

VERTEBROBASILAR DOLICHOECTASIA AS A CAUSE OF TRIGEMINAL NEURALGIA

THE ROLE OF MICROVASCULAR DECOMPRESSION

Case report

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ABSTRACT - Our purpose is to report a case of trigeminal neuralgia caused by vertebral dolichoectasia treated with microvascular decompression. A 63-year-old man sought treatment for a recurrent lancinating left facial pain in V2 and V3 trigeminal territories. The computed tomography angiography revealed a mechanical compression of the left trigeminal nerve due to vertebral dolichoectasia. The patient was submitted to a left suboccipital craniotomy. Shredded Teflon[®] was introduced in the conflicting neurovascular area, achieving a satisfactory decompression. The patient's pain resolved immediately. Vertebral dolichoectasia is a rare cause of trigeminal neuralgia and a successful outcome can be achieved with microvascular decompression.

KEY WORDS: trigeminal neuralgia, vertebral dolichoectasia, microvascular decompression.

Dolichoectasia vertebral como causa de neuralgia trigeminal: o papel da descompressão microvascular. Relato de caso

RESUMO - O objetivo desse estudo é relatar um caso de neuralgia trigeminal causado por dolichoectasia vertebral tratado com descompressão microvascular. Um homem (63 anos) consultou por neuralgia trigeminal recorrente na hemiface esquerda (territórios V2 e V3). A angiotomografia cerebral revelou compressão mecânica do nervo trigêmeo esquerdo devido à dolichoectasia vertebral. O paciente foi submetido à craniotomia suboccipital esquerda. Introduziu-se Teflon[®] na área de conflito neurovascular, obtendo-se uma descompressão satisfatória. O paciente apresentou remissão da dor imediatamente. A dolichoectasia vertebral é uma causa rara de neuralgia trigeminal e uma excelente evolução pode ser alcançada com a descompressão microvascular.

PALAVRAS-CHAVE: neuralgia trigeminal, dolichoectasia vertebral, descompressão microvascular.

Trigeminal neuralgia is a common facial pain syndrome which usually affects middle-aged and elderly people. The syndrome consists of paroxysms of lancinating pain, usually in the distribution of the mandibular and maxillary divisions of the trigeminal nerve. Patients often involuntarily wince when experiencing this severe pain, providing the derivation of the term *tic douloureux*¹. The most common cause of idiopathic trigeminal neuralgia is microvascular compression of the nerve². A compressing vessel is iden-

tified for most patients who undergo microsurgical decompression, being the superior cerebellar artery responsible for 75% of cases³. Other arteries, such as the anteroinferior cerebellar artery (10%), posteroinferior cerebellar artery (1%), vertebral artery (2%), basilar artery (1%), and primitive trigeminal artery or its variants, have also been identified as the cause of this condition^{4,5}. Tumors, aneurysms and vascular malformations are observed in only a few cases³. Vertebral dolichoectasia is also rarely a cause

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Fig 1. Standard CTA (coronal view) showing the dolichobasilar ectasia.

of trigeminal neuralgia^{3,6}. Many surgical or nonsurgical modalities of treatment have been proposed for trigeminal neuralgia. Microvascular decompression is the most effective surgical modality available. It is nondestructive, mortality and morbidity rates are low when properly performed, and it confers the best short and long-term quality of life to the patients⁷.

The purpose of this study is to report and discuss a rare case of trigeminal neuralgia due to vertebrobasilar dolichoectasia successfully treated with microvascular decompression and documented by computed tomography angiography (CTA).

CASE

A 63-year-old man with a past medical history of hypertension sought treatment after experiencing a recurrent lancinating left facial pain in trigeminal territories (V2 and V3) for almost five years. The pain was described as sharp and electrical and was exacerbated by talking, chewing and sometimes was spontaneously triggered. These symptoms resolved by October 2001, after a percutaneous surgical procedure (radiofrequency lesioning of the gasserian ganglion). After a pain-free period of almost 4 years, the pain recurred with the same characteristics. High doses of carbamazepine and amitriptyline did not relieve the pain adequately. The patient was referred with clinically intractable symptoms and subsequently considered for microsurgical decompression after neurological reinvestigation.

