



Tearing with mastication in relation to facial paralysis: a case report of the crocodile tears syndrome

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ABSTRACT

Introduction: Abnormal lacrimation with mastication is an uncommon complaint, which may follow paralysis of the facial nerve of any cause. The mechanism behind this symptom is not clearly established, and there is no consensus about the best treatment option. This case highlights the role of family doctors in patient education and reassurance, fundamental for acceptance and management of the emotional impact associated.

Case report: Male, 56 years old. Presented to his family doctor in February 2020 complaining of crying every time he ate. This affected only the left eye and started a year before, while the patient was undergoing rehabilitation therapy for peripheral facial paralysis secondary to an acoustic neuroma removal surgery. Profuse nasal congestion affecting only the left nostril occurred simultaneously. The left side of the face showed a higher muscle tone than the right. Symptoms began concurrently with the recovery of motor function. At the follow-up neurosurgery consultation, the patient mentioned the abnormal tearing and was told that it was a sequela of his lesion which required no further investigation. The patient wanted to know more about this symptom and what could be done to solve it, so he consulted his family doctor. The diagnostic hypothesis was explained to the patient and treatment options were briefly approached. The patient felt that an invasive procedure was unnecessary and felt reassured by the diagnosis.

Comments: Even though lachrymation while eating is not so impactful as facial paralysis as a sequela of acoustic neuroma surgery, it is associated with a great emotional burden on patients, who frequently avoid social eating. Sufficient information should be provided to the patient, as it may be enough to give the patient relief, or, if treatment is needed, the patient should be correctly oriented.

Keywords: Tears; Adverse effects; Facial paralysis; Acoustic neuroma; Case report.

INTRODUCTION

Abnormal lacrimation with mastication is an uncommon complaint in the medical setting. According to Chorobski,¹ this phenomenon of paroxysmal lachrymation while eating, also known as the gusto-lachrymal reflex, was first noted by Oppenheim in 1913, even though Bogorad was the first to describe it in further detail and to term it Crocodile Tears syndrome, in 1928.²

This phenomenon of paroxysmal lachrymation while eating may follow paralysis of the facial nerve of any cause.¹ Most reported cases are due to Bell palsy, but it also happens after trauma, cancer, stroke, herpes zoster infection, neuro-sarcoidosis, or surgery (such as resection of the greater petrosal nerve, ear surgery, or acoustic neuroma resection),³⁻¹⁰ as well as a congenital form associated with Duane's syndrome.¹¹

Many patients complain of concomitant ipsilateral rhinorrhoea while eating, which may be secondary to the excessive tearing or result from abnormal secretion of nasal mucosal glands.^{5,12} Synkinesias of the facial muscles can also be present, which are considered to

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be due to abnormal regeneration of the motor fibres.⁵

Many mechanisms have been proposed to explain this phenomenon. The most accepted theory is that two different mechanisms coexist, depending on the timing of onset. Early-onset Crocodile Tears syndrome is thought to arise from transaxonal transmission of impulses, with a cross-stimulation between salivary and lachrymal fibres due to scar tissue.^{1,3,5,10,13} This means that impulses in salivary fibres reach the peripheral lachrymal fibres, thus inducing lachrymation.¹ On the other hand, late-onset Crocodile Tears syndrome appears to result from abnormal regeneration of the fibres after lesion, a process that takes several months to develop.^{3,10,12} After an injury, the secretory nerve fibres destined for the salivary glands regenerate in a false direction and reach the lachrymal gland, thus establishing a new reflex arc, beginning in the taste buds and ending in the lachrymal glands instead of in the salivary glands. As a result, gustatory stimuli that normally give rise to salivation are now producing lachrymation.^{1,5}

Regarding prognosis, recovery is rare, with only 6% of individuals stating that the symptom improved significantly or resolved completely.¹² However, one study reported a higher recovery incidence of 71%.³ The existing methods of treatment intend to abolish lachrymation either by removing the lachrymal gland or by blocking the nerve impulses to lachrymation.^{5,10} Botulinum toxin injection into the lacrimal gland is the most widely accepted treatment.¹⁰ There is no consensus about the best treatment option, regardless of its cause (Table 1).

