



Frey's Syndrome as an Unusual Consequence of Surgery for TMJ Ankylosis

**Aritra Chatterjee^{1*}, Subhasish Burman¹, Soumen Mandal¹,
Abira Chattopadhyay¹, Md. Arif Hossain¹ and Debabrata Gayen¹**

¹*Department of Oral and Maxillofacial Surgery, Dr. R. Ahmed Dental College and Hospital, Kolkata, India.*

Authors' contributions

This work was carried out in collaboration among all authors. All the authors were involved in designing and conceptualization of the study. All were involved in the drafted and editing of the manuscript and literature search. All authors read and approved the final manuscript.

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Case Study

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ABSTRACT

Frey's syndrome is a rare, unwanted complication of surgery/injury to the parotid gland, occasionally seen following surgery in the temporomandibular joint region, neck dissection and faces lift procedures. It is characterised by sweating, flushing and a sense of warmth over the distribution of the auriculotemporal nerve and/or greater auricular nerve while eating foods that produce a strong salivary stimulus. As a result this can be a source of great discomfort and embarrassment to the patient. Here, we present a case of Frey's Syndrome which developed after an inter positional arthroplasty in the right TMJ region. In the follow up period he reported to us with complaints suggestive of gustatory sweating. A positive Minor's Starch Iodine test confirmed the diagnosis. We reassured him and also mentioned the treatment options available. The symptoms being mild, he decided on no further interventions. The literature does not speak much of such a complication arising after surgery for TMJ ankylosis. This case should be an eye opener for many surgeons as to the possibilities of Frey's syndrome developing after such surgeries and the means to manage them.

*Corresponding author: Email: c.aritra@gmail.com;

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ABBREVIATIONS

ATN : Auriculotemporal nerve
SCM : Sternocleidomastoid Muscle
ATS : Auriculotemporal Syndrome
TMJ : Temporomandibular Joint
BTA : Botulinum Toxin A

1. INTRODUCTION

The first case of Auriculotemporal Syndrome (ATS) was described by Duphenix in 1757, following surgical drainage of a parotid abscess [1]. Baillarge, in 1853, reported bilateral involvement of the Auriculotemporal nerve (ATN) following a parotid infection [2]. In 1897, Weber observed the syndrome after suppuration in the parotid lymph nodes [3]. In 1923, Lucja Frey was the first to observe that the sweating and flushing occurred over the distribution of the ATN after a bullet traumatized a patient's parotid gland [4] and she coined the term "Auriculotemporal Syndrome" or gustatory sweating. It has, since then been also referred to as Frey's syndrome.

Frey's syndrome is commonly found in patients who have undergone parotidectomy or those with extensive injuries to the parotid glands. The classical symptom of Frey's Syndrome is gustatory sweating characterised by excessive sweating on the forehead, cheek and around the ears, after eating foods, specifically those with a strong stimulus such as spicy, salty and sour foods. Additionally flushing, warmth and pain in the affected areas may also be reported. In most of the cases, the symptoms are mild and are ignored by the patient. In some cases however, the symptoms may become so troublesome that the patients may require some form of intervention.

Diagnosis of Frey syndrome is based on clinical history, but confirmatory testing can be done with a Minor starch-iodine test. It involves the painting of the ipsilateral face with an iodine solution 3-5% and allowing it to dry. Subsequently dusting of starch powder is done on the same side. The patient is then given a sialogogue and observed. Dark blue staining on the same side confirms the initial diagnosis.

Development of such a condition following surgery for release of TMJ ankylosis is rare. Not much has been written in the literature about such an occurrence. Frey's syndrome following

interposition arthroplasty adds to the list of conditions that the surgeon should remain careful about, not just during the surgical procedure, but also during follow ups.

