstrated a hyperintense area which is a new bleeding. There was no contrast enhancing part of the lesion. The diagnosis was cerebral cavernous hemangioma. Because of she had a clinically overt bleeding from the cavernoma Gamma knife radiosurgery was recommended to her. The treatment was performed for the lesion with 12 Gy marginal doses. After 7 years of follow-up she is still using carbamazepine 200 mg/day without any new bleeding attack and neuralgic pain.

**Case 2:** A 37-year-old woman was admitted to our hospital with the complaint of severe electric-shock like pain in the right lower jaw for 3 months. Physical and neurological examinations revealed normal signs. Neuroradiological evaluations with MRI showed a pontine hyperintense lesion in T1-weighted and T2-weighted images. No contrast enhancement was found. MR spectroscopy was normal. (Fig 2a,b) She was diagnosed as having capillary telangiectasia in the pons. Carbamazepine 400 mg/day for three months relieved the complaints. After 2 years of follow-up she has no complaints with any medication.

![Fig 1: The T2-weighted MR image demonstrates the pontine cavernoma.](image1)

![Fig 2: The capillary telangiectasia is seen in T1-weighted sagittal (a) and T2-weighted axial (b) MR images.](image2)
DISCUSSION

The mechanism of TN due to lesions that involve the central trigeminal pathways is still a matter of debate. Central theories on the etiology of TN mainly focus on the increased neuronal activity in the trigeminal nucleus. King hypothesizes that the irritation of the descendant tracts and nucleus of the trigeminal nerve may cause excessive reactivity to electrical stimulation of the peripheral parts of the trigeminal nerve\(^{(3)}\). Thus, he believes that TN is due to an irritation at the trigeminal nucleus. List and Williams stated that TN is due to a pathologic multineuronal reflex of the trigeminal system in the brain stem\(^{(4)}\). Ferroli et al argue that the demyelization of the trigeminal fibers in multiple sclerosis (MS) patients triggers electrophysiological alterations in the trigeminal system\(^{(1)}\). This may result in ephaptic transmission, spontaneous firing, and mechanosensitivity, both before and after discharge. We think that an infarction that involves the intramedullary trigeminal fascicular fibers may have the same effects as a chronic MS plaque.

To the best of our knowledge there are 4 cases in the literature with brainstem vascular malformations causing trigeminal neuralgia (Table 1). Shimpo reported a patient with pontine cavernoma and he concluded that the damage to the intra-axial trigeminal nerve caused neuralgia\(^{(6)}\). In their patient with brachium pontis localized cavernoma Vitek and Tettenborn supposed that the reason was direct compression of the intra-axial trigeminal nerve root\(^{(8)}\). Stellmann et al reported a patient with brain stem cavernoma who responded well to medical treatment\(^{(7)}\). Kapnadak et al reported a patient with medullary telangiectasia that causes glossopharyngeal and trigeminal neuralgia\(^{(2)}\). They advocated that the reason for pain in this patient is due to involvement of spinotrigeminal nucleus and tract (CN V), the solitary nucleus (CN VII, IX and X) and the nucleus ambiguous (CN IX and X).

In conclusion we think that the damage to the intra-axial trigeminal nerve pathways resulted with the neuralgia. In the patient with cavernoma, bleeding may cause the damage. However, in the other patient with telangiectasia there was no bleeding. The mass effect on the trigeminal system may cause to the symptoms. But we still do not know why brainstem glial tumors do not cause TN even they compress the trigeminal pathways and the nuclei.

<table>
<thead>
<tr>
<th>Author</th>
<th>Case</th>
<th>Type of VM</th>
<th>Localisation</th>
<th>Treatment</th>
</tr>
</thead>
<tbody>
<tr>
<td>Shimpo 2000</td>
<td>67/M</td>
<td>Cavernoma</td>
<td>Pons</td>
<td>Carbamazepine</td>
</tr>
<tr>
<td>Vitek 2002</td>
<td>61/M</td>
<td>Cavernoma</td>
<td>Brachium pontis</td>
<td>Gabapentin</td>
</tr>
<tr>
<td>Stellmann 2007</td>
<td>55/F</td>
<td>Cavernoma</td>
<td>Bulbus</td>
<td>Pregabalin</td>
</tr>
<tr>
<td>Kapnadak 2010</td>
<td>72/F</td>
<td>Telangiectasia</td>
<td>Bulbus</td>
<td>No</td>
</tr>
<tr>
<td>Present case 1</td>
<td>52/F</td>
<td>Cavernoma</td>
<td>Pons</td>
<td>Carbamazepine</td>
</tr>
<tr>
<td>Present case 2</td>
<td>37/F</td>
<td>Telangiectasia</td>
<td>Pons</td>
<td>Carbamazepine</td>
</tr>
</tbody>
</table>

Table 1: Brainstem vascular malformations (VM) with trigeminal neuralgia
Correspondence to:
Selcuk Peker
E-mail: peker@selcukpeker.com

Received by: 03 April 2012
Revised by: 12 April 2012
Accepted: 13 April 2012

The Online Journal of Neurological Sciences (Turkish) 1984-2012
This e-journal is run by Ege University Faculty of Medicine,
Dept. of Neurological Surgery, Bornova, Izmir-35100TR
as part of the Ege Neurological Surgery World Wide Web service.
Comments and feedback:
E-mail: editor@jns.dergisi.org
URL: http://www.jns.dergisi.org
Journal of Neurological Sciences (Turkish)
Abbr: J. Neurol. Sci.[Turk]
ISSNe 1302-1664

REFERENCES