Medical literature is scarce on this syndrome, and most articles are from many decades ago, even though an increasing number of cases are being reported. The reported case reflects the impact of a careful investigation of initially unexplained symptoms and correct and complete transmission of information to the patient. This case highlights the role of the family doctor in the education and reassurance of the patient regarding the symptoms, which was fundamental for patient acceptance and management of the emotional impact associated with the syndrome.

This case report follows the CARE guidelines.¹⁴

CASE REPORT

Male, 56 years old, caucasian, totally independent

on the Barthel Index for Activities of Daily Living (100 points). Married, with good family support, in the VI stage of Duvall's Family Life Cycle. Employed, working as a sales agent, of a high social class according to the Grafar Scale (8 points). Living in an urban area with all commodities, no animals in the house.

The patient's previous medical history includes an uncomplicated appendectomy in 2004 and a left acoustic neuroma diagnosed in 2018, treated with surgery followed by radiotherapy, with residual left peripheral facial paralysis and unilateral hearing impairment. The patient presents no tobacco use, alcohol abuse, or drug consumption. The patient does not take any medication on a regular basis. Family history includes mother and grandfather from the mother's side with colorectal cancer and father with hypertension and a stroke at age 55.

In February 2020, the patient presented to his family doctor complaining of crying every time he ate. This symptom affected only the left eye and had started over a year before, while the patient was undergoing rehabilitation therapy for his left peripheral facial paralysis. The patient reported no additional symptoms and did not have excessive tearing in the absence of food ingestion. Initially, the patient showed no concern for the symptom as he thought of it as a compensatory response from the eye after being excessively dry due to facial paralysis. However, the symptom did not improve over time, and the patient felt at that moment that it affected both his social life and his occupation as a salesman, especially in the context of meals with family, friends, customers, or associates, as it frequently elicited questions and comments.

After a deeper inquiry, it was ascertained that the excessive lachrymation never failed to appear when the patient ate and never occurred under other circumstances. With a latency of only a few seconds after starting to eat, tears would start overflowing the left eye and running down the cheek. All types of food caused lachrymation, sometimes with different degrees, but the patient did not correlate any specific food with more tear production. Simultaneously to the tearing, profuse nasal congestion would also occur, affecting only the left nostril. Movements of the facial muscles and mimicking mastication in the absence of food did not produce tears. Emotional and reflex lachrymation were

TABLE 1. Reported treatment methods to abolish the gusto-lachrymal reflex in the crocodile tears syndrome¹⁸

Author	Treatment Method	Outcomes
Boyer & Gardner (1949) ¹⁹	Section of the greater superficial petrosal nerve	Description of one case in which complete remission was obtained for at least 1 year; description of two cases who developed the syndrome after undergoing this procedure for another cause
	Section of the glossopharyngeal nerve	Description of two cases due to section of the greater superficial petrosal nerve who achieved complete remission
Golding-Wood (1963) ²⁰	Resection of the tympanic branch of the glossopharyngeal nerve	Description of three cases in which complete remission was achieved
Gottesfeld & Leavitt (1942) ²¹	Blockage of the postganglionic fibres of the sphenopalatine ganglion	Description of one case of 1-hour remission with cocaine injection of the posterior nares; description of one case of 4-month remission with alcohol injection in the sphenopalatine ganglion
Borojerdj et al. (1998) ²²	Injection of botulinum toxin into or near the lachrymal gland	Description of 10 cases of 6-month complete remission
Ho (2018) ⁶		Description of one case of 6-month complete remission
Hofmann (2000) ²³		Description of two cases of 4 to 5-month complete remission
Montoya et al. (2002) ²⁴		Description of one case of 6-month complete remission; description of three cases of partial remission for 3.5-6 months
Nava-Castañeda et al. (2006) ²⁵		Description of 15 cases of complete remission up to 6-months
Riemann et al. (1999) ⁸		Description of one case of 6-month complete remission
Savin (1939) ²⁶	Resection of the palpebral portion of the lachrymal gland	No case reports
Spiers (1970) ²⁷	Anticholinergic drugs to reduce lachrymal secretion	Eye-drops and oral anticholinergic drugs were not effective; local cholinergic blockade was effective but induced visual disturbances

normal. The taste was preserved. The patient reported occasional minor muscular spasms on the left side of the face, such as trembling of the left upper lid.

