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Case Report

Trigeminal neuralgia with rare solitary pontine lesion: A case report and literature review ☆,☆☆

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ABSTRACT

Trigeminal neuralgia (TN) associated with brainstem lesions as revealed by Magnetic resonance imaging (MRI), is a rare condition. The MRI often shows a distinctive single pontine in cases of TN (SPL-TN). While the significance of this MRI finding remains unclear, various case reports suggest a potential link to chronic injury in the pontine pathways of the trigeminal nerve. In this report, we present the case of a 42-year-old female who was referred for TN that is refractory to medical treatment with an ipsilateral MRI lesion over the pons who had an excellent response to a trigeminal nerve bock, shedding light on the intriguing interplay between TN and pontine lesions.

Introduction

Trigeminal neuralgia (TN) is the commonest type of chronic facial neuropathic pain [1]. Characterized by sudden, severe, brief, and recurrent stabbing pain within the branches of the trigeminal nerve [2], TN is typically attributed to vascular compression of the trigeminal nerve root. However, a minority of cases, identified through MRI, have revealed the presence of brainstem lesions [3,4].

Case

This is a 42-year-old female whom has been diagnosed of TN for the last 11 years. Initially, her symptoms started with several episodes of severe electrical shock-like pain involving the right maxillary branch (V2) distribution that lasted for a maximum duration of 1 minute and reoccurred every 15 minutes. These episodes were triggered by eating, stress, and exposure to cold. After 5 years of taking carbamazepine, the fre-

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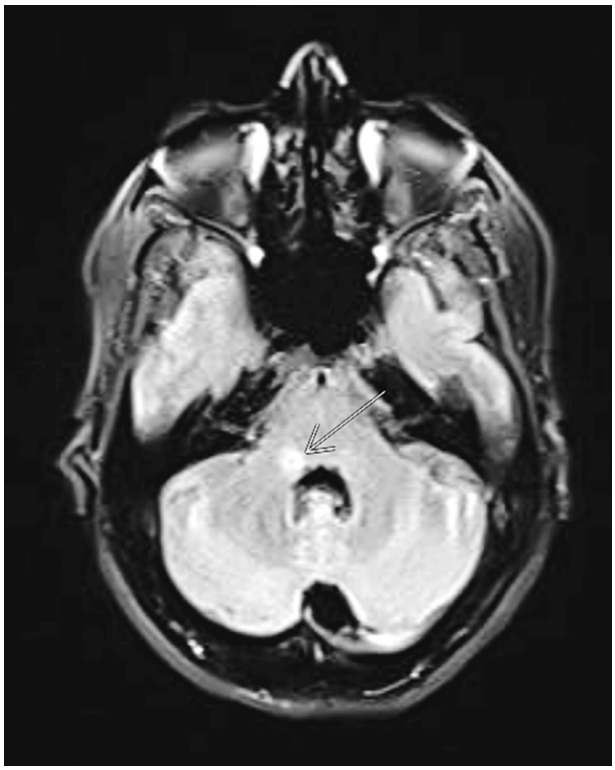

Fig. 1 – Brain MRI T2-weighted-fluid-attenuated inversion recovery (FLAIR) axial view. There is an oval shaped right middle cerebellar peduncle focus of hyper-intense signal.

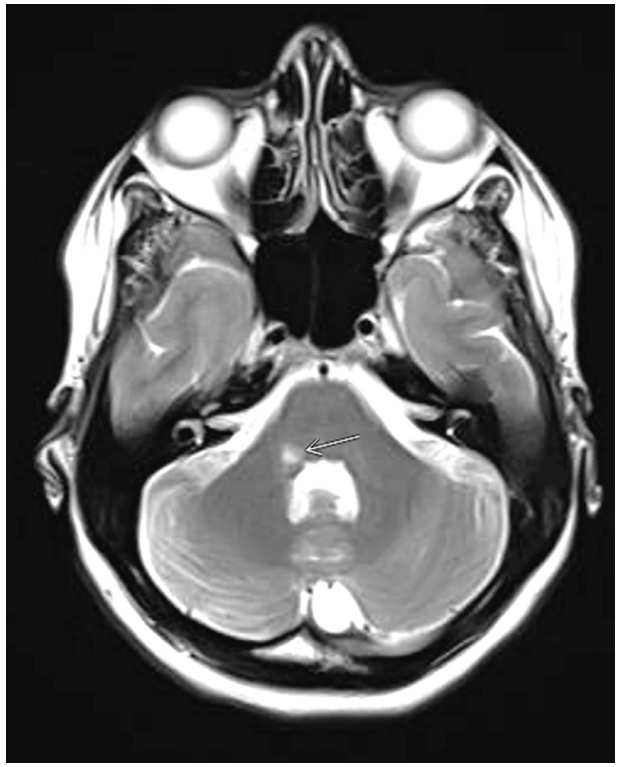

Fig. 2 – Brain MRI T2 axial view. There is an oval shaped right middle cerebellar peduncle focus of hyper-intense signal.

quency and severity of pain worsened and started to involve mandibular branch (V3) territory.

