Thirteen maxillary full dentures over 10 years: A case of peripheral painful traumatic trigeminal neuropathy

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There are multiple reasons for failure of full maxillary dentures. It is necessary for clinicians to consider the rationale behind the failure of the prosthesis before remaking a maxillary full denture. Diagnostic categories related to pertinent maxillofacial and oral neuropathies and psychosocial issues are also discussed. Here, we present a case with a natural history that supported a diagnosis of peripheral painful traumatic trigeminal neuropathy (PPTTN). Furthermore, the clinical oral examination revealed findings that were consistent with atrophic glossitis, necessitating an investigation of diagnoses of nutritional deficiency–induced neuropathy and/or oral candidiasis. (Oral Surg Oral Med Oral Pathol Oral Radiol 2019;128:e180–e186)

We as dentists have heard during our dental education years or in conversations with other dentists about a patient presenting with numerous dentures and complaints regarding difficulty utilizing those dentures for normal oral functions. Stefanac and Nesbit[1] listed "previous disease experience" as an issue in evidence-based decision-making risk assessment. Certainly, the remaking of a denture after the first 2 or even more treatment outcome failures should alert a dentist that fabricating another denture would probably not be in the patient's best interests because there must be some biologic or psychological reasons (or both) for this type of clinical failure. In this article, we report a case involving a patient who experienced 12 remakes of her maxillary denture despite being told at various points along the way that she might have a nontechnical problem that would not be resolved by repeated attempts at making new prostheses. Because the main complaint, as reported by this patient when she tried to wear her denture, was the presence of specific pain symptoms (including burning pain) that could only be relieved by removing the denture, there was a high probability that she might have a neuropathic oral pain condition. However, despite being told this by several dentists, she insisted that a better-made prosthesis could probably solve her problem. The details of those interactions and the failed attempts at finding a permanent solution for this challenging problem are the main points of discussion in this unusual case report.

With respect to orofacial chronic pain, the most important issue is to determine the diagnosis so that appropriate therapy can be instituted.[2] This patient presented an interesting and difficult challenge with respect to determining the specific diagnosis or diagnoses. Her presenting psychological make-up, her problematic personal established concepts of oral health and dentistry, and her lack of trust resulting in lack of compliance certainly complicated the diagnostic process and ultimately limited the ability of a successful therapeutic outcome. But is it fair to blame such a treatment failure on the patient? There is little doubt that we can learn more from our failures than from our successes.

Patients with orofacial chronic pain conditions, such as painful post-traumatic trigeminal neuropathy (PPTTN) and other oral neuropathies, may require critical diagnostic evaluations[2-8] that may not be readily available outside of metropolitan locations. Metropolitan areas offer a variety of medical and dental specialists, whereas rural areas tend to offer fewer options.

We present a particularly unusual case of a patient who had all of her remaining maxillary teeth extracted and an immediate maxillary full denture placed; she then presented with consistent complaints of oral pain (continuing pain only relieved by removing the denture) and experienced 12 remakes of the original denture over a 10-year period.

CASE REPORT

The 75-year-old female patient reported to an oral medicine clinician in February 2017 with a chief complaint of "13 sets of teeth made since 2008." The patient had had all of her upper teeth extracted in 2008 and replaced by an immediate maxillary full denture. The patient noted that she was adamant about never having her husband see her without her teeth and would only remove her denture before going to sleep. The patient reported that she had been found to be allergic to the monomer of acrylic in a previous denture and that curing of the monomer had improved the burning condition. The patient reported that her lips tingled, and that "the mouth feels like a mouth full of bumblebees." The patient previously reported to a

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university dental school clinic for treatment of burning mouth syndrome, and the treatment had not been successful. The patient had discontinued lisinopril, a hypertensive medication, 6 months ago but did not notice any change in her oral pain condition. The patient reported that the pain was alleviated by taking out her denture at night. She noted that the pain while wearing her upper denture was centered at the upper left posterior area of the denture. One of the patient's previous dentists had prescribed an unknown antifungal drug, with no treatment success. The patient believed that the oral pain occurs along a nerve trunk. The patient's primary care physician had diagnosed a seizure condition and had prescribed levetiracetam, but the patient was intolerant of this medication and she discontinued taking it. The patient was taking low-dose aspirin therapy and coenzyme Q and curcumin supplements. The patient's oral and maxillofacial surgeon had referred the patient to the oral medicine clinician 6 years ago; however, the patient had misplaced the referral information until she found it recently.

