Persistent Idiopathic Facial Pain Following Dental Implant Placement: A Case Report

Elena Queral-Godoy, DDS, MS[1]/Eduardo Vazquez-Delgado, DDS, MS[2]/
Jeffrey P. Okeson, DMD[3]/Cosme Gay-Escoda, MD, DDS, PhD[4]

The present case report depicts the management of a patient with persistent idiopathic facial pain following the placement of 2 dental implants in the mandibular anterior alveolar ridge. After 15 months of unsuccessful diagnosis and management, the patient was seen at the Orofacial Pain Unit of the Oral Surgery and Implantology master's degree program of the University of Barcelona. Seven months after treatment onset, a combination of nortriptyline, clonazepam, and relaxation procedures has successfully controlled the patient's facial pain symptoms. INT J ORAL MAXILLOFAC IMPLANTS 2006;21:136–140

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Persistent idiopathic facial pain (PIFP) is reported to be one of the most frustrating facial pain conditions that challenge medical and dental clinicians.[1] To date, the prevalence of PIFP in the general population is still unknown. The available literature, however, indicates a higher prevalence of PIFP among women, with a concentration of cases of women in their late forties.[1] Although this facial pain condition was first described in 1947 by McElin and Horton by the term *atypical facial pain* (AFP),[2] so far there is still a lack of consensus in the medical and scientific community regarding terminology, classification, and diagnostic criteria. The International Classification of Headache Disorders of the International Headache Society[3] includes PIFP, in subgroup 13.18, "Central causes of facial pain," together with other entities such as anesthesia dolorosa, central poststroke pain, facial pain attributed to multiple sclerosis, and burning mouth syndrome. PIPF is included in group 13, "Cranial neuralgias and central causes of facial pain," as entity 13.18.4.

PIFP is diagnosed by excluding all other pathologies that may provoke facial pain in the affected area. Despite the disparity of diagnostic criteria, most authors agree that PIFP is best described as a chronic form of facial pain that is normally continuous, deep, and poorly localized, of low to moderate intensity, with sporadic episodes of intense pain. Furthermore, in cases of PIFP, investigations such as radiography of the face and jaw do not demonstrate any relevant abnormality.[4,5] Some patients, however, may describe their pain as throbbing, burning, or even stabbing. The pain is not associated with sensory loss

[1]Resident, Orofacial Pain Unit, Oral Surgery and Implantology master's degree program, College of Dentistry, University of Barcelona, Spain.
[2]Professor, Orofacial Pain Unit, Oral Surgery and Implantology master's degree program; Associate Professor, Department of Oral and Maxillofacial Surgery, College of Dentistry, University of Barcelona, Spain.
[3]Director, Orofacial Pain Center; Professor and Chair, Department of Oral Health Science, College of Dentistry, University of Kentucky, Lexington, Kentucky.
[4]Director, Oral Surgery and Implantology master's degree program; Chairman, Department of Oral and Maxillofacial Surgery, College of Dentistry, University of Barcelona, Spain.

or other physical signs, unlike burning mouth syndrome, in which pain is frequently accompanied by subjective complaints of dryness of the mouth, paraesthesia, or altered taste.[3] PIFP may frequently be initiated by surgery or injury to the face, unlike burning mouth syndrome, which is usually more unclear with respect to onset.[1,3] Hence, the majority of clinicians consider PIFP a form of deafferentation neuropathic pain.[6] The mechanisms through which pain is generated in PIFP remain unclear. Certain individuals may report that their pain started without any recognizable lesion to the facial structures; thus, the term *idiopathic* is used to describe this form of pain. The absence of any visible tissue injury, nevertheless, does not rule out a neuropathic origin for the disease, since several studies have demonstrated that tissue damage is not always necessary to cause a disruption of the central nervous system (CNS) pain modulatory mechanisms.[1] On the other hand, several authors have suggested a psychological origin for this pathology.[7,8] This hypothesis is based on the high prevalence of psychological conditions in PIFP patients. Indeed, several studies have shown a strong correlation between PIFP and several psychological conditions, such as depression, anxiety, and somatization disorders.[8–10] Still, as in other chronic pain populations, it is not clear whether psychological distress is the cause or the result of the pain. To date, for that reason, the pathophysiological mechanisms of PIFP remain to be elucidated. Most authors, nonetheless, identify PIFP as a neuropathic pathology.

Once PIFP is diagnosed, appropriate therapy should be established, avoiding any further damage to the facial structures. Thus far, there is a lack of high-quality clinical trials (randomized clinical trials) that would support the use of any specific form of therapy for the treatment of PIFP. Nonetheless, clinical experience and the available scientific literature indicate that certain medications used for the treatment of neuropathic pain seem to be at least modestly effective in the treatment of PIFP. Among these medications, tricyclic antidepressants (TCAs) appear to be the most effective.[1,5] Amitriptyline, at doses ranging from 25 to 100 mg a day,[6] has been used more frequently than any other medication. Other studies also demonstrate that phenothiazine,[11] β-blockers,[12] and certain anticonvulsants, such as clonazepam,[13] gabapentine,[14–16] and baclofen,[12] seem to be fairly effective in the treatment of PIFP. Topical medications such as capsaicin[6] at a concentration of 0.025% and minor opiate analgesics, such as tramadol or codeine, may also give good results in certain patients.[17]

When pain is extreme and cannot be controlled with these medications, opiate analgesics such as morphine sulphate, methadone, or fentanyl may be used in selected individuals.[13,14] These patients, however, are best managed at a multidisciplinary facility, where thorough psychological screenings and random blood tests are performed on a regular basis.

