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# SELF-INDUCED ULCERATION OF ALA NASI IN TRIGEMINAL TROPHIC SYNDROME – OPERATIVE PSYCHIATRIC APPROACH

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SUMMARY – Self-induced ulceration of ala nasi in trigeminal trophic syndrome is a rare complication secondary to trigeminal nerve peripheral or central lesion, which occurs as a sequel of anesthesia and facial paresthesias associated with compulsive touching of the nose. Two patients with trigeminal trophic syndrome associated with facial and statoacoustic nerve lesion following statocoustic neurinoma surgery are described. Successful ala nasi reconstruction with psychiatric treatment for prevention of compulsive nose touching was performed in one patient.

Key words: *Skin ulcer – etiology; Skin ulcer – surgery; Skin ulcer – psychology; Reconstructive surgical procedures – methods; Nose; Trigeminal nerve injuries; Self mutilation psychology*

## Introduction

Trigeminal trophic syndrome (TTS) is a rare clinical entity that most commonly occurs in elderly patients with trigeminal neuralgia. The syndrome has also been described in patients with brain tumors, vertebrobasilar circulation disorders, following meningoencephalitis, and brain trauma, whereas the cause of TTS may occasionally remain obscure. Most TTS patients complain of facial pruritus, tingling and burning sensation, redness and insensitivity that lead to the development of neurotrophic ulcers, mostly on the wing of the nose[1-4]. The diagnosis of TTS is based on typical history of nerve lesion and pathognomonic crescent ulceration of ala nasi, generally caused by compulsive touching of the nose as an obsessive-compulsive disorder. The latent period for the occurrence of ulcer in TTS may range from 2 to 60 months, while the mean time from ulceration to the diagnosis of TTS is 6 months[5,6]. Surgical therapy for ala nasi ulceration in TTS is extremely difficult and can only be performed in highly motivated patients, whereby patient education on the nature of the disease is of utmost importance[7].

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## Case Report and Ala Nasi Reconstruction

***Case 1.*** A 67-year-old male, operated on for left-sided statocoustic neurinoma at age 55. In addition to facial skin and eye lesions, a nasal wing defect developed two years after the surgery. The patient also had left-sided facial palsy, loss of hearing, and left eye areflexia (Fig. 1).

***Case 2.*** A 32-year-old male, operated on for statoacoustic neurinoma at age 22. Clinically, complete defect of the right nasal wing, the rest of the facial skin dry, reddish and scaly; severe keratoconjunctivitis of the right eye; facial nerve lesion of peripheral type; loss of hearing and right ear areflexia. Ala nasi ulceration occurred as early as two months postoperatively, whereas complete defect developed in two years. The patient was treated at psychiatry on several occasions for depression following cerebral neurinoma surgery and TTS.

Both patients denied compulsive touching of the nose, yet the younger one reported he was disturbed by his nose appearance. Both patients were proposed surgical reconstruction of the nasal wing defect. Only the younger patient gave his consent for the operation, as he was highly motivated for the procedure after a number of psychiatric treatments.

As the right side of the face was neurologically dead, the island transposition flap from the intact side of the

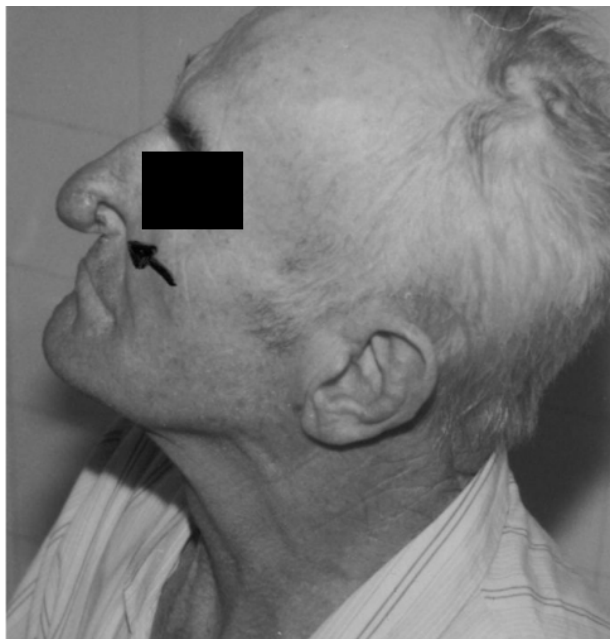

*Fig. 1. The elderly patient with trigeminal trophic syndrome; the arrow denotes the right nasal wing defect.*

