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Case Report

Delayed blink R1 latency in a patient with trigeminal neuralgia due to a contralateral vestibular schwannoma: An illustrative case

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ABSTRACT

Background: Although the blink reflex (BR) is effective in objectively evaluating trigeminal neuropathy, few studies have demonstrated its effect on trigeminal neuralgia (TN). The authors report a patient with TN due to contralateral vestibular schwannoma (VS) functionally diagnosed by delayed R1 latency of the BR.

Case Description: A 36-year-old man presented with left-sided deafness and paroxysmal facial pain in the right V1-3 area. Magnetic resonance imaging (MRI) showed a solid cystic mass compressing the right pons and left brainstem at the left cerebellopontine angle. Although preoperative BR evoked by right supraorbital nerve stimulation-induced delayed ipsilateral R1 latency and normal ipsilateral and contralateral R2 responses, the BR latency evoked by left supraorbital nerve stimulation was normal, indicating deficits in the principal nucleus of the trigeminal nerve in the right pons. The symptoms of TN disappeared after the removal of the VS. Postoperative MRI showed subtotal removal of the tumor and sufficient decompression of the pons and cerebellopontine cistern. The R1 latency returned to normal 50 days after surgery.

Conclusion: The perioperative BR test was not only useful for objective evaluation of the localization of trigeminal neuropathy but also correlated with the symptoms of TN.

Keywords: Blink reflex, Contralateral vestibular schwannoma, Trigeminal neuralgia

INTRODUCTION

Trigeminal neuralgia (TN) can occur due to compression of the root entry zone (REZ) of the trigeminal nerve.[2] Vestibular schwannomas (VSs) are relatively rare causes of TN; only 1.2–3.3% of VSs have been reported to cause TN.[6,10,14] Most of these cases are associated with ipsilateral VSs. However, TN has also been reported in some patients with contralateral VSs.[4,5,13,14,16,17] Since a massive mass effect from contralateral lesions could limit the accuracy of preoperative evaluation of trigeminal nerve compression, the causal relationship between TN and contralateral VS remains unclear.[4,5,13,14,16,17] Blink reflex (BR) testing is a noninvasive electrophysiological method

used to evaluate the localization and degree of TN.[18] Here, we report the case of a patient with TN due to contralateral VS diagnosed by delayed R1 latency in the BR test.

CASE DESCRIPTION

A 36-year-old man with an unremarkable medical history experienced recurrent right facial pain in the second and third division of the trigeminal nerve for 27 months. Facial pain was triggered by chewing and tooth brushing, which occurred about 6 times a day with each episode lasting for 10 min. Nine months after the initial onset, facial pain spread to the first division of the trigeminal nerve. Carbamazepine administration slightly alleviated the pain but did not eliminate it; therefore, the patient was referred to our department for further evaluation and treatment. Neurological examination revealed left-sided deafness, left Bruns' nystagmus, bilateral papilledema, and paroxysmal electric pain in all right trigeminal nerve divisions. Magnetic resonance imaging (MRI) showed a heterogeneously enhanced mass measuring 4.8 × 3.3 cm in the left cerebellopontine angle (CPA) and a tight cerebellopontine cistern of the right CPA [Figure 1].

For further evaluation of right facial pain, the bilateral BR was obtained using bipolar stimulation by placing the cathode in the supraorbital crease and the anode 2 cm above the cathode. Recording electrodes were placed on the lower eyelids, with a reference electrode near each lateral cantus. Stimulation with 0.2 ms square-wave pulses was measured using Neuropack X1 (Nihon Kohden, Japan). The latency of the R1 and R2 waves ipsilateral and contralateral to the stimulus and the difference between the R1 latency ipsilateral and contralateral to the lesion were measured. We referred to previous study indices of normal neurophysiology values expressed as the mean ± SD in ms: R1, 10.5 ± 1.6; R2 ipsilateral, 30.5 ± 6.4; R2 contralateral, 30.5 ± 3.4; the difference between right and left R1, 1.2; and mean upper limit, +2 SD.[10] The BR was recorded immediately after the neurological examination. Preoperative evaluation of the BR showed a delayed R1 latency after stimulation of the right supraorbital nerve, despite a normal R2 latency and normal R1 and R2 latencies after right and left supraorbital nerve stimulation, respectively, suggesting neuropathy in the afferent pathway between the right REZ and the principal nucleus of the trigeminal nerve in the pons [Figure 2].

