

Crocodile Tear Syndrome Post Microvascular Decompression of the Trigeminal Nerve: A Case Report and Literature Review

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ABSTRACT

Background: Crocodile Tear Syndrome (CTS) is a condition characterised by excessive tear secretion in response to eating, drinking, or smelling foods. Traditionally, acquired cases are most commonly reported following facial nerve trauma or paralysis, or in slow-growing facial nerve tumours. More recently, it has been reported following vestibular Schwannoma surgery. We report the first case of crocodile tear syndrome following microvascular decompression of the trigeminal nerve. **Case Presentation:** A 61-year-old lady presented with excessive lacrimation and clear rhinorrhoea one month post-operatively from a re-do trigeminal microvascular decompression (MVD) surgery. The patient experienced similar symptoms following her initial surgery two years prior, which had resolved spontaneously. CT and MRI head, and comprehensive clinical examination showed no evidence of CSF leak from her retromastoid wound site. An ENT opinion was sought, and the patient was diagnosed with post-operative crocodile tear syndrome. Surgical technique and relevant imaging were reviewed for any possible explanation for the condition. We discuss the possible aetio-pathogenesis for the development of the condition following MVD procedure. **Conclusion:** We conclude that CTS should be considered in patients presenting with CSF rhinorrhoea following MVD of Trigeminal nerve after excluding CSF leak from the middle ear via eustachian tube. Treatment for CTS in this context may pose a challenge. The patient has undergone botulinum toxin injection of the lacrimal gland and will likely need long term follow up. This is the first documented case of CTS post microvascular decompression of the trigeminal nerve.

Keywords: Crocodile tear syndrome, Microvascular decompression, Trigeminal nerve/neuralgia, Teflon sponge.

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INTRODUCTION

Crocodile Tear Syndrome (CTS) is a condition of gustatory lacrimation - an excessive secretion of tears as a result of eating, drinking, or smelling foods.¹ Previous reports most commonly associate this condition with recovery following facial nerve trauma or palsy.¹ Sporadic cases have been reported in the context of slow-growing tumours affecting the facial nerve, directly or indirectly.² Congenitally, it is been associated with Duane's retraction syndrome.² We describe a case of iatrogenic CTS in a patient following Microvascular Decompression (MVD) of the trigeminal nerve for Trigeminal Neuralgia (TN).³⁻⁵ The possible aetio-pathogenesis including a review of literature with anatomical and surgical factors are discussed in this report.

CASE DESCRIPTION

A fit and well 61-year-old lady presented with a history of excessive right sided lacrimation and rhinorrhoea whilst eating, two weeks following revision right-sided MVD for recurrent classic type 1 TN. The patient had experienced transient gustatory lacrimation, along with transient sensorineural hearing loss following the initial surgery two years prior. The excessive lacrimation was not brought to our attention at the time, and the patient recovered well from all post-operative symptoms without any long-lasting side effects.

At this presentation, the patient was investigated with CT and MRI brain to rule out CSF rhinorrhoea. There was no evidence of breach of mastoid air cells to support this diagnosis. Comprehensive cranial nerve examination and imaging scans showed no specific cause of her symptoms. She had developed a temporary sensory neural hearing loss following the surgery. An opinion was sought from otolaryngology for the hearing loss and continued clear nasal discharge. This shed light on an interesting case of 'Crocodile Tear Syndrome' (CTS).



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In this case, CTS has been diagnosed following MVD of the trigeminal nerve. Subsequently, an intra palpebral botulinum toxin injection was recommended by the Ophthalmology team for the management of CTS.

