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Case Report

Navigating the neurovascular maze of trigeminal neuralgia ☆,☆☆

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ABSTRACT

Tic douloureux, also known as trigeminal neuralgia, is distinguished by recurrent episodes of severe, lancinating pain that affects one or more branches of the trigeminal nerve, representing a prevalent pain syndrome. This condition has an annual incidence rate of 27 per 100,000 individuals. Nevertheless, direct compression caused by vertebrobasilar dolichoectasia (VBD) represents a considerably less frequent etiology of trigeminal neuralgia, with an estimated overall incidence of about 1%. A 65-year-old female patient with a history of diabetes mellitus and hypertension presented with a severe, paroxysmal headache and lancinating pain localized to the right facial regions corresponding to the V2 and V3 trigeminal distributions, persisting for 3 years. MRI imaging indicated that the right trigeminal nerve is sandwiched between right superior cerebellar artery and hypertrophied right transverse pontine vein s/o left vertebral and basilor dolichoectasia with entrapment of trigeminal nerve between right SCA and right transverse pontine vein- (TYPE IV neurovascular compression). Vertebrobasilar dolichoectasia represents an uncommon etiology of neurovascular compression affecting the trigeminal and facial nerves, which may result in the development of trigeminal neuralgia and facial hemispasm. Magnetic resonance imaging (MRI) is the optimal modality for elucidating TN aetiology. Precise preoperative detection of neurovascular conflict enhances surgical efficiency and minimizes operative time. Initially, medical management should be pursued, as it may yield significant therapeutic benefits and potentially eliminate the necessity for surgical interventions.

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Introduction

Tic douloureux, also known as trigeminal neuralgia, is distinguished by recurrent episodes of severe, lancinating pain that affects one or more branches of the trigeminal nerve, representing a prevalent pain syndrome. This condition has an annual incidence rate of 27 per 100,000 individuals [1]. Trigeminal neuralgia is a clinical entity marked by transient, recurrent episodes of severe, unilateral shock-like pain characterized by abrupt onset and cessation, confined to the sensory territory of the trigeminal nerve [2]. Although numerous pathological conditions affecting the anatomical segments of the trigeminal nerve may elicit trigeminal neuralgia, neurovascular compression at the cisternal segment of the trigeminal nerve is regarded as the predominant etiology [3].

Nevertheless, direct compression caused by vertebrobasilar dolichoectasia (VBD) represents a considerably less frequent etiology of trigeminal neuralgia, with an estimated overall incidence of about 1% [4].

Magnetic resonance imaging (MRI) is particularly valuable as it can visualize the entire trajectory of the trigeminal nerve, with particular emphasis on the cisternal segment. Preoperative magnetic resonance imaging (MRI) assessment of the posterior fossa is instrumental in elucidating the primary etiological factors contributing to trigeminal neuralgia. Although pharmacological management is efficacious for pain alleviation during the initial 2 years following symptom onset, a significant number of patients ultimately necessitate surgical intervention. In the current investigation, we present a case involving a patient who experienced trigeminal neuralgia resulting from vertebrobasilar dolichoectasia (VBD) and was managed conservatively.

Case presentation

A 65-year-old female patient with a history of diabetes mellitus and hypertension presented with a severe, paroxysmal headache and lancinating pain localized to the right facial regions corresponding to the V2 and V3 trigeminal distributions, persisting for 3 years. The patient characterized the headache as a sharp, electric shock-like sensation lasting approximately 30 seconds, occurring multiple times throughout the day. Notably, there were no prodromal symptoms preceding the onset of headache episodes. The patient indicated that these episodes were triggered by light tactile stimulation of the facial region. Although the patient had been enduring these headaches for 2 years, there was a recent exacerbation in both the severity and frequency of the episodes. The accompanying symptoms included involuntary contractions of the right facial musculature and mild nausea. The patient affirmed that she experienced complete relief from pain during the intervals between episodes. By performing activities such as talking, chewing, shaving, and occasionally exposure to cool air, symptoms of the trigeminal nerve are exaggerated. Our patient had a pain severity score of 9 out of 10 and had little relief on oral opioids, tricyclic antidepressants, or dual antidepressant therapy.

