Volume-6, Issue-2, February - 2017 • ISSN No 2277 - 8160 IF : 3.62 | IC Value 80.26

Original Research Paper Medicine

FREY'S SYNDROME – A CASE REPORT

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ABSTRACT

Frey's syndrome is a disorder characterized by unilateral sweating and flushing of the facial skin in the area of the parotid gland occurring during meals. (Malatskey, Rabinovich, Fradis, & Peled, 2002)

Frey's syndrome is most often the sequelae of parotidectomy but also may follow other factors like post-surgical, traumatic and other inflammatory injuries of the parotid and submandibular glands and the cervical and upper thoracic portions of the sympathetic trunk. (Malatskey, Rabinovich, Fradis, & Peled, 2002)

Although most patients with Frey's syndrome have only mild to moderate symptoms around 6% of the patients may develop severe symptoms. (Malatskey, Rabinovich, Fradis, & Peled, 2002)

The present article highlights a case report and a brief overview of Frey's syndrome.

KEYWORDS : Frey's syndrome, Auriculotemporal syndrome, parotidectomy, anticholinergics.

INTRODUCTION

Frey's syndrome is a disorder characterized by unilateral sweating and flushing of the facial skin over the parotid bed following gustatory stimuli and is accompanied by pain or generalized discomfort in the region causing considerable distress and social embarrassment to the patient. (Sonny, Sunder, & Trikha, 2008)

CASE REPORT

A 46 year old male patient reported to the Department of Oral Medicine and Radiology for undergoing a routine dental checkup. Patient gave a history of slowly progressing; asymptomatic, swelling on the right middle 3rd of the face about 30 years back. It was diagnosed then as a benign parotid tumor and a superficial parotidectomy was performed. After 4 years of initial surgery, the swelling recurred which was again treated by superficial parotidectomy. The swelling recurred, again after a long remission of 9 years and this time it was treated by total parotidectomy.

After the total parotidectomy, patient had developed facial paralysis of the right side of the face which was managed by physiotherapy. After a month of total parotidectomy the patient noticed profuse perspiration preceded by a feeling of warmth on the right side of his face in the preauricular, cheek and temple region while eating food. This phenomenon consistently occurred while eating or occasionally at the thought or sight of food and stopped after food. Presently the patient continues to have the same symptoms. The residual signs of facial paralysis such as mild deviation of the right angle of the mouth are clinically evident. Wrinkling of the forehead, blowing of the cheeks and closing of the eyes showed no apparent abnormality.

On the right middle third of the face, an oblique scar is seen on the lateral side of the neck extending behind and just below the right ear lobe upto about 1 cm below the angle of the mandible along the posterior aspect of the mandible which is indicative of the scar for incision of parotidectomy.

Intra oral examination revealed that the orifice of Stenson's duct on the right side showed no flow of saliva into the oral cavity and the salivary pooling in the oral cavity appeared to be normal.

A chair-side investigation of 'The Minor's iodine starch test' was performed. The right side of the patient's face was painted with tincture iodine solution and was allowed to dry. A dry starch powder was then sprinkled in abundance over the dried iodine solution. The patient was then given concentrated juice of lemon to drink slowly, and the starch powder sprinkled on the cheek was observed closely. After about 5 minutes, the starch powder slowly started changing its colour from white to purple. The colour change from white to purple continued to become more conspicuous with increasing duration, suggestive of sweating on the right side of the face. As the patient started sweating on the right side of the face, the resultant moisture caused the dried iodine and dry starch powder to react, resulting in purple colour (Figure 1, 2).

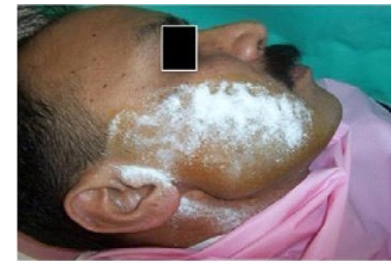

Figure 1 – Starch sprinkled on the right side of the face

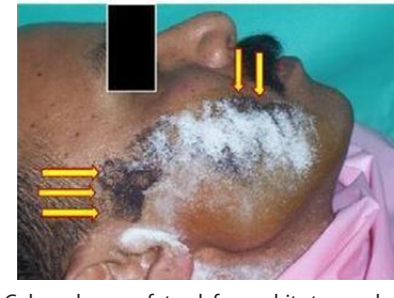

Figure 2 – Colour change of starch from white to purple

DISCUSSION

Auriculotemporal syndrome was first reported by French surgeon M. Duphenix in 1757 and later by Baillarger in 1847. This phenomenon was recognized as a distinct entity by a Neurologist Lucia Frey in 1923, who proposed the hypothesis of its pathology and since then, the syndrome bore the name. (Ahmed, 1995)

The Synonyms of Frey's syndrome are Auriculotemporal Syndrome, Baillarger's Syndrome, Frey-Baillarger syndrome, Dupuy's Syndrome, Salivosudoriparous Syndrome, Sweating Gustatory Syndrome and Von Frey's Syndrome.

