Bangladesh Journal of Neuroscience 2020; Vol. 36 (2): 113-116

Vertebrobasilar Dolichoectasia as A Rare Cause of Trigeminal Neuralgia - A Case Report

KABIR MS[1], CHOWDHURY A[2], DEY SK[3], MUZAHID MAA[4], SAHA S[5], ROY U[6], BHUIYAN MM[7], ISLAM MR[8]

Abstract:

We present a case of 24 years old young male with trigeminal neuralgia due to vertebrobasilar dolichoectasia, a rare condition characterized by enlargement, tortuosity, or elongation of intracranial arteries. Dolichoectatic vessels can cause dysfunction of cranial nerves through direct vascular compression. Vertebrobasilar dolichoectasia is a rare cause of trigeminal neuralgia and a successful outcome can be achieved with microvascular decompression. The relationships of vertebrobasilar dolichoectasia with the particularities of neurovascular conflict and images findings are discussed.

Key words: *Trigeminal neuralgia, vertebrobasilar dolichoectasia, microvascular decompression.*

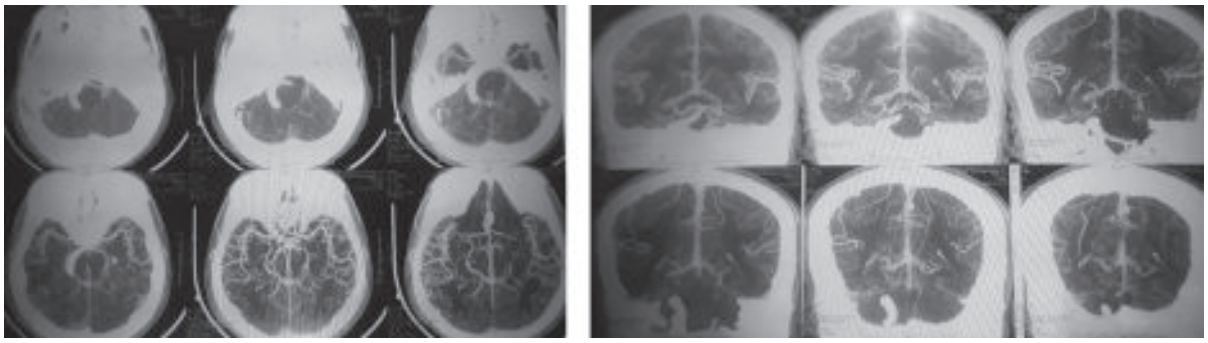
Introduction:

Trigeminal neuralgia (TN) is a well-known clinical entity characterized by paroxysmal hemifacial pain[1]. Vertebrobasilar dolichoectasia (VBD) is a very unusual cause of TN associated to vascular compression due to characteristic conformation of VBD[2]. The most common cause of idiopathic trigeminal neuralgia is microvascular compression of the nerve[2]. A compressing vessel is identified for most patients who undergo micro surgical decompression, being the superior cerebellar artery responsible for 75% of cases[3]. 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]. Tumors, aneurysms, and vascular malformations are observed in only a few cases[5]. Vertebrobasilar dolichoectasia is also rarely a cause of trigeminal neuralgia[6]. In the present study, we describe a young patient who developed trigeminal neuralgia caused by VBD.

Case Summary

Our patient was a 24-year-old Bangladeshi male, who was normotensive, non-diabetic, right- handed electrician, presented with a 1-year history of severe paroxysmal and lancinating right facial pain in V2 and V3 trigeminal territories. The pain used to come in sudden bursts lasting 1-5 minutes and recurs 10-30 times a day. The pain was described as sharp and electrical and was exacerbated by talking, chewing, and sometimes was spontaneously triggered. The pain was not satisfactorily controlled by carbamazepine, tricyclic, or dual antidepressant. The patient's neurological examination revealed mild hyperesthesia in the V2 and V3 distribution of the trigeminal nerve on the right side. All other aspects of the neurological examination were normal. Contrast CT scan, MRA of brain, and cerebral DSA demonstrated an elongated and tortuous vertebro-basilar artery causing mechanical compression at the right trigeminal nerve root (Figures 1(a,b),2,3). Many surgical and non-surgical modalities of treatment

