Self-induced Nasal Ulceration

Travis T. Tollefson, MD; J. David Kriet, MD; Tom D. Wang, MD; Ted A. Cook, MD

Background: Nasal ulcerations have many causes. Ulcerations that are self-induced are difficult to diagnose and treat. Two rare conditions with self-induced nasal ulceration are trigeminal trophic syndrome (TTS) and factitious disorder (FD). Trigeminal trophic syndrome is characterized by trigeminal anesthesia, nasal alar ulceration, and facial paresthesia. Appearance of the nasal ulcer after trigeminal ablation for neuralgia is diagnostic. Self-induced nasal lesions that occur in FD are primarily distinguished from those in TTS by the presence of normal trigeminal nerve function and frequent patient denial of lesion manipulation.

Objectives: To increase physician awareness of the disorders leading to self-induced nasal ulceration and to discuss management issues in our patient series.

Design: A retrospective review of 7 cases in which the patients presented for reconstructive consultation between March 1985 and October 1997 and were found to have self-induced nasal ulcerations.

Setting: Tertiary university medical center.

Results: Five patients were identified with TTS and underwent nasal reconstruction an average of 43 months (range, 4-72 months) after nasal ulcer presentation. Four of the 5 patients developed ulcer recurrence between 1 and 58 months after reconstruction; secondary reconstruction resulted in recurrence in 2 of these patients. Two patients were identified with FD and self-induced nasal ulceration. One of these 2 patients underwent total nasal reconstruction 15 months after ulcer occurrence and developed recurrence 2 weeks after surgery.

Conclusions: Self-induced nasal ulceration remains a difficult condition to diagnose and treat. Readily treatable conditions should be excluded, and diagnostic workup should include tissue biopsy and laboratory studies. Patients with TTS may have associated ocular findings, and those who do should be referred for ophthalmologic consultation. Surgical reconstruction can be considered in the highly motivated patient with TTS; however, delayed ulcer recurrence is common. Patients with FD should be treated primarily with local wound care and referred for psychiatric intervention. We strongly recommend nasal prosthetic devices as the primary means of aesthetic correction and discourage surgical repair in the patient with FD.

Arch Facial Plast Surg. 2004;6:162-166
We describe 5 patients with TSS and 2 with FD to increase physician awareness of the high risk of ulcer recurrence after nasal reconstruction. Management options for self-induced nasal ulcerations are also discussed.

METHODS

Seven cases that presented with self-induced nasal ulcerations between March 1985 and October 1997 were retrospectively reviewed. A review of the patients’ medical records included age, sex, diagnosis, reason for trigeminal sensory deficit, associated illness, ulcer characteristics, recurrence, surgical treatments, and surgical outcome. Preoperative and postoperative photographs were evaluated when available.

RESULTS

From March 1985 through October 1997, 7 patients presented with nasal ulceration. All patients underwent laboratory testing and biopsy to rule out malignant, granulomatous, or infectious processes. Each of the patients was determined to have self-induced nasal ulceration. Five patients were identified with TTS and 2 with FD. Patient demographics are outlined in Table 2. The mean age was 50 years (age range, 16-77 years). The mean follow-up was 48 months (range, 18-114 months). In the 5 patients with TTS, the initial ulceration first appeared several months to more than 10 years after the facial anesthesia-producing event. The mean time from presentation with nasal ulcer to reconstruction was 43 months (range, 4-72 months). All patients had complete epithelialization of their wounds before reconstruction.

Six patients (5 with TTS and 1 with FD) underwent nasal reconstruction; 2 underwent multiple reconstructions. Five of the 6 patients ultimately developed recurrent nasal ulcerations after a mean of 14 months (range, 0.5-58 months).

Trigeminal sensory deficit, which was identified in the 5 patients with TTS, resulted from motor vehicle accident (n=2), trigeminal nerve ablation (n=2), and combat injury (n=1). No identifiable sensory defect was seen in the 2 patients with ulceration from FD. Ipsilateral eye abnormalities were identified in 4 of the 5 patients with TTS.

Additional psychiatric conditions were identified in our patient population. Both patients with FD were treated for depression, and 1 was treated for narcotic dependence. One patient with TTS had undergone previous treatment for alcohol dependence.