The oral medicine clinician had clinically evaluated the patient and noted that the dorsal tongue was smooth and denuded of filiform papillae (Figure 1). The remaining oral tissues, including the palate and edentulous maxillary arch, appeared to be normal. A panoramic radiograph, obtained in January 2014 (Figure 2), revealed several teeth treated with root canal therapy, post and cores, and crowns, but no evident radiographic pathology. Complete edentulism of the maxilla was noted. The patient had an existing maxillary complete denture, as well as other maxillary full dentures, most of which she brought with her. Also, she had a photograph that displayed several of her previous dentures (Figure 3). Several of her dentures appeared to have the palatal aspect of the denture reduced. (This was also evident on the photograph.) A direct examination of the dentures also confirmed

Fig. 1. The dorsal tongue demonstrates denuded filiform papillae characteristic of atrophic glossitis.

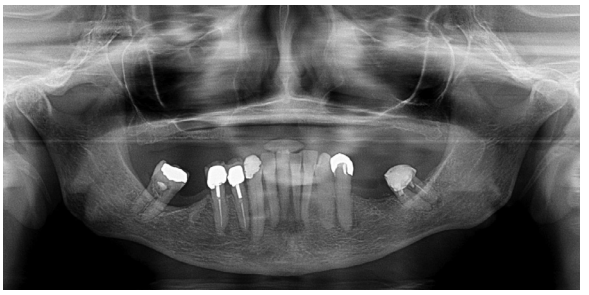

Fig. 2. A panoramic radiograph demonstrating an edentulous maxilla.

reduction of the palatal aspect of several of these dentures. The differential diagnosis included oral dysesthesia, oral candidiasis, deficiency-induced neuropathy, and PPTTN/traumatic neuroma. The clinician referred the patient back to her primary care physician for relevant blood studies (complete and differential blood counts, B_{12}, folic acid, iron, and hemoglobin A_{1C}), as well as providing a prescription for fluconazole. The patient was to report her progress in 2 weeks, but the patient did not report her progress and her phone was found to be disconnected.

In October 2017, the office was able to contact the patient. The patient had not filled the fluconazole prescription and, instead, had her physician call in a prescription for nystatin suspension (100,000 units per

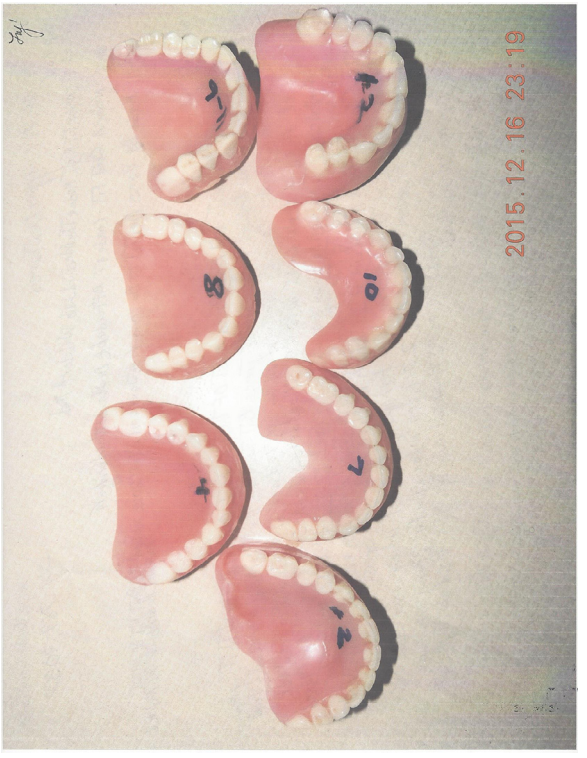

Fig. 3. A photograph of 7 failed maxillary full dentures.