The patient's neurological examination revealed hyperesthesia in the V2 and V3 distribution of the trigeminal nerve on the left side. All the others aspects of the neurological examination were normal. The CTA revealed a

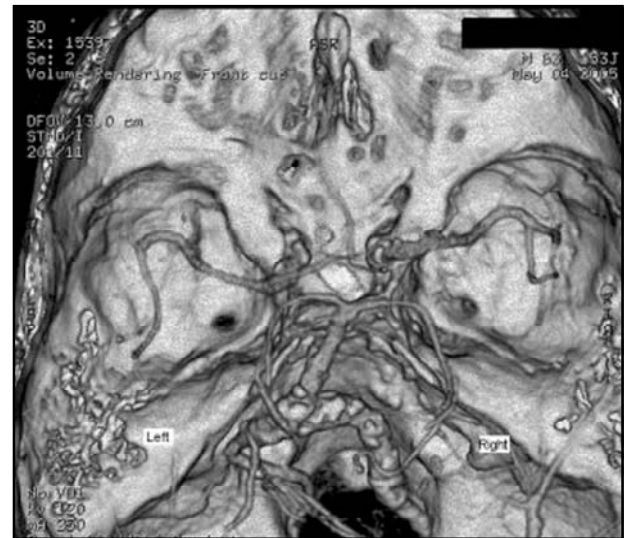


Fig 2. Reconstruction 3D - CTA showing the dolichobasilar ectasia and its relations to skull base.



Fig 3. Transoperative photography showing the anatomy of the left ponto-cerebellar angle and the dolichobasilar ectasia and its relations to the trigeminal nerve and other anatomical structures (V.A., vertebral artery; C.N., cranial nerve; Bas. A., basilar artery; D.V., Dandy's vein).

mechanical compression at the left trigeminal nerve due to vertebrobasilar dolichoectasia (Figs 1 and 2). Surgery was indicated.

The patient was placed in the prone oblique (*park bench*) position, and a left suboccipital craniotomy was performed. The dura was opened, and cerebrospinal fluid was released at the cisterna magna to provide a capacious working environment. Arachnoid dissection revealed a large vascular structure, later identified as the basilar dolichoectatic artery, dislocating and compressing the left trigeminal nerve at its root entry zone (Fig 3). Shredded Teflon[®] was introduced in the conflicting neurovascular area (between the artery and the trigeminal nerve), achieving a satisfactory decompression. There was no other vascular or nerve microsurgical manipulation.

The patient's lancinating facial pain resolved immediately after surgery. He initially presented with mild disequilibrium, but it was completely resolved at a 3-month follow-up examination.

DISCUSSION

Vertebrobasilar dolichoectasia is an uncommon vasculopathy of unclear etiology which affects the arterial wall of vertebral and/or basilar arteries⁸. Traditionally, vertebrobasilar dolichoectasia has been regarded as atherosclerotic in nature, although recently Mizutani and Aruga have suggested that some cases represent a dissecting process^{9,10}. This disease causes arterial elongation and enlargement, with subsequent haemodynamic and haemostatic changes, which, in turn, lead to thrombosis, microembolisation, and brainstem compression, with or without aneurysm formation¹¹. A variety of clinical syndromes have been associated with ectatic vertebral arteries. These include a number of isolated or combined brainstem/cranial nerve syndromes, cervicomedullary junction compression, transient or permanent motor deficits, cerebellar dysfunction, central sleep apnea, hydrocephalus and ischemic stroke¹¹⁻¹³.

