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The Wounds of a Past Insult! - A Rare Case Report

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Abstract

Trigeminal trophic syndrome is a rare disease presenting with unilateral painless persistent facial ulcers which occurs after an insult to trigeminal nerve. Patients typically present with persistent ulceration, paraesthesia, numbness, or self-manipulation of the affected area. We report a case of 40-year-old male who was diagnosed as trigeminal trophic syndrome with atypical presentation. The diagnostic process required careful exclusion of infectious, inflammatory, and neoplastic causes of chronic facial ulceration. A multidisciplinary approach was employed for accurate diagnosis and effective management. The patient showed significant improvement with a combination of protective measures, behavioral modification, and pharmacological therapy.

This case highlights the importance of early recognition of TTS and the role of coordinated multidisciplinary care in optimizing outcomes.

Keywords: Trigeminal trophic syndrome; Herpes zoster

1. Introduction

Trigeminal trophic syndrome (TTS) is a rare disorder occurring after an insult to trigeminal nerve, resulting in self-manipulation of the skin. The syndrome consists of a triad of anaesthesia, paraesthesia, and a secondary persistent or recurrent facial ulceration [1]. It was first described as a cutaneous ulceration in the trigeminal dermatome by Wallenberg, in 1901, and later by Loveman in 1933 [2,3].

The etiology typically includes trauma, infections such as herpes zoster, neurosurgical procedures, or vascular insults that disrupt trigeminal pathways. Clinically, patients may present with unilateral, well-demarcated, non-healing ulcers often accompanied by abnormal sensations leading to repetitive rubbing or picking. Management requires accurate identification of

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the underlying nerve injury and a multidisciplinary approach incorporating protective measures, behavioral modification, and medical therapy. We report an atypical presentation of trigeminal trophic syndrome following herpes zoster ophthalmicus.

2. Case Report

A 40-year-old male patient presented with complaints of multiple non healing, persistent painless raw areas over right side of the forehead and scalp associated with tingling sensation secondary to which the patient was constantly scratching and picking the skin since 1 month. There was a history of herpes zoster ophthalmicus right side 3 months back for which the patient was treated and healed completely. The patient had no history of immunodeficiency, chronic systemic illness, or smoking.

On examination, there were multiple well defined, sharply demarcated superficial ulcers, few having crescent shape largest measuring 3 cm × 2 cm × 0.5 cm and smallest 1 cm × 1 cm × 0.1 cm, distributed over right half of forehead and scalp in the distribution of ophthalmic branch of trigeminal nerve. No vesicles/ hypo pigmented patches around the ulcers. Neurological examination revealed absent sensation over ophthalmic division of trigeminal nerve of right side. Corneal reflex was diminished over right side. Examinations of other cranial nerves were normal. General physical, systemic and neurological examinations were normal.

Tzanck smear was done from the ulcer which didn't show any acantholytic or multinucleate giant cells. Slit skin smear was done which was negative. ANA, Hepatitis B, C, VDRL, HIV were negative. Routine blood investigations were within normal limits. ECG, Chest X ray, CT scan and MRI of the brain didn't reveal any abnormality.

Patient was diagnosed to have trigeminal trophic syndrome after excluding other causes. The ulcers were treated with foam dressing, topical antibiotics mupirocin and colloidal silver. Psychiatry consultation was given and was treated with pregabalin 75 mg OD, diazepam 5mg OD for a month along with counselling of the patient and behaviour modification. The ulcers healed in 3 weeks; the patient is being followed up and there is no recurrence since a year.



FIG. 1. Image of the patient with multiple well defined, sharply demarcated superficial ulcers, few having crescent shape largest measuring 3 cm × 2 cm × 0.5 cm and smallest 1 cm × 1 cm x 0.1 cm, distributed over right half of forehead and scalp in the distribution of ophthalmic branch of trigeminal nerve.



FIG. 2. Image of the patient after 3 weeks of treatment showing healing of ulcers.

3. Discussion

Trigeminal trophic syndrome is a rare complication after an injury to trigeminal nerve either in central or peripheral pathway. Cases are most commonly reported in elderly women [4]. The pathogenesis is poorly understood. The common dermatological causes are Hansen's disease and herpes zoster [5,6]. Bell's palsy, post encephalitis, acoustic neuroma, vertebra-basilar insufficiency, following treatment for trigeminal neuralgia which includes injection of alcohol to Gasserian ganglion, Trigeminal rhizotomy, surgical ablation, amyloid deposits in CNS and trigeminal nerve [2] are the other possible etiological factors.

Clinical features include formation of crescent shaped ulcer from small crusts [2]. The common sites of ulcers reported are sides of the nose including alae nasi, cheeks, lips, scalp, eyes, inside of mouth, forehead, and jaw [7]. There will be characteristically sparing of the nose [2]. The ulcers are usually multiple, painless and unilateral. Anaesthesia and paraesthesia of the affected area led to constant rubbing, scratching, and picking leading to persistent non healing ulcers. The duration between trigeminal nerve injuries to ulceration varies from weeks to years, average of 2 years [6].

The differential diagnosis to be considered includes dermatitis artefacta and compulsive skin picking which lacks identifiable neurological abnormality. Basal cell carcinoma, squamous cell carcinoma, destructive lethal midline granuloma, syphilis, varicella zoster or herpes simplex virus infection, leishmaniasis, leprous trigeminal neuritis, yaws, blastomycosis, paracoccidioidomycosis, pyodermagangrenosum, Wegener's granulomatosis, tuberculosis, arteritis temporalis [7] are to be differentiated.

This condition is diagnosed clinically. Histopathological examination doesn't have any diagnostic value [2].

Treatment is difficult, a multidisciplinary approach is needed. Proper patient education, behavioural modification, occlusive wound dressings, topical antibiotics are used. Surgical reconstruction and transcutaneous electrical nerve stimulation may be beneficial [8]. Drugs which are used with varying success include diazepam, carbamazepine, vitamin B supplementation, pregabalin, chlorpromazine, amitryptalline [8,9].

4. Conclusion

Trigeminal trophic syndrome should be considered in patients with persistent, unilateral, non-healing facial ulcerations. Early recognition and timely intervention are crucial to prevent progressive tissue damage. This case is presented to highlight its rarity, atypical presentation, and the need for greater clinical awareness for prompt diagnosis and management.

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