



## Case Report

# Trigeminal neuralgia caused by an arachnoid cyst in Meckel's cave: A case report and literature review

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## ABSTRACT

**Background:** We present a rare case of trigeminal neuralgia (TN) caused by an arachnoid cyst (AC) in Meckel's cave (MC).

**Case Description:** A 35-year-old man presented with facial pain in the left maxillary and mandibular regions. Since the initial magnetic resonance (MR) imaging showed no apparent offending vessels or tumors, the patient was diagnosed with idiopathic TN, for which carbamazepine was initially effective. When his pain worsened, he was referred to our hospital. A slightly asymmetric shape of MC and distorted course of the trigeminal nerve was confirmed on the initial and repeat MR images. His pain was characterized as electric-shock-like pain, which was triggered by touching the face. Under the tentative diagnosis of an AC confined to MC compressing the trigeminal nerve, the exploration of MC through suboccipital craniotomy was performed. Intraoperatively, the AC was identified in the rostral portion of MC. The indentation of the trigeminal nerve was also observed at the orifice of MC, indicating severe compression by the AC. The wall of the AC was fenestrated. The patient's pain was relieved immediately after surgery. Postoperative MR images showed that the course of the trigeminal nerve was straightened. Although our literature review found five similar cases, the size of the AC was the smallest in our case.

**Conclusion:** Although it is rare, the AC confined to MC can cause TN. The findings of this study emphasize the importance of evaluating subtle radiological findings of compression on the trigeminal nerve in cases of TN seemingly without neurovascular compression.

**Keywords:** Arachnoid cyst, Meckel's cave, Trigeminal neuralgia

## INTRODUCTION

Trigeminal neuralgia (TN) is known as facial pain with a wide range of severity. Although it can be idiopathic, TN also occurs secondary to the compression of the trigeminal nerve by vascular structures or tumor masses at the cerebellopontine angle extending to areas adjacent to the trigeminal nerve. It is widely accepted that microvascular decompression confers favorable surgical outcomes for TN with vascular compression of the trigeminal nerve.<sup>[1,5,13]</sup> Despite improvements in radiographic imaging technology, visual detection of the offending vessels is still difficult in some cases. In fact, 3.1–20.8% of patients with TN have been reported to have no visible signs of vascular compression based on imaging.<sup>[6,9,11,12]</sup> The precise mechanism underlying trigeminal pain in cases without vascular compression is still unclear.

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In the present case, we present a rare case of TN caused by an arachnoid cyst (AC) confined to Meckel's cave (MC). Preoperative magnetic resonance (MR) imaging demonstrated a severe torsion of the trigeminal nerve due to the AC. We emphasize the importance of careful evaluation of the subtle laterality of MC and the distorted course of the trigeminal nerve in cases without apparent vascular compression or tumors.

## CASE PRESENTATION

### Patient history and examinations

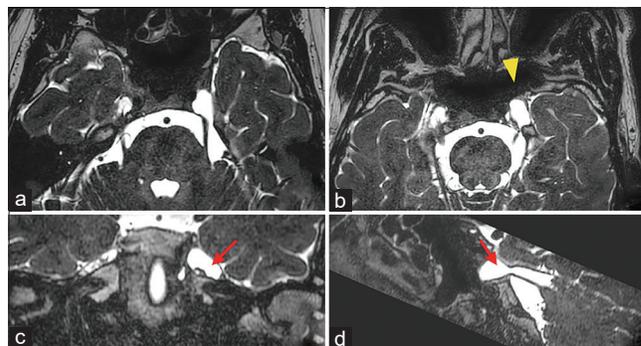
A 35-year-old man presented with facial pain of the left cheek that was similar to that of TN with neurovascular compression, that is, electric-shock like pain with a trigger point. Initial MR images showed no apparent vascular compression of the trigeminal nerve. Medical treatment with carbamazepine allowed gradual improvement of TN; however, the pain worsened within 1 year and remained for 6 months. He was, therefore, referred to our department for further investigation. Repeat constructive interference in steady state MRI revealed slightly expanded MC on the left side with a diameter of  $11 \times 11$  mm compared to that on the right side ( $6 \times 7$  mm) [Figure 1a and b] and distortion of the trigeminal nerve toward caudally in MC, suggesting the possible existence of cystic structure [Figure 1c and d]. This cystic structure was also retrospectively confirmed at the initial MR imaging, which had been overlooked at the initial diagnosis. After a thorough discussion of the treatment options, he wished to undergo surgical exploration of the trigeminal nerve and MC.

