

TRIGEMINAL TROPHIC SYNDROME (A case report with review of literature)

K. M. ACHARYA* AND B. H. SHAH †

Summary

A rare case of trigeminal trophic syndrome in an adult male is reported and the literature on the same is reviewed.

Trophic lesions in the trigeminal field, described as a complication of pathological processes involving the first and second neurones, are surprisingly uncommon after surgical division of the trigeminal nerve for relief of trigeminal neuralgia. Still uncommon are trophic lesions on the forehead, scalp, and malar regions.

Herewith, we are reporting a rare case of trigeminal trophic syndrome in a 35 years old male with trophic changes on the left side of the scalp and forehead following left intracranial trigeminal sensory rhizotomy.

Case Report

A 35 years old male clerk was admitted on 6—12—'77 to the New Civil Hospital, Ahmedabad, for treatment of nephrotic syndrome of 8 years' duration. The following day, patient was referred to the Skin O. P. D., for the lesions on the scalp and forehead of 2 years' duration.

A detailed history revealed that patient had dull pain in the left second upper molar tooth in 1972. As the intensity of pain increased he went to the Dental Hospital where a "decayed tooth" was extracted. The dull ache, however persisted and kept increasing in intensity. He later began to get intractable and lightening pain with numbness which started in the left supra-orbital region. Although patient later developed pain all over the face it was intolerable on the left side, where it was lancinating leading to inability to swallow or chew. Paroxysms of pain were precipitated, particularly, during shaving or opening the mouth. Within two months the frequency of pain increased from 2 to 3 times a day to 20 to 30 times a day. Patient consulted a neurosurgeon who diagnosed trigeminal neuralgia and advised tab. tegretol; to start with one tablet which was stepped up to three times a day. Tab. Epsolin was also given t. i. d. After three months of treatment patient had not improved and he was admitted to the hospital under another neurosurgeon on 16—4—'75. At the time of admission, routine examination of blood, urine, stool, x-ray chest & skull (A. P., lateral, base & Town's view) were all normal.

On 23—4—'75, patient had left intracranial trigeminal sensory rhizotomy.

* Assistant Professor and Head of the Deptt. of Dermatology, M. P. Shah Medical College and Irwin Group of Hospitals, Jamnagar

† Senior Professor and Head of the Deptt. of Dermatology, B. J. Medical College and New Civil Hospital, Ahmedabad

Received for publication on 8—4—1978

Pain completely disappeared immediately after operation. On the third post-operative day, patient developed ulceration on the left forehead and scalp in the distribution of the ophthalmic branch of the trigeminal nerve. A dermatologist advised neosporin ointment and patient was discharged ten days after the operation.

When the patient attended the Skin O. P. D. of Civil Hospital, Ahmedabad, he complained of tingling and numbness of the skin in the distribution of all three divisions of the left trigeminal nerve. Examination revealed weakness and wasting of the temporalis muscle, weakness of the masseter muscle, complete loss of superficial sensations, in the distribution of left trigeminal nerve, and absent left corneal reflex. Taste sensation and autonomic functions were normal. Rest of the C. N. S. examination was normal. Clinical examination of the eye and funduscopy did not reveal any abnormality. Investigations at this time showed Haemoglobin-8.5 gms%, E. S. R. - 50 m.m./hour, trace of albumin in urine, blood urea was 30mgms%, serum cholesterol-696 mgms%, serum creatinine 1.1 mgm%, V. D. R. L. - non reactive and x-ray skull (A. P., lateral, and Town's view) — normal. Total & differential count and stool examination were normal.

Examination of skin showed a well defined plaque, 1.5 x 1.5 cms. with a hyperpigmented border, ulceration and crusting, a smaller plaque, 1 cm. x 1 cm, just lateral to the above and also an alopaecic plaque on the left side of the scalp, 2 cms. x 2 cms., with ulceration and crusting.

(Fig. No. 1 & 2).

Excepting for a mild hypertension (B. P. - 140/100 m.m. of Hg.), systemic examination was normal.

Discussion

The most common cause of trigeminal denervation is surgical transection



Fig. 1 Showing trophic changes in the ophthalmic branch of left trigeminal nerve

of the sensory root done as a therapeutic measure for trigeminal neuralgia^{1,2}. Trophic lesions commonly beginning in the ala nasi and in rare instances on the forehead scalp, malar regions following minor trauma to the anaesthetic skin within the trigeminal area have been reported^{3,4,5}. The interval between operation and the appearance of trophic lesions may vary from few weeks to several years.



Fig. 2 Showing trophic changes in the ophthalmic branch of left trigeminal nerve

The portion of the face which is supplied by the trigemenuis may be denervated by various procedures viz, sectioning of its peripheral branches, the gasserian ganglion, the posterior root or more selectively cutting the descending spinal tract in the medulla. Opinions vary regarding the observation that trophic changes did not occur if sensory root was completely destroyed⁶. In a review of 107 cases, Becker⁷ was able to find only 5 cases in which cutaneous complications had developed after trigeminal neurectomy. He himself reported the case of a 47 years old female who developed erythematous, vesicular eruptions after surgery. In another report on 587 cases of trigeminal neuralgia treated by surgery, trophic changes were not mentioned⁸. Harris^{9,12} in 1940 studied a series of 1433 cases of trigeminal neuralgia and stated that trophic lesions may occur "Very occasionally". In 1940, Karnosh and Scherb reported three cases with trophic lesions in trigeminal area⁶. All these cases were studied by competent dermatologists who found no explanation for the appearance of the lesions except a neuropathic one. Among these three cases one patient had developed trophic lesions as a result of occlusion of the posterior inferior cerebellar artery. The authors stated that trophic changes are uncommon on the face because it is instinctively better protected from trauma than other parts of the body which become anesthetic.