mL, 4 times a day; rinsing with 5 mL for 1 minute). Also, the patient reported to another medical facility, rather than to her primary care physician, for the blood studies and blood chemistry, which showed normal results; the requested laboratory studies were not undertaken. The oral medicine clinician had called in a prescription for fluconazole and sent the request for the blood studies. The blood studies demonstrated essentially normal values. The patient sent in a large folder with information and pictures concerning her oral pain and prosthetic failures. She reported longstanding severe oral burning pain. She also reported having undergone the Clifford Dental Allergy Test, which had revealed an allergy to acrylic monomer, as well as to cadmium and mercury. Therefore, she had at least 3 dentures fabricated with hypoallergenic flexite silicone. She noted that her condition may have been secondary to a local anesthetic injection into the left side of the soft palate, which she believed caused nerve damage and secondary chronic nerve pain. She reported having had the area of the posterior left denture border trimmed and that this caused loss of suction and decreased denture retention but that it did not alleviate the oral chronic pain condition. She also reported trying a relining product on several of her failed dentures but then complained about the leeching out of "awful tasting," burning chemicals, which could not be tolerated. She reported soaking these dentures with the home relining in baking soda and water to treat noxious chemicals but that this was also not helpful. Of all the dentists the patient saw with a request to have a new upper denture fabricated, only 1 refused to do so, with the dentist stating that he did not expect that his results would be any different from those of the previous dentists.

In November 2017, the patient was referred once again for blood studies and again prescribed fluconazole antifungal drug therapy (this time for 4 weeks). The blood studies demonstrated low serum iron. The patient was referred back to her primary care physician for obtaining a prescription for an iron supplement. However, after a few days, she discontinued the iron supplement, complaining that it caused palpitations. In a December 2017 telephone interview, she reported that the antifungal therapy was not successful in alleviating her symptoms. The patient was referred back to her oral and maxillary surgeon for a local anesthetization procedure to aid in evaluating the presumed diagnosis of PPTTN/traumatic neuroma. However, the oral and maxillofacial surgeon refused to complete the procedure. Because of the results of the antifungal drug therapy trial and previous history, a diagnosis of PPTTN was suggested, and a referral to a neurologist was made. The patient reported that she had spoken with a dentist to consider laser therapy. In May 2018, the patient reported for a neurology evaluation, and, according to the patient, the neurologist refused to provide a diagnosis or offer any further referral advice. The neurologist suggested that the patient see a dentist.

In June 2018, the patient was referred to an orofacial pain clinician. The orofacial pain clinician, re-evaluated the patient with respect to both history and an oral neurologic evaluation and confirmed the diagnosis of PPTTN. The orofacial pain clinician reported that pressure to the greater palatine foramen region elicited increased pain triggered to the anterior maxilla. The application of 20% topical benzocaine to the posterior left palate resulted in alleviation of the patient's pain. These results confirmed the diagnosis of PPTTN of the left greater palatine nerve. In view of the success of the topical local anesthetic, the orofacial pain clinician prescribed topical capsaicin therapy. The patient initially tried the therapy but quickly became noncompliant. The patient set up an appointment to have the maxillary anterior frenum surgically relieved by another dentist. She claimed that she thought that pain from the frenum was causing pain in the left posterior palate.

In December 2018, the patient reported to a general dentist requesting to have another maxillary full denture fabricated. The dentist referred the patient to a local periodontist to have another local anesthetization procedure completed. The periodontist reported that he numbed the left greater palatine area and that it resulted in a partial decrease in the patient's pain. However, the patient continued to complain of pain in the anterior left central incisor area. The periodontist reported that the patient's maxillary burning pain was completely relieved after local anesthetization of the maxillary left central incisor. The oral medicine clinician (R.S.B.), orofacial pain clinician (J.A.), and the prosthodontist (I.B.) conferred and suggested that the patient treat the condition with topical benzocaine (20%). The patient complied and reported that the denture was now comfortable, but this lasted only for a limited time. In late December 2018, the oral medicine clinician (R.S.B.) counseled the patient to reapply the topical local anesthetic every 2 hours and to wait at least 1 month of successful resolution of her symptoms before having a new denture fabricated. The patient reported that her present dentist suggested that utilizing digital prosthetic impressions would result in a more accurate impression and in a superior maxillary full denture, which could remedy her pain condition. The patient was then lost to follow-up.

