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## Case Report

# Absent Meckel's cave as a possible cause of trigeminal neuralgia: A case report <sup>☆</sup>

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## ARTICLE INFO

## Article history:

Received 21 November 2022

Accepted 9 December 2022

## Keywords:

Trigeminal Neuralgia

Meckel's Cave

Case Report

## ABSTRACT

Trigeminal neuralgia (TN) is a debilitating yet potentially treatable facial pain disorder. TN is difficult to miss clinically, as patients' clinical presentation is often strikingly stereotypical: unilateral, paroxysmal, stimulus-dependent pain involving the trigeminal territory. Magnetic resonance imaging (MRI), which is used for further evaluation of an underlying etiology of TN, most commonly shows neurovascular compression of the trigeminal nerve to be the culprit. Secondary etiologies, though less common, do exist. An absent Meckel's cave with ipsilateral TN was reported in a few case reports and series, and whether an etiological relationship exists is yet to be established. We herein present a case of a 22-year-old female patient who presented with typical TN clinical manifestations. MRI was ordered to assess for the underlying cause and an ipsilateral absent Meckel's cave was the only significant finding. This case report adds to the scarcity of literature highlighting this entity, further larger clinical studies are needed to establish a causal relationship.

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## Introduction

Trigeminal neuralgia (TN) is an uncommon facial neuropathic pain disorder, characterized by sudden, brief paroxysms of excruciating pain along the distribution of one or more divisions of the trigeminal nerve. The recurrent unilateral pain attacks are often described by patients as having electric-shock-like or stabbing quality and can be provoked by innocuous triggers, like chewing, touching the face, smiling, eating, drink-

ing, or brushing teeth [1]. In most cases, vascular compression of the trigeminal nerve is proposed to cause TN [2]. However, other etiologies exist, including tumors, cysts, multiple sclerosis, facial injury, or structural anomalies of the cranium [3,4]. Although the TN is a clinical diagnosis, imaging studies, such as magnetic resonance imaging (MRI) are unequivocally useful to evaluate for secondary causes of TN. MRI, however, is only useful after clinically establishing a TN diagnosis [5].

Meckel's cave is a dural recess in the posteromedial portion of the middle cranial fossa. It is attached to the lateral wall of

<sup>☆</sup> Competing Interests: The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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<https://doi.org/10.1016/j.radcr.2022.12.018>

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the cavernous sinus where it harbors the root of the trigeminal nerve [6]. The rare phenomenon of an absent Meckel's cave, which has been described in only a few case reports and series, is mostly found in conjunction with TN. This raises the possibility of an etiologic role of absent Meckel's cave in TN, which has been previously described in the literature.

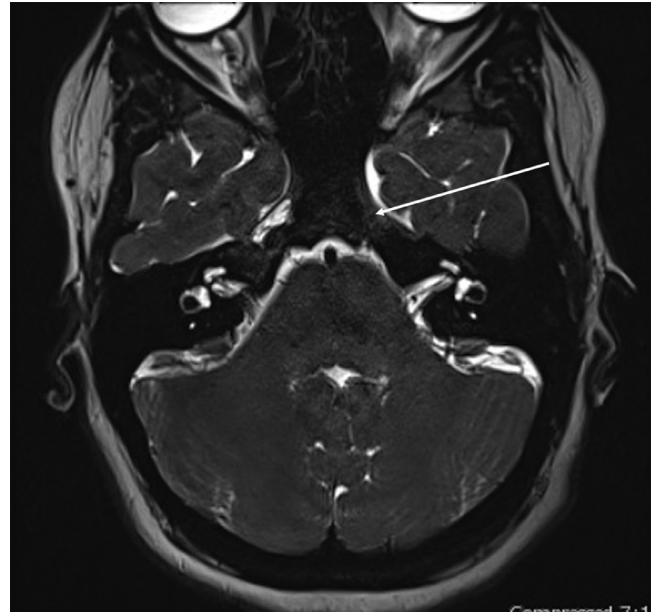
Herein, we report a rare case of an absent Meckel's cave in a 22-year-old female patient with TN. This case report aims to highlight this rare association and its possible etiological implications.