Surgical division of the trigeminal nerve can give rise to a number of complications (post-operative). The commonest is herpes simplex occurring in the anaesthetic area and some surgeons look for this as the hallmark of a successful operation. Neuropathic keratitis is the second commonest complication of trigeminal sensory rhizotomy. Dott reported the value of cervical sympathectomy, both in prevention and in the treatment of neuropathic keratitis^{1,2}. Vasomotor distur-

bances have been reported only after injection of alcohol into the gasserian ganglion or after electro-coagulation.

Spillone and Wells stated, "the rarest complication of which we are aware of in surgical denervation of the skin of the face is the ulceration en arc". In a study of 16 cases with trigeminal neuropathy, they found one case with ulceration of the right ala nasi which developed 12 years after sensory root section. McKenzie found two cases with ulcerations of the nose among 78 patients surgically treated for trigeminal neuralgia. In a third case the lesions appeared after the removal of an acoustic neuroma. Loveman¹⁰ reported a case with ulcers on the forehead and alopecia. Jaeger¹¹ reported three cases in 1950. Howel² reported three cases with ulcerations of the ala nasi in 1955.

Trophic lesions of the face as a sequel to the interruption in the trigeminal sensory pathway within the brain-stem have been reported by Karnos and Scherb⁶ (1940) and Savitsky and Elpern¹³ (1948). The earliest report was that of Wallenberg himself in 1901, who had first described the "Posterior inferior cerebellar artery syndrome". His patient, a 55 years old female had trophic lesions of the ala nasi and right side of the nose. Rosenberg and Solovay¹⁴ reported two cases in which trophic lesions occurred as a complication of post encephalitic parkinsonism.

Neuropathic changes in trigeminal areas follow diseases, injury, or surgery which destroys fibres conveying pain and temperature sensations^{3,5}. Trauma to the analgesic face is probably the most frequent precipitating cause for the occurrence of the ulcers.

The differential diagnosis in this condition are basal cell epithelioma, post radiation ulcers and dermatitis artefacta.

The diagnosis is obvious when history is adequate^{3,4}.

Prevention of trauma and protective dressings of existing lesions are the mainstay in the management of the disease. Even binding the limbs may be necessary⁴. Secondary infection may be treated with antibiotics. Prosthesis has been found helpful as a protective measure³.

Acknowledgment

We are thankful to Dr. D. N. Chhatrapati, Superintendent, New Civil Hospital, Ahmedabad for allowing us to publish this paper.

References

1. Spillane JD, Wells CEC : Isolated trigeminal neuropathy, *Brain* 82 : 391, 1959.
2. Howell RG : Trophic changes in the skin after operations on the trigeminal nerve : *Bri J Derm*, 67 : 444, 1955.
3. Rook A, Wilkinson DS, Ebling FJG : Text book of Dermatology : 2nd Ed, Blackwell Sci Publ, Oxford, London, 1972, p1808.
4. Demis DJ, Dobson RL, McGuire J : Clinical Dermatology : 2nd Ed, Harper and Row Publ, New York, London, Vol. IV 1975.
5. Brain, Walton JN : Diseases of the nervous System, 7th Ed, E.L.B.S., London, 1975, p163.
6. Karnosh LJ, Scherb RF : Trophic lesions in the distribution of the trigeminal nerve *JAMA*, 115 : 2144, 1940.
7. Becker SW : Dermatitis in Association with Diseases or injury of the peripheral nerve, *Arch Derm Syph*, 12 : 235, 1925.
8. Howell JB : Neuropathic changes in the trigeminal nerve, *Arch Derm*, 86 : 442, 1962.
9. Harris W : Analysis of 1433 cases of paroxysmal trigeminal neuralgia (Trigeminal-Tic) and end-results of gasserian alcohol injections : *Brain*, 63 : 209, 1940.
10. Loveman AB : An unusual dermatitis following section of the 5th cranial nerve : *Arch Derm Syph*, 28 : 369, 1933.
11. Jaeger HC : New type of neuropathic ulcer of the ala nasi following retrogasserian neurotomy: *Dermatologica*, 100:201, 1950.
12. Harris W : Rare forms of paroxysmal trigeminal neuralgia and their relation to disseminated sclerosis : *Bri Med J*, 2 : 1015, 1950.
13. Savitsky N, Elpern SP : Gangrene of face following occlusion of posterior inferior cerebellar artery : *Arch Neurol Psychiat*, 60 : 388, 1948.
14. Rosenberg SJ, Solovay : Trophic ulcers following encephalitis lethargica : *Arch Derm Syph*, 39 : 825, 1939.