CLINICAL CASE

A 55-year-old Caucasian woman with a 15-month history of orofacial pain following the placement of 2 dental implants in the mandibular anterior alveolar ridge was seen at the Orofacial Pain Unit of the Oral Surgery and Implantology master's degree program at the University of Barcelona. The patient reported a constant burning pain localized in the mandibular anterior alveolar ridge, with an intensity of 9 of 10 on a visual analog scale (VAS). According to the patient, the pain started after the placement of 2 dental implants in the anterior mandibular alveolar ridge (Figs 1 and 2). These implants were removed 2 weeks later because of the patient's pain complaints. Their removal did not improve the patient's symptoms. The patient underwent a thorough radiographic examination of the painful area including panoramic radiographs (Fig 3), computer tomography, and bone gammagraphy. These examinations did not show any relevant abnormalities.

The patient's past medical history revealed thalasemia minor, which had been successfully managed by her family physician. Medication trials such as carbamazepine, oxcarbazepine, gabapentine, and amitriptyline failed to solve her current symptoms because of inadequate pain control or side effects. Psychological therapy had also been tried with modest results. At presentation, the patient was taking ibuprofen (600 mg) and lorazepam (0.5 mg) QHS. The patient was evaluated using the Symptom Check List-90-Revised (SCL-90-R)[18] at her initial visit. Her scores were within 1 standard deviation of the average scores of the general population and thus not clinically significant. The results of a cranial nerve examination were also within normal limits. Mandibular and cervical ranges of motion were also normal. Palpation of the cervical and mandibular muscles did not provoke any pain or increase the existing pain. There was also no pain when the temporomandibular joints were palpated. The intraoral examination was within normal limits. However, palpation of the mandibular anterior alveolar ridge increased the patient's pain.

Based on the patient's history and clinical examination the diagnosis of persistent idiopathic facial pain was established. Initial treatment began with educating the patient about the condition and teaching her some basic physical self-regulation

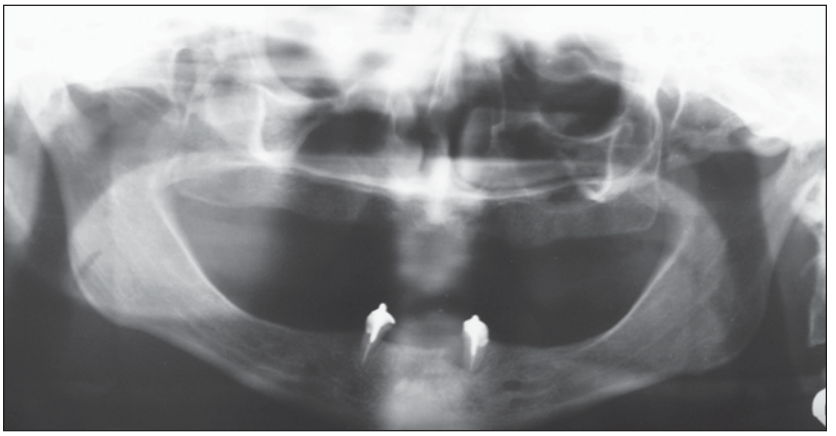

Fig 1 Panoramic radiograph 1 week before the placement of the implants.

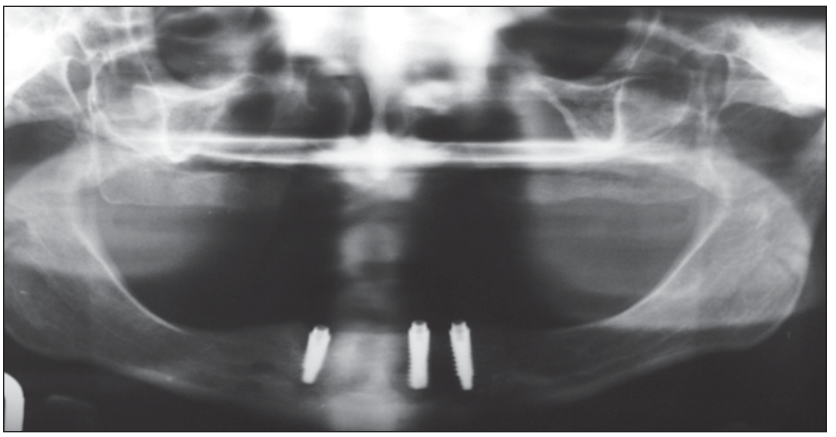

Fig 2 Panoramic radiograph 1 week after the placement of the implants.

