CASE REPORT

Trigemino-cardiac reflex caused by selective angiography of the middle meningeal artery

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SUMMARY

We describe an interesting case of trigemino-cardiac reflex (TCR) caused by selective angiography of the middle meningeal artery (MMA). A 28-year-old woman presented with a symptomatic meningioma. Preoperative tumour embolisation was performed. In the procedure, when selective MMA angiography was done with Omnipaque 300 mg I/mL for 3 mL by manual injection, the patient complained of flashing lights in her eye followed by vomiting and bradycardia down to 40 bpm without increased intracranial pressure signs. On selective MMA angiography, the choroidal crescent and arteries of the periorbital region were opacified by anastomosis from the MMA via the meningo-ophthalmic artery. We diagnosed that her symptoms were caused by selective MMA angiography leading to high pressure stimulation towards the ophthalmic nerve innervation around the orbit as a TCR. We suggest that the operator should be prepared to manage TCR during treatment with expected selective MMA angiography, and gentle low pressure contrast injection should be attempted.

BACKGROUND

Owing to continued developments in devices used in neurointerventional surgery, the limits of neurointerventional treatment have been progressively improved. Accordingly, meticulous delineation of cranial fine vessels has been required for appropriate treatment. Similarly, selective angiography focusing on small vessels is increasing. In particular, in the preoperative embolisation of meningioma, selective fine angiography and procedures in the dural artery are necessary. In the skull base area trigeminal nerve innervation, care is needed because of the possibility of trigemino-cardiac reflex (TCR). In some cases TCR may develop into severe bradycardia, or even asystole and arterial hypotension which require intervention. We describe a TCR caused by selective angiography of the middle meningeal artery (MMA).

CASE PRESENTATION

A 28-year-old woman presented with morning headaches. Her MRI showed a suboccipital giant tumour that enhanced homogeneously. The brainstem was compressed by the tumour. From the imaging investigations a peritumoral giant meningioma was suspected. Preoperative tumour embolisation was scheduled for the day prior to planned tumour excision.

TREATMENT

The procedure took place under local anaesthesia with 1% lidocaine at the site of the punctured common femoral artery. A guiding catheter, 5 Fr 90 cm Flexor Shuttle Guiding Sheath (Cook Medical, Bloomington, Indiana, USA), was introduced into the left external carotid artery. Diagnostic angiography showed an occipital branch from the left MMA and a meningeal branch from the left occipital artery as feeding arteries to the tumour. To increase deliverability and navigability of the microcatheter, a 4 Fr 125 cm Cerulean G (Medikit, Tokyo, Japan) was introduced into the MMA to the level of the foramen spinosum. Selective angiography from this Cerulean G microcatheter was done with non-ionic contrast (Omnipaque 300 mg I/mL) for 3 mL by manual injection. Immediately following the injection the patient experienced vomiting and bradycardia down to 40 bpm without headache, altered consciousness or increased intracranial pressure signs. The patient's vomiting and bradycardia disappeared quickly and did not recur for the left-sided manipulations. An Exelsior 1018 microcatheter (Stryker, Kalamazoo, Michigan, USA) was therefore inserted into the occipital branch from the left MMA as close as possible to the tumour. Intratumour embolisation was then performed with particles (Embosphere 300–500 µm; Merit Medical Systems, South Jordan, Utah, USA). Next, a Marathon microcatheter (Covidien, Dublin, Ireland) was inserted into the meningeal branch from the left occipital artery as close as possible to the tumour. Sufficient intratumoural embolisation at the left side of the tumour was then achieved with liquid embolic material (17% n-butyl cyanoacrylate).

To approach the right side of the tumor, the 5 Fr 90 cm Flexor Shuttle Guiding Sheath was introduced into the right external carotid artery. Diagnostic angiography showed an occipital branch from the right MMA and a meningeal branch from the right occipital artery as feeding arteries of the tumour. Similar to the left, a 4 Fr 125 cm Cerulean G was introduced into the right MMA up to the level of the foramen spinosum. Selective angiography from this Cerulean G was again performed with Omnipaque 300 mg I/mL for 3 mL by manual injection. Again, immediate but temporary vomiting and bradycardia occurred. This time the patient also complained of flashing lights in her right eye. We noted that her choroidal crescent and arteries of the periorbital region were opacified by

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Figure 1 Selective middle meningeal artery (MMA) angiography from a 4Fr Cerulean G at the level of the foramen spinosum (ellipse). The choroidal crescent and arteries of the periorbital region are opacified by an anastomosis from the MMA via the meningo-ophthalmic artery (black arrow). Simultaneously, tumour opacification from an MMA occipital branch (white arrow) is seen. (A) Right MMA angiography, anteroposterior view. (B) Left MMA angiography, anteroposterior view. (C) Right MMA angiography, lateral view.

anastomosis from the MMA via the meningo-ophthalmic artery bilaterally (figure 1). We diagnosed that her paroxysmal vomiting and bradycardia was a TCR caused by selective MMA angiography leading to stimulation of the ophthalmic nerve innervation around the orbit. We refrained from additional selective MMA angiography. Without recurrence of the symptoms, sufficient embolisation of the right side of the tumour was achieved in a similar fashion to the left side.