Direct compression by vertebral dolichoectasia is an uncommon cause for trigeminal neuralgia. The incidence, as estimated in previous reports, ranges from 0.9% to 5.7%¹⁴. Piatt et al.¹⁵ reported 2 cases in a series of 105 patients. Bederson et al.¹⁶ related 4 cases in a group of 256 operated cases. Klun et al.¹⁷ reported 2 cases in a group of 220 operated patients. Vascular compression usually occurs at or near the root entry zone (REZ) of the trigeminal nerve, as reported by some authors. Hamlyn¹⁸ observed that 42 out of 46 patients who underwent posterior fossa surgery for treatment of trigeminal neuralgia had a vessel in contact with the nerve. Of those, 28 had a vessel in contact at the REZ, 12 had a vessel in contact lateral to the REZ (the point of contact with the nerve was more than one-half of the vessel's diameter away from the brainstem), and 2 had a vessel in contact at the REZ as well as lateral to it. Sindou et al.¹⁹ observed the presence of a contacting vessel in 97% of 579 patients with idiopathic trigeminal neuralgia. The site of contact was at the REZ in 52% of cases, in the mid-third of the nerve in 54%, and at the exit of the nerve from Meckel's cave in 10%. In the present case, the conflicting neurovascular area was located at the REZ.

Several operative treatments for trigeminal neuralgia are in current use, including radiofrequency

gasserian rhizotomy, glycerol postgasserian rhizolysis, balloon compression of the gasserian ganglion, and microvascular decompression of the trigeminal root. When cranial nerve dysfunction, especially trigeminal neuralgia, is caused by anomalies of caliber, length, and tortuosity of the vertebral arteries, alternative techniques, such as repositioning of the tortuous vertebral artery by pulling it toward the nearby dura mater²⁰ and encircling method of trigeminal nerve decompression¹⁴ have been reported recently. In the present case, the authors thought that these techniques would not bring advantages over the microvascular decompression²⁰.

Microvascular decompression for hyperactive dysfunction of cranial nerves was initially developed by Gardener and Miklos²¹ and Gardner and Sava²² and was perfected and popularized by Jannetta²³⁻²⁵ after the introduction of the microsurgical technique under an operative microscope²⁶. Microvascular decompression for trigeminal neuralgia has proven to be a highly effective and safe surgical procedure in alleviating the effects of neurovascular compression²⁷. Compared to alternative treatments, microvascular decompression offers significant advantages for trigeminal neuralgia²⁸. There is a growing body of evidence suggesting microvascular decompression as the best surgical modality for trigeminal neuralgia⁷. The rates of success (free of pain, without medication) are superior or at least equal to those of other reported treatments, with substantially lower rates of facial numbness²⁸.

The majority of the series in the literature reports a percentage of pain relief between 63% and 94%⁷ with well-defined follow-ups (mean time ≥ 2 years). However, the incidence of recurrence has been reported to range from 3 to 30%²⁹. Long-term follow-up studies revealed that most postoperative recurrences of trigeminal neuralgia occurred in the first 2 years after surgery²⁹. Mendoza and Illingworth³⁰ reported that 90% of recurrences occurred within 2 years. The annual rate of recurrence for trigeminal neuralgia decreases below 2% within 5 years after surgery and below 1% within 10 years after surgery²⁹. Twenty-year follow-up data demonstrated that 30% of successfully treated patients experienced trigeminal neuralgia recurrences²⁹.

It was previously reported that female sex, symptom duration of more than 8 years, and a lack of immediate postoperative cessation of trigeminal neuralgia were significant predictors of eventual recur-

rence. Preoperative sensory deficits, a history of a trigeminal ablative procedure, and the number of trigeminal divisions affected by trigeminal neuralgia were not significant predictors²⁹.

In the present case, the patient's lancinating facial pain resolved immediately after surgery. He initially presented with mild disequilibrium, but it was completely resolved at a 3-month follow-up examination. We attribute it to the manipulation of the vestibular nerve, and the complete resolution of this symptom after a 3-month follow-up reinforces this suspicion. The follow-up period in the present case is certainly short, but the patient does not present any of the previously reported predicting recurrence factors, so we strongly believe that the cure with microvascular decompression in this case is very likely to be obtained.

In conclusion, vertebral basilar dolichoectasia is a rare cause of trigeminal neuralgia and a successful outcome can be achieved with microvascular decompression.

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