Frey's Syndrome Following an Unexpected Cause

Beklenmeyen Bir Nedene Bağlı Görülen Frey Sendromu

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ABSTRACT

Frey's syndrome is a known rare complication of surgery to Otorhinolaryngologists. Here, we outline the case report of a 23-year-old gentleman who had Frey's syndrome as a complication from bilateral sagittal split osteotomy surgery.

Key Words: Frey's syndrome, bilateral sagittal split osteotomy, mandible, orthognathic

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ÖZET

Frey sendromu, Kulak Burun Boğaz uzmanları için ameliyatın bilinen nadir bir komplikasyonudur. Burada, bilateral sagital split osteotomi ameliyatından kaynaklanan bir komplikasyon olarak Frey sendromuna sahip olan 23 yaşındaki bir beyefendinin vaka raporunu özetledik.

Anahtar Sözcükler: Frey sendromu, bilateral sagital split osteotomi, mandibula, ortognatik

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INTRODUCTION

Frey's syndrome was first described by Lucie Frey in 1923 as being commonly associated with the postoperative complications of parotid surgery, characterized by gustatory sweating and flushing in response to mastication or a salivary stimulus(1,2). Also known as an auriculotemporal syndrome, it is due to aberrant reinnervation of postganglionic parasympathetic neurons to nearby denervated sweat glands and cutaneous blood vessels(3).

CASE REPORT

A 23-year-old healthy gentleman was presented to our clinic with unilateral, right facial sweating while eating, which usually occurred several minutes after starting a meal. The condition was painless, not associated with weather changes or eating spicy food. The symptom occurred even when eating plain rice. The gentleman required 2 to 3 pieces of tissue to wipe the sweat off; which were generally fully soaked after the meal.

The condition was accompanied with numbness over his right lower lips and right cheek area with occasional muscle ache at the right submental region. There was no facial asymmetry or excessive salivation and drooling. He had no history of hyperhidrosis and no family history of a similar condition. He had no history of facial trauma and was not taking any substances or medications. However, eight months before the symptoms began, he had Bilateral Sagittal Split Osteotomy (BSSO) to correct dentofacial and biting alignment abnormalities. The operation was deemed uneventful, and he was discharged home well.

Upon review in the clinic, there was no facial asymmetry. There was no visible external surgical scar. He had normal oral cavity examination with the absence of his bilateral lower 3rd molar. Other ENT examinations were also unremarkable. However, on cranial nerve examinations, the sensation of bilateral mandibular distribution of the trigeminal nerve was affected. Other cranial nerves were intact.

In the clinic, we proceeded with Minor starch iodine testing to confirm the diagnosis (Figure 1). The test was positive, the patient was diagnosed with Frey's syndrome, and was treated conservatively; as agreed by the patient.

Figure 1: Positive iodine starch test on the patient

DISCUSSION

Bilateral Split Sagittal Osteotomy (BSSO) is an important component of orthognathic surgery to correct excess, deficiency and/or asymmetry of the horizontal mandible(4). BSSO, which aims to free the segment and place it in its desired position, involves soft tissue dissection and osteotomies of the mandible.

In our patient, soft tissue dissection was done from the first premolar extended up to the wisdom tooth. Meanwhile, lateral osteotomy and screw fixation was done in between the first and second molars of the mandible bilaterally. Postoperatively, the sensation of the bilateral lower lip and chin were affected, which indicated the involvement of the mental nerve; a branch of the inferior alveolar nerve. Anatomically, the inferior alveolar nerve gives off to the mental nerve at about the level of the mandibular second premolar; which exits the mandible via mental foramen to supply the sensory branches to the chin and lower lips(5). Therefore, we were speculating that the injury to the mental nerve was at the area of the mental foramen.

The right auriculotemporal nerve was also affected with the presence of Frey's syndrome on the right lateral cheek. In this case, the damage was possibly in the area of the right parotid gland. During the procedure, the mandible or the osteotome might have been pushed backwards during manipulation and pressed against the parotid gland; hence, causing damage to the gland(6).

Monson LA (2013) describes in the literature, that complications related to BSSO include bleeding from injury to the inferior alveolar artery or mesenteric artery, unanticipated fractures and unfavourable splits, avascular necrosis, condylar resorption, malposition of the proximal segment and worsening of temporomandibular joint (TMJ) symptoms. The diagnosis of Frey's syndrome can be done clinically from history as the symptoms include flushing, sweating, burning, neuralgia, and itching[1], and confirmed by Minor starch-iodine testing.

For management, medical treatment can be given via injection therapy or topical application, which involves topical antiperspirants or injection of alcohol, scopolamine, and glycopyrrolate or botulinum toxin A (BTA)(1). BTA, which is the current treatment of choice, can be given as an intradermal injection and may require repeat injections to control the symptoms.

Surgical management, which may be used in refractory cases, is described as a surgical transection of auriculotemporal nerve, tympanic nerve and greater auricular nerve. However, it is not commonly practiced.

CONCLUSION

This is the case study report of the rare complication of Frey's syndrome in a gentleman with bilateral sagittal split osteotomy. The diagnosis was clinically confirmed with starch iodine testing. Further data and study are required for the evaluation of the association and mechanism of the injury.

Conflict of interest
No conflict of interest was declared by the authors.

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