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Two rare causes of Frey syndrome

Frey syndrome (gustatory sweating) is a well recognised potential postoperative complication following parotidectomy. It has also been documented following other surgical procedures which can potentially disrupt the parasympathetic carriage within the auriculo-temporal nerve (ie. both temporo-mandibular joint surgery and trauma). Reports of this syndrome following other head and neck procedures are exceedingly rare. In this paper we document the unusual causes of postoperative gustatory sweating and cutaneous vasodilation in two patients. One patient had undergone submandibular adenectomy, while the other had undergone excision of a cystic hygroma. These cases presented are interesting because Frey syndrome occurred in an unusual neural distribution. In the first case of submandibular adenectomy; the parasympathetic secretomotor nerve is likely to be the Chorda Tympani and in the second case of cystic hygroma excision from the neck, branches of the cervical plexus. We briefly review both the aetiology and contemporary management of this syndrome.

Summary of cases

Case 1: A 71-year-old female underwent excision of the left submandibular gland due to chronic recurrent obstructive sialadenitis. A standard operative

technique was used. The incision was placed >3cm below the inferior border of the mandible to avoid injury to the marginal branches of the facial nerve. The gland was exposed by a combination of sharp and blunt dissection having first divided the subcutaneous tissue, platysma and cervical fascia. The submandibular gland was removed after identification of the lingual nerve and ligation of the Wharton's duct. The surgical site was closed in layers, with the platysma muscle sutured with resorbable sutures. Postoperatively there was no facial nerve deficit. Six months following surgery, she complained of gustatory sweating in the operative site consistent with Frey syndrome. The patient did not feel that her gustatory sweating was sufficiently severe to warrant treatment.

Case 2: A 19-year-old female complained of long history of gustatory sweating and flushing in the neck. She had previously undergone excision of a congenital cervical cystic hygroma in the same anatomical location. The cystic mass was excised at one year of age. There was no documented intra-operative damage to adjacent neuro-vascular structures. The status of the lingual, hypoglossal, marginal mandibular and accessory nerve were intact postoperatively. Her postoperative recovery was uneventful and without recurrence of lesion. In view of her long history gustatory sweating and the fact that the patient was unconcerned about the problem, no treatment was recommended.

Our cases are interesting because Frey syndrome occurred presumably within the cutaneous sensory distribution of the cervical plexus [anterior and posterior branches of greater auricular nerve (C2, C3) and transverse cervical nerve (C2 & C3)]. This is in contrast to post-parotidectomy gustatory sweating, which would typically result from disruption of parasympathetic carriage within the auriculo-temporal nerve. These observations highlight the fact that Frey syndrome may result from any operative procedure within the head and neck, which potentially may disrupt post ganglionic para sympathetic secreto-motor - vasodilator fibres.

References

1. Linder TE, Huber A, Schmid S (1997) Frey's syndrome after parotidectomy: a retrospective and prospective analysis. *Laryngoscope* 107(11): 1496–1501
2. Mellor TK, Shaw RJ (1996) Frey's syndrome following fracture of the mandibular condyle: case report and literature review. *Injury* 27(5): 359–360
3. Teague A, Akhtar S, Phillips J (1998) Frey's syndrome following submandibular gland excision: an unusual postoperative complication. *ORL J Otorhinolaryngol Relat Spec*. 60(6): 346–348

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