### Case presentation

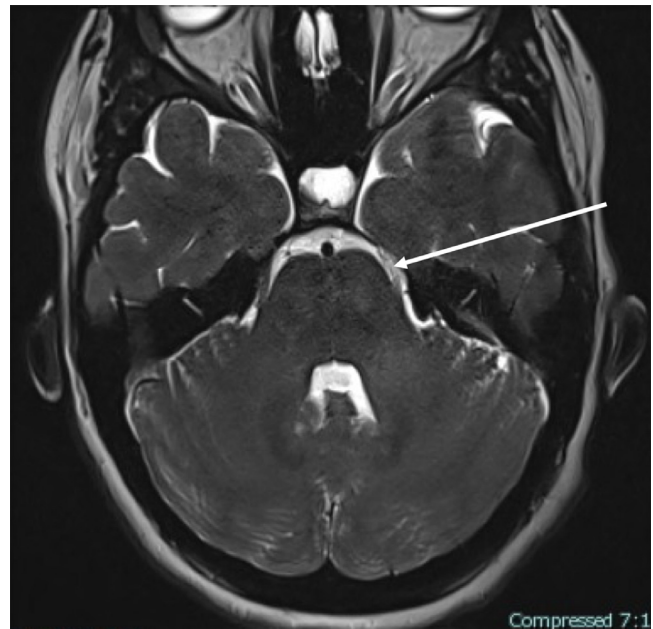
A 22-year-old female patient presented complaining of left-sided facial pain of 2 months duration. The pain was excruciatingly painful, with a sudden onset and stabbing character. The pain began at the corner of her left mouth and spread to encompass the entire left side of her face. The patient experienced multiple painful episodes every day, each lasting less than a minute. These episodes were typically triggered by light touch or cold air, though they may occasionally arise spontaneously. Accordingly, she was advised to stay away from the triggers. Analgesia, however, did not alleviate the pain. She has no medical history apart from right-sided Bell's palsy 6 years prior to presentation which was managed with corticosteroids. She has no family history of neurological diseases, and she is currently taking a vitamin B12 supplement. On physical examination, she was in distress due to the pain and was vitally stable. There was a slight mouth angle deviation as a residual defect from Bell's palsy episode 6 years ago. The oral cavity examination excluded any dental cause, and the neurological examination was normal throughout. However, we were not able to examine the left side of the face due to the patient's fear of stimulating pain episodes. A diagnosis of TN was made based on the clinical picture, and a brain MRI was ordered for further investigation into possible causes. The MRI showed an absent left Meckel's cave (Fig. 1). In addition, an atrophic left cisternal segment of the trigeminal nerve was demonstrated (Fig. 2). Based on these findings and given that the contralateral (unaffected) side had no demonstrable abnormalities, TN, in this case, was presumed to have been caused by the absence of Meckel's cave. A complete blood count test and liver function tests were ordered, which came back normal. She was started on carbamazepine 100 mg twice daily for symptomatic relief and maintenance therapy.

### Discussion

Although TN is a clinical diagnosis, neuroimaging studies, particularly magnetic resonance imaging (MRI) studies are of paramount importance in identifying structural etiologies affecting the trigeminal nerve anywhere along its course from the proximal origin of its root in the brainstem to its distal sensory branches in the face. Therefore, MRI should be used for further classification after an initial clinical diagnosis of TN.



**Fig. 1** – A thin slice axial T2 MRI of the cerebellopontine angle shows an absent left Meckel's cave.



**Fig. 2** – A thin slice axial T2 MRI of the cerebellopontine angle shows atrophic left cisternal trigeminal nerve.

In most cases, TN can be etiologically traced back to a neighboring blood vessel (usually an aberrant loop of the superior cerebellar artery) compressing the trigeminal nerve (classical TN) [7]. Of note, the mere compression of the trigeminal nerve root is not sufficient to establish a diagnosis of classical TN. That is, the compressed nerve must demonstrate morpho-

logical changes, namely dislocation, distortion, and/or atrophy on MRI imaging. In contrast, idiopathic TN refers to cases wherein the neurovascular contact is lacking or insufficient to produce morphological changes [8]. According to the European Academy of Neurology guideline on trigeminal neuralgia, classical and idiopathic TN are collectively termed primary TN. When other neurological diseases trigger TN (including tumors, arteriovenous malformations, and demyelinating diseases like multiple sclerosis), it is called secondary TN. This classification has bearing on the appropriate medical and surgical options for each particular case of TN [9]. Classical TN typically manifests in adults in their 50s, whereas secondary TN presents at a younger age [5].

An absent Meckel's cave as a possible cause of TN is a rarely described and poorly understood entity. Such a phenomenon has been described in only 2 case series, 7 patients and 3 patients, respectively, and 2 case reports, adding up to a total of 12 cases [10–13]. Interestingly, all cases reported in the literature, including ours, were female. Remarkably, our patient presented at a younger age (22 years) than previous cases reported in the literature (fourth and fifth decades of life). In the present case, similar to all previous cases, Meckel cave was absent ipsilateral to the side affected by TN and was normal on the unaffected side, implying a causal relationship. This is bolstered by the findings of Jain et al., who retrospectively reviewed their institutional records for a 20-year period and found no instance of absent Meckel's cave in patients without TN or contralateral TN, and neither did they find asymptomatic absent Meckel's cave [12].

Two theories exist on the etiological basis of an absent Meckel's cave. Primary failure of formation of the subarachnoid CSF-containing space within Meckel's cave is one theory. The other theory proposes that the absence of Meckel's cave is due to its secondary contraction. The CSF is important to drain away any inflammatory markers like TNF-alpha in the vicinity of the nerve. Thus, absence of the Meckel's cave, with the subsequent absence of CSF, allows the inflammatory markers to accumulate and cause Schwann cell damage and trigeminal nerve demyelination [12].

The first-line therapy in TN is anticonvulsants, namely carbamazepine and oxcarbazepine [14]. Other alternatives include lamotrigine, gabapentinoids (gabapentin and pregabalin), baclofen, and botulinum toxin type A, which can either be used as monotherapy or in combination with carbamazepine or oxcarbazepine. Surgery may also be considered but should be reserved for cases refractory or intolerant to pharmacotherapy [15]. Surgical options include microvascular decompression (MVD) as well as transcutaneous and non-transcutaneous neurosurgical procedures that produce controlled destruction of sensory trigeminal nerve fibers [16].

Lastly, an absent Meckel's cave is a rare entity associated with TN. Our case report adds to the existing paucity of literature discussing this association, yet larger clinical studies are still needed to establish a possible causal relationship.

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## Patient consent

Written informed consent for the publication of this case report was obtained from the patient.