### Surgery

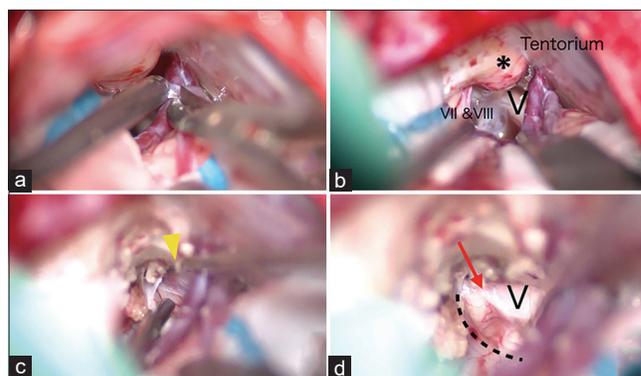
A standard retrosigmoid approach was performed. The trigeminal nerve was found behind the superior petrosal vein and its tributaries [Figure 2a]. After intradural resection of the suprameatal tubercle, the entire length of the trigeminal nerve was successfully exposed [Figure 2b and c]. There was no vascular compression along the trigeminal nerve. The AC in MC severely compressed the trigeminal nerve toward caudally [Figure 2c, arrowhead], resulting in the indentation of the nerve surface [Figure 2d]. After fenestration and collapse of the cyst, the trigeminal nerve compression was successfully released.

### Postoperative course

The patient's facial pain resolved immediately after the surgery. Postoperative MR imaging revealed that the course of the trigeminal nerve in MC was straightened with no compression [Figure 3]. The patient had no recurrence of pain at 1-year follow-up.



**Figure 1:** Preoperative constructive interference in steady state MRI. (a) No visible vascular compression on the trigeminal nerve. (b) Enlarged Meckel's cave (MC, yellow arrow head) detected with the isointense cerebrospinal fluid. (c and d) Stretched trigeminal nerve detected at the floor of MC (red arrow), on coronal and sagittal view.



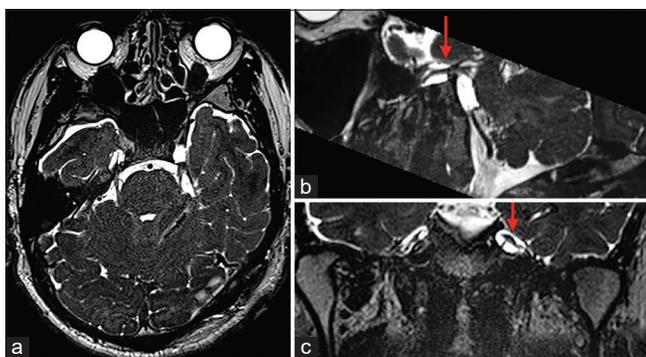
**Figure 2:** Intraoperative photographs. (a) The arachnoid sleeve enveloping the petrosal vein was cut. (b) The suprameatal tubercle (SMT; asterisk) obstructed the trigeminal nerve root (v) in direction to MC. (c) After resection of the SMT, the transparent wall of arachnoid cyst (yellow arrow head) was detected above the trigeminal nerve and fenestrated using a micro dissector. (d) The trigeminal nerve was transposed rostrally, an indentation (red arrow) was detected.

