

Post Herpetic Frey's Syndrome

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Abstract

Frey's syndrome is characterized by unilateral sweating and flushing of the skin in the area of the parotid gland, occurring during meal, or on salivary stimulation. It is a common complication following surgery/injury in the parotid gland region/temporomandibular joint surgery; however, very few cases have been reported of its occurrence following herpetic infection. It is important for the clinician to recognize its unusual symptoms as early as possible to prevent its potential negative social and psychological effect on the patient. It is diagnosed on clinical presentation and confirmed by objective testing methods. The treatment ranges from topical application of glycopyrrolate to local injections of botulinum toxin and lastly to surgical intervention. An interesting case of Frey's syndrome is presented in a young patient having the previous history of herpes zoster infection, which was diagnosed clinically and confirmed by starch-iodine test and was successfully treated with topical application of glycopyrrolate.

Keywords: Frey's syndrome, glycopyrrolate, herpes infection, minor-starch iodine test

INTRODUCTION

Frey's syndrome is a disorder characterized by unilateral sweating and flushing of facial skin in the area of parotid gland occurring during meals. The syndrome was initially termed "auriculotemporal nerve syndrome" and also been referred to as "gustatory sweating."^[1,2]

Frey's syndrome has been reported to occur more frequently following parotid gland surgery. Some authors believed that the incidence vary from 10% to 30% of symptomatic patients, others report it to vary from 30% to 50%. Over 90% of the patients test positive for gustatory sweating many of whom are asymptomatic. Less frequently, it follows fracture of mandible, surgery or fracture of temporomandibular joint, radical neck dissection, submandibular gland excision, and thyroidectomy or after thoracic sympathectomy. Beale discussed the development of Frey's syndrome after chemotherapy for testicular teratoma. It is very rare to occur after herpetic infection.^[3-5]

The symptoms of Frey's syndrome may be precipitated by any type of food, especially during mastication and include flushing, sweating, erythema, or general discomfort about the skin in the area anterior to the ear over the angle of the mandible and over the site of the parotid gland.^[5]

Various tests to assess the presence of gustatory sweating have been described. The most widely used is minor's starch-iodine test. Other test include bio-sensing method using enzymatic electrodes to detect L-lactate levels on the skin of affected area, use of thin facial tissue paper to demonstrate areas of sweating, one step method using dyes such as bromophenol blue, powder ferric hydroxide or quinizarin, and infrared medical thermography.^[1,3,5,6]

Its differential diagnosis includes crocodile tear syndrome, gustatory sweating associated with diabetes and food allergy in case of children.^[7,8]

The treatment of Frey's syndrome includes medical treatment and surgical measures. Medical treatment includes systemic or topical application of anticholinergics such as scopolamine, glycopyrrolate, aluminum chloride hexahydrate, diphenhydramine methyl sulfate, and intra-cutaneous injections of botulinum toxin type A. Surgical measures include interposition of sternocleidomastoid flap, superficial musculo-aponeurotic system, alloderm, fascia lata or silastic

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sheeting, trans-meatal tympanic neurectomy, intracranial neurolysis of glossopharyngeal nerve or transaction of Jacobson's anastomosis.^[5,6,8]

This paper focuses on a case of Frey's syndrome reported in a male patient with a history of herpes zoster, a rare cause. The patient manifested sweating, flushing, and pain in the pre-auricular and temporal region for 7 years.

CASE REPORT

A 27-year-old male patient reported to Oral Medicine and Radiology Department of Government Dental College and Hospital, Ahmedabad with the chief complain of unusual sweating on right side of the face during mastication for 6–7 years. The patient also had complain of pain in the right side of ear for 2 years, and this complaint restricted his social life.

The patient was giving history of herpes zoster on right side of the face before 8 years. At that time patient had developed vesicles and blisters on the affected side of the skin and also had ulcers in the buccal mucosa and tongue on the same side and it was cured completely after proper medication. The patient was normal for 1 year, suddenly after 1 year patient developed complaint of sweating over the scarred area at the pre-auricular and temporal regions during taking some food. Subsequently, sweating increased which was associated with intense pain, warmth, and flushing in that area. For that, the patient consulted Otolaryngologist, but no intra-auricular cause was found. Hence, the patient visited our department with the same complaint. Except herpes zoster, past medical and dental history were non-significant. No history of oral destructive habits or history of allergy was reported.

Extraoral examination revealed the presence of scar marks over the right side of face, particularly in the preauricular and temporal region [Figure 1]. Intra-oral examination revealed discolored mandibular right central incisor and impacted third molars. Orthopantomogram showed periapical changes in relation with lower right central incisor and impacted lower third molars [Figure 2]. Magnetic resonance imaging excluded the possibility of intracranial cause for complaint [Figure 3].

The diagnosis of "Frey's syndrome" was suspected which was confirmed by performing minor starch-iodine test. The affected area was coated with 1% iodine solution and allowed to dry [Figure 4a]. Then starch powder was applied to the skin, [Figure 4b] and the patient was given a tablet of ascorbic acid 500 mg as a salivary stimulant. Subsequently, after taking the tablet, the bluish-black color was noted over the right side of the face [Figure 4c] which suggested a positive starch-iodine test.

The patient was treated with topical 0.5% glycopyrrolate solution roll on method. From the first day patient had decreased sweating on the affected side. Total ten applications were done and the patient had complete relief from the complaint. Patient's mandibular right third molar was extracted and also scaling was carried out.

