

Gustatory itching: an unusual complication following superficial parotidectomy

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Abstract

Objective: We report the previously undocumented complication of gustatory itching following superficial parotidectomy.

Method: Case report and review of the English literature concerning Frey's syndrome, complications of superficial parotidectomy and the pathophysiology of itching.

Results: A 49-year-old woman developed gustatory itching following a superficial parotidectomy. Her symptoms were satisfactorily managed with topical and oral antihistamine preparations. We propose a neurophysiological pathway involving acetylcholine and histamine to explain this phenomenon.

Conclusion: To our knowledge, this is the first documented case of gustatory itching following superficial parotidectomy. The use of antihistamine preparations appears to effectively manage this symptom, without the need for invasive procedures.

Key words: Parotidectomy; Itching; Frey's Syndrome

Introduction

Frey's syndrome is a well described side effect of parotid surgery.¹ It consists of gustatory stimulation of sweating, flushing and warmth in the temporal and preauricular areas.²

We present a case of gustatory itching following surgery for benign parotid pathology. This symptom has not previously been described in the English literature.

Materials and methods

Case-based discussion and review of the English literature concerning Frey's syndrome and the neurophysiology of itching.

Case report

A 49-year-old woman presented to the clinic with a two-month history of a left-sided facial lump. She had no swallowing difficulty.

On examination, she was found to have a 2 × 2 cm, hard, deeply fixed lump at the left angle of the mandible. This was associated with left-sided otalgia.

A fine needle aspiration biopsy was performed; cytological analysis suggested a diagnosis of pleomorphic adenoma of the left parotid gland.

The patient subsequently underwent a left superficial parotidectomy. Histological examination of the resected mass confirmed the diagnosis of a completely excised pleomorphic adenoma.

At the first post-operative review one week after surgery, the patient was satisfied with her progress. Her scar was healing well and she had retained full facial nerve function. She did complain of numbness over her left ear lobe, which

was expected as the great auricular nerve had been divided during surgery. She denied any symptoms of Frey's syndrome or gustatory itching.

However, 14 months post-operatively the patient was referred to our clinic again. She had developed symptoms of pain and itching over her left cheek and preauricular area after eating. She reported that these symptoms lasted for approximately 30 minutes after eating. Itching would also be stimulated by salivation in the absence of food. The itching could be so intense that she reported episodes of scratching the affected area until it bled.

A trial of non-sedating antihistamine tablets and topical antihistamine creams was undertaken, and a marked improvement in symptoms was reported. The topical creams had a greater effect than the tablets. Subsequently, symptoms persisted but were reduced to a tolerable level.

Discussion

Frey's syndrome, or gustatory sweating, is a common and well recognised phenomenon following parotid surgery. It was first described by Lucja Frey, who named it the auriculotemporal syndrome. Its reported incidence varies from 18 to 45 per cent.^{1,3} It has also been described following most facial and neck surgical procedures in the preauricular and mandibular region. The syndrome consists of a range of symptoms including sweating, flushing and warmth over the preauricular and temporal region shortly after eating. It is often most marked after eating foods that cause a greater salivary response, e.g. acidic foods. The most commonly used diagnostic test is Minor's iodine starch test.²

There are many hypotheses concerning the cause of Frey's syndrome. The most widely accepted theory

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postulates the development of a parasympathetic–sympathetic anastomosis following surgery. During parotidectomy, the parasympathetic fibres supplying the salivary glands are inevitably transected, and this is also true for the sympathetic fibres supplying the sweat glands to the skin overlying the area. Despite the parotid innervation being parasympathetic and the skin innervation sympathetic, both pathways use acetylcholine as their neurotransmitter. It is postulated that the parasympathetic fibres regenerate after surgery and anastomose with the remaining sympathetic fibres. When there is gustatory stimulation and excitation of the parasympathetic fibres, the common neurotransmitter allows cross-excitation of the sympathetic chain, resulting in sweating.³

The presented case is notable because of the unusual nature of the patient's symptoms. Although Frey's syndrome is very well documented in the literature, gustatory itching was unheard of in our department. Indeed, a search of the literature revealed no previous reports or descriptions of such a phenomenon.

In order to understand the presented case, it is beneficial to review the neurophysiology of itching. Recent studies have suggested a separate central neural pathway for itching, along the lateral spinothalamic tract.^{4,5} These fibres are highly sensitive to histamine release, and cause a sensation of intense itching.⁵ However, the lateral spinothalamic tract does not use acetylcholine as a neurotransmitter; rather, it uses glutamate.⁵ Therefore, the neuronal anastomosis and shared neurotransmitter theory of Frey's syndrome would not appear to be relevant: as the two pathways do not share a common neurotransmitter, it is difficult to appreciate how cross-excitation could occur. It was interesting to observe that our patient's symptoms were relieved by the use of chlorphenamine, an antihistamine.

- **Frey's syndrome is a common side effect of parotid surgery, caused by cross-excitation of the sympathetic nervous system with acetylcholine released by the parasympathetic nervous system**
- **The reported patient complained of gustatory itching after parotidectomy, a previously undescribed symptom**
- **Itching is mediated by the spinothalamic tract, which uses glutamate as a neurotransmitter**
- **Antihistamine preparations reduced the intensity of gustatory itching in this case**

This leads to the possibility that our patient's symptoms were propagated by histamine release. In cholinergic urticaria, a dermatological condition, intense itching and weal formation are propagated by cholinergic (i.e. acetylcholine) induced histamine release.⁶ If acetylcholine can be responsible for a histamine release sufficient to produce urticaria, it certainly seems possible for it to produce a milder degree of itch. There is good evidence to support the ability of the parasympathetic nervous system, and acetylcholine, to cause mast cell degranulation and therefore histamine release. *In vitro* studies have shown that the addition of acetylcholine to solutions containing mast cells increases the concentration of free histamine.^{7,8} Indeed, one study has demonstrated the ability of free acetylcholine to provoke histamine release from mast cells in guinea pig auricle tissue.⁹ This study also demonstrated that direct electrical stimulation of the parasympathetic nervous

system resulted in increased concentrations of free histamine in solution.

Therefore, we propose that our patient's symptoms were caused by gustatory stimulation of the parasympathetic nervous system, leading to the release of free acetylcholine in the tissues underlying the symptomatic region. This in turn caused mast cell degranulation, leading to an increase in free histamine and resulting in stimulation of the lateral spinothalamic tract, causing our patient's symptom of gustatory itching.

Conclusion

Gustatory itching following superficial parotidectomy has not previously been described. We hypothesise that this symptom is caused by gustatory stimulation of the parasympathetic nervous system, leading to release of free acetylcholine in the tissues underlying the symptomatic region. This free acetylcholine then causes mast cell degranulation, leading to stimulation of the lateral spinothalamic tract, resulting in gustatory itching.

The reported case highlights the importance of nervous chain recovery after surgical disruption, a possibly much overlooked source of post-operative dissatisfaction among patients lacking adequate follow-up. This case serves to elucidate the management of gustatory itching following superficial parotidectomy.

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