The tumor was removed using the left retrosigmoid approach under general anesthesia with auditory brainstem response (ABR), motor-evoked potential (MEP), and sensory-evoked potential monitoring. The MEP was used to monitor brainstem injury. During surgery, the preservation of facial nerve function was confirmed using a neural integrity monitor. The ABR and MEP results did not change postoperatively. Finally, complete tumor resection was performed, while avoiding facial nerve and brainstem dysfunction. The tumor was diagnosed as a grade 1 schwannoma based on histopathological examination (the World Health Organization criteria). Because his right facial pain disappeared 5 days after surgery, carbamazepine was gradually tapered off until completely discontinued on the 45th postoperative day. Postoperative MRI demonstrated a significant reduction in brainstem compression and widening of the right cerebellopontine cistern [Figure 3]. The BR test performed on the 50th postoperative day showed normalization of the ipsilateral R1 latency, followed by right stimulation and enlargement of the amplitude of the R1 wave, indicating improvements in right trigeminal nerve

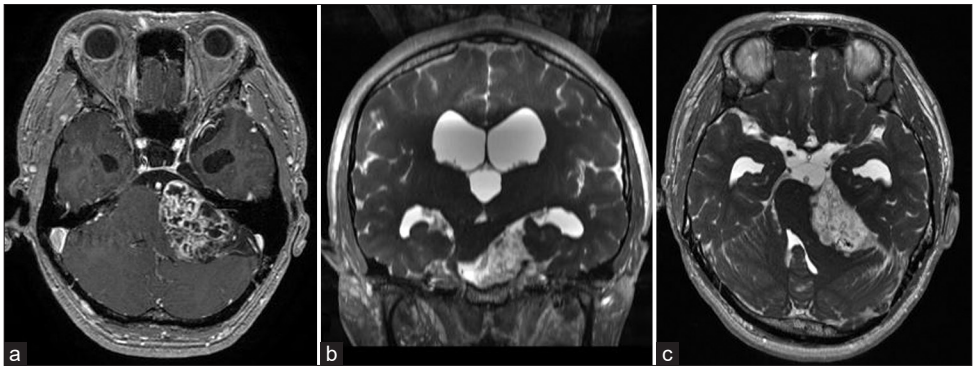

Figure 1: Preoperative axial T1-weighted enhanced magnetic resonance imaging showing a well-margined, heterogeneous enhancing mass at the left cerebellopontine angle (48 × 33 × 34 mm), compatible with a vestibular schwannoma (a). Preoperative coronal fast imaging employing steady-state acquisition (FIESTA) showed that the brainstem was compressed by the tumor and deviated contralaterally (b). Preoperative axial FIESTA revealed a tight cerebellopontine cistern on the right side, and the trigeminal nerve was deviated and compressed toward the petrous bone. There were no obvious vessels responsible for the compression of the right trigeminal nerve, such as the superior cerebellar artery (c).

function [Figure 4]. The patient has been free of facial pain for 8 months after surgery without medication.

DISCUSSION

BR testing has been performed to evaluate the function of the trigeminal and facial nerves, which elicit two distinct responses to supraorbital nerve stimulation: R1 and R2 waves.[7,8,12] The R1 wave is a unilateral early response evoked approximately 10 ms after ipsilateral stimulation and is conducted through the main sensory nucleus of the trigeminal nerve in the pons. The R2 wave is a bilateral late response evoked approximately 30 ms after stimulation that reveals afferent impulses conducted through the descending spinal tract of the trigeminal nerve and efferent impulses ascending to reach the bilateral facial nuclei in the pons.[3,8,9,12] Previous reports have shown a total of six lesions localization within the brainstem and corresponding BR response abnormalities in the right and left orbicularis oculi muscles after stimulation of the supraorbital nerves. The six locations are the REZ of the trigeminal nerve, the main sensory nucleus of the trigeminal nerve and descending spinal trigeminal tract in the pons, the spinal trigeminal nucleus and crossed fibers that ascend ipsilaterally to the side of lesion, the crossed and uncrossed ascending trigemino-facial connections mediated through the lateral tegmental field, and the facial nucleus or the intrapontine part of the facial nerve fibers.[1,7-9,12,18] Tanaka *et al*. reported a case of right TN due to ipsilateral trigeminal root schwannoma. In the preoperative BR test, the latency of the R1 and R2 response was absent on both sides after stimulation of the right supraorbital nerve. On left supraorbital nerve stimulation, the R1 and R2 on the left side and the R2 response on the right side were normal. These results suggested that the function of the REZ of the trigeminal nerve was impaired on the right side and that the function of the facial nerve was preserved. The BR reappeared on both sides after right-side stimulation after the removal of the tumor.[18] Nurlu *et al*. reported five cases in which BR tests were performed preoperatively for assessing trigeminal nerve dysfunction due to CPA tumors. The combination of abnormal ipsilateral and contralateral R1 and R2 responses suggested nerve dysfunction at the REZ of the trigeminal nerve in four of five cases, and in the main sensory nucleus of the trigeminal nerve in the pons in five cases.[11] Cenzato *et al*. reported a case of TN caused by an intra-axial cavernous hemangioma along the primary sensory nucleus of the pons. Preoperative BR showed the