The patient had a medical history of poorly controlled hypertension, diabetes mellitus, and dyslipidemia for 30 years, and her past history revealed suboptimal management of these conditions. There was no documented history of transient ischemic attacks or seizure disorders.

Laboratory evaluations revealed mild anaemia [red blood cell count of 4.10×10^{12}/L (normal range: 4.2 to 5.4 $\times$ 10^{12} /L), a haemoglobin level of 126 g/L (normal range: 121 to 151 g/L)] and hypokalemia [serum potassium concentration of 3.6 mmol/L (normal range: 3.5 to 5.5 mmol/L)], with renal and liver function remaining within normal limits. There was slight deviations in these laboratory parameters which was attributed to the patient's anxiety and suboptimal dietary habits exacerbated by the facial pain. The patient's pharmacological regimen included aspirin at a dosage of 75 mg, metformin at 1000 mg, bisoprolol at 5 mg, perindopril at 5 mg, and atorvastatin at 20 mg. Family history was unremarkable. The patient, a retired educator, lived independently with her husband. Upon clinical examination, the patient exhibited signs of distress. Vital signs were recorded as follows: a normal temperature of 38°C, a respiratory rate of 15 breaths per minute, a pulse rate of 86 beats per minute, blood pressure of 148/98 mmHg, and an oxygen saturation level of 97% in ambient air. The patient was found to be alert, conscious, and fully oriented. Neurological examination did not reveal any focal neurological deficits. Assessment of the upper and lower extremities demonstrated normal muscle tone, strength, and reflexes. Upon summarising the clinical presentation, the patient was diagnosed of trigeminal neuralgia.

Given the protracted history of headache episodes and the presence of localized findings during the physical examination, the patient was referred for a magnetic resonance imaging (MRI) scan of the brain to rule out any potential space-occupying lesions.

The imaging results indicated that the intracranial course of the left vertebral artery appeared aberrant. It was crossing the midline in front of the medulla reaching into the right CP angle cistern with resultant lateral displacement of right SCA, right vertebral artery which are seen compressing over right trigeminal nerve at the transition point causing its thinning (Figs. 1 A and B).

Right trigeminal nerve is sandwiched between right SCA and hypertrophied right transverse pontine vein s/o left vertebral and basilor dolichoectasia with entrapment of trigeminal nerve between right SCA and right transverse pontine vein-(TYPE IV neurovascular compression) (Fig. 1C).

The clinical diagnosis, along with the radiological interpretations, was thoroughly discussed with the patient.

The patient was advised of both medical and surgical options. Considering the advanced age and comorbid conditions she was initiated on medical management and was prescribed carbamazepine for pain management and was discharged with a plan for regular follow-ups at the outpatient clinic. The patient gave a positive response to the medical intervention and reported considerable improvement in her symptoms.