1. Dr. Md. Suman Kabir, Resident, Dept. of Neurology, BSMMU
2. Dr. Ashish Chowdhury, Resident, Dept. of Neurology, BSMMU
3. Dr. Subash Kanti Dey, Associate Professor, Dept. of Neurology, BSMMU
4. Dr. Md. Abdullah Al Muzahid, Resident, Dept. of Neurology, BSMMU
5. Dr. Sujan Saha, Resident, Dept. of Neurology, BSMMU
6. Dr. Uttam Roy, Resident, Dept. of Neurology, BSMMU
7. Prof. Dr. Md. Moniruzzaman Bhuiyan, Dept. of Neurology, BSMMU
8. Prof. Dr. Md. Rafiqul Islam, Ex.Chairman, Dept. of Neurology, BSMMU



(a) (b)

Fig.-1: *(a) (axial) and (b) (coronal) Contrast CT scan of brain of the patient shows enlarged and tortuous basilar artery crossing and displacing upper pons with compression of the right trigeminal nerve at the root entry zone.*

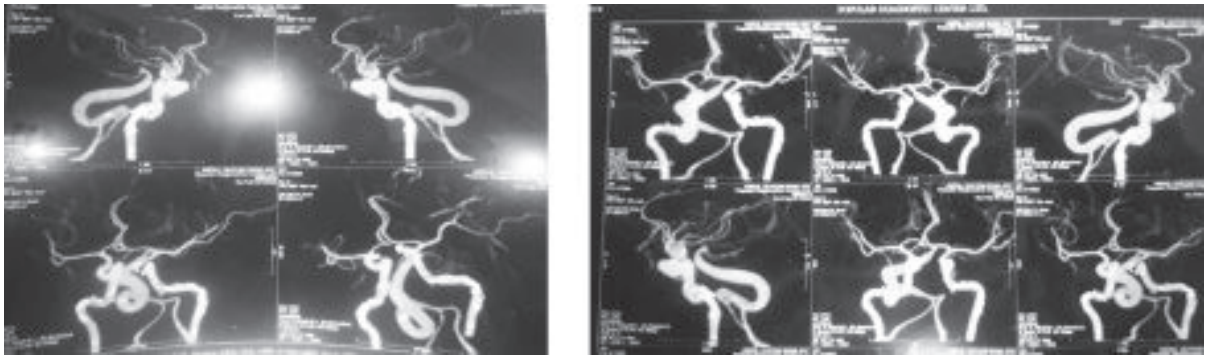


Fig.-2: *MRA of brain of the patient shows dilated and tortuous basilar artery.*

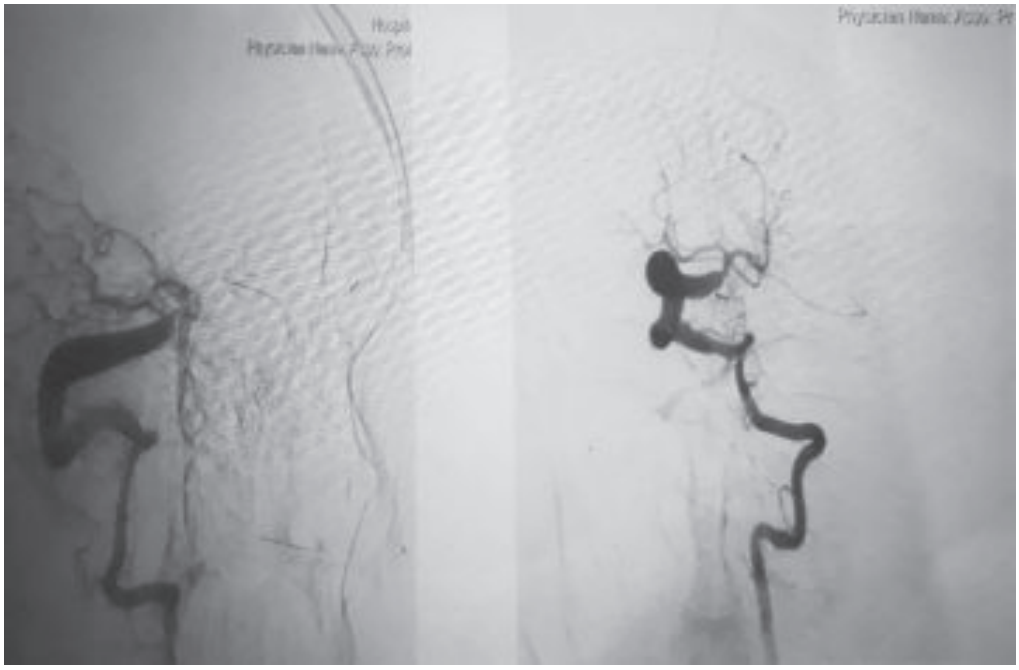


Fig.-3: *Cerebral DSA shows hugely dilated and tortuous basilar artery.*

have been proposed for trigeminal neuralgia. Microvascular decompression (MVD) is the most effective surgical modality available. We have planned for Surgical procedure (MVD) due to refractoriness and images findings.

Discussion:

Intracranial arterial dolichoectasia is a condition characterized by enlargement, tortuosity, or elongation of major arteries at the base of the brain. The most common localization of dolichoectasia is the vertebrobasilar system[3]. Vertebrobasilar system is considered to be elongated if the basilar artery lies lateral to the margin of the clivus or dorsum sellae or if it bifurcates above the plane of the suprasellar cistern. Ectasia is considered to be present if the basilar artery has a diameter greater than 4.5mm (Figures 1 and 2)[4]. The degeneration of the vascular wall due to atherosclerosis in association with hypertension is suggested as the pathogenic factor. However, other authors consider dolichoectasia to be a congenial vascular anomaly on the basis of histological observations of defect in the internal elastic lamina and thinning of the media secondary to smooth muscle atrophy[5]. In fact, dolichoectasia seems to be due to a congenital anomaly, and its evolution may be influenced by arterial hypertension and superimposed atherosclerosis.