DISCUSSION

Microvascular decompression of the trigeminal nerve is a neurosurgical operation in which a blood vessel compressing the trigeminal nerve is mobilised; and a Teflon sponge inserted between the two: decompressing the nerve, and alleviating neuralgia.^{6,7} A few neurosurgical practices now advocate the use of fibrin glue to maintain the Teflon sponge's position.^{8,9} The technique is not widely practiced, owing to the belief that adequate Teflon sponge adhesion is generally achieved due to the nature of Teflon fibres.⁹ Increasing evidence in the literature suggests fibrin glue successfully prevents Teflon sponge migration and subsequent granuloma formation, a known cause of recurrent trigeminal neuralgia and hemi-facial spasm.⁸

Review of initial procedure

A draining vein was found to be splitting the sensory division of the trigeminal nerve in two. It was dissected clear off the nerve. Additionally, an arterial vascular loop was found adhering to the trigeminal nerve, stuck with extensive arachnoid adhesions. This was also dissected, and a Teflon sponge was inserted around the vein and arterial loop, decompressing the nerve. Tisseel™ fibrin glue drops were used to keep the Teflon sponge in place. The 7th and 8th nerve complex were noted to be clear, and standard closure was performed. Bone dust collected from the craniectomy was replaced with Tisseel™ Fibrin glue to perform a water tight closure.¹¹

MRI FIESTA sequencing of the brain during the current presentation revealed the absence of a Teflon sponge near the trigeminal nerve with granulomatous appearances close to the 7th/8th nerve complex. This was suggestive of a dislodged Teflon sponge.

Review of re-do procedure

Significant arachnoid adhesions were seen around the trigeminal nerve. The Teflon sponge from the initial procedure was not visualised, and is likely displaced. A new vein cluster was found adjacent to the 7th/8th nerve complex. This was not explored. The arachnoid adhesions were dissected, and a new Teflon sponge was inserted between the vein and nerve complex, and Tisseel™ was used. Fibrin glue was utilised in this patient for both procedures. Unfortunately, the sponge dislodged following the initial operation regardless. Dural closure was performed with 5'0 prolene and the bone fragments collected from the redo craniectomy was replaced with Tisseel™ Fibrin glue to perform a water tight closure similar to the initial procedure.¹¹

In the immediate post-operative period, the patient experienced transient dysphagia-like symptoms which spontaneously resolved. She remains free of TN symptoms following the second surgery, and is currently off all medications for TN. Her sensorineural hearing loss had improved at a 6 month follow-up. In this context, we discuss the relevant neuroanatomy and physiology of the trigeminal nerve and facial-auditory nerve complex. Figure 1 summarises the sequence of clinical events in chronological order. Figure 2A describes the T2 weighted sequence MRI brain at the level of Internal Auditory meatus Post MVD and Figure 2B shows the preoperative appearance of the 7 and 8 cranial nerve complex.

Relevant neuroanatomy of the trigeminal nerve and facial-auditory nerve complex

The facial nerve (CN VII) consists of sensory, motor, and parasympathetic nerve fibres, the latter of which originate at the superior salivatory and lacrimatory nuclei.^{3,4} Secreto-motor fibres originating at salivatory nuclei supply the sublingual and submandibular glands which, in normal physiology, stimulate saliva production in response to the gustatory reflex.³ Fibres originating at the lacrimatory nuclei supply the lacrimatory glands and stimulate tear production in response to sensory input from the conjunctiva and cornea, or complex emotional stimuli.⁵ Each of the nuclei supplies the ipsilateral side of the face.³⁻⁵

The trigeminal nerve consists of motor fibres stimulating the muscles of mastication and tensor tympani muscles of the ear, and sensory fibres which innervate the head and face.³ The trigeminal nerve has sympathetic innervation in its first two divisions; and parasympathetic innervation in the third.⁶ The neuro-vascular conflict culprit of trigeminal neuralgia at the Dorsal Root Entry Zone (DREZ) is usually caused by one of the following vascular structures: the superior cerebellar artery, anterior inferior cerebellar artery, and the trigeminal & superior petrosal veins.⁶⁻⁸