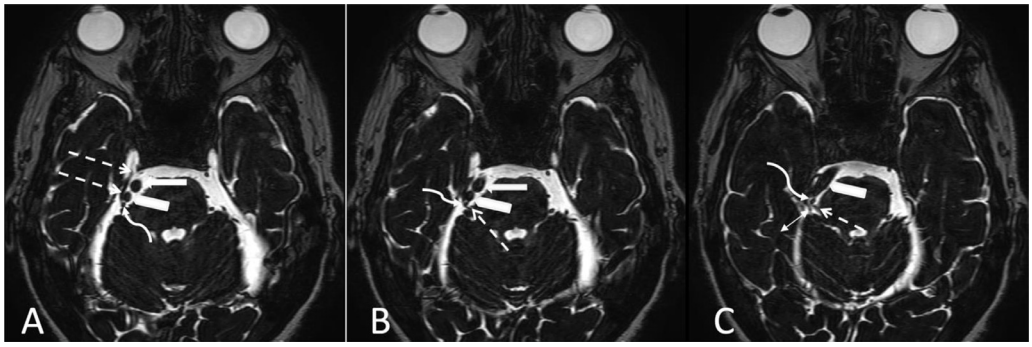

Fig. 1 – (A and B) T2WI 3D DRIVE sequence axial section brain reveals the aberrant intracranial course of the left vertebral artery (white arrow) and normal right vertebral artery (white pentagon arrow). It was crossing the midline in front of the medulla, reaching into the right cerebellopontine angle cistern with resultant lateral displacement of the right superior cerebellar artery (white curved arrow), and right vertebral artery (white pentagon arrow), which are seen compressing over the right trigeminal nerve (dash white arrow) at the transition point causing its thinning. (C) T2WI 3D DRIVE sequence axial section brain reveals right trigeminal nerve (dash white arrow) is sandwiched between right superior cerebellar artery (white curved arrow) and hypertrophied right transverse pontine vein (double head white arrow) s/o left vertebral (white arrow) and basilar dolichoectasia with entrapment of right trigeminal nerve (dash white arrow) between right superior cerebellar artery (white curved arrow) and right transverse pontine vein (double head white arrow)- (TYPE IV neurovascular compression).

Discussion

Intracranial arterial dolichoectasia is typified by the enlargement, tortuous configuration, or elongation of significant arterial structures at the base of the cranial cavity. Dolichoectasia most commonly occurs at the vertebrobasilar system [5]. When the basilar artery is positioned laterally to the boundaries of the clivus or dorsum sellae, or when bifurcation occurs above the suprasellar cistern plane, the vertebrobasilar system is deemed elongated. Ectasia is the diameter of the basilar artery more than 4.5 mm [4].

Vertebrobasilar dolichoectasia may manifest asymptomatically. The clinical presentations of symptomatic instances exhibit considerable variability [6]. The predominant clinical manifestation is ischemic stroke. Additional manifestations may encompass symptoms attributable to cranial nerve compression, brainstem compression, hemorrhagic events, and obstructive hydrocephalus [7].

The degeneration of the vascular wall attributed to atherosclerosis in conjunction with hypertension is posited as a potential pathogenic mechanism. However, alternative scholarly perspectives regard dolichoectasia as a congenital vascular anomaly, based on histological findings indicative of defects in the internal elastic lamina and media thinning secondary to atrophy of smooth muscle [8]. Indeed, dolichoectasia appears to stem from a congenital anomaly, with its progression potentially influenced by arterial hypertension and superimposed atherosclerotic changes. In the case presented herein, we concurred that the etiology of vertebrobasilar dolichoectasia was multifactorial.

Although infrequent, it is occasionally observed that an ectatic vertebrobasilar vessel may be the sole etiological factor for trigeminal neuralgia (TN). In comparison to other TN patients, those with TN instigated by vertebrobasilar compressions tend to be older, predominantly male, and more frequently afflicted by hypertension [9].

Furthermore, autoimmune and inflammatory mechanisms are also regarded as risk factors for vertebrobasilar dolichoectasia [10].

Moreover, several of the etiological factors of vertebrobasilar dolichoectasia previously delineated are also implicated as determinants of arterial dissection. Furthermore, it has been documented that fusiform aneurysms represent a type of arterial dissection characterized by localized ballooning along the entirety of the vessel wall for a brief segment [11].

Trigeminal neuralgia (TN) episodes typically endure for only a few seconds but may recur frequently within a brief timeframe. These episodes are frequently triggered by minor sensory stimulation of designated trigger zones, which can be situated anywhere within the distribution area of the affected nerve. TN predominantly impacts the second and third divisions of the trigeminal nerve [4].

The incidence of TN is observed in both sexes, albeit with a slight predominance in females, and the diagnosis is predominantly established in individuals over the age of 50, even though younger adults and children may also present with this condition [12].

Cranial nerves are composed of a root entry zone (REZ), a transitional zone, and a peripheral zone. The REZ is myelinated by glial cells, the peripheral zone is myelinated by Schwann cells, and the transitional zone is myelinated by both types of cells. The REZ's length varies among cranial nerves, but it typically measures between 3 to 5 mm in the trigeminal nerve [13]. The site of neurovascular conflict significantly impacts the presentation of typical versus atypical neuralgia symptoms. Neurovascular conflict occurring at the REZ is associated with typical TN, whereas conflict at the peripheral zone may result in atypical manifestations.

It is hypothesized that with advancing age, arterial elongation occurs alongside a descent in brain structure.