DISCUSSION

As it turned out, atrophic glossitis (AG) was a red herring, but certainly it was necessary for the clinician to

further diagnose and treat this condition and to investigate a possible diagnosis of neuropathy and whether or not the patient was nutritionally deficient. Other similar terms for AG include bald tongue, shiny tongue, and red beefy tongue. Oral burning is a symptom of oral candidiasis, and painful neuropathy can be the symptom of nutritional deficiency. AG (denudation of the filiform papillae of the dorsal tongue) can be the clinical finding to elucidate a diagnosis of either nutritional deficiency, or oral candidiasis.[9-11]

Some of the earliest published articles regarding the oral manifestations of nutritional deficiencies were by Dreizen.[12-14] Dreizen[11-14] reported that oral mucosal conditions, such as atrophic filiform papillae of the tongue, may be an early sign of systemic disease, particularly within the geriatric community. Patients with AG may report to dentists and physicians with secondary complaints of oral burning or oral discomfort. AG is typically caused by either oral candidiasis or nutrient deficiencies, such as vitamin B_{12} or iron deficiency. Other vitamin deficiencies are also known to have an association with AG.[9,10,15-17]

The difficult denture patient (DDP) is an accepted conceptual description of the difficulty found with some denture patients. DDPs may present a psychologically driven urge to seek denture treatment. These patients may seek and leave clinicians because of trust issues and lack of trust and may seek new clinicians again and again. Another major issue is the DDP's failure to comply with prescribed therapies and, at times, referrals.[18,19]

Durham and Nixdorf,[20] who described the patient with persistent dentoalveolar pain (PDAP) expounded on the biopsychosocial impact in PDAPs and noted the effect of consistent negative reinforcement of multiple ineffective dental therapies. In the present case, the antifungal medication resulted in resolution of the patient's oral candidiasis but failed to resolve her pain condition. The blood studies failed to determine a treatable diagnosis of a deficiency-related neuropathy or diabetic neuropathy. Topical capsaicin therapy failed to immediately resolve the chronic pain condition (even though the treating clinician explained to the patient that such therapy takes time to demonstrate positive results). Manfredini et al.,[21] Burke et al.,[22] Merrill et al.,[23] and Dworkin et al.[24] reported on the importance of psychological factors and psychological functioning of individuals suffering from chronic pain.

This patient was and is a difficult patient. The patient disappeared for over 8 months after the initial presentation and failed to follow the recommended antifungal drug regimen and the referral to her physician for the suggested blood studies. She had initially been referred by an oral and maxillofacial surgeon to an oral medicine clinician 3 years before her initial presentation. There were transportation issues because the patient lived in a rural area 2 hours away from the oral medicine and orofacial pain clinicians' offices located within an urban metropolitan area and she was not able to drive herself to these offices. She was the sole caregiver to her bedridden husband, and this added to her stress issues and further limited her ability to travel for receiving necessary care for herself. The oral medicine clinician tried to find a neurologist close to the patient's area of residence and found and referred her to one, with the hope of assistance with regard to her diagnosis of neuropathy and also made a suggestion for a referral to either a psychologist or psychiatrist. These recommendations were made in the detailed referral documents sent to the neurologist. However, the neurologist was not helpful. The patient was referred back to the original oral and maxillofacial surgery practice within the rural area for treatment with local anesthetization[25] and further diagnostic evaluations, but unfortunately, the original referring clinician was not available because the clinician was on extended leave. The oral and maxillofacial surgeons refused to accomplish the procedures for which the patient was referred.

Capsaicin therapy relies on irritating the irritated area to downregulate pain receptors, which initially results in more pain with the hope that the pain will be controlled in the future by increasing the pain threshold.[4,26] Pastre et al.[27] reported successful treatment with topical capsaicin in a 62 year-old female patient with posttraumatic peripheral neuropathy caused by her denture. Unfortunately, the patient in the case presented here refused further treatment related to the presumptive diagnosis of PPTTN/traumatic neuroma and may yet continue to attempt further solutions with similar removable prosthetics. The failure to remedy this patient's pain complaints is essentially a failure of all the treating clinicians.

The patient persisted in wanting what she thought was best, sometimes contrary to what clinicians advised, and remained convinced that merely having another full denture fabricated would alleviate her problematic painful denture condition. She did accept certain modifications, such as denture relining procedures, further acrylic curing, and nonallergenic denture materials, but she steadfastly attempted and reattempted to solve her problem by acquiring new full dentures.