Her MRI brain (Fig. 1), showed oval shaped right middle cerebellar peduncle focus of hyperintense T2/FLAIR signal. No evidence of diffusion restriction or enhancement or mass effect. Bilateral trigeminal nerves seen with normal signal, and no vascular compression.

In an attempt to manage the worsening symptoms, the patient was administered a higher dose of carbamazepine and Gabapentin. Given her poor response to the treatment and with the newly diagnosed pontine lesion on MRI, the patient was referred to neuro-immunology clinic to rule out inflammatory demyelinating disease.

To address the persistent pain, the patient then underwent ultrasound-guided trigeminal nerve block via the pterygopalatine fossa with almost complete recovery of her pain. She had no history of varicella zoster (VZ) infection nor any other neurological complaints. This case underscores the complexities of TN management, especially in instances where conventional treatments prove ineffective, prompting a multidisciplinary approach for a thorough assessment and tailored intervention.

Discussion

In this report, we present an exceptionally rare case of trigeminal neuralgia with a single pontine lesion (SPL-TN). The patient, who had no history of prior VZ infection exhibited clinical symptoms consistent with typical trigeminal neuralgia, albeit with a distinctive single pontine lesion evident in the MRI scan. Intriguingly, unlike the majority of trigeminal neuralgia cases, her MRI did not reveal any neuro-vascular compression (Fig. 2). Despite receiving conventional treatment with a poor response and the emergence of the pontine lesion on MRI, the patient was referred to our neuro-immunology clinic to explore the possibility of inflammatory demyelinating disease (Figs. 3–5). Radiologically, she did not meet the criteria for multiple sclerosis (MS) diagnosis, as the MRI displayed a nonenhancing, single pontine lesion. Clinically, the patient did not report any neurological symptoms indicative of an MS attack. Importantly, the patient did not fulfill the Macdonald's criteria for MS, ruling out this particular neuroinflammatory condition.

Despite 2 medications, her pain persisted until an ultrasound-guided trigeminal nerve block via the pterygopalatine fossa led to nearly complete pain relief. SPL-TN, a rare condition, has limited cases reported in the literature [4]. The lesion's etiology encompasses postherpetic infection, demyelination, and ischemic changes [5–8].

Tohyama et al. retrospectively analyzed 481 TN patients, finding 24 cases with a single pontine lesion, constituting 5% of TN cases [4]. Of the 18 patients with sufficient follow-up, all underwent various surgical interventions, yielding a single case achieving 100% pain reduction through Gamma Knife radiosurgery over an 8-year period [4]. Notably, the location

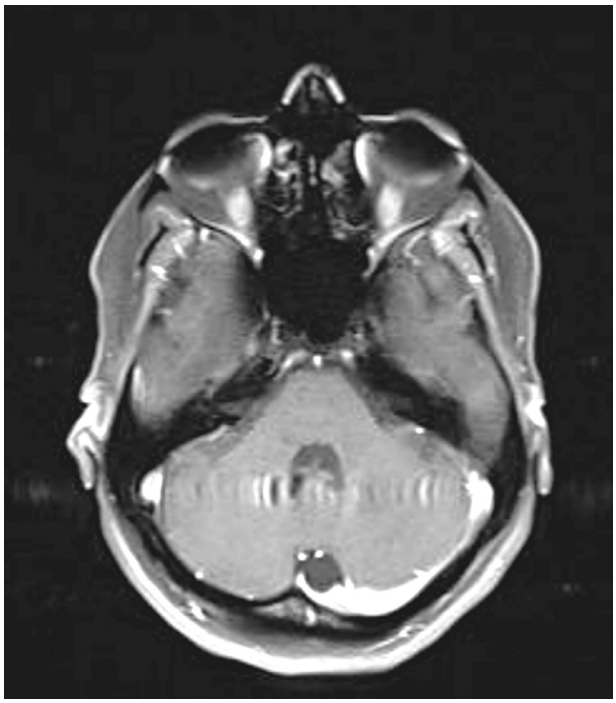

Fig. 3 – Brain MRI T1 axial view postcontrast. Post contrast images showed no pathological enhancement.

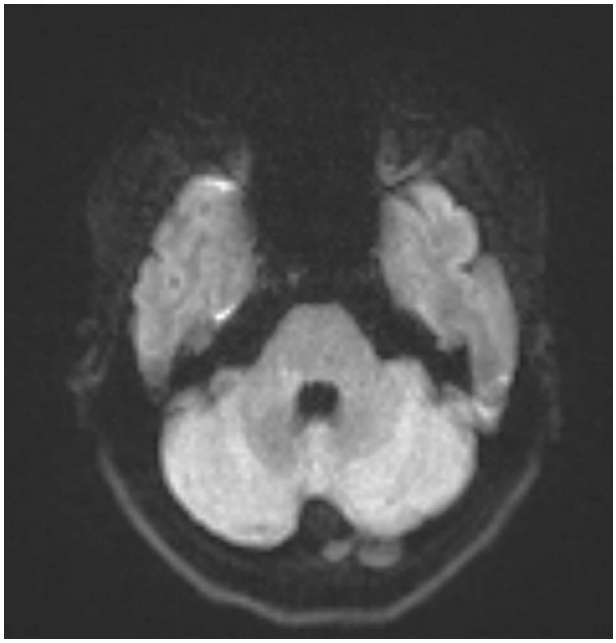

Fig. 4 – Brain MRI DWI sequence. No evidence of diffusion restriction.

of the lesion played a crucial role; nonresponders had more centrally located lesions, while responders showed lesions at the trigeminal root entry zone periphery [4]. Remarkably, our patient exhibited an outstanding response to ultrasound-guided trigeminal nerve block via the pterygo-palatine fossa.

