



Trigeminal Trophic Syndrome

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Abstract

Trigeminal Trophic Syndrome (TTS) is a rare, well-documented neurocutaneous condition characterized by the triad of paresthesia, analgesia, and ulcerations caused by self-manipulation, most commonly affecting the ala nasi. It typically arises following injury to the trigeminal nerve, either centrally or peripherally.

We report the case of a 75-year-old woman who developed TTS following a cerebral stroke. Initially, her symptoms were misinterpreted as a psychiatric disorder, but the diagnosis of TTS was ultimately made based on the combination of prior trigeminal nerve injury and the classical symptom triad. This case highlights the diagnostic challenges of TTS and emphasizes the need to consider neurological etiologies in patients with atypical facial ulcerations.

Introduction

Trigeminal Trophic Syndrome (TTS) is a rare condition characterized by lesions in the area of the face supplied by the trigeminal nerve, typically resulting from central or peripheral damage to the trigeminal nerve^{1,2}. Fewer than 200 cases have been described in the medicine literature³.

The hallmark presentation of TTS typically involves a unilateral, crescent-shaped ulcer located over the ala nasi. However, lesions may also appear in other regions innervated by the trigeminal nerve, including the scalp, forehead, cheeks, ears, and even the hard palate¹⁻⁵. The pathogenesis of TTS is directly linked to damage to the trigeminal nerve (cranial nerve V), resulting in sensory deficits such as analgesia in the affected area. This loss of sensation often leads to abnormal sensory experiences—burning, itching, or tingling paresthesias. Patients may misinterpret these sensations as nasal congestion due to disrupted sensory feedback^{5,6}.

The sensory deficit associated with trigeminal trophic syndrome (TTS) plays a central role in the development of atrophic lesions, which are perpetuated by a cycle of self-inflicted trauma. Patients frequently engage in unconscious or compulsive manipulation of the affected area, exacerbating both the severity and persistence of the ulcers. Although some individuals may recognize their behavior, many remain unaware of the extent of their self-mutilation, which may be driven by either conscious or subconscious mechanisms. These lesions often become chronic and refractory, with some cases persisting for decades despite patient awareness of the underlying cause¹.

TTS most commonly occurs following trauma to the trigeminal nerve, such as in cases of cerebral infarction (stroke), trigeminal

rhizotomy, or alcohol injections administered for trigeminal neuralgia. Less frequently, TTS may be triggered by physical trauma, herpesvirus infections, acoustic neuromas, or as a postoperative complication of craniotomy^{4, 5}. The onset of symptoms can range from as early as two weeks to several decades after the initial nerve injury. The condition predominantly affects older adults, with a higher prevalence among women^{4,5}.

Case Presentation

A 75-year-old woman was admitted to a psychiatric ward. She had experienced several years of itching, pain, and ulceration on the left side of her face (Figure 1 and Figure 2). Her medical history included recurrent depressive episodes, multiple psychiatric hospitalizations, and a prior cerebral stroke, which had resulted in sensory deficits on the left side of her face.



Figure 1: Clinical presentation of a 75-year-old patient showing ulceration and tissue loss on the left side of the face, with complete destruction of the left ala nasi

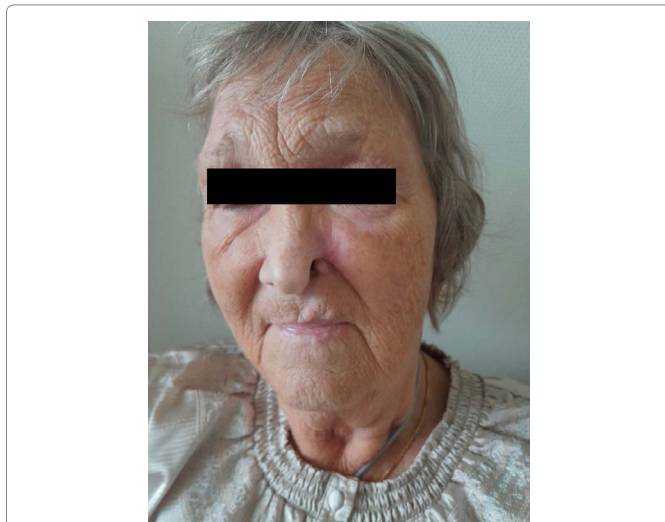


Figure 2. lacks follow-up photos after the ulcer healing

Following the stroke, the patient reported shooting pains and intense pruritus, primarily affecting the left ala nasi. For several years, these symptoms were attributed to her psychiatric condition, delaying appropriate evaluation. A comprehensive dermatological and neurological assessment was subsequently initiated and brain MRI demonstrated post-infarct sequelae consistent with prior cerebrovascular insult. Symptoms began several months after the cerebrovascular event. Earlier assessments interpreted the symptoms as psychiatric in origin, contributing to delayed diagnosis. The manipulation appeared largely unconscious and driven by neuropathic discomfort. Differential diagnoses considered included malignancy, vasculitis, infection, and factitial dermatitis.

The patient was started on gabapentin at 300 mg daily, titrated to 900 mg daily.

Discussion

This case highlights the complex diagnostic challenges that can arise when trigeminal trophic syndrome (TTS) is overlooked. The patient, with a history of cerebrovascular disease and psychiatric disorders, experienced persistent pain, pruritus, and ulceration on the left side of her face, particularly around the ala nasi. For years, these symptoms were attributed solely to her psychiatric condition, delaying accurate diagnosis and effective management. A thorough dermatological and neurological assessment ultimately led to the identification of TTS, emphasizing the importance of interdisciplinary collaboration in recognizing and diagnosing this rare condition. Differential diagnoses considered included malignancy, vasculitis, infection, and factitial dermatitis. Trigeminal denervation disrupts normal sensory feedback, leading to neuropathic sensations and self-induced ulceration.

It can be difficult to diagnose TTS. Patients are often referred to an otologist specialist due to bloody nasal discharge, an ophthalmologist due to suspicion of a tear duct disorder, an oncologist due to suspicion of basal cell carcinoma, and a neurologist for pain management.

The triad of paresthesia, analgesia, and ulceration particularly in the context of a relevant neurological history should prompt immediate consideration of TTS. In this case, the patient's history of stroke, which resulted in sensory deficits on the left side of her face, provided a key clue to the underlying pathophysiology. While psychiatric disorders such as depression can present with somatic symptoms, including chronic pain or itching, it is essential that these complaints are not prematurely attributed solely to psychiatric causes without thorough investigation.

Conclusion

Trigeminal Trophic Syndrome (TTS) is a condition of particular importance to clinicians in general practice,

plastic surgery, and psychiatry, as it can easily be mistaken for malignancy or a primary psychiatric disorder. Early recognition is essential. A dermatologist to ensure accurate diagnosis and appropriate treatment should evaluate patients presenting with chronic facial ulceration and sensory disturbances. Early multidisciplinary collaboration can reduce diagnostic delay and morbidity.

Treatment of TTS is multifaceted, requiring both pharmacological and, in some cases, behavioral interventions. Patient education and engagement are critical. Pharmacological therapy may include neuroleptics to manage underlying neuropathic symptoms. In cases with significant tissue loss, referral for plastic surgical reconstruction may be warranted. Alternatives such as carbamazepine, pregabalin, and behavioral strategies may also be beneficial.

Patient Consent Statement

The authors obtained written consent from the patient for the publication of their photographs and medical information to be published.

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