

Short communication

An unusual cause of Frey syndrome

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Accepted 25 July 2008

Available online 19 September 2008

Abstract

We present the case of a young woman who complained of gustatory sweating of the skin of the submandibular region after resection for oral cancer, neck dissection and reconstruction with a radial forearm free flap. This is unusual after neck dissection and particularly so after selective neck dissection that did not need adjunctive radiotherapy.

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Keywords: Frey syndrome; Selective neck dissection

Case report

A 26-year-old white woman presented with a T2N0Mx squamous cell carcinoma of the right lateral border of the tongue. Otherwise, she was fit and healthy, did not smoke cigarettes or drink alcohol. She had a tracheostomy, right level I-IV selective neck dissection (including right submandibular salivary gland removal and diathermy of the submandibular ganglion), resection of tumour, reconstruction of the resection site with a left radial forearm free flap, and reconstruction of the forearm defect with a full thickness skin graft from the anterior abdominal wall. All resection margins were clear postoperatively and there were no neck metastases, so post-operative radiotherapy was not required. Almost 8 months after operation, she had gustatory sweating or hyperhidrosis and flushing (otherwise known as Frey syndrome) in the right submandibular region. Minor's starch-iodine test had not been done for confirmation; because it has been reported that there can be a discrepancy between subjective complaints and objective data.¹ She was not concerned by this phenomenon and did not seek any active treatment.

Discussion

Gustatory sweating was first described by Duphenix in 1757 in association with parotid gland trauma, then by Baillarger in 1853, and subsequently by Frey in 1923; not only in association with parotid gland trauma, but confined to the distribution of the auriculotemporal nerve.^{1–4} It has since been reported after parotid gland surgery.^{1–3} Frey syndrome is an unusual complication after removal of the submandibular salivary gland^{1,4,5} and neck dissection.¹ When reported in relation to neck dissection it has been associated with radical dissections,^{1–3,6,7} particularly for thyroid cancer,² and has tended to occur in the submandibular area on the upper skin flap.³ It has only recently been reported after a selective type anterolateral neck dissection (for thyroid medullary carcinoma) in a 30-year-old man in Argentina.⁸ The theory of aberrant or crossed innervations^{1–3,4,8} is the accepted explanation; this is where injury to the lingual nerve (along with the chorda tympani parasympathetic secretomotor branch of the facial nerve and sympathetic nerve fibres) at the submandibular ganglion leads to aberrant reinnervation of cutaneous sweat glands and blood vessels, and then to Frey syndrome of the skin of the submandibular region. This theory is verified by the fact that a temporary lingual nerve block resolves the condition,⁴ so has been termed the chorda tympani syndrome.⁴ It is therefore possible that this

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complication occurs more often than is reported by patients after selective neck dissections, particularly where level I is removed and in cases in which this is not followed by radiotherapy. Perhaps we should be more concerned about this complication in such patients, as it can be distressing and is treatable, for example by injection of botulinum A toxin.^{1,8}

References

1. Laskawi R, Ellies M, Rödel R, Schoenebeck C. Gustatory sweating: clinical implications and etiologic aspects. *J Oral Maxillofac Surg* 1999;**57**:642–9.
2. Spiro RH, Martin H. Gustatory sweating following parotid surgery and radical neck dissection. *Ann Surg* 1967;**165**:118–27.
3. Myers EN, Conley J. Gustatory sweating after radical neck dissection. *Arch Otolaryngol* 1970;**91**:534–42.
4. Teague A, Akhtar S, Phillips J. Frey's syndrome following submandibular gland excision: an unusual postoperative complication. *J Otorhinolaryngol Relat Spec* 1998;**60**:346–8.
5. Berini-Aytes L, Gay-Escoda C. Morbidity associated with removal of the submandibular gland. *J Craniomaxillofac Surg* 1992;**20**:216–9.
6. Parkash S, Ramakrishnan K. Gustatory syndrome following radical neck dissection. *Aust NZ J Surg* 1983;**53**:491–2.
7. Coldwater KB. Treatment of the auriculotemporal syndrome. *AMA Arch Surg* 1954;**69**:54–7.
8. Báez A, Paleari J, Durán MN, et al. Síndrome de Frey por submaxilectomía y tratamiento con toxina botulínica (Frey syndrome secondary to submaxillectomy and botulinic treatment). *Medicina (B Aires)* 2007;**67**:478–80.