The removal of the posterior palatal border of the patient's maxillary denture certainly would tend to decrease denture retention and the patient's satisfaction with several of her dentures because of the loss of the palatal seal.[28] However, it appears that the reduction was secondary to the patient's complaints of pain emanating from the posterior left palate.

Some of the patient's dentists considered a diagnosis of acrylic allergy or irritation, which may be caused by acrylic allergy or secondarily by incomplete curing of the monomer. Completing the curing or using nonallergenic denture material was not successful. Furthermore, denture allergy typically demonstrates erythema of the supporting tissues, and this was not the case in the case presented here.[29]

It is very important not to minimize the psychosocial issues related to chronic pain in the context of its diagnosis and management. The patient's persistent identification of chronic pain may be both biologic and psychosocial. Patients with chronic pain, in time, may adopt the identity of a chronic pain patient or a chronic denture patient.[19,23,24] The psychosocial issue of identification as a chronic problem denture patient may be a significant factor with respect to the patient in the case presented.

The head and neck region with its confluence of blood vessels, nerves, and muscles presents a difficult challenge with respect to the diagnosis and treatment of chronic pain. Dentists, for the most part, are involved within the context of deep somatic pain, which comprises pain mainly of muscles and teeth but also includes vascular pain. The diagnoses and treatment of neuropathic pain, such as atypical odontalgia (AO) and PPTTN, presents even more challenges.[2]

The International Classification of Headache Disorders III (ICHD III), under secondary headaches caused by another disorder, lists such diagnostic categories as posttraumatic pain, vascular disease, nonvascular intracranial pathology, pain associated with substances, central nervous system infection, homeostatic disorder, cranial neck pain (eye, ear, nose, and throat; sinus; tooth; jaw; and psychiatric pains).[3,4,30] According to Baad-Hansen,[5] AO is a subgroup of persistent idiopathic facial pain (PIFP) (also including atypical facial pain [AFP]). AO and AFP share the description of persistent facial pain that does not have the characteristics of cranial neuralgias and is not attributed to another disorder.[5] Adibi et al.[2] listed AFP and PIFP as the same condition. Nixdorf and Moana-Filho[7] noted that although such neuropathic orofacial pain conditions as AO, phantom tooth pain, neuropathic tooth pain, AFP, and PIFP are unlikely to be characterized as occurring within the same disorder, it is safe to assume that these conditions share more commonalities than not and to consider the overall diagnostic entity of PDAP. Nixdorf and Moana-Filho,[7] Adibi et al.,[2] Baad-Hansen,[6] Nixdorf et al.,[3] Baad-Hansen and Benoliel,[5] and Benoliel et al.[4] all supported avoiding irreversible dental therapy, such as endodontic therapy or tooth extraction, when an orofacial neuropathy is within the differential diagnosis. See Tables I through IV, for the criteria and characteristics of PPTTN, AO, PDAP, and PIFP, respectively.

Table I. Peripheral painful traumatic trigeminal neuropathy (PPTTN)—criteria and characteristics[3-5]

- Clinically evident neurologic dysfunction
- Imaging or neurophysiology demonstrating a neurologic lesion and its location
- Spontaneous or stimulus-dependent pain which lasts from seconds to minutes and is relatively constant
- The pain tends to be unilateral facial and/or oral pain
- History of a traumatic event to the trigeminal region with clinically evident signs of nerve dysfunction
- Pain located in the distribution area of the involved trigeminal nerve
- Pain developing within 3 to 6 months after the traumatic event
- Another ICHD-3 diagnosis does not apply
- Pain characteristically constant, lasting most of the day and typically present on most days
- Pain may be described as shooting and/or burning pain
- Pain does not usually interfere with sleep
- Occurs mainly in women age greater than 45 years

