

## Case Report:

### A case report of Frey's syndrome following TMJ surgery

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#### ABSTRACT

Frey's syndrome is a well recognized complication of surgery in preauricular region. It is characterized mainly by recurrent episodes of hyperesthesia, flushing and warmth or sweating limited to cutaneous distribution of auriculotemporal nerve while eating foods that produce strong salivary stimulus. Although commonly encountered as a complication of total or partial parotidectomy, on rare occasions it follows surgery or fracture of temporomandibular joint. A case of Frey's syndrome in a patient who developed symptoms 3 yrs after TMJ surgery is reported.

Keywords : Frey's syndrome

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#### INTRODUCTION

Frey's syndrome is a disorder characterized by unilateral sweating and flushing of facial skin in area of parotid gland occurring during meals.<sup>1,2</sup> The syndrome was initially termed 'auriculotemporal nerve syndrome' and also been referred to as 'gustatory sweating'.<sup>1,2,3</sup> Duphenix first reported it in 1757 and it was Lucia Frey, a French neurologist who in 1923 implicated auriculotemporal nerve in pathogenesis of syndrome and used the term 'auriculotemporal nerve syndrome'.<sup>2,3,4</sup>

Frey's syndrome has been reported to occur more frequently following parotid gland surgery. Some authors believe that incidence vary from 10-30% of symptomatic patients<sup>5</sup> others report it to vary from 30-50%<sup>6,7</sup>. Over 90% of the patients test positive for gustatory sweating many of whom are asymptomatic.<sup>8</sup> Less frequently it follows fracture of mandible, surgery or fracture of TMJ, radical neck dissection, submandibular gland excision, and thyroidectomy or after thoracic sympathectomy.<sup>5,8</sup>

Although relatively common in adults the condition in rare instances has been reported in children and infants as a sequel of perinatal birth trauma from assisted forceps delivery.<sup>9,10</sup> Through the years several theories have been proposed regarding the pathophysiology of the syndrome. Lucia Frey believed that the damaged auriculotemporal nerve is invaded and irritated by healing tissue.<sup>2,5</sup> Freedberg suggested damage to nerve may cause distribution of sympathetic fibers leading to parasympathetic hypersensitivity and stimulation. The theory of aberrant regeneration says that there is defective nervous regeneration following injury to auriculotemporal nerve. The misdirection of regenerating parasympathetic fibers to denervated sweat glands results in simultaneous activation of parotid and sweat glands.<sup>4,5,6</sup>

Various tests to asset the presence of gustatory sweating have been described. The most widely used is minor's starch iodine test.<sup>1,2,3,11</sup> Other test include biosensing method using enzymatic

electrodes to detect L-lactate levels on skin of affected area, use of thin facial tissue paper to demonstrate areas of sweating, one step method using dyes like bromophenol blue powder, pyrogallol, ferric hydroxide or quinazarin and infrared medical thermography.<sup>4,12</sup>

Differential diagnosis include crocodile tear syndrome, gustatory sweating associated with diabetes<sup>8</sup> and food allergy in case of children.<sup>9,10,13</sup>

Frey's syndrome in children usually resolves spontaneously and no treatment is required. Several treatment options have been described in adults who aim at reducing incidence of Frey's syndrome but none of them have given promising results. There are mainly three options- surgical measures, medicinal treatment and radiation therapy. Surgical measures include interposition of sternocleido mastoid flap,<sup>5,14</sup> superficial musculoaponeurotic system, alloderm,<sup>15</sup> fascia lata or sialasthic sheeting, transmeatal tympanic neurectomy, intracranial neurolysis of glossopharyngeal nerve or transaction of Jacobson's anastomosis.<sup>14,15,16</sup> Medicinal treatment includes systemic or local anticholinergics such as scopolamine, glycopyrrolate,<sup>16</sup> aluminium chloride hexahydrate, diphemanil methyl sulphate and intracutaneous injections of botulinum toxin type A.<sup>6,17</sup>

#### **CASE REPORT**

A 26 years old male patient reported to oral medicine and radiology dept. of G.D.C.H. Ahmedabad with chief complaint of pain and sweating in left preauricular region while eating since fifteen days.

There was history of trauma when patient was four years of age. His mouth opening gradually decreased after that so he was operated for left TMJ ankylosis at oral surgery dept. of G.D.C.H.