On physical examination, the patient was well nourished. Neurological examination revealed sensorineural deafness on the left side. Ocular motility was normal. The left side of the face showed a higher muscle tone than the right, with mild ptosis of the left eye,

and a deeper nasolabial fold on the left. Voluntary and involuntary movements of the left side of the face resulted in a slight contraction of all facial muscles on the same side. When the patient closed his eyes, the left side of the mouth was retracted. There were no changes in strength or sensibility in any other part of the body. The patient had been previously observed by a private ophthalmologist who detected no ocular



disturbances, Schirmer's test was negative, and the lachrymal duct showed no obstruction.

Through a deeper look into the chronological evolution of the symptoms, a relation appeared to have been found. In November 2017, the patient consulted a private otorhinolaryngologist due to complaints of fullness in the left ear, soon followed by vertigo and hearing loss on the left side. The symptoms did not respond to corticotherapy and an MRI was requested. The patient was diagnosed with an acoustic neuroma in January 2018. After the diagnosis, the patient was referred to a neurosurgery consultation and a partial resection of the acoustic neuroma was performed in February 2018. After the surgery, the patient reported paralysis of the left side of the face. The left eye was dry and did not fully close, so there was a need to keep the eye closed with a pad. Severe hearing loss on the left side remained. Since the lesion was not completely removed, the patient underwent stereotaxic radiotherapy for the remaining lesion, between April 2018 and June 2019. After several months of rehabilitation physiotherapy – the patient cannot precise but believes it happened at least six months after the surgery – power began to return to the left side of the face and the facial asymmetry became less prominent. Facial paralysis gradually recovered, leaving only a slight facial asymmetry. This was temporally related to the beginning of the tearing symptoms. At the one-year post-operation follow-up consultation with the neurosurgeon, the patient mentioned the abnormal tearing and was told that it was a sequela of his lesion which requested no further investigation. The patient wanted to know more about this symptom and what could be done to solve it, so he consulted his family doctor.

At the time of the first consultation with the family doctor, no explanation could be given to the patient as to what was causing his symptoms, but an agreement was made that further information would be pursued and a new consultation scheduled to discuss diagnostic and therapeutic options. After an extensive literature review on the subject, a diagnosis of Crocodile Tears syndrome or Bogorad syndrome seemed most likely. About a month after the first consultation, the patient returned for orientation. The symptoms remained identical. The diagnostic hypothesis was explained to the patient and treatment options were briefly approached.

The patient felt that an invasive procedure with uncertain results was unnecessary and felt reassured by the diagnosis, as now he had a valid explanation for his symptoms and knew nothing hazardous was causing the tearing. Upon realizing that the tearing would probably persist, the patient made the decision to adapt to the situation and opted to carry a handkerchief to use during meals to clean the tears. The patient opted for periodic monitoring of symptom evolution, being reassured of the possibility to pursue another treatment option if he later changed his mind.

COMMENTS

In the reported case, both the clinical presentation and the epidemiology are congruent with the diagnosis of Crocodile Tears syndrome, as the patient reported the phenomenon of paroxysmal lachrymation while eating, associated with concomitant ipsilateral rhinorrhoea while eating and synkinesias of the facial muscles, beginning around six months after a surgery that has been described as a cause for this syndrome. The acoustic neuroma could be a cause of the syndrome by itself, but in this patient, the temporal relation does not support this hypothesis, as the tearing and facial paralysis only appeared after the removal of the tumour. The radiotherapy could also be a cause for this syndrome, but it seemed less likely because, even though the tearing started after the radiotherapy, the facial paralysis, which is causally related to the abnormal tearing, developed after the surgery but before the radiotherapy.