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 the trigeminal nerve. It is mixed nerve and has sympathetic and parasympathetic fibers. The secreto-motor activity of parotid gland is controlled through parasympathetic fibers of this nerve.^[1,2,6]

In post sympathetic gustatory sweating, the auriculotemporal syndrome appears to be caused by faulty regeneration or collateral sprouting of parasympathetic fibers after degeneration of sympathetic fibers traveling with peripheral branches of trigeminal nerve. In our case, the cause for Frey's syndrome is herpes zoster infection. As in herpes zoster infection, nerve damage is present, with skin areas affected by herpetic scarring in the distribution of auriculo-temporal nerve which receives sympathetic innervation from plexus around the middle meningeal artery. Some sympathetic fibers traverse the otic ganglion without synapsing before joining these branches of the mandibular nerve. Parasympathetic and sympathetic fibers are thus adjacent in the otic ganglion in auriculo-temporal nerve. Collateral sprouting of fibers after the partial damage is a well-known phenomenon in the autonomic nervous system.^[6,8,9]

After herpetic infection, the damaged parasympathetic fibers regenerate to connect damaged distal sympathetic fibers that innervate subcutaneous sweat glands and cutaneous blood vessels. When the inferior salivary nucleus is stimulated, the output previously destined for the salivary gland now stimulates sweat glands in the distribution area of the auriculotemporal nerve. This cross-regeneration between parasympathetic and sympathetic nerve fibers is only possible because both use acetylcholine as a neurotransmitter in this area.^[4,5,9]

The features of Frey's syndrome can be observed by any type of food, especially during mastication and include sweating, flushing, erythema, or general discomfort about the skin in the area anterior to the ear over the angle of the mandible and over the site of the parotid gland.^[4] Our patient had complained of sweating and feeling of warmth over the skin near the preauricular region. As such, as symptoms of Frey's syndrome usually present in six weeks to several months or may present as late as 5–8 years. In the present case, the patient had symptoms for 7 years after the herpetic infection.

Irrespective of the cause, Frey's syndrome can be diagnosed clinically and confirmed by a minor starch-iodine test. The sweat which gets produced combines with the iodine and reacts with overlying starch to give the bluish-black color. A variant of this test uses an iodinated starch powder spray. The minor starch test is considered to be very accurate and is capable of identifying Frey's in asymptomatic patients.^[5,9] In our case, minor starch-iodine test was positive, and hence, the patient's diagnosis was confirmed as Frey's syndrome.

The patient was successfully treated with 0.5% glycopyrrolate solution by roll on method. Glycopyrrolate is a quaternary



Figure 1: Lateral profile of the patient showing the presence of scar marks over the preauricular region and temporal region (arrows)

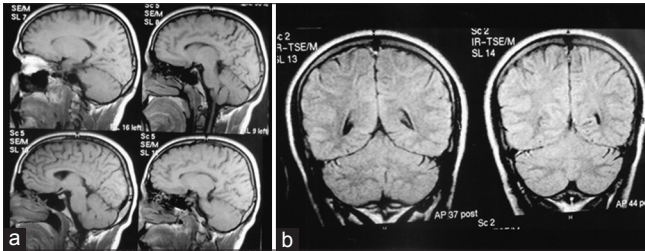


Figure 3: Sagittal (a) and coronal (b) magnetic resonance imaging section of the patient with no intracranial abnormality

ammonium anticholinergic agent which does not cross blood-brain barrier. It penetrates biological membrane more slowly than other anticholinergics and has very low incidence of side effects. It has been shown to be an effective treatment of Frey's syndrome.^[7]

In this case, proper therapy was instituted and hence, the patient was free from the symptoms. Above that, the patient was free from psychological distress and enjoying normal social life.

An ideal treatment would be one that could produce permanent suppression of the symptoms while offering success, minimal invasiveness, and few side effects. The literature reveals three options for treatment: surgical measures, chemotherapy, and radiation therapy. The intensity of symptoms is very important in choosing the correct treatment. Radical surgeries, such as an intracranial resection of the glossopharyngeal nerve and a Jacobson anastomosis are extremely drastic in view of the degree of disability. Drug treatment strategies involve the topical and systemic administration of anticholinergics. However, topical application can lead to skin irritation and systemic administration at the required doses could cause side effects (accommodation difficulty, the onset of glaucoma, tachycardia). In addition to causing skin irritation, topical treatment with anti-hyperhidrotics has also proved to be problematic and unpleasant for patients due to its ineffectiveness despite frequent application. von Lindern *et al.* reported that treatment with type A botulinum toxin is a promising therapeutic approach.^[2,8,10]



Figure 2: Orthopantomograph of the patient with periapical radiolucency in relation to lower central incisors and impacted lower third molars



Figure 4: Application of 1% iodine over the affected area (a), application of starch powder over the skin (b), bluish-black discoloration over the right side of face suggestive of positive starch iodine test (c)

CONCLUSION

A very rare case of post-herpetic Frey's syndrome is reported in which patient has manifested the symptoms for 7 years. The patient was diagnosed clinically and confirmed by a minor starch-iodine test. Topical application of glycopyrrolate was selected as therapy. The patient was completely free from symptoms and psychological distress allowing him to enjoy his social life in a better way. Topical application of glycopyrrolate is very safe, cost effective, and provides complete relief from the symptoms and hence should be considered as the first line of treatment approach for Frey's syndrome.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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