CASE REPORT

Atypical hemifacial spasm due to vertebral artery dolichoectasia: rare cause of a rarer clinical entity

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SUMMARY

A 32-year-old man presented with atypical left hemifacial spasm (HFS) with MRI showing compression of left facial nerve at the root exit zone. HFS rarely presents atypically with onset in orbicularis oris and later spreading upward to involve the orbicularis oculi as was seen in our case. Atypical presentation of hemifacial spasm is rare and vertebral artery dolichoectasia as the underlying aetiology, like in our case is even rarer and has not been described in literature so far.

BACKGROUND

Hemifacial spasm (HFS) is a movement disorder characterised by involuntary paroxysmal facial movements that usually involve the orbicularis oculi and then spread to the other facial muscles over months to years. In cases of atypical HFS, spasm starts with the orbicularis oris and buccinator muscles, and gradually spread upward to involve the orbicularis oculi muscle, reverse of what is seen in cases of typical HFS. Such atypical presentation of hemifacial spasm is rare and even rarer is vertebral artery dolichoectasia as the underlying aetiology.

CASE PRESENTATION

A 32-year-old man presented with 1.5-month history of painless intermittent twitching of the left side of the face which started with deviation of left angle of mouth and over a period of 25 days extended to involve left eye and upper chick. He had no significant medical or family history. Neurological examination revealed ipsilateral,

involuntary, paroxysmal contraction of the facial musculature, particularly provoked during activity (speech, blinking and so forth), consistent with hemifacial spasm.

The ophthalmological evaluation was normal and hearing was intact. Corneal and facial sensations were bilaterally normal. There was no facial weakness. No long tract signs were observed.

INVESTIGATIONS

Three Tesla MRI of the brain with focused cuts of brainstem revealed the presence of a dolichoectatic left vertebral artery impinging on the root exit zone of the left facial nerve (figure 1).

TREATMENT

Patient was managed conservatively with carbamazepine. Initially there was some symptomatic relief but again few days later symptoms recurred. The patient was later referred to the neurosurgeon.

OUTCOME AND FOLLOW-UP

Patient was later lost to follow-up.

DISCUSSION

HFS is a complex symptom comprising involuntary, painless spasms of the orbicularis muscle that may progress to involve all facial muscles. It is frequently the result of compression of the facial nerve at its root exit zone from the brain stem by vascular loops or aneurysms of posterior inferior cerebellar artery, anterior inferior cerebellar artery or vertebral artery.¹

Figure 1 (A) Three Tesla MRI of the brain showing left vertebral artery (arrow) impinging on the left facial nerve at the root exit zone. (B) CT angiogram of the same patient showing dilated and elongated left vertebral artery.

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Atypical hemifacial spasm, as was seen in our case, is rare. Very few cases of HFS caused by vertebral dolichoectasia are found in medical literature. In a series of 155 cases of HFS, the authors found only two cases of atypical HFS in which spasm started with the orbicularis oris and buccinator muscles and gradually spread upward to involve the orbicularis oculi muscle.² Cases of atypical hemifacial spasm due to vertebral artery dolichoectasia are even rarer and to the best of our knowledge have not been described so far. MRI not only excludes other aetiologies such as tumour or arteriovenous malformation, but also demonstrates cranial nerve compression by ectatic vertebral and basilar arteries.³

In conclusion, diagnosis of HFS requires MRI of the brain and MR angiography. Neurovascular contact is the commonest cause of HFS, and vertebrobasilar dolichoectasia is a very rare form. Ipsilateral dolichoectasia with HFS indicates existence of a direct neurovascular contact, clearly shown by MRI and MR angiography. Microsurgical vascular decompression is the curative treatment of choice, and is effective in 70–95% of patients.⁴

Learning points

- ▶ Atypical presentation of hemifacial spasm is rare and due to vertebral artery dolichoectasia has not been reported so far.
- ▶ MRI of the brain not only excludes other aetiologies but also demonstrates cranial nerve compression by any vascular loop.
- ▶ Microvascular decompression in the hands of experienced neurosurgeon is the treatment of choice.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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