

Trigemino-cardiac Reflex as a Complication of Excision of Schwannoma of the Trigeminal Nerve - A Rare Clinical Case Report

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Abstract

Rationale: Trigeminal schwannoma (TS) is a very rare tumour in the head-and-neck region and the occurrence of intraoperative trigemino-cardiac reflex (TCR), during surgery, should not be underestimated. The physiological function of this rare brainstem reflex is yet to be fully established. **Patient Concerns:** TCR occurs in a variety of surgeries, such as neurosurgical, maxillofacial, dental and skull base surgeries with bradycardia as the presenting sign. **Diagnosis:** This is a clinical profile of two patients who presented with schwannoma of the trigeminal nerve. **Outcomes:** Intraoperatively, during dissection of tumour, both patients had bradycardia with hypotension. **Treatment:** The first patient had a spontaneous recovery but the other required intervention with vasopressors. **Take-Away Lessons:** One must be aware of the rare occurrence of TCR mainly while operating on a rare occurring TS. Ceaseless intraoperative monitoring and being prepared with adequate measures while manipulating close to the nerve will prevent serious complications.

Keywords: Bradycardia, schwannoma, trigeminal schwannoma, trigemino-cardiac reflex, vagal reflex

INTRODUCTION

Trigemino-cardiac reflex (TCR) is a reflex response of hypotension, bradycardia and gastric hypermotility which occurs due to mechanical stimulation of the trigeminal nerve (TN) during surgery. It is defined as a drop of >20% in mean arterial blood pressure (MABP) and heart rate (HR) <60 beats/min after stimulation of the TN.^[1]

TCR is reported to occur, especially in maxillofacial and craniofacial surgeries during the manipulation of the TN on cerebellopontine angle, cavernous sinus and peripheral branches of the TN.^[2]

The occurrence of schwannoma in the head and neck is a rare event and constitutes 25%–45% of all reported schwannomas.^[3] Although the lateral neck is the most common site, facial involvement is, even more, rarer, which usually arises from the TN root or any one of the three peripheral branches of the TN.^[4] The occurrence of TCR due to the manipulation of peripheral branches of trigeminal schwannoma (TS) has not yet been reported, to the best of our knowledge.

In the present study, we report our experience and management of TCR in two patients who underwent surgical excision for TS. The existence of this rare reflex in TS, which is also a very rare tumour makes this case report a distinctive clinical entity.

CASE REPORTS

Case report 1

A 31-year-old female with no co-morbidities presented with a progressive swelling in the right cheek for the past two years, associated with tingling sensation in the right side of the face.

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A magnetic resonance imaging (MRI) scan was consistent with peripheral nerve sheath tumour involving the maxillary division of the right TN [Figure 1].

She underwent excision of the tumour through the Caldwell-Luc approach under general anaesthesia (GA).

During dissection of the tumour, there was significant bradycardia (>40% fall), hypotension (>60% fall) and fall in mean arterial pressure [Figure 2]. The surgery was withheld for a few minutes, following which the parameters returned to normal without intervention [Figure 3]. The histopathological examination report revealed a benign nerve sheath tumour suggestive of schwannoma.

No other intraoperative and post-operative complications were noted and no recurrence was noted in a follow-up of two years.

Case report 2

A 54-year-old female with no co-morbidities presented with a gradually progressive left cheek swelling for the past four years with loosening of the left upper premolar teeth.

MRI showed a well-delineated lobulated spindle-shaped lesion measuring epicentred along the course of the left infraorbital nerve [Figure 4].

The tumour was approached by sublabial incision and surgical excision was done under GA.

During excision, significant bradycardia (>50% fall) and hypotension (>60% fall) were noted in spite of the instillation

of local anaesthesia to the nerve [Figure 2]. The surgery was temporarily withheld, and vasopressors were administered following which the HR returned to normal limits [Figure 3]. Surgery was resumed and no other significant intraoperative and post-operative complications were experienced.

The patient is disease-free for a follow-up of 1 year.

DISCUSSION

TCR occurs due to intraoperative surgical stimulation of the TN by physical or electrical stimuli.^[5] Although TCR was witnessed in 1870 by Kratschmer, it was explained as trigeminal depression response only in 1975.^[2]

The possible reflex arc of TCR would be: from the maxillary division of TN, impulses are sent to the Gasserian ganglion and then to the sensory nucleus of the TN, which constitutes the afferent pathway. These fibres reach the reticular formation and then to the motor nucleus of the vagus nerve (VN) from which it is carried by VN to the myocardium. The end result of stimulation would be sudden-onset bradycardia, asystole and hypotension.^[6]

TCR is classified into central, which is activated by stimulus proximal to the Gasserian ganglion in the intracranial course, and peripheral TCR, which includes stimulus distal to the ganglion.^[7] Both patients in the present study belonged to the peripheral type of TCR.

Meuwly *et al.* observed that deceleration of HR is a more constant variable than MABP for its diagnosis. Furthermore, the anatomical location of stimulation of the nerve has an

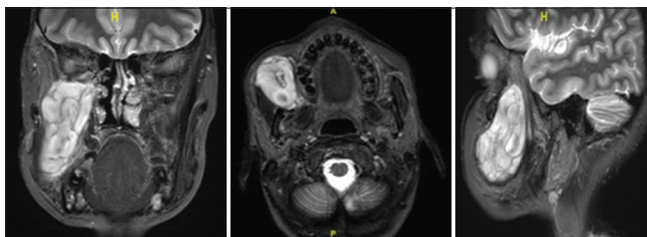


Figure 1: MRI of patient 1, showing well-defined soft-tissue density lesion in the right buccal space, extending into ipsilateral infratemporal fossa, pterygomaxillary fissure and pterygopalatine fossa with widening of the right vidian canal and foramen rotundum

