CASE REPORT

Trigeminal trophic syndrome complicating a case of borderline tuberculoid leprosy

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Accepted for publication 28 October 2011

Summary An example of trigeminal trophic syndrome presenting as ulceration of ala nasi in a case of borderline tuberculoid leprosy is reported. To the best of our knowledge, this is only the second case report of this manifestation in leprosy to be documented.

Introduction

Cranial nerve involvement is commonly seen in patients with leprosy. Facial, trigeminal and other nerve involvement has been emphasised.\(^1\)\(^-\)\(^5\) The Trigeminal nerve is probably the second most common cranial nerve to be affected in leprosy. Though such involvement tends to be more common in patients with the lepromatous type of leprosy of longer duration, it can occur in other forms of leprosy and also in short-duration disease.\(^4\) Hypoaesthesia and anaesthesia, in the maxillary divisions of the trigeminal nerve are the most frequent findings.\(^4\) Trigeminal trophic syndrome (TTS) is a rare entity in which nasal ulceration occurs following sensory impairment in the trigeminal area.\(^6\) Trigeminal nerve involvement manifesting as trophic ulceration of the nose, in a case of borderline tuberculoid leprosy, is reported here.

Case Report

A 60 year-old male, resident of Uttar Pradesh, India, presented with persistent, asymptomatic ulcer over the right ala nasi for 1 month. The patient was a clinico-histopathologically diagnosed case of borderline tuberculoid leprosy with hypoanesthetic erythematosus plaques present over left arm, back (Figure 1) and right leg and thickened peripheral nerves including right ulnar, left common peroneal and right supra-orbital nerves.

Patient consent: informed written consent was obtained from the patient.

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The patient has been on multibacillary multidrug therapy for the last 5 months. Three months earlier he started having paraesthesiae over the right side of his nose and cheek. He indulged in self picking at the nose due to the paraesthesiae, which subsequently resulted in ulceration on the right side of nose. He denied any history of topical applications. There was no history of any red raised lesion in that location prior to the onset of ulceration, nor a history of flare-up of the pre-existing lesions. He did not suffer any trauma, and denied any significant medical, surgical history or alcohol intake.

Examination revealed a sickle-shaped, well defined, ulcer measuring 2 × 2.5 cm present over the right ala nasi with some extension on to the philtrum (Figure 2).

The skin surrounding the ulcer was normal, and an examination of the nasal cavity was normal. Neurological examination revealed a decreased level of pain and thermal perception over the ophthalmic and maxillary divisions of the trigeminal area, as well as a diminution of corneal reflex. Jaw jerk and mastication were normal. Cranial nerve examination was otherwise normal. There was no sign of acute neuritis. Slit lamp examination of both eyes revealed no evidence of any corneal changes.

A biopsy of a cutaneous plaque revealed well-defined epithelioid cell granulomas in the upper and mid dermis (Figure 3).

A biopsy taken from the edge of the ulcer showed hyperplasia of the dermis with neither malignant-cellular proliferation nor granulomatous infiltration. Antinuclear antibody (ANA) assay by indirect immunofluorescence and antineutrophilic cytoplasmic antibody (ANCA) test, and VDRL done on serum were negative. To rule out dermatitis artefacta, a psychiatric opinion was obtained according to which he was not suffering from anxiety and/or depression. The patient was followed up every week with the provision of protective gloves, topical antibiotics and normal saline bandaging of the ulcer at each visit. The ulcer healed completely over a period of 2 months leaving a residual scar (Figure 4).

Discussion

Cranial nerve involvement has frequently been detected in patients with leprosy, the facial (VIIth) nerve, being the most frequently afflicted.\(^1-5\) The trigeminal nerve (Vth) is probably
the second most common cranial nerve to be involved.\(^4\) Although cranial nerve involvement is more often a manifestation of lepromatous leprosy, it may occur in other types too. Loss of sensation in the form of hypoesthesia and/or anaesthesia, in the maxillary divisions of the trigeminal nerve is the most common sign.\(^4\) Our patient had reduced sensation along the ophthalmic and maxillary divisions, and diminished corneal reflex, however, the jaw jerk was normal, conforming to trigeminal nerve involvement.

Trigeminal trophic syndrome (TTS) is a fascinating, rare entity in which nasal ulceration occurs following sensory impairment in the trigeminal area. Loveman\(^6\) and McKenzie\(^7\) first described TTS in 1933. In a majority of TTS cases the ulcers occur following destruction of the gasserian ganglion after alcohol injection for trigeminal neuralgia or rhizotomy. Other neurological associations include vertebro-basilar insufficiency, acoustic neuroma and syringobulbia.\(^8\) There is a single report in which Thomas et al.\(^9\) described a

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**Figure 2.** Crescent-shaped ulceration of the right nasal ala.

**Figure 3.** Section of a cutaneous plaque depicting well-defined epitheloid cell granulomas in the upper and mid dermis (H & E \(\times\) 40).
case of self inflicted nasal ala ulceration as a result of trigeminal nerve involvement in a case of leprosy.

The characteristic lesion of TTS, crescent-shaped lateral nasal ala ulceration, is associated with trigeminal nerve anaesthesia and facial paraesthesia. The margins of the ulcer are free, and the ulcer base has scant crust. Other areas may be involved with these ulcers, including the frontal scalp, ears, cheek, temple, and cornea. Patients complain of picking, rubbing, or scratching sensations in the affected areas; these sensations are attributed to paraesthesias, or pain following damage of the sensory trigeminal fibres or nuclei. In the case under review, neurological examination revealed decreased sensation of pain and thermal perception over the trigeminal area. This sign is very important for making a diagnosis.

The other differential diagnosis of TTS includes basal cell carcinoma, infectious diseases (tertiary syphilis, recurrent herpes simplex, leishmaniasis, cutaneous tuberculosis) or systemic disease (Wegener’s granulomatosis, Horton disease). In the current case ANA (by IIF), ANCA (by IIF), serum VDRL and histo-pathological examination were negative. Self-induced nasal lesions that occur in factitious dermatitis are primarily distinguished from those in TTS by the presence of normal trigeminal nerve function and frequent patient denial of lesion manipulation.

The management of trigeminal trophic syndrome is often difficult. Treatment regimens including amitriptyline and diazepam in addition to chlorpromazine and pimozide have met with some success. Carbamazepine is an effective therapy in trigeminal neuralgia and atypical facial pain because it may influence both the paraesthesiae and the behavioural factors in this syndrome. Application of occlusive dressings can reduce trauma of the affected area and hydrocolloid dressings help the ulcers heal. The use of finger bandages and protective gloves has been suggested to prevent digital manipulation. In the current case hand gloves were provided and normal saline bandaging of the ulcer was done along with topical antibiotics. The response was favourable with complete healing of the ulcers.

To our knowledge there is a single previous case report of TTS in leprosy. This case is a unique presentation of self inflicted nasal ulceration consequent to trigeminal nerve involvement in a leprosy patient, resolving completely with protective measures. In conclusion one needs to be punctilious when examining a leprosy patient, anticipating possible cranial nerve involvement.
References