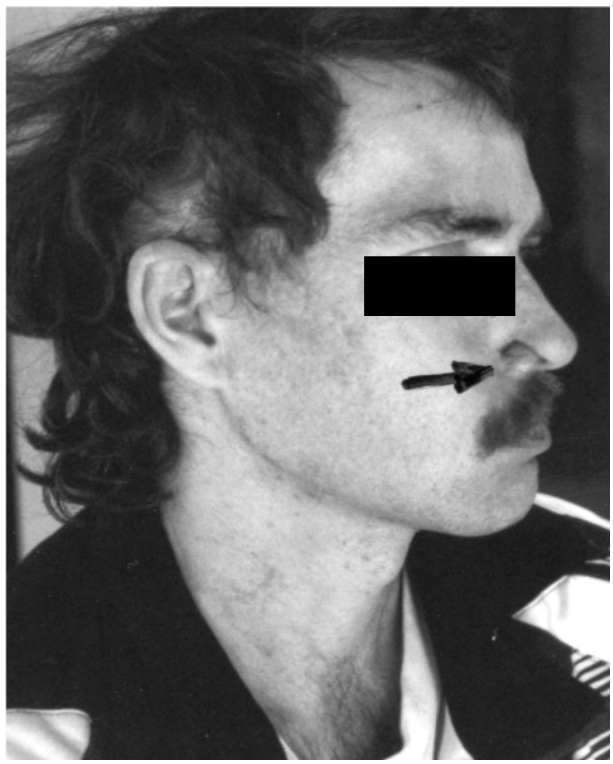

*Fig. 2. The younger patient at four years of reconstructive surgery; the arrow denotes the "new" nasal wing.*

forehead, including the supratrochlear artery with accompanying nerve, was used on the reconstruction. The nasal wing thus formed is independently innervated and vascularized, thus being independent of the recipient region. Four years after the procedure, there was no ulcer recurrence, while the patient has been under regular psychiatric control (Fig. 2).

## Discussion

Trigeminal trophic syndrome as a clinical entity was first described in 1933 in patients suffering from trigeminal neuralgia and treated with alcohol injections into the gasserian ganglion region or by surgical nerve dissection. TTS due to other factors such as vertebrobasilar insufficiency, tumors or degenerative brain diseases is less commonly reported[1-6]. Our search of Anglo-Saxon literature 1982-2003 yielded 102 TTS patient reports, including our two patients with facial and statoacoustic nerve lesion in addition to trigeminal nerve lesion, which has not been found in other TTS reports[4,8-10]. Interestingly enough, only three patients with TTS due to a different mode of treatment for trigeminal neuralgia were described in Japan until 1991[12]. The treatment of TTS is difficult and rarely successful, from various measures of self-care, physical therapy, prosthetic management, through surgical reconstruction of ala nasi defects[13-15]. The initial therapeutic success is frequently followed by ulcer recurrence due to repeat self-mutilation, which occurs as a compulsive disorder in most patients. Tollefson *et al.* describe five reconstructive procedures with four relapses within 1 to 58 months of reconstruction, caused by repeat self-mutilation during the 1985-1997 period[2]. On differential diagnosis of nasal wing ulceration, basal cell carcinoma should be excluded first. In seven TTS patients treated at Mayo Clinic until 1997, chronic inflammation but not malignancy was confirmed by multiple biopsies[11].

As TTS is associated with severe physical and cosmetic handicap, especially in young patients, it also entails changes in the patient's mental state. It is only persistent psychiatric treatment that can motivate the patient to undergo operative treatment, as it is the only way to prevent self-mutilation and ulcer recurrence. Independently innervated and vascularized tissue that does not depend on the recipient region should be used on reconstructive surgery[7,14].

## Conclusion

The treatment of self-induced nasal wing ulceration in TTS requires team approach by a surgeon and a psychiatrist, to achieve high patient motivation for the operative procedure and eliminate the tendency to self-mutilation. The tissue to be used in the reconstructive procedure should not be dependent on the recipient region.