Consequently, the presence of redundant arterial loops and the bridging of intrinsic hindbrain veins contribute to the compression of cranial nerve root entry zones [4].

Vertebrobasilar dolichoectasia represents a relatively prevalent cause of TN in comparison to aneurysms [14]. Furthermore, approximately 5% of TN cases exhibit bilateral involvement [15]. A familial predisposition to TN, the occurrence of other cranial nerve compressions, and essential hypertension constitute significant risk factors for bilateral manifestations of the disease [16].

Beyond arterial compression, TN may also be attributed to venous compression. It is known that the transverse pontine vein exerts pressure on the trigeminal nerve within Meckel's cave [17]. Prior to microvascular decompression (MVD) surgery this knowledge is important. The transverse pontine vein might not be visible in conventional suboccipital approach which can lead to surgical failure.

Preoperative identification of such cases through magnetic resonance imaging (MRI) is thus important [18]. Lateral mesencephalic veins, cerebellopontine fissure, middle cerebellar peduncle and pontotrigeminal vein are the venous structures that may induce compression on the trigeminal nerve. A patient may have simultaneous arterial and venous compression of a nerve.

In this case, the patient was managed medically with carbamazepine and demonstrated a favorable response to conservative treatment for trigeminal neuralgia. Surgical intervention was deemed unnecessary in this patient as there was elevated anesthetic risk associated considering the age.

Vertebrobasilar dolichoectasia is frequently managed conservatively [7]. Surgical procedures includes the placement of ventriculoperitoneal shunts for concomitant hydrocephalus or microsurgery aimed at decompression of the trigeminal or facial nerves [6]. An alternative therapeutic strategy that has demonstrated favorable outcomes is gamma knife radiosurgery represents [19].

In this case, the patient, the patient exhibited trigeminal neuralgia and facial spasms resulting from neurovascular compression impacting the trigeminal and facial nerves; however, no brainstem injury or hydrocephalus was observed.

Historically, surgical options for medically refractory pain include percutaneous or microsurgical rhizotomy and microvascular decompression (MVD), with MVD applied to patients with and without trigeminal neuralgia linked to dolichoectatic artery anomalies. Currently, various operative techniques for trigeminal neuralgia are utilized, including radiofrequency gasserian rhizotomy, glycerol postgasserian rhizolysis, balloon compression of the gasserian ganglion, and microvascular decompression of the trigeminal root. In cases where trigeminal neuralgia is caused by anomalies in the vertebrobasilar artery, alternative approaches, such as repositioning the tortuous artery and wrapping the trigeminal nerve, have been explored recently. A growing body of evidence supports microvascular decompression as the leading surgical method for trigeminal neuralgia, with success rates either superior or comparable to other treatments and significantly lower rates of facial numbness [20].

Although trigeminal neuralgia from vertebrobasilar dolichoectasia is rare and its management well-defined, such patients require more clinical attention compared to their counterparts without this condition, necessitating the expertise of a radiologist and neurosurgeon for accurate arterial dissection identification.

Conclusion

Vertebrobasilar dolichoectasia represents an uncommon etiology of neurovascular compression affecting the trigeminal and facial nerves, which may result in the development of trigeminal neuralgia and facial hemispasm.

Magnetic resonance imaging (MRI) is the optimal modality for elucidating TN aetiology. Precise preoperative detection of neurovascular conflict enhances surgical efficiency and minimizes operative time. Identifying the specific compressive vascular structure is vital for determining the suitable surgical approach for each patient. Time-of-flight (TOF) MR angiography and high-resolution 3D T2-weighted imaging are reliable methods for identifying arterial compression.

Initially, medical management should be pursued, as it may yield significant therapeutic benefits and potentially eliminate the necessity for surgical interventions, particularly in patients with concomitant medical conditions who are unsuitable candidates for surgical procedures.

Ethics approval and consent to participate

Written consent taken.

Availability of data and material

None.

Author contributions

DN and PHP was involved in providing clinical details of the patient. RP discussion on the pathology. SD accumulated the results of the patient's radiological investigations. PNB and RK was involved in collecting images and formatting data. All authors have read and approved the manuscript.

Patient consent

Informed and written consent was obtained from the patient.

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