Ahmedabad using gap arthroplasty with preauricular incision. There were no substantial complaints till 3 yrs after surgery but before 15 days patient complained that when beginning to eat he felt a sharp pain, intense warmth and sweating in preauricular area on left side. The rest of his medical history was non contributory.

Examination revealed scar in left preauricular region (fig-1), restricted mouth opening of 30mm, and deviation of face to left side (fig-2). Intraoral examination showed posterior cross bite on left side and deviation of anterior midline to left side. An orthopantomogram showed condylectomy of left side (fig 3). The diagnosis of Frey's syndrome was made and confirmed by performing minor's starch iodine test. The affected areas were coated with 1% iodine solution and were allowed to dry. Starch powder was applied on the skin (fig-4) and patient was given a tablet of ascorbic acid as a salivary stimulus. After a few minutes patient complained of sharp pain and heat sensation and areas of blue black colorations could be observed (fig-5). These areas represented the combination of secretion of sweat glands diluted with iodine which reacted with starch producing this coloration. Although the areas were small, the diagnosis of Frey's syndrome was confirmed. Available treatment options were explained to the patient. Patient opted for topical application and was prescribed topical use of Glycopyrrolate twice daily. After fifteen days of follow up the patient stated improvement in pain and gustatory sweating in the preauricular area which he was experiencing pretherapeutically. However he experienced slight dryness of mouth for less than an hour after the topical application of glycopyrrolate cream.

## DISCUSSION

The specific mechanism involved in Frey's syndrome is yet unknown. However the theory of aberrant regeneration is the most acceptable theory. Auriculotemporal nerve is the terminal of mandibular branch of trigeminal nerve. It is mixed nerve and has sympathetic and parasympathetic fibers. The secretomotor activity of parotid gland is controlled via parasympathetic fibers of this nerve. Injury to the branches of auriculotemporal nerve during preauricular surgery damages it. In process of nerve regeneration parasympathetic secretomotor fibers may become misdirected and grow along distal cut ends of sympathetic fibers to the skin vessels and sweat glands. As both parasympathetic and sympathetic fibers are cholinergic, a new stimulus is made possible and a gustatory stimulus produces sweating and flushing.<sup>1,2,3,4,5</sup>

The symptoms of Frey's syndrome usually present 6 weeks to several months after surgery in parotid gland but can present as late as 5 years after surgery.<sup>2,3,4</sup> In present case patient presented with symptoms 3 years postoperatively. Although rarely but Frey's syndrome do follow surgery of TMJ<sup>2,7,12</sup> as is seen in present case.

Symptoms of Frey's syndrome include flushing and warmth with overheating of affected areas of the skin which in some cases is associated with pain<sup>2,6</sup> as is seen in this case. Minor's starch iodine test performed in the present case is most widely used test. The test is accurate, easy to perform, provide usual confirmation of gustatory sweating and can identify asymptomatic patients of Frey's syndrome.<sup>4,8,11</sup>

Only 10-15% of patients with Frey's syndrome have symptoms severe enough to seek

treatment.<sup>2,4,6</sup> The mainstay of the treatment lies in reassurance and an explanation of the condition. Various forms of treatment have been advocated with varying reported degrees of success.<sup>4,5</sup> Surgical treatment procedures have possible complications and no guarantees of permanent success.<sup>5,11</sup> Medicinal treatment provides temporary relief and can cause local irritation to skin and sweat glands. Injections of botulinum toxin are costly, have adverse side effects and there are reports of recurrence after intracutaneous injections.<sup>6,8,17</sup> The use of 0.5% aqueous solution of topical glycopyrrolate for hyperhidrosis is well documented. Glycopyrrolate, an anticholinergic drug, is a quaternary ammonium compound that does not cross the blood brain barrier. It also penetrates biological membrane more slowly than other anticholinergics and appears to lead to fewer side effects.<sup>4,16</sup>

## CONCLUSION

Frey's syndrome is an interesting illustration of how misdirected nerve regeneration can manifest clinically. Frey's syndrome has the potential to cause great social distress for the patient and potential exist for negative psychological consequences. For this reason it is important not only for dentist but also for general practitioner to be aware of this disorder and to be able to counsel patient on available treatment options.

Fig.1 Photograph of patient's face showing scar in left preauricular region.

Fig.2 Deviation of patient's face towards left side

Fig.3 OPG showing condylectomy of left side.

Fig.4 Iodine and starch powder applied on skin

Fig.5 Black discoloration in area of scarring.

Figure 1



Figure 2

Figure 5



Figure 3



Figure 4



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