Cases of Crocodile Tears syndrome after acoustic neuroma removal are being increasingly reported, with an estimated incidence between 9.5% and 44%.^{3,12,15} Approximately two-thirds of cases arise within six months after surgery (early-onset), and a second peak occurs approximately twelve months after surgery (late-onset).³

Studies on the impacts of acoustic neuroma removal surgery on the facial nerve have generally ignored the sensory component of the nerve, focusing mainly on facial nerve motor function.¹² This may result from the fact that facial paralysis is more obvious to the doctor and causes greater distress to the patient,^{12,15} but the occurrence of lachrymation while eating is also reported to have a great emotional impact on patients, who frequently avoid social eating.¹² Since it appears to be



more common than it was previously considered, all patients undergoing acoustic neuroma removal should be given sufficient information about this syndrome prior to surgery.^{3,12} Even after surgery and symptom development, sufficient information should be provided, as it may be enough to give the patient relief, making an invasive procedure unnecessary.¹⁶

Sometimes, patients present to the doctor with complaints for which no cause can be immediately found. These symptoms are often chronic and cause a high negative impact on quality of life. In these cases, perception of physician caring is associated with overall patient satisfaction,¹⁷ and this can help the patient accept their symptoms and provide them adequate relief. This deconstruction of the physiology behind the symptom may provide the patient with adequate relief regarding alternative diagnostic options that could have a worse prognosis, thus reducing the anxiety associated with fear of the unknown. By providing the patient with all relevant information regarding the prognosis and the existing methods of treatment, the patient felt empowered, and the feeling of control over his health provided him the relief to adapt and adjust to the symptom.

In conclusion, doctors should be aware of Crocodile Tears syndrome as a possible and fairly common complication of facial paralysis of any cause, in order to counsel the patient regarding its causes, potential treatments, and prognosis. Providing information is usually sufficient to give the patient relief, but if great emotional impact derives from the syndrome, invasive procedures can be tried, and the patient should be correctly oriented.

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AUTHORS' CONTRIBUTION.

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DECLARATIONS

This case report follows the CARE guidelines.

CONFLICTS OF INTEREST

The author declares not to have any conflict of interest.

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RESUMO

LACRIMEJO DURANTE A MASTIGAÇÃO ASSOCIADO A PARALISIA FACIAL: RELATO DE CASO DA SÍNDROMA DE LÁGRIMAS DE CROCODILO

Introdução: O lacrimejo anormal com a mastigação é uma queixa incomum, que pode surgir após uma paralisia do nervo facial de qualquer etiologia. O mecanismo por trás deste sintoma não está claramente estabelecido e não existe consenso quanto à melhor opção terapêutica. Este caso realça o papel do médico de família na educação e tranquilização do doente, fundamentais para a aceitação e controlo do impacto emocional associado.

Descrição do caso: Homem, 56 anos. Recorreu ao médico de família em fevereiro de 2020 por chorar quando comia. Afetava apenas o olho esquerdo e teria começado cerca de um ano antes, enquanto realizava reabilitação de paralisia facial periférica secundária a cirurgia por neurinoma do acústico. Simultaneamente apresentava congestão nasal profusa da narina esquerda. A hemiface esquerda apresentava tónus muscular superior à direita. Os sintomas iniciaram-se concomitantemente à recuperação da função motora. Na consulta de seguimento de neurocirurgia, o paciente mencionou o lacrimejo anormal e foi-lhe dito que era uma sequela sem necessidade de investigação adicional. O paciente queria saber mais sobre o seu sintoma e possíveis soluções, tendo consultado o seu médico de família. Após revisão da literatura, a hipótese diagnóstica foi encontrada e explicada ao utente e as opções terapêuticas foram abordadas. O paciente considerou desnecessário submeter-se a um procedimento invasivo com resultados incertos, tendo optado por ajustes no seu dia-a-dia. O esclarecimento do diagnóstico foi suficiente para reduzir significativamente a sua preocupação.

Comentário: Apesar de o lacrimejo com ingestão de alimentos não ser tão impactante como a paralisia facial, enquanto sequelas da cirurgia a neurinoma do acústico, associa-se a grande impacto emocional, com frequente evitamento de refeições sociais. Deve fornecer-se informação completa, pois poderá ser suficiente para proporcionar alívio ou, caso o paciente pretenda tratamento, permitir a sua orientação adequada.

Palavras-chave: Lágrimas; Efeitos adversos; Paralisia facial; Neurinoma do acústico; Caso clínico.