2018	<p>Initial operation</p> <p>Microvascular decompression of the TN for trigeminal neuralgia. The patient experienced transient gustatory lacrimation and sensorineural hearing loss following this procedure. This was not brought to our attention at the time</p>
2019	<p>Recurrence of trigeminal neuralgia</p> <p>MRI Brain shows a misplaced Teflon sponge near the trigeminal nerve with granulomatous appearances by the 7th/8th nerve complex</p>
October 2020	<p>Re-do operation</p> <p>Re-do microvascular decompression of the TN for recurrent trigeminal neuralgia</p>
November 2020	<p>Post-op review</p> <p>The patient presented with excessive lacrimation and clear rhinorrhoea ipsilateral to operative side. CT and MRI brain showed no evidence of CSF rhinorrhoea. Comprehensive cranial nerve examination and imaging scans showed no specific cause for this phenomenon. Her sensorineural hearing loss on the right had also recurred. ENT opinion was sought. This shed light on an interesting case of crocodile tear syndrome. Her trigeminal neuralgia has resolved, and she remains pain-free</p>
2021	<p>Treatment of CTS</p> <p>Referral for ophthalmology opinion. She is due trans-lacrimal botulinum toxin injection for treatment of her CTS</p>

Figure 1: Case description and timeline of events.

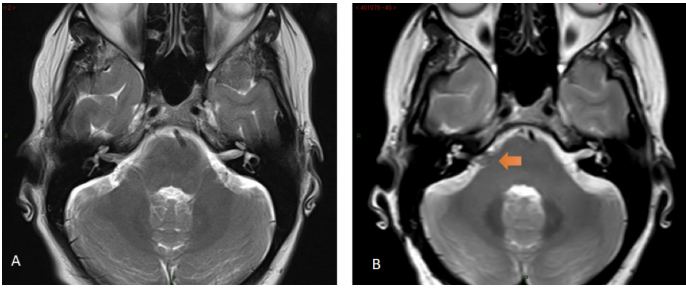


Figure 2: A. Axial MRI brain scan of patient at level of Internal Auditory Meatus (IAM). B. Right IAM prior to primary procedure.

Table 1: Suggested aetio-pathogenesis in discussed case.

Suggested Aetio-pathogenesis
Surgical manipulation.
Arachnoid granulations
Dislodged Teflon sponge
Vascular origin (new venous cluster noted during re-do MVD)
Idiopathic

It is understood that compression of the sensory division of the trigeminal nerve by one of these vessels gives rise to the classical symptoms of TN.^{7,8}

Pathogenesis of CTS

The most widely accepted theory behind the pathogenesis of crocodile tear syndrome attributes the condition to misdirected regeneration of gustatory fibres destined from the facial nerve to the salivary glands, to the lacrimal glands of the ipsilateral eye. As a consequence, the stimulation of the gustatory reflex stimulates lacrimation.² The aetiology of acquired CTS is usually associated with facial nerve paralysis secondary to trauma or an idiopathic Bell's palsy.^{1,2} Figure 3 describes the pathway.

Hypothesised aetio-pathogenesis in the case

In Table 1, we summarise the possible contributing factors for this case of CTS following MVD of the trigeminal nerve. Although the patient had no other manifestations of facial nerve trauma, surgical intervention in the skull base and ensuing granuloma formation on the 7th/8th nerve complex may have stimulated misdirected regeneration of facial nerve gustatory fibres to the patient's right lacrimal gland. Iatrogenic CTS following CN VIII surgery has also been reported, a structure also in close proximity to both the facial and trigeminal nerves.^{3,4,10} It is possible that this patient's CTS could be secondary to surgical intervention.