OUTCOME AND FOLLOW-UP

Following embolisation, gadolinium-DTPA-enhanced MRI showed disappearance of enhancement of most of the tumour (figure 2). No lasting orbital or meningeal injury was seen. The next day the tumour was completely resected with minimal bleeding and no neurological deficits.

DISCUSSION

The clinical features of TCR include sudden onset of parasympathetic dysrhythmia, sympathetic hypotension, apnoea and gastric hypermotility. The physiological mechanism of the TCR is elucidated as follows. As the afferent pathway of the reflex arc, neuronal signals are conducted from the sensory nerve endings of the trigeminal nerve via the Gasserian ganglion to the sensory nucleus of the trigeminal nerve. In the reticular formation of the brainstem, via the short internuncial nerve fibres,

these neuronal signals are sent to the efferent pathway in the motor nucleus of the vagus nerve.¹ Therefore, TCR triggered during the procedure at the site of trigeminal nerve innervation has been reported. The oculocardiac reflex, which is a physiological subtype of TCR, was triggered by mechanical stimulation of ocular and periocular structures that are innervated by the ophthalmic division of the trigeminal nerve. In addition, cranio-facial surgery,² tumour resection in the cerebellopontine angle³ or trans-sphenoidal adenomectomy⁴ have the potential to cause a TCR because of innervation by the maxillary and mandibular divisions and the root of the trigeminal nerve. Current developments in devices used in neurointerventional surgery have improved the limits of treatment. With this progression, TCR has recently been seen during neurointerventional procedures in the skull base region. For instance, TCR was reported during embolisation of a dural arteriovenous fistula with Onyx and embolisation of a juvenile nasopharyngeal angiofibroma with Onyx.^{5–7}

To the best of our knowledge, TCR occurring during selective MMA angiography, such as in our case, has not previously been reported. In the current case, a robust anastomosis between the MMA and the orbital arteries was demonstrated. In cadaveric specimens the orbital branch of the MMA has been reported in 57.4–64.9%.^{8–9} The angiographic appearance along with our patient's complaint of flashing lights led us to believe that the

Figure 2 Gadolinium-DTPA enhanced MRI demonstrates the tumour and the disappearance of most of the enhancement after embolisation. Left, before embolisation. Right, after embolisation.

TCR was caused by high pressure stimulation from manual injection of contrast into the orbital arteries that interacted with ophthalmic nerve innervations. The reflex might have been mediated by nerve connections between the ophthalmic division of the trigeminal cranial nerve via the ciliary ganglion and the vagus nerve of the parasympathetic nervous system (figure 3).¹⁰ An alternative explanation is manipulation of the meningeal branch of the mandibular nerve in the foramen spinosum, but

this is less likely as we have never seen this with any prior MMA access.

For the management of TCR, risk factor identification and modification are important. The risk factors already known include hypercapnia, hypoxaemia, light general anaesthesia, age (more pronounced in children), the nature of the provoking stimulus and drugs. Drugs known to increase TCR include potent narcotic agents (eg, sufentanil and alfentanil), β -blockers and calcium channel blockers. When a TCR occurs, manipulation should be stopped and, if needed, vagolytic agents (atropine) should be administered followed by adrenaline (epinephrine) if no resolution occurs. Fortunately, in our case the symptoms were temporary but, in some cases, a TCR may develop into severe bradycardia or even asystole and arterial hypotension which require intervention. Coronary artery spasms provoked by TCR have been reviewed.¹¹ Care should be taken with ST changes, ventricular fibrillation, ventricular tachycardia, atrioventricular block and myocardial infarction as accompanying manifestations of TCR. We suggest that the operator should be prepared to manage a TCR during treatment involving expected selective MMA angiography, and permissible and gentle low pressure contrast injection should be attempted.

Figure 3 Drawing illustrating the pathway of the trigeminocardiac reflex (TCR) speculated in the current case. A, long ciliary nerve; B, short ciliary nerve; C, ciliary ganglion; D, ophthalmic nerve; E, maxillary nerve; F, pterygopalatine ganglion; G, mandibular nerve; H, otic ganglion; 1, Gasserian ganglion; 2, trigeminal nerve; 3, sensory nucleus of the trigeminal nerve; 4, short internuclear fibres; 5, motor nucleus of the vagus nerve; 6, vagus nerve. Red ellipse represents the foramen spinosum. Red solid lines represent the external carotid artery, middle meningeal artery and orbital arteries. Red dashed line represents the meningo-ophthalmic artery. Yellow solid line represents the 4Fr 125 cm Cerulean G. Adapted from Lang *et al.*¹⁰

Learning points

- ▶ Trigemino-cardiac reflex (TCR) may occur during selective middle meningeal artery (MMA) angiography.
- ▶ At selective MMA angiography, permissible and gentle low pressure contrast injection should be attempted.
- ▶ The operator should be prepared to manage a TCR during treatment involving expected selective MMA angiography.

Contributors TT, DER, MGM, ASP, MJG and AKW critically reviewed the study proposal.

Competing interests None declared.

Patient consent Obtained.

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Data sharing statement We agree with data sharing.

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