In the present case, we have agreed that the VBD origin was multifactorial. Two types of symptoms were found associated with intracranial arterial dolichoectasia: those resulting from the compression of structures adjacent to the abnormal vessel and those resulting of ischemic events. Trigeminal and facial nerves are the commonest cranial nerves involved[6]. However, direct compression by VBD is an uncommon cause of TN with an estimated general incidence of approximately 1%[4]. In patients with VBD, the compression is slowly progressive, so the brainstem can functionally tolerate severe distortion without overt clinical manifestations, which may explain why most patients with VBD are asymptomatic[7]. The proposed mechanism for TN is vascular compression at a specifIc portion of the cisternal segment of the nerve known as the root entry zone (REZ). There have been suggested that REZ is particularly vulnerable to continued pulsatile pressure, which may result in focal demyelination and “short-circuiting” of impulses[8]. Traditionally, the surgical options for patients with medically refractory pain include percutaneous or microsurgical rhizotomy and microvascular decompression (MVD). However, based on neurovascular confliction, MVD has been practiced for the treatment of patients with/without TN associated to dolichoectatic artery[9,10]. In fact, decompression of the nerve root produces rapid relief of symptoms in most patients with neurovascular conflicting, probably due to the resulting separation of demyelinated axons and their release from focal distortion reduce the spontaneous generation of impulses and prevent their ephaptic spread[11]. Recent technological advancement in radiosurgery has revolutionalised all traditional surgical approaches in patients with TN. To date, Gamma knife surgery has become a keyhole to the minimally invasive approaches to TN associated with or not with VBD. However, some authors have shown that pain control rates of Gamma knife surgery in patients with TN associated with VBD were inferior to those of patients without VBD[12]. The natural history of VBD shows that patients with VBD may experience mainly cerebrovascular event with high incidence after the initial diagnosis. This may be explained by the fact that there are various mechanisms by which VBD may promote brain ischemia, including occlusion of small perforating vessels, reduction of anterograde flow in the dilated artery, distortion and stretching of the branches of the basilar artery (Duret’s Hemorrhages), and superimposed atheromatous changes[16]. This information is very essential in-patient outcome with TN caused by VBD because of natural tendency to overvalue facial pain instead of dolichoectasia and its potential complication. In the present case, we planned for MVD which may be a safe and effective treatment for TN.

Conclusion:

Vertebrobasilar dolichoectasia (VBD) is a rare cause of trigeminal neuralgia. So, to evaluate facial pain VBD should also be considered in the

differentials as it can be treated by microvascular decompression surgery.