2. CASE HISTORY

A 22 year old man was referred to the Department of Oral and Maxillofacial Surgery of our institution. He had come with the chief complaint of inability to open his mouth. History taking revealed that he had undergone surgery for temporomandibular joint ankylosis, at the age of 14, at an outside centre. His condition had not improved much post surgery and he had developed a debilitating mouth opening at the present time.

Extraoral examination revealed a scar in the right preauricular region, a mouth opening of 2 mm, a deviation of face to the right side along with fullness of the same side. On intraoral examination, crowding and labial inclination of lower anteriors was seen along with a posterior cross bite on the right side and a marked deviation of the dental midline to the right. Subsequent radiographic (orthopantomogram and CT scan) evaluation, added to the clinical scenario affirmed a diagnosis of reankylosis of the right TMJ (Fig. 1).



Fig. 1. Preoperative facial profile

After obtaining informed consent from him, an interposition arthroplasty procedure was planned to increase his mouth opening. Access to the ankylotic mass was made utilising the scar of the Al-Kayet and Bramley incision that had been used in the previous surgery (Fig. 2). The operative procedure was carried out with care to avoid injury to any of the nerves, especially the facial nerve found in the region. We succeeded in giving him a mouth opening of 42 mm and his immediate post operative period passed without any trouble. During a routine follow up procedure on the 8th postoperative month, the patient reported with a complaint of sweating and a feeling of warmth in the surgical area during meals. An initial diagnosis of gustatory sweating was made based on the clinical presentation and this was confirmed by a positive Minor's Starch-Iodine test (Fig. 3). A second detailed history taking was done as to whether the condition was present prior to the second surgical procedure, but the patient categorically denied having any such complaints following the first surgery. We explained to the patient the condition in detail and presented him with all the possible treatment options. However, happy with the mouth opening that he had achieved post surgery, (which was his primary complaint) he opted for no further interventions. The decision taken by him was based on the fact that his current condition was not too troublesome for him and we respected the same.



Fig. 2. Scar along the line of incision from the previous surgery



Fig. 3. Positive minor's starch iodine test

3. DISCUSSION

The ATN is one of the terminal branches of the mandibular division of the trigeminal nerve. It is a mixed nerve and it carries both sympathetic and parasympathetic fibres. The postganglionic sympathetic fibres are derived from the superior cervical ganglion and travel along the external carotid artery and its branches, joining the ATN via the facial artery, middle meningeal artery, and internal maxillary artery. These sympathetic fibres are responsible for salivary inhibitory impulses and vasoconstriction of the vessels supplying the parotid gland. The nerve also provides branches to the temporomandibular joint and the skin over temporal area and external acoustic meatus. The inferior salivary nucleus gives rise to the parasympathetic fibres, which exit the skull through the jugular foramen and pass successively through the tympanic branch (nerve of Jacobson) of the IX cranial nerve, tympanic plexus and lesser petrosal nerve and relay into otic ganglion. After synapsing at the otic ganglion, the postganglionic fibres from the ganglion pass through the ATN and reach the parotid gland and provide secretory innervation [5,6].

Though there are several theories but none explain the specific mechanism of the Frey's syndrome. Frey suggested that the ATN was

irritated by scar tissue [7]. The Aberrant Regeneration Theory, proposed by Ford and Woodhall, is the mostly acceptable theory [8]. This theory is based on the fact that the ATN is a mixed nerve containing both sympathetic and parasympathetic fibers. When the nerve is injured, these fibers become damaged. It is postulated that a misdirected regeneration of the parasympathetic fibers to denervated sweat glands in the skin results in simultaneous activation of parotid and sweat glands when there is a salivary stimulus, causing inappropriate gustatory sweating.

It is always a possibility that the condition developed due to scarring after the first surgery [7], but a history after the second surgery refuted any such theory. The patient categorically denied having this condition after the first surgery. History taking prior to the second surgery did not reveal any such condition too.