In our case, the patient had symptoms of facial and auditory nerve involvement post MVD in the form of sensorineural hearing loss and CTS. Sensorineural hearing loss is a well recognised complication in microvascular decompression of the trigeminal and facial nerve, and occurs in up to 5-8% of patients undergoing MVD.¹² We therefore suggest an inflammatory reaction of

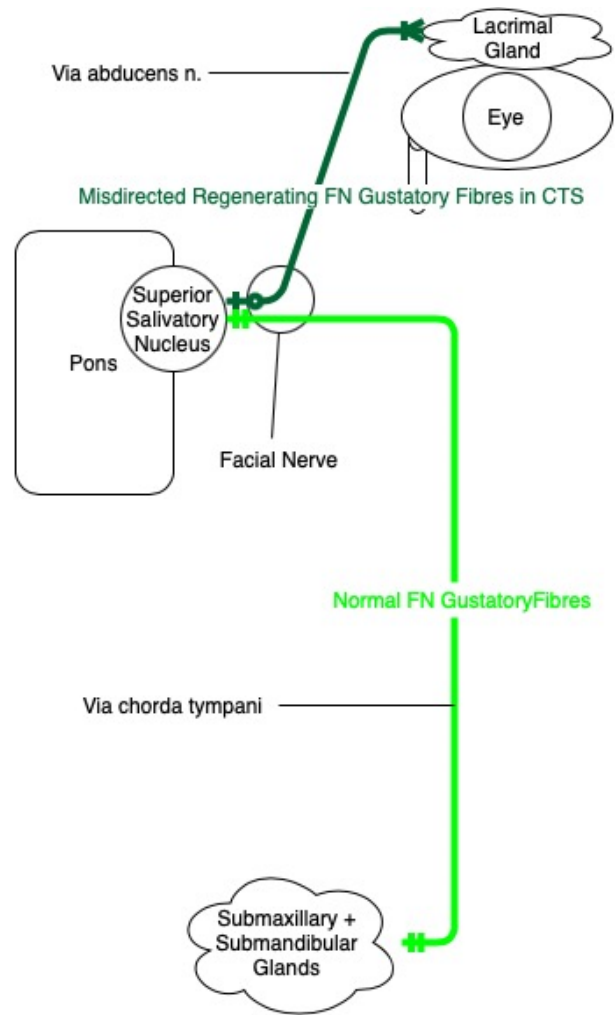


Figure 3: Schematic diagram of pathogenesis of CTS.

the 7th/8th nerve complex due to a dislodged Teflon sponge is a possible pathogenesis.

CONCLUSION

CTS is an under-reported condition, with poor pick-up rates.^{10,13} The discussed case highlights this, in that the patient had not presented following her initial operation, despite experiencing symptoms of mild CTS. There is no evidence in current literature which reports CTS following MVD of the trigeminal nerve.^{10,13}

The most widely accepted treatment option for CTS is lacrimal gland botulinum toxin injection.¹⁴ The toxin, an acetylcholine release inhibitor, acts at the neuromuscular junction by preventing parasympathetic stimulation of the lacrimal gland. It can be injected trans-cutaneously or into the palpebral part of the lacrimal gland.¹

The management of CTS depends on the severity of the case. In mild cases, it can be managed expectantly and with counselling after doing a Schirmer's test.¹⁵ In severe cases, subtotal resection of the palpebral part of the lacrimal gland may be offered. In our

case, Her sensorineural hearing loss had improved at a 9 month follow up.

We report the first case of CTS following MVD for TN. This may be a rare complication of the procedure. In patients presenting with clear rhinorrhoea post operatively, CTS should be considered as part of the differential diagnosis after ruling out a CSF leak.

CONFLICT OF INTEREST

The authors declare that there is no conflict of interest.

ABBREVIATIONS

MRI: Magnetic resonance imaging; **CT:** Computed tomography; **CTS:** Crocodile tear syndrome; **MVD:** Microvascular decompression; **TN:** Trigeminal neuralgia; **CSF:** Cerebrospinal fluid; **IAM:** Internal auditory meatus; **ENT:** Ear, Nose and Throat (Otolaryngology); **DREZ:** Dorsal root entry zone.

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