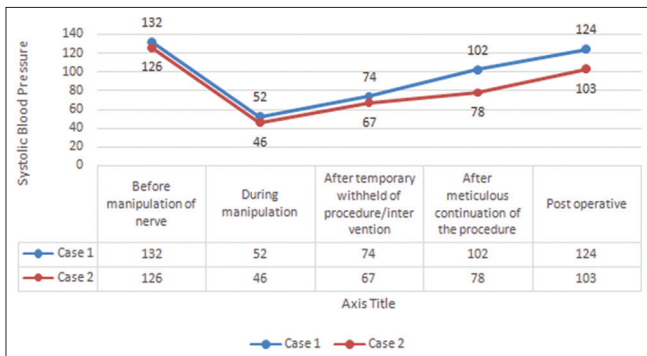


Figure 3: Trend in changes in systolic blood pressure at various phases of the procedure

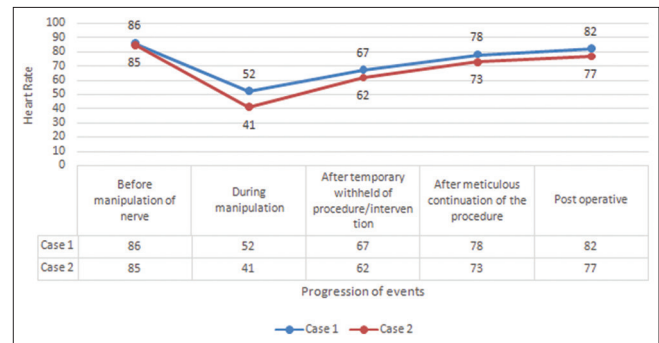


Figure 2: Trend in changes in heart rate at various phases of the procedure

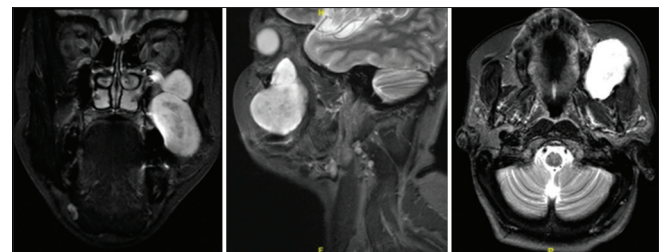


Figure 4: MRI of patient 2, showing a well-delineated lobulated spindle-shaped lesion epicentred along the course of the left infraorbital nerve, which extends from the pterygopalatine fossa

effect on the variation of MABP.^[8] In both our patients, a drop of >20% in MABP and HR was observed.

Predisposing factors for increased risk of TCR include hypoxemia, cardiac risk, insufficient depth of anaesthesia, high sympathetic activity, hypercapnia and pharmacology agents such as beta-blockers and calcium channel blockers.^[9] Occasionally, TCR may present as a fatal sudden cardiac arrest, without prior warning signal of bradycardia or hypotension.^[9]

Prophylactic use of atropine is advocated in some studies. Effective communication between surgeon and the anaesthetist plays a vital role in TCR management.^[9] In our study, although no prophylactic anaesthetic medication had been used, TCR was managed successfully by early recognition of bradycardia due to constructive knowledge both by the anaesthetic and surgical teams.

Strabismus surgery has the highest incidence of TCR (32%–90%) followed by blepharoplasty (25%).^[9] Lübbers *et al.* proposed orbital exenteration, orbital floor fractures, strabismus as high-risk surgeries and zygomatic/temporomandibular joint surgeries, Le Fort I osteotomies as low-risk surgeries in encountering TCR.^[9] Precious and Skulsky reported the incidence of bradycardia to be 1.6% during surgery for maxillofacial trauma, in which two patients (2/11) developed asystole, requiring atropine.^[1] Joshi *et al.* also reported the incidence of TCR is highest (91.3%) during the reduction of midfacial fractures.^[6] In the present study, the first patient had a spontaneous recovery by temporarily pausing the manipulation, whereas the other required intervention in the form of vasopressors. Surgery was resumed successfully by gentle manipulation and no further complications were experienced.

Management of TCR as mentioned by Arasho *et al.*^[10] includes prompt identification of risk factors, use of prophylactic vagolytic drugs during nerve manipulation and continuous intraoperative monitoring.

Although there are a handful of case reports reporting central TCR, the strength of this report rests on the fact that this is one of the few which elaborates the phenomenon of peripheral TCR (manipulation of trigeminal nerve) and to add to its uniqueness during the surgical excision in a rare tumour of TS.

The limitation of this study is the diagnosis of TCR was based on haemodynamic variations (HR and MABP). Even though the clinical reliability of this variation is validated as per previous literature, a more precise diagnostic tool to efficiently document this reflex is yet to be identified.

The basic definition of TCR, which is haemodynamic alteration with 20% drop of HR and MABP, is still a debate.^[5] Subtle TCR reflexes, which do not meet this drop, must be considered. This report will serve its importance in classifying the subtypes of TCR based on the trigger point, especially on the peripheral

TN. Future studies focusing on the peripheral TCR due to manipulation of the TN will have more insights into this reflex.

CONCLUSION

On operation for peripheral TN schwannoma, it would be prudent to keep TCR in mind. An initial indication of TCR is bradycardia. Ceaseless intraoperative monitoring with adequate depth of anaesthesia, gentle manipulation of the nerve, timely diagnosis, especially while manipulating the nerve, and rapid intervention with vasopressors wherever necessary is the key measure in the management of TCR. In most cases, TCR resolves spontaneously without medical intervention.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patients have given their consent for all concerned images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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