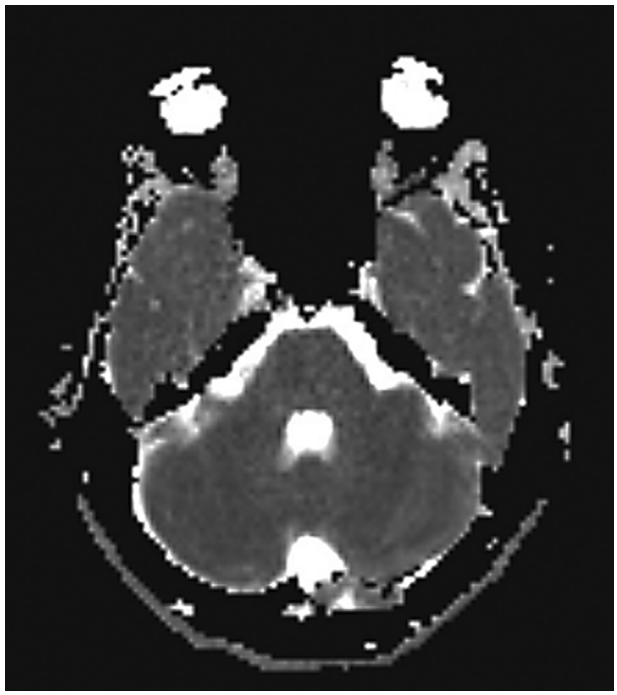

Fig. 5 – Brain MRI ADC sequence. No evidence of diffusion restriction.

Anita et al. studied MRI images of 7 patients with TN. Two female patients were found to have trigeminal pontine sign with an MRI T2 linear hyper-intensity along the trigeminal tract in the ipsilateral pons with no enhancement. One of these patients had a history of childhood varicella infection, while the other was not sure if she had it during childhood [9].

Another study by D'Amico et al. found 7 patients with trigeminal pontine sign. The majority were females, and all patients had herpes zoster infection without neuro-vascular compression. The MRI findings were always consistent with TN [5]. Another reported case described a lady with post herpes zoster infection TN with complete imaging recovery after 4 months [6].

Katsuno and colleagues reported a 68-year-old man with a sudden onset of TN associated with a nonenhancing MRI lesion, which was hypo-intense on T1 and hyper-intense on T2 sequence, with the impression of pontine infarction because of small branch occlusion [10]. Another report a case of solitary pontine lesion with trigeminal neuralgia in a 72-year-old female previously healthy with a sudden onset of facial pain typical for TN, yet her magnetic resonance angiography (MRA) demonstrated significant narrowing of left vertebral artery, ipsilateral to the TN [7].

SPL-TN is a rare condition with uncertain MRI significance. Literature case reports suggest a potential link to chronic trigeminal nerve injury in pontine pathways. Clinicians should differentiate this from CNS inflammatory demyelinating disease. Prognostic implications remain unclear.

Conclusion

Single pontine lesion in trigeminal neuralgia may signify chronic nerve injury, but its precise significance is yet to be defined.

Availability of data and materials

The datasets used and analyzed during the current study are available from the corresponding author upon reasonable request.

Author contributions

AA, AB, RA, AA, RA designed the study. AA, AB, RA, AA, collected the clinical data. AA, AB, RA, AA, RA analysed and interpreted the data. AA, AB, RA, AA, RA, and SB drafted the manuscript. AA, and SB checked and approved the authenticity of the clinical data. All authors read and approved the final manuscript.

Ethics approval and consent to participate

The study was carried out in accordance with the code of international and local Ethics (Declaration of Helsinki). This study was reviewed and approved by the local ethics committee of the King Fahad Specialist Hospital Dammam (Dammam, Saudi Arabia).

Patient consent

I, [Patient's Name], hereby give my consent for the publication of my medical information, including but not limited to my case history, diagnostic findings, treatment plans, and any associated images or data. I understand that my personal information will be anonymized to protect my privacy, and only relevant clinical details will be included in the publication. I authorize the use of this information for scientific and educational purposes, with the understanding that it may be accessed by other healthcare professionals and researchers. I acknowledge that I have been provided with the opportunity to review the manuscript or article before publication and have had the opportunity to ask questions or request modifications if needed. I understand that my participation in this publication is voluntary, and I may withdraw my consent at any time before the publication process is complete.

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