As patients with VBD may be presented with devastating cerebrovascular complications and our patient is very young we planned for flow reduction by ligating RVA then microvascular decompression (MVD) for further management.

Conflict of interest

The authors have no conflict of interest.

References:

1. P. J. Jannetta, "Cranial nerve vascular compression syndromes (other than tic douloureux and hemifacial spasm)," Clinical Neurosurgery, 1981;28:445–456.
2. E. E. Ubogu and O. O. Zaidat, "Vertebrobasilar dolichoectasia diagnosed by magnetic resonance angiography and risk of stroke and death: a cohort study," Journal of Neurology, Neurosurgery and Psychiatry, 2004;75(1): 22–26.
3. B. Ince, G. W. Petty, R. D. Brown Jr., C. P. Chu, J. D. Sicks, and J. P. Whisnant, "Dolichoectasia of the intracranial arteries in patients with first ischemic stroke: a population-based study," Neurology, 1998;50(6):1694–1698, .
4. S. Love and H. B. Coakham, "Trigeminal neuralgia: pathology and pathogenesis," Brain, 2001;124(12):2347–2360.
5. S. G. Passero and S. Rossi, "Natural history of vertebrobasilar dolichoectasia," Neurology, 2008;70(1):66–72.
6. M.Baquero, "Vertebro-basilar dolichoectasia, "Revista de Neurologia, vol. 26, no. 149, pp. 143–148, 1998.
7. A. de Azambuja Pereira-Filho, M. deBarros Faria, C.Bleil, and J. L. Kraemer, "Brainstem compression syndrome caused by vertebrobasilar dolichoectasia," Arquivos de Neuro-Psiquiatria, vol. 66, no. 2, pp. 408–411, 2008.
8. A. R. Moller, "The cranial nerve vascular compression syndrome: II. A review of pathophysiology," Acta Neurochirurgica, vol. 113, no. 1-2, pp. 24–30, 1991.
9. R.Garcia DeSola and M.EscosaBage," Microvasculardecompression of trigeminal neuralgia caused by vertebrobasilar dolichoectasia," Revista de Neurologia, vol. 32, no. 8, pp. 742– 745, 2001.
10. T. J. Lovely and P. J. Jannetta, "Microvascular decompression for trigeminal neuralgia. Surgical technique and long-term results," Neurosurgery Clinics of North America, vol. 8, no. 1, pp. 11–29, 1997.
11. P. J. Hamlyn and T. T. King, "Neurovascular compression in trigeminal neuralgia: a clinical and anatomical study," Journal of Neurosurgery, vol. 76, no. 6, pp. 948–954, 1992.
12. K. J. Park, D. Kondziolka, O. Berkowitz et al., "Repeat gamma knife radiosurgery for trigeminal neuralgia," Neurosurgery, vol. 116, pp. 73–81, 2011.
13. S. Passero and G. Filosomi, "Posterior circulation infarcts in patients with vertebrobasilar dolichoectasia," Stroke, vol. 29, no. 3, pp. 653–659, 1998.
14. Deshmukh VR, Hott JS, Tabrizi P, Nakaji P, Feiz-Erfan I, Spetzler R. Cavernous malformation of the trigeminal nerve manifesting with trigeminal neuralgia: case report. Neurosurgery 2005;56:E623.
15. Du R, Binder DK, Halbach V, Fischbein N, Barbaro N. Trigeminal neuralgia in a patient with a dural arteriovenous fistula in Meckel's cave: case report. Neurosurgery 2003; 53:216-221.
16. Barker FGII, Jannetta PJ, Bissonette DJ, Larkins MV, Jho HD. The longterm outcome of microvascular decompression for trigeminal neuralgia. N Engl J Med 1996; 334:1077-1083.
17. Morita A, Fukushima T, Miyazaki S, Shimizu T, Atsuchi M. Tic doul o u reux caused by primitive trigeminal artery or itsvariant. J Neuro s u rg 1989;70:415-419.
18. Kirsch E, Hausmann O, Kaim A, Gratzl O, Steinbrich W, Radu EW. Magnetic resonance imaging of vertebrobasilar ectasia in trigeminal neuralgia.Acta Neurochir (Wien) 1996;138:1295-1299.