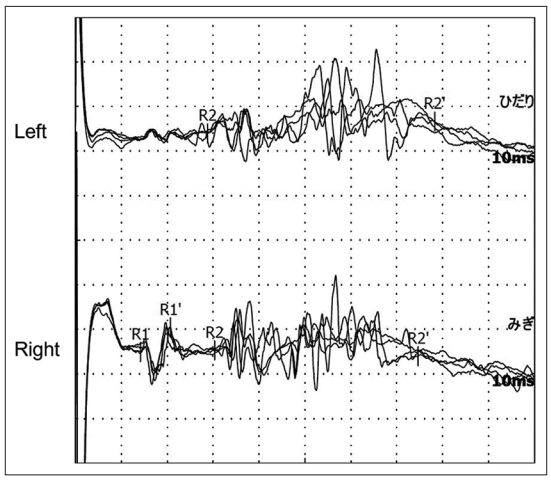

Figure 2: Blink reflex with a lesion elicited with right supraorbital nerve stimulation. Reflex responses were recorded on the left (upper) and right (lower) eyelids. Stimulation of the right supraorbital nerve elicited an ipsilateral R1 response (lower) with a delayed latency of 14.2 ms, an ipsilateral R2 response with a normal latency of 30.3 ms, and contralateral R2 response (upper) with a normal latency of 28.8 ms.

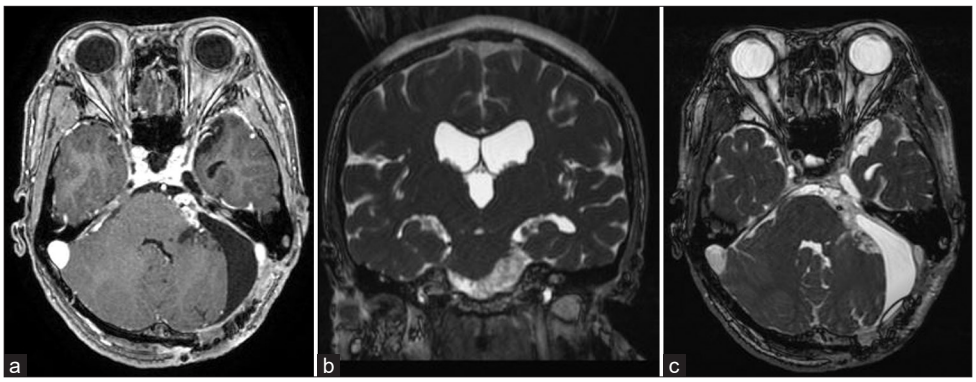

Figure 3: Postoperative T1-weighted enhanced magnetic resonance (MR) imaging showed a small residual tumor and enlargement of the cerebellopontine cistern (a). Postoperative coronal fast imaging employing steady-state acquisition (FIESTA) MR images showed that the brainstem deviation was improved (b). Postoperative axial FIESTA MR images showed normalization of the right cerebellopontine cistern, normalization of the brainstem, and trigeminal nerve position compared to before tumor resection (c).

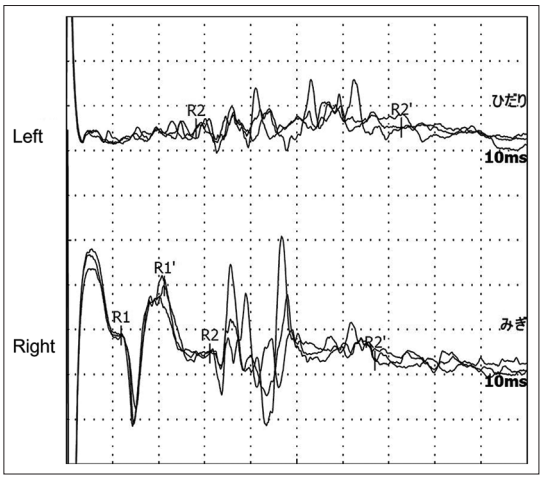

Figure 4: Blink reflex elicited with a lesion on the right supraorbital nerve. Reflex responses were recorded on the lower left (upper) and right (lower) eyelids. Stimulation of the right supraorbital nerve elicited an ipsilateral R1 response (lower) with a normal latency of 11.9 ms, an ipsilateral R2 response with a normal latency of 31.1 ms, and contralateral R2 response (upper) with a normal latency of 28.1 ms.

disappearance of R1 wave latency and normalization of the wave postoperatively, with recovery from TN.[3] However, since extra-axial mass effects from contralateral lesions make the detection of the site of trigeminal nerve compression challenging, even with high-resolution MRI, there have been no reports objectively indicating the site of TN.[4,5,12,13,15,16] In the present case, right supraorbital nerve stimulation evoked a delayed ipsilateral R1 latency despite normal R2 responses to ipsilateral stimulation and R1 responses to contralateral stimulation. These findings indicate damage to the main sensory nucleus of the trigeminal nerve in the right pons. Based on the electrophysiological and radiographic findings, we concluded that compression of the right brainstem toward the pyramidal bone by the contralateral VS caused the TN. Interestingly, the TN disappeared after the removal of the contralateral VS, which was consistent with the normalization of the BR test results.

CONCLUSION

Pre- and postoperative BR testing might be a feasible modality to objectively evaluate the localization of the TN and perioperative changes in trigeminal nerve function, such as neuralgia.

Disclosures

The authors have no personal, financial, or institutional interests in any of the drugs, materials, or devices described in this article.

Declaration of patient consent

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

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Conflicts of interest

There are no conflicts of interest.

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