According to Benoliel et al.,[4] in their proposed diagnostic criteria, the diagnosis of PPTTN can be determined on the basis of spontaneous or stimulus-dependent pain affecting one or more divisions of the trigeminal nerve, lasting from seconds to minutes, being constant—greater than 8 hours per day and greater than 15 days per month—developing within 3 months of an identifiable trauma to the painful area or relevant innervation, continuing for greater than 3 months, and not being attributed to another disorder. With the evident overlapping diagnostic criteria between PPTTN, AO, PIFP, and PDAP, it is difficult to determine specific clinical cases which would not demonstrate a difficulty in separating these diagnostic categories from one another.[3] Certainly, the specificity of imaging may demonstrate a neurologic lesion and its location, more definitively indicating a diagnosis of PPTTN. In the case presented here, another positive

Table II. Atypical odontalgia (AO)—criteria and characteristics[2,3,5,6]

- Pain in a tooth or persistent pain after a tooth extraction with no evident pathology
- Pain ongoing at or beyond 6 months
- Pain in and around teeth or where teeth were previously located
- Maxillary teeth tend to be affected more often than mandibular teeth, and posterior teeth more often than anterior teeth
- Pain present most every day
- Nonparoxysmal pain—variable pain, mild to severe, continuous or near continuous
- Typically the condition does not interfere with sleep
- Occurs mainly in women, often ages between 35 and 45 years
- Dull, aching, and/or throbbing pain

Table III. Persistent dentoalveolar pain (PDAP)—criteria and characteristics[3,5,7]

- Persistent pain of at least 8 hours daily for at least 15 or more days per month and at least 3 months' duration
- Pain according to the International Association for the Study of Pain (IASP) criteria, which includes dysesthesia
- Pain localized within a defined anatomic area
- The extent of the evaluation not specified but can include dental and neurologic evaluations and imaging modalities, such as dental intra- and extra-oral radiography and magnetic resonance imaging (MRI)
- Not associated with paroxysmal pain
- Cannot be explained by another diagnostic condition
- May or may not include a history of trauma
- A sensory abnormality may or may not be present

Table IV. Persistent idiopathic facial pain (PIFP)—criteria and characteristics[2-8]

- Recurs daily for greater than 2 hours a day over 3 months
- Does not follow a peripheral nerve distribution
- Debilitating pain that exists along the distribution of the trigeminal nerve, that does not follow a dermatomal distribution, and that is typically without periods of remission
- Can be bilateral or unilateral pain
- No autonomic signs of pain
- Typically occurs in women age greater than 40 years
- Dull, aching, burning, nagging pain
- Poorly localized pain
- Possible psychogenic association

diagnostic criterion provided by Benoliel et al.[4] was evident neurologic dysfunction. With regard to the case reported here, it appears that the nerve tissue of the left greater palatine nerve was traumatized and that the chronic pain described by the patient was consistent with a diagnosis of PPTTN.

In the case reported here, the most important issue was making a definitive diagnosis so that a relevant therapeutic plan could be determined and initiated. We evaluated the patient and ruled out both deep somatic pain and superficial pain (pain from mucosal inflammation). Judging from the patient's history and clinical findings (local anesthetization of the painful area), it was our opinion that the best diagnostic fit for a working diagnosis was PPTTN. Treatment was initiated on the basis of the diagnosis.[26,27] Unfortunately, the patient did not have sufficient trust to continue the recommended therapy, even though it may have proven successful. Furthermore, we failed to address her psychological issues. Regardless of the outcome in this particular case, we feel that the cooperation between the oral medicine, orofacial pain, and prosthodontic clinicians was beneficial and necessary in our attempts to help the patient.

Also, the diagnosis of PPTTN was previously defined as traumatic neuroma. Traumatic neuroma/PPTTN cases documented traumatic injury from tooth extraction and local anesthesia resulting in orofacial neuropathic pain within the palate and maxilla, consistent with the findings in the case presented here.[4,6,31-37]

CONCLUSIONS

Our patient with multiple prosthetic therapy failures should serve as a learning example with regard to the importance of attaining a definitive diagnosis and evaluating psychosocial issues before repeating similar failing therapies. Hopefully, the lack of diagnostic expertise with respect to this case was an aberration, but it points to the failure of general dentists within a rural setting to refer patients to prosthodontists, oral medicine clinicians, and orofacial pain clinicians located in a more urban area. Furthermore, the responsibility for the treatment failure, as reported here, is shared by all of the treating clinicians; however, it is hoped that this report will help improve patient care in the future.

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