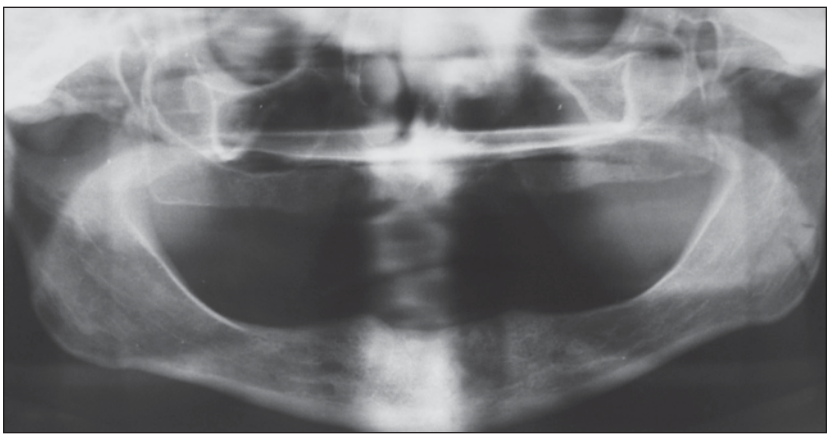

Fig 3 Panoramic radiograph 1 year after extraction of the implants.

strategies aimed at reducing sympathetic nervous system up-regulation.[19] A series of blood tests was requested to obtain baseline parameters before the administration of any medication. Medical management began with 25 mg nortriptyline before bedtime. The patient's evening dose of lorazepam was also replaced by 0.5 mg clonazepam. The patient was instructed to increase her dose of nortriptyline by half a tablet every week until pain control was reached. She was also advised to contact the clinic if she experienced any major side effects, eg, tremors, tachycardia, sedation, drowsiness, rash, or fatigue.

After 2 weeks of treatment the patient reported a 50% reduction (on a VAS) of her orofacial pain. She was taking 50 mg of nortriptyline and 0.5 mg of clonazepam at bedtime. The patient reported no side effects from the medications. Given the partial response and lack of adverse side effects, the decision was made to increase the daily dose of nortriptyline to 75 mg and the daily dose of clonazepam to 1 mg (0.5 mg twice a day). Two months later the patient reported an overall 75% improvement (on a VAS) of her orofacial pain. Since the patient did not complain of any side effects from the administered

medications, the daily dose of clonazepam was then gradually increased to 0.5 mg 4 times a day. She was scheduled for a follow-up visit in 4 months.

The patient was re-evaluated 7 months after treatment onset. She reported that her current pain level was 0/10 (on a VAS). There was still some tenderness on palpation of the mandibular anterior alveolar ridge. There were no side effects from the medications. The patient was instructed to continue with her present medical treatment (nortriptyline 75 mg at bedtime and clonazepam 0.5 mg 4 times a day), and she was scheduled for a follow-up visit in 6 months.

DISCUSSION

The prevalence of PIFP following dental implant placement is unknown. Furthermore, a MEDLINE search on the topic of "dental implants" and "PIFP" provided no reports regarding PIFP following dental implant surgery. However, the prevalence of PIFP following other dental procedures, such as endodontic treatment, is estimated to be around 3% to 6%.[20,21]

The treatment of PIFP is predominantly pharmacological. According to the available literature, TCAs seem to be the most effective medications for the treatment of PIFP.[1,5] However, the mechanisms by which TCAs inhibit pain are far from being understood. It is well established that TCAs inhibit the recapture of serotonin and norepinephrine. These neurotransmitters are known to be present in CNS sites involved in pain inhibition, such as the locus ceruleus, the periaqueductal grey matter, and the raphe nucleus.[22] Therefore, it is suggested that TCAs could mediate therapeutic effects by increasing the activity of CNS pain inhibitory mechanisms.

Additionally, TCAs have affinity for muscarinic, histaminic, and β-adrenergic receptors.[23] This lack of receptor selectivity is responsible for the majority of their side effects. While amitriptyline presents a high affinity for these receptors, other TCAs, such as nortriptyline and desipramine (secondary amines), present a lower affinity.[24] In the present case the patient reported intolerable side effects, such as high sedation and tremors, when taking amitriptyline. For that reason nortriptyline was the medication of choice.

Clonazepam, a benzodiazepinic anticonvulsant drug that acts on GABAergic receptors and chloride ion channels, is known to be moderately effective in the treatment of neuropathic facial pain conditions.[13,15] However, there is a lack of scientific literature regarding its use for the treatment of PIFP. Clonazepam, like other benzodiazepines, is also used for the treatment of sleep and anxiety disorders.[23] In the present case, the patient reported sleep improvement when taking lorazepam at bedtime. However, lorazepam is not known to be effective in the treatment of neuropathic pain. Therefore, clonazepam was substituted for lorazepam. As previously mentioned, clonazepam adds to its sedative properties a moderate effectiveness in the treatment of neuropathic facial pain disorders.

Long-term administration of medications is a major concern for many clinicians when managing lifelong chronic pathologies. However, thoughtful drug selection, careful dosing, and judicious monitoring of side effects will improve patient safety and increase treatment efficacy. According to the available scientific literature, chronic administration of TCAs for pain management is generally well tolerated by patients.[25] For long-term administration, nortriptyline and desipramