

An Uncommon Cause of Facial Ulcers: Trigeminal Trophic Syndrome

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Abstract

Background: Trigeminal trophic syndrome (TTS) is one of the rare causes of ulceration over the face, which occurs due to self-manipulation or rubbing of the facial skin after an injury to the trigeminal nerve. **Case Report:** We report a case of trigeminal trophic syndrome in a 60 years old lady, known case of trigeminal neuralgia, who presented with ulcer and swelling of the right side of the face. She was treated, counseled and is being followed up. **Conclusion:** Trigeminal trophic syndrome, though a rare complication, should be suspected as a cause of unilateral facial ulceration in a patient with trigeminal neuralgia.

Keywords: Case Report, Pain, Skin Diseases, Trigeminal neuralgia, Ulcer.

Introduction

Trigeminal trophic syndrome which was previously called “Trigeminal neurotrophic ulceration” is a rare condition which presents as ulcer in the dermatome of the trigeminal nerve as a consequence of anaesthesia of the dermatome, either due to a procedure in the nerve, done to relieve the pain of trigeminal neuralgia or due to compression of the nerve by a vessel, which in turn alters the structure of the nerve.

Case Report

A 60-year-old woman presented with sharp, shooting, intermittent pain on the right side of the face, which was aggravated on touching the face or on exposure to cold water or air, relieved spontaneously after 2-3 minutes. She had developed ulcer on the right cheek near the nasolabial fold over the past one month. This was accompanied by swelling of the right cheek and right side of the upper lip. There was history of vigorous rubbing of the right side of the face, during each episode of sharp, shooting pain.

She is a known case of trigeminal neuralgia, which was diagnosed in 2018, when she had similar pain. Electromyography and magnetic resonance imaging (MRI) done in 2018 were normal. She was treated for 8 months as trigeminal neuralgia, after which she had discontinued the treatment and had been asymptomatic for the past 2 years. Pain recurred in January 2021 during which MRI brain was repeated. MRI showed the right superior cerebellar artery impinging the right trigeminal nerve at the root entry zone.

On examination, there was a superficial ulcer of size 2×2 cm over the right side of the face, near the nasolabial fold. The right side of the upper lip appeared swollen. However, there were no ulcers in the oral mucosa [Fig.1]. Ulcer was treated with oral and topical antibiotics and potassium permanganate soaks. Amitriptyline was also added. She was started back on tablet carbamazepine for trigeminal neuralgia and dose was titrated based on symptoms. She improved symptomatically and is being followed up.

Discussion

Trigeminal trophic syndrome is a rare condition, which is a consequence of damage to the trigeminal nerve either due to surgical procedures like nerve ablation or due to alteration in the structure of the nerve due to impingement by a vessel or tumor. It was first noticed in the 20th century, by neurosurgeons after doing trigeminal rhizotomies in patients with trigeminal neuralgia [1]. There are reports of similar occurrences in patients who had undergone cyberknife radiosurgery for trigeminal neuralgia [2]. TTS is also found to occur in patients with herpes zoster, herpes simplex, Bell's palsy, syphilis and leprosy [3].

TTS is characterized by a triad of anesthesia, paresthesia and recurrent facial ulceration. The average time duration between nerve injury and the development of ulcer is 2 years. Patients present with history of frequent rubbing of the face due to the sharp shooting pain that develops. Since the ulcer is painless, patients tend to rub it more, which makes the ulcer more persistent [4]. The management includes, management of the underlying condition, trigeminal neuralgia with carbamazepine and treatment of the ulcer with antibiotic creams and oral antibiotics. Carbamazepine at the dose of 200 mg three times a day, has been found to be effective in curing the ulcer as well [5]. Occlusive dressings have found to be effective to hasten healing and to prevent recurrence.

Conclusion

Trigeminal trophic syndrome should be suspected in a patient with history of trigeminal neuralgia, who presents with ulcer over the face, with typical history of frequent rubbing of the face, associated



Fig.1: Clinical picture of our patient.

with sensory loss over that dermatome. Early diagnosis and treatment will prevent worsening and recurrence of the ulcer.

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