

Perioral gustatory sweating: case report

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Abstract

Objective: Presentation of a case of perioral Frey syndrome.

Design: Case report.

Subject: A 72-year-old woman with hyperhidrosis around the mouth and chin.

Results: This patient suffered from bilateral perioral gustatory sweating following a mandibular osteotomy; such a case has not previously been described. Possible pathophysiological hypotheses are discussed in relation to the anatomy and innervation of the salivary glands.

Conclusion: Perioral gustatory sweating is a rare complication of osteotomy.

Key words: Gustatory sweating; Frey Syndrome; Perioral; Hyperhidrosis

Introduction

Frey syndrome, also known as auriculotemporal syndrome, is a well-known complication of parotid surgery. Approximately 24 per cent of patients undergoing parotidectomy experience gustatory sweating, although the reported incidence varies greatly.¹ Frey syndrome appears following a latency period of one to 36 months (or longer) after surgery.^{2,3}

The aetiology of Frey syndrome is explained by 'aberrant nervous regeneration'. During parotidectomy, the post-ganglionic parasympathetic fibres, which arise from the auriculotemporal nerve and innervate the parotid gland, are frequently damaged. They may regenerate in an aberrant way and connect with sympathetic nerve fibres. This can lead to parasympathetic innervation of sweat glands and small blood vessels. Once salivation is stimulated, these parasympathetic fibres innervate cutaneous sweat glands and blood vessels. This results in gustatory sweating and cutaneous local vasodilation in the distribution area of the auriculotemporal nerve. Regeneration between parasympathetic and sympathetic nerves in this area is only possible because both use acetylcholine as a neurotransmitter. This common neurotransmitter is the main focus of therapy.

Frey syndrome is named after the Polish neurologist Lucja Frey, who described the underlying pathogenesis of this phenomenon in 1923. It has mostly been reported due to iatrogenic injury to the auriculotemporal nerve after parotidectomy or temporomandibular joint surgery.⁴ It has also been reported after trauma, carotid endarterectomy⁵ and obstetric trauma due to forceps use,⁶ idiopathic cases have also been described.^{7,8}

Case report

A 72-year-old woman was referred to the ENT clinic by her general practitioner. She had suffered for 32 years from

perioral excessive sweating and flushing which only occurred during (and not preceding) eating. Her complaints had begun following bilateral osteotomy of the mandible in 1960, at the age of 25 years. The indication for this procedure had been prognathism. Her recovery had been complicated by inadequate bone healing and loss of the right and left inferior alveolar nerves (Figure 1).

The diagnosis of hyperhidrosis was made using the iodine-starch test as described in 1927 by Minor. In this test, liquid iodine is applied to the affected region, followed after drying by a layer of starch powder. After an appropriate stimulus (in our case eating), the hyperhidrotic area becomes visible as a sharply demarcated, violet patch (Figure 2a). This method enables the clinician to precisely define the intensity of hyperhidrosis as well as the region of treatment.⁹

Our patient was treated with intradermal botulinum injections. Because of the risk of paresis of the orbicularis oris and mentalis muscle, a test injection was made median and low on the chin, with good results (Figure 2b).

Discussion

Both the submandibular and lingual glands are innervated by fibres from the facial nerve. These fibres derive from the superior salivary nucleus and are connected via the chorda tympani to the lingual nerve (a branch of the mandibular division of the trigeminal nerve) at the infratemporal fossa. Together they travel to the floor of the mouth, where the chorda tympani fibres leave the lingual nerve. In the floor of the mouth, beneath the lateral border of the tongue, lies the submandibular ganglion. Here the preganglionic parasympathetic secretomotor fibres of the chorda tympani synapse with postganglionic fibres which are distributed to the submandibular glands as well as the lingual glands.^{10,11}



FIG. 1

Orthopantomogram taken 44 years after the patient's original osteotomy. Note the bone deformation on the left side (white arrow). The bone defect on the right side is probably scar tissue due to incomplete bone healing (black arrow). Too much bone has been removed on the right side; hence, the chin points towards the right side.

Injury to the lingual nerve at the submandibular ganglion, along with the chorda tympani parasympathetic secretomotor branch of the facial nerve and sympathetic fibres, leads to aberrant reinnervation of sweat glands and blood vessels. In such cases, a Frey-like syndrome of the skin in the submandibular region has been documented, termed the 'chorda tympani syndrome'.¹²⁻¹⁴ Due to disruption of the

chorda tympani, gustatory otorrhoea has also been described after modified radical mastoidectomy.^{15,16}

In our case, gustatory sweating occurred around the mouth and chin area. The infraorbital and mental nerves are responsible for sympathetic innervation in this area. The infraorbital nerve emerges at the infraorbital foramen as a terminal branch of the maxillary nerve, and innervates the upper lip. The mental nerve arises from the mandibular canal at the mental foramen as a terminal branch of the inferior alveolar nerve, and innervates the skin of the chin and lower lip. Our patient had undergone an osteotomy in 1960, for which an external (extraoral) approach had been used. We hypothesise that this osteotomy damaged not only both inferior alveolar nerves but also the parasympathetic fibres from the submandibular and lingual glands. These latter two sets of nerve fibres subsequently regenerated and united with the sympathetic fibres of the infraorbital and mental nerves. This resulted in gustatory sweating, also termed Frey-like syndrome, in the skin around the mouth and chin. Unfortunately, we were not able to obtain a detailed surgical report of our patient's original procedure. However, the anatomical position of the submandibular ganglion in relation to the mandible, the infraorbital nerve, the mental nerve and the patient's two external submandibular scars (each about 10 cm in length) supports our hypothesis.

- The first described case of perioral gustatory sweating is presented
- A hypothesis is postulated based on Frey syndrome pathophysiology
- Perioral gustatory sweating is a rare complication of external mandibular osteotomy
- In this case, botulinum toxin was helpful in treating perioral hyperhidrosis



(a)



(b)

FIG. 2

Clinical photographs taken during an iodine-starch test, (a) after a gustatory stimulus, with the hyperhidrotic area becoming visible as a dark patch; and (b) after injection with botulinum toxin, showing minor asymmetry of the right corner of the mouth as a side effect of treatment.

A temporary, diagnostic lingual nerve block, as described by Graham and Baldwin,¹³ could have proven this hypothesis. Unfortunately, due to personal circumstances our patient was not able to undergo any subsequent investigation or treatment.

Classic Frey syndrome is pathophysiologically explained by aberrant reinnervation of postganglionic parasympathetic fibres, which previously innervated the parotid gland, with sympathetic fibres of the auriculotemporal nerve. In our patient's case, it is highly unlikely that the parasympathetic fibres of the parotid were damaged and regenerated, because the parotid gland was not in the operation area. Furthermore, there is no close proximity of the parotid gland to the mental nerve.

Conclusion

Perioral Frey syndrome can be a long-term complication of mandibular osteotomy. Trauma to the postganglionic parasympathetic fibres can occur during this type of surgery, during complicated recovery, or both. In combination with trauma to both infraorbital and mental nerves, this gives rise to a symmetrical Frey syndrome, via regeneration and sprouting of nerve fibres in the distribution area of these nerves. To our knowledge, no previous case of perioral gustatory sweating has ever been published. We postulate a mechanism similar to classic Frey syndrome. In our case,

treatment was with intradermal injections of botulinum toxin.

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