## DISCUSSION

We describe a rare case of TN caused by an AC confined to MC. To date, several cases of ACs affecting MC have been reported in the literature [Table 1].<sup>[2-4,7,14]</sup> Excluding reports of ACs of the petrous apex, leading to bony erosion toward MC,<sup>[3]</sup> our literature search found five similar cases of AC in MC causing trigeminal symptoms. The diagnosis may have been easier in those cases because the cyst was large and clearly discernible on MR images or CT scans. However, there was diagnostic delay in our case because the size of cyst was much smaller than that in previous cases and the bony structure constituting MC was almost preserved. Based on our findings, observing slight changes in MC size on MR images may be of diagnostic value. Besides this, we

**Table 1:** Summary of the previous reports of arachnoid cyst in Meckel's cave causing trigeminal symptoms.

Author /year	Age/sex	Symptoms	Radiological findings	Treatment	Improvement of symptoms
Barta <i>et al.</i> , 2002	55/F	Toothache Facial paresthesia Partial sensory loss	CT: Expansile lesion with scalloping of the adjacent bony cortex MRI: Lobulated CSF-intensity lesion with no contrast enhancement	Temporal craniotomy	Resolved in 6 weeks
Wörner <i>et al.</i> , 2002	44/M	Oral dysesthesia Vertigo Diplopia	MRI: T2 high-intensity cystic lesion	Suboccipital craniotomy (twice)	No recurrence during 15 months
Jacob <i>et al.</i> , 2008	32/F	Periocular pain Sixth nerve palsy	CT: No erosion of the petrous apex MRI: Lobulated lesion of CSF intensity	Observation	Resolved in 3 months
Fois <i>et al.</i> , 2011	42/F	Facial pain	CT: Bone erosion of the petrous apex MRI: CSF-intensity cystic lesion with no contrast enhancement	No description	No description
Bigder <i>et al.</i> , 2017	57/F	Shooting pain Facial paresthesia	CT: No abnormal finding MRI: CSF-intensity cystic lesion	Percutaneous drainage	Immediately pain-free No recurrence during 2 years
Our case	35/M	Facial pain	CT: Slight expansion of the Meckel's cave MRI: CSF-intensity lesion with no contrast enhancement	Suboccipital craniotomy	Immediately pain-free No recurrence during 1 year



**Figure 3:** Postoperative imaging. (a-c) CISS MRI revealed relief of compression of the trigeminal nerve in axial, sagittal, and coronal view. Note: the trigeminal nerve (red arrow) traverses the center of MC.

believe that distortion of the axis of the trigeminal nerve should be carefully evaluated on preoperative MR images when the nerve compression is not apparent. In the present case, a sagittal MR image demonstrated a constriction of the trigeminal nerve at the orifice of MC because the AC severely compressed the trigeminal nerve from rostrally. Kimura *et al.* also reported a case of TN caused by nerve constriction due to a fibrous ring and emphasized the need for careful evaluation of preoperative MR images.<sup>[8]</sup> Furthermore, the characteristics of pain might be indicative of the presence of nerve compression. Classical TN presentation of a brief electric-shock-like sensation with partial or temporary relief conferred by carbamazepine was noted in the present case. These are favorable factors for pain resolution after microvascular decompression.<sup>[10]</sup> For patients with these pain

characteristics, the presence of some organic etiology should be rigorously investigated on multiple plane MR imaging.

## CONCLUSION

Although such cases are rare, it is worth considering that TN can be caused by a small AC confined to MC. When there is subtle asymmetry of MC, the torsion or constriction of the trigeminal nerve inside MC might be indicative of this condition. Our report indicates the importance of careful evaluation of preoperative MR images for subtle evidence of compression on the trigeminal nerve in cases of TN seemingly without neurovascular compression.

## Declaration of patient consent

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

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Nil.

## Conflicts of interest

There are no conflicts of interest.

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