Crocodile tear syndrome post microvascular decompression of the trigeminal nerve: a case report and literature review

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Case report

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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 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 rhinorrhoea or cause of her symptoms. An ENT opinion was sought, and the patient was diagnosed with post-operative crocodile tear syndrome.

Literature review revealed no reported cases of CTS following microvascular decompression of the trigeminal nerve. Surgical technique and relevant imaging were reviewed for any possible explanation for the condition. Considering the accepted pathogenesis of CTS, we discuss the aetio-pathogenesis for the development of the condition following this procedure.

Conclusions

We conclude CTS should be considered in patients presenting with rhinorrhoea following microvascular decompression of the trigeminal nerve. In patients presenting with post-operative rhinorrhoea after MVD, after excluding CSF leak, CTS should be considered as a potential differential diagnosis. Treatment for CTS in this context may pose a challenge. The patient has undergone botulinum toxin injection of the lacrimal gland and will need long term follow up. This is the first documented case of CTS post microvascular decompression of the trigeminal nerve.

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). The possible aetio-pathogenesis is described in context of relevant anatomical and surgical factors, in addition to current literature.

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. Her right-sided sensorineural hearing loss had also recurred. 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).

In this case, CTS has been diagnosed following MVD of the trigeminal nerve. Following diagnosis, the patient was referred to ophthalmology for an opinion, who recommended CTS treatment with intra-palpebral botulinum toxin injection.

Fig. 1. Case description & timeline of events

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 [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. TisseelTM 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.

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.

Fig. 2. Axial MRI brain scan of patient at level of internal auditory meatus (IAM)

Fig. 2a. Right IAM prior to primary procedure

Fig. 2b. Right IAM following primary procedure. Granulation tissue highlighted with orange arrow.

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 TisseelTM was used. Fibrin glue was utilised in this patient for both procedures. Unfortunately, the sponge dislodged following the initial operation regardless.

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. In this context, we discuss the relevant neuroanatomy and physiology of the trigeminal nerve and facial-auditory 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 lacrimal 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 lacrimal nuclei supply the lacrimal 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 and superior petrosal veins [6-8]. 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.

Fig. 3. Schematic diagram of pathogenesis of CTS

Hypothesised aetio-pathogenesis in the case

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

Table 1. Suggested aetio-pathogenesis in discussed 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 the 7th/8th nerve complex due to a dislodged Teflon sponge is a possible pathogenesis.

Conclusions

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, the patient is due a Botox injection to the lacrimal gland, and remains pain free from her TN.

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.

Declarations

Ethics approval and consent to participate: not applicable

Patient consent for publication: obtained

Availability of data and materials: data sharing is not applicable to this article as no datasets were generated or analysed during the current study.

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Authors' information and contributions:

Dr Mohammad Abul, Senior House Officer: Manuscript write-up and submission

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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 & Throat (otolaryngology)

DREZ - dorsal root entry zone

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Figures

2018	Initial operation 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
2019	Recurrence of trigeminal neuralgia MRI Brain shows a misplaced Teflon sponge near the trigeminal nerve with granulomatous appearances by the 7th/8th nerve complex
October 2020	Re-do operation Re-do microvascular decompression of the TN for recurrent trigeminal neuralgia
November 2020	Post-op review 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
2021	Treatment of CTS Referral for ophthalmology opinion. She is due trans-lacrimal botulinum toxin injection for treatment of her CTS

Figure 1

Case description & timeline of events

Figure 2

Fig. 2. Axial MRI brain scan of patient at level of internal auditory meatus (IAM) Fig. 2a. Right IAM prior to primary procedure Fig. 2b. Right IAM following primary procedure. Granulation tissue highlighted with orange arrow.

Figure 3

Schematic diagram of pathogenesis of CTS