## References


1. MONRAD SU, TERREL JE, ARONOFF DM. The trigeminal trophic syndrome: an unusual cause of nasal ulceration. J Am Acad Dermatol 2004;50:949-52.
2. TOLLEFSON TT, KRIET JD, WANG TD, COOK TA. Self-induced nasal ulceration. Arch Facial Plast Surg 2004;6:162-6.
3. SAGEGHI P, PAPAY FA, VIDIMOS AT. Trigeminal trophic syndrome – report of four cases and review of the literature. Dermatol Surg 2004;30:807-12.
4. ELLOUMI-JELLOULI A, BEN AMMAR S, FENNICHE S, ZGHAL M, MARRAK H, MOKHTAR I. Trigeminal trophic syndrome: a report of two cases with review of literature. Dermatol Online J 2003;9:26.
5. VINKE JG, HOORWEG JJ, De HAAS ER, POUBLON RM. Trigeminal trophic syndrome: 2 patients with a non-healing ulcer on the ala nasi. Ned Tijdschr Geneeskd 2003;147:1866-9.
6. LITSCHEL R, WINKLER H, DAZERT S, SUDHOFF H. Herpes zoster-associated trigeminal trophic syndrome: a case report and review. Eur Arch Otorhinolaryngol 2003;260:86-90.
7. MIKO K, BAUER K, SCHOP RE. Neurotrophic ulcer as late sequela of trigeminal nerve excision. Hautarzt 2001;51:1043-4.
8. LYON CC, MUGHAL MZ, MUSTON HL. Herpetic trigeminal trophic syndrome in an infant. J R Soc Med 2001;94:135-7.
9. FERRARA G, ARGENZIANO G, CICARELLI G, CUSANO F, DELFINO M. Post-apopletic trigeminal trophic syndrome. J Eur Acad Dermatol Venereol 2000;15:153-5.
10. DATTA RV, ZEITOUNI NC, ZOLLO JD, LOREE TR, HICKS WL Jr. Trigeminal throphic syndrome mimicking Wegener's granulomatosis. A case report with a review of the literature. Ann Otol Rhinol Laryngol 2000;109:331-3.
11. DICKEN CH. Trigeminal trophic syndrome. Mayo Clin Proc 1997;72:543-5.
12. TADA J, UEDA M, YOSHIKO A, FUJIWARA H, ARAKAWA K, ARATA J. Trigeminal trophic syndrome – a report of three patients. J Dermatol 1991;18:613-5.
13. MAYER RD, SMITH NP. Improvement of trigeminal neurotrophic ulceration with pimozide in a cognitively impaired elderly woman – a case report. Clin Exp Dermatol 1993;18:171-3.
14. McLEAN NR, WATSON AC. Reconstruction of the defect of the ala nasi following trigeminal anaesthesia with an innervated forehead flap. Br J Plast Surg 1982;35:201-3.
15. WESTERHOF W, BOS JD. Trigeminal trophic syndrome: a successful treatment with transcutaneous electrical stimulation. Br J Dermatol 1983;108:601-4.



Sažetak

SAMOIZAZVANA ULCERACIJA NOSNOG KRILA U TRIGEMINALNOM TROFIČNOM SINDROMU – KIRURŠKO PSIHIJATRIJSKO LIJEČENJE

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Samoizazvana ulceracija nosnog krila u okviru trigeminalnog trofičnog sindroma rijetka je komplikacija koja nastaje nakon periferne ili središnje ozljede trigeminalnog živca, kao posljedica anestezije i parestezija lica udruženih s prisilnim diranjem nosa. Opisuju se dvojica bolesnika s trigeminalnim trofičnim sindromom i ozljedom ličnog i statoakustičnog živca nakon operacije statoakustičnog neurinoma. Kod jednog bolesnika učinjena je uspješna rekonstrukcija nosnog krila u kombinaciji s psihijatrijskim liječenjem radi sprječavanja prisilno uvjetovanog diranja nosa.



Ključne riječi: *Kožni ulkus – etiologija; Kožni ulkus – kirurgija; Kožni ulkus – psihologija; Rekonstrukcijski kirurški zahvati – metode; Nos; Ozljede trigeminalnog živca; Psihologija samoozljeđivanja*