Anatomically, after origin from posterior division of mandibular nerve ATN encircles the middle meningeal artery and passes backward under cover of lateral pterygoid muscle between the neck of the condyle and sphenomandibular ligament. Then, ATN turns laterally behind the temporomandibular joint, close to the upper part of the parotid gland. Ultimately it ascends behind the superficial temporal vessel and reaches the temporal region. Thus the ATN is vulnerable to injury in TMJ surgeries. In our case, it is this vulnerability that may have led to an injury to the ATN during the removal of the ankylosis mass. Additionally, a smaller incision and less anterior tissue reflection seem to reduce the possibility of development of this condition [9]. Thus, had we used a smaller incision, this condition might have been avoided to the benefit of the patient.

ATS has also been documented after subcondylar osteotomy for mandibular setback [10], sagittal split osteotomy, condylar fracture, submandibular gland and cervical lymph node surgery [11]. Tuinzing et al described the syndrome as a complication following a sagittal ramus osteotomy [12]. Bohdan Kryshtalsky J et al reported this syndrome in 3 out of 16 patients (20 TMJs), after temporomandibular surgery through the preauricular approach [13]. Zöller et al reported Frey's syndrome secondary to a subcondylar fracture [14]. In 1996, Mellor and Shaw encountered the syndrome following a condylar fracture [15]. The following year, a similar case was reported by Sengezer et al after closed treatment of bilateral condylar fractures

[16]. More recently, however, Tue et al made an extensive literature review and concluded that trauma of considerable impact, the existence of more than one mandibular fracture site, dislocation of the condyle, and altered sensibility in the preauricular region, all appear as major risk factors for Frey's syndrome [17].

The time in which the symptoms of Frey's syndrome first appear varies. Although commonly seen 6 weeks after surgery in parotid gland, it can present as late as 5 years after surgery [18]. In our case patient presented with symptoms after 8 months postoperatively.

There are several medical and surgical treatment strategies to treat Frey's syndrome. Most of the medical therapies used are given via injection therapy or by topical application. Previously alcohol, scopolamine, glycopyrrolate, or BTA were used in different forms. Currently, BTA is the most widely used agent for intradermal injection and it has been seen that patients undergoing BTA injection demonstrate significant improvement in symptoms over time. Surgical transection of the auriculotemporal nerve, tympanic nerve, and greater auricular nerve has been described for the management of Frey syndrome, but they are rarely done.

A variety of surgical procedures have been proposed for prevention of Frey's syndrome by incorporating a barrier between the underlying postganglionic parasympathetic nerve endings within the transected parotid and the overlying cutaneous tissue. Dia and colleagues performed a cohort study of 17 patients with post parotidectomy Frey's syndrome who underwent both SCM and temporalis fascia transposition. They reported that greater than 50% of patients who underwent that surgery had complete resolution by starch-iodine testing [19]. However, surgical management of post parotidectomy Frey syndrome should be reserved for severe and refractory cases, as there is limited data to support its use.

In the last decade Botulinum A toxin has become established as a therapy for individuals with bothersome Frey syndrome. It is minimally invasive and provides relief for a period of 9-12 months. Though the effects seem to reduce with time, sweating seems to reduce with successive sets of injections to the extent that it is no longer troublesome [20].

4. CONCLUSION

Frey's syndrome is an unwanted complication of surgeries which are related to the parotid gland and the TMJ region. It may cause considerable social embarrassment and incapacity to the patient because of the profuse flushing and sweating while eating. Although there are numerous medical and surgical treatment modalities, significant risks or side effects after the treatment along with incomplete cure, are worries both for the patient as well as the treating surgeon. The mainstay of treatment, even today, lies in explanation of the condition to the patient and reassurance. To conclude, preventive measures should always be kept in mind during any surgical procedure in the said region, in order to avoid the development of this bothersome condition.

CONSENT

As per international standard patient's written and Informed consent has been collected and preserved by the author(s).

ETHICAL APPROVAL

As per international standard or university standard written ethical approval has been collected and preserved by the author(s).

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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