REPORT OF 3 CASES

CASE 1

A 36-year-old man was referred for reconstruction of a septal perforation and a left nasal alar defect that had been treated conservatively by his dermatologist for 4 years (Figure 1A). Left hemifacial anesthesia and other cranial neuropathies resulted from a motor vehicle crash. Three-layered nasal reconstruction was performed with an epithelial turn-in flap, conchal cartilage, and a paramedian forehead flap. Photographs taken 18 months after surgery revealed adequate reconstruction with no recurrence of ulceration at the last follow-up visit (Figure 1B).

CASE 2

A 76-year-old woman presented with hemifacial anesthesia, nasal paresthesia, a corneal scar, and an epithelialized nasal alar defect. Over a 15-year period, the patient suffered from unremitting trigeminal neuralgia and facial pain. Treatment attempts included alcohol injec-

Table 1. Differential Diagnosis of Nasal Ulceration

<table>
<thead>
<tr>
<th>Differential Diagnosis of Nasal Ulceration</th>
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<tbody>
<tr>
<td>Malignancy: Basal cell carcinoma</td>
</tr>
<tr>
<td>Squamous cell carcinoma</td>
</tr>
<tr>
<td>Infectious process: Staphylococcus aureus</td>
</tr>
<tr>
<td>Syphilis</td>
</tr>
<tr>
<td>Leprous trigeminal neuritis</td>
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<tr>
<td>Paracoccidiomycosis</td>
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<tr>
<td>Blastosporosis</td>
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<tr>
<td>Laishmaniasis</td>
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<tr>
<td>Herpetic reactivation</td>
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<tr>
<td>Granulomatous disease</td>
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<tr>
<td>Sarcoïdosis</td>
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<tr>
<td>Wegener granulomatosis</td>
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<tr>
<td>Angiocentric immunoproliferative lesions</td>
</tr>
<tr>
<td>Polymorphic reticulosis</td>
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<tr>
<td>Lethal midline granuloma syndrome</td>
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<tr>
<td>Midline destructive granuloma</td>
</tr>
<tr>
<td>Pyoderma gangrenosum</td>
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<tr>
<td>Trigeminal trophic syndrome</td>
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<tr>
<td>Factitious disorder with ulceration</td>
</tr>
</tbody>
</table>

Table 2. Patient Characteristics

<table>
<thead>
<tr>
<th>Patient No./Sex/Age, y</th>
<th>Side of Lesion</th>
<th>Cause of Trigeminal Dysfunction</th>
<th>Corneal Lesion</th>
<th>Psychiatric Disorders</th>
<th>Waiting Period Before Surgery, mo</th>
<th>Overall Follow-up, mo</th>
<th>Recurrence, mo</th>
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<tbody>
<tr>
<td>1/M/36</td>
<td>Left</td>
<td>Motor vehicle crash</td>
<td>Yes</td>
<td>None</td>
<td>48</td>
<td>18</td>
<td>None</td>
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<tr>
<td>2/F/76</td>
<td>Right</td>
<td>Trigeminal ablation</td>
<td>Yes</td>
<td>None</td>
<td>60</td>
<td>60</td>
<td>25, 5</td>
</tr>
<tr>
<td>3/F/59</td>
<td>Midline</td>
<td>None</td>
<td>None</td>
<td>Narcotic dependence</td>
<td>15</td>
<td>18</td>
<td>0.5</td>
</tr>
<tr>
<td>4/M/63</td>
<td>Left</td>
<td>Combat injury</td>
<td>None</td>
<td>None</td>
<td>72</td>
<td>22</td>
<td>4</td>
</tr>
<tr>
<td>5/F/77</td>
<td>Right</td>
<td>Trigeminal ablation</td>
<td>Yes</td>
<td>None</td>
<td>60</td>
<td>114</td>
<td>58</td>
</tr>
<tr>
<td>6/F/16</td>
<td>Right</td>
<td>Motor vehicle crash</td>
<td>Yes</td>
<td>Alcohol dependence</td>
<td>4</td>
<td>72</td>
<td>4, 1</td>
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<tr>
<td>7/F/27</td>
<td>Right</td>
<td>None</td>
<td>None</td>
<td>Depression</td>
<td>No surgery</td>
<td>32</td>
<td>No surgery</td>
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tion and radiofrequency trigeminal ganglion ablation, as well as suboccipital craniotomy with microsurgical nerve decompression. Nasal reconstruction was successfully performed with a superiorly-based melolabial flap. Twenty-five months later, a self-induced right nasal excoriation was identified, and the patient did not return for follow-up. Three years later, a nasal reconstruction performed elsewhere also resulted in nasal ulcer recurrence.