This syndrome presents in a classic form, characterized by facial sweating and flushing in the distribution of the auriculotemporal nerve over the temporal area, malar region and the region of parotid gland3. The symptoms are typically unilateral, although a couple of

bilateral cases have been reported. Males and females are equally affected. In males the most obvious symptom of Frey's Syndrome is excessive sweating, while in females it is the frequency of flushing in the process of eating food. Sometimes it may be associated with accompanying pain which may last for few seconds or longer, and result either from an artificial synapse between sympathetic efferent and the sensory afferent nerves, or from traumatic neuritis of the auriculotemporal nerve (Ahmed, 1995). The onset of symptoms ranges from as early as few days after insult (degenerative concept) to late onset of months and years post operatively (regenerative concept) (Ahmed, 1995).

The clinical incidence of Frey syndrome after parotidectomy has been reported to be as high as 53%. About 30 to 50% of the patients who undergo total or partial parotidectomy report having the typical symptoms of Frey's syndrome and in approximately 15% of the patients the symptoms are severe. (Spiro, & Martin, 1967)

Though it frequently occurs as sequelae of parotidectomy, it may also occur after other surgical, traumatic and inflammatory injuries of the parotid or submandibular glands and of the cervical and upper thoracic portions of the sympathetic trunk. (Malatskey, Rabinovich, Fradis, & Peled, 2002)

The etiology of Frey's syndrome remains uncertain, but it has been noted to occur following a variety of conditions which are traumatic in nature. These include direct trauma and partial or complete parotidectomy, parotitis, central nervous system diseases such as such as syringomyelia, encephalitis and secondary to dorsal sympathectomy, subcondylar osteotomies, condylar fracture, food allergy, diabetic neuropathy and post sympathetic nerve section. (Ahmed, 1995).

There are various schools of thought regarding the pathogenesis of Frey's syndrome which include:
1. Misdirected regeneration theory. (Ahmed, 1995)
2. Sensitization theory. (Spiro, & Martin, 1967)
3. The damage to the nerve may cause destruction of sympathetic fibers leading to parasympathetic hypersensitivity and stimulation. (Sverzut, Trivellato, Serra, Ferraz, & Sverzut, 2004)
4. The damaged auriculotemporal nerve is invaded and irritated by healing tissue. (Sverzut, Trivellato, Serra, Ferraz, & Sverzut, 2004)

Of which the misdirected regeneration theory by Ford and Woodhall in 1938 is most widely accepted. This theory is based on the aberrant regeneration of sectioned parasympathetic fibers, which occurs after the loss of their connection to the parotid tissue. The resultant innervation of blood vessels and sweat glands of the skin leads to cutaneous local vasodilation and sweating during meals. (Sonny, Sunder, & Trikha, 2008).

Other most commonly accepted theories of pathogenesis are

- Sensitization Theory - The area in which the sweating occurs demonstrates a hypersensitivity to intradermally injected acetylcholine which is characteristic of the increased sensitivity that follows denervation of effector structures anywhere in the body.

Despite the fact that Frey's syndrome is a benign condition, explanation and reassurance are usually of adequate therapy. Some cases warrant treatment, especially if the syndrome causes distressful situation for the affected individual.

Various modalities of management have been suggested in literature which includes medical management and surgical intervention. The intensity of symptoms is very important in choosing the correct treatment modality. (Sverzut, Trivellato, Serra, Ferraz, & Sverzut, 2004)

Medical management primarily consists of topical and systemic administration of drugs mainly anticholinergics. The remarkable response to topical application of anticolinergic agents makes them the first treatment of choice. Anticholinergic agents act by blocking the muscarinic acetylcholine receptors on the sweat glands. (Sonny, Sunder, & Trikha, 2008).

Other modalities are topical application of 3 % scopalamine cream which is very effective with repeated administration regardless of sensitization3. Intracutaneous injections of botulinum toxin a minimally invasive therapy have been reported to have shown favourable results. (Sonny, Sunder, & Trikha, 2008).

Stellate ganglion blocks with local anaesthesia, might show significant results with repeated administration. (Ahmed, 1995)

Surgical treatment for Frey's syndrome includes resection of auriculotemporal nerve, excision of a small localized skin of the affected area, tympanic neurectomy, insertion of a graft under the involved skin and transfer of "Superficial Musculoaponeurotic System (SEAS) Application", and upper dorsal sympathectomy. (Ahmed, 1995)

Biofeedback training, hypnosis and different types of relaxation techniques have been used to treat hyperhidrosis. (Haider, & Solish, 2005).

CONCLUSION

Freys' syndrome can sometimes cause irritability and discomfort to the patient. Hence the patient needs to be taken care of so that it will not impair the normal function and improve the quality of life of the patient. Thus these patients most often need reassurance and counselling as the mainstay of the management rather than extensive medical and surgical treatment protocols.

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