CASE 3

A 59-year-old woman presented with pruritis, epistaxis, and a severe nasal deformity that had developed after she underwent a septoplasty 16 years earlier. Nasal crusting and self-induced erosion were treated with local wound care, and a nasal prosthesis was successfully used. During this period, the patient underwent chronic pain management and psychiatric consultation. Despite the satisfactory appearance of the prosthetic rehabilitation, the patient insisted on nasal reconstruction. The ulceration recurred 12 days after 3-layer nasal reconstruction was performed.

COMMENT

Loveman and McKenzie first described TTS in 1933. The characteristic crescent-shaped lateral nasal ala ulceration is associated with trigeminal nerve anesthesia and facial paresthesia. This triad most commonly develops after chemical or surgical ablation of the trigeminal sensory root in the gasserian ganglion. The characteristic triad is also present in patients with traumatic trigeminal impairment. Weintraub et al described 63 cases of TTS, the majority of which were caused by surgical trigeminal ablation (46%) and alcohol injection (29%) of the gasserian ganglion. The rest were caused by cerebral vascular disorders and infarctions, acoustic neuroma, postencephalitic parkinsonism, and syringobulbia. Self-induced nasal lesions often result from persistent digital trauma. Complaints of ipsilateral nasal congestion, burning, and severe pruritis may precipitate self-induced nasal defects. A peculiar sensation localized to the affected nasal area has been described as “things crawling under [the] skin” that creates the urge to pick at the area. The pathogenesis of these nasal paresthesias has been suggested to relate to an overlap of sensory input from the ophthalmic and maxillary distributions of the disrupted trigeminal nerve. This theory is supported by the presence of the characteristic crescent-shaped alar erosion at the junction of these dermatomes. Another theory suggests that autonomic vasomotor control is altered when sympathetic innervation is altered by trigeminal nerve injury, which in turn results in unfavorable wound-healing conditions owing to vasodilatation, venous stasis, and hypothermia. The nasal tip skin is commonly spared from ulceration and separately innervated by the external ethmoidal branch of the ophthalmic division.

Figure 1. A 36-year-old man with trigeminal trophic syndrome before (A) and 18 months after (B) 3-layered nasal reconstruction. Nasal ulcer recurrence did not develop.
The reported incidence of TSS after surgical or injection therapy for trigeminal neuralgia ranges from 0% to 16%. The interval between sensory deficit and ulceration has been reported to range from 2 weeks to 23 years. This wide range is consistent with the findings in our 5 cases of TTS.

Trigeminal neuropathy after nonpenetrating cranial injury is a rare complication. Anatomically, the gasserian ganglion is the most vulnerable site of traumatic trigeminal nerve injury because of its fixed location within Meckle’s cave. It is the same anatomical site that is targeted during alcohol injection or trigeminal rhizotomy and may suggest the mechanism for TTS in the 3 patients with previous facial trauma in our series.

Eye lesions associated with TTS include neurotrophic keratitis, iritis, and corneal ulceration and opacification. Normally, the trigeminal nerve’s sensory input to the cornea promotes corneal epithelialization through a trophic effect. Absence of this effect can lead to impaired corneal healing and ulceration. In 1954, Sigelman and Friedenwald suggested the importance of this neurotrophic effect by describing persistent corneal changes in a neurotrophic keratitis animal model even after tarsorrhaphy was performed to protect the animals’ eyes from corneal exposure. Current trials are studying the benefit of treating patients with neurotrophins such as epithelial growth factors. Also, corneal insensitivity, decreased tearing, and absent corneal reflexes can contribute to dry, painless eye irritation. Four of our 5 patients with TTS were found to have ipsilateral corneal lesions, suggesting that ophthalmologic consultation should be an integral part of the evaluation of patients with TTS. Eye lesions were not identified in the patients with FD.

The psychiatric conditions in patients with self-induced ulceration range from neurosis to psychosis, and even to malingering, in which lesions are consciously produced to obtain a consciously desired secondary gain. Factitious disorder is thought to fall somewhere between a neurosis and a psychosis. These conditions in patients with factitial diseases make treatment difficult. The patients will frequently have undergone multiple previous evaluations by physicians in different specialties. Such patients commonly are manipulative and dependent and deny responsibility for the self-induced lesions. This behavior may evoke a sense of frustration in the treating physician but may point the physician toward the diagnosis of FD. Excessive psychological stressors are also commonly present in patients with FD and may serve as a trigger for lesion manipulation.

Most psychiatrists recommend that nonpsychiatric physicians maintain a supportive and cautious relationship with the patient with FD to develop and retain trust. Psychological intervention should not be attempted, but rather relegated to a mental health professional. Attempts to bring about patient insight and acceptance of guilt may lead to vehement denial and termination of the physician’s care.

Previous treatments for TTS have included psychotropic medications, radiation therapy, transcutaneous electrical stimulation, and even stellate ganglionectomy. Initially, the nasal lesion should be treated with local wound care, including topical and systemic antibiotic therapy when indicated. The use of finger bandages and protective gloves has been suggested to prevent digital manipulation. Nasal casts or prostheses,
which can also be used to minimize unconscious manipulation of the nasal lesion, were used in 2 of our patients.

If surgical treatment of TTS nasal ulceration is considered, it should be restricted to wounds that have healed with conservative wound care and cessation of manipulation. Abyholm and Eskeland recommended the use of contralateral, sensate flaps, such as a tunneled paramedian forehead flap, as a deterrent to self-manipulation. Despite their recommendation, ulcer recurrence remains high and has been seen up to 3 years after nasal reconstruction. In our series, all 5 patients with TTS underwent reconstructive surgery. Four of them ultimately developed recurrent nasal ulceration 1 to 38 months after surgery, a range that is consistent with that reported in the literature. The high recurrence rate after surgical reconstruction emphasizes the need for thorough preoperative patient counseling and strong consideration of nonoperative intervention.

Surgical intervention in cases of FD is contraindicated until the wound is epithelialized and the patient has been evaluated and cleared by a mental health professional. In our series, we declined surgical intervention in one patient with FD and performed total nasal reconstruction in the other patient only after the ulceration remained epithelialized and without manipulation for a period of 1 year (Figure 2A). During this time, the patient reluctantly wore a nasal prosthesis (Figure 2B). Despite the delay in reconstruction, she developed recurrent self-induced nasal ulceration 2 weeks after surgery (Figure 2C). The home health nurse reported compulsive nasal manipulation. The ear and forehead donor sites also displayed signs of manipulation. Three months after reconstruction, the patient continued to deny manipulation of these sites and was unavailable for follow-up.

CONCLUSIONS

Self-induced nasal ulcerations are difficult lesions to diagnose and treat. Trigeminal trophic syndrome is an uncommon disorder that is characterized by trigeminal anesthesia, nasal alar ulceration, and facial paresthesia. The appearance of a characteristic ulcer after trigeminal ablation is diagnostic. Associated ocular findings may also be present and should be evaluated by an ophthalmologist. A biopsy can provide important information to exclude other causes.

Self-induced nasal lesions in FD are primarily distinguished from TTS nasal lesions by normal trigeminal nerve function and frequent patient denial of lesion manipulation. This rare disorder should be addressed primarily with psychiatric treatment and local wound care. We strongly recommend nasal prosthetic devices as the primary means of aesthetic correction and discourage surgical repair in the patient with FD. If surgical correction must be performed, psychiatric clearance is necessary, but does not guarantee long-term success. Surgical reconstruction can be considered in highly motivated patients with TTS; however, delayed ulcer recurrence in these patients is common.

Accepted for publication October 1, 2003.

This study was presented in part at the Seventh International Symposium of Facial Plastic Surgery; June 17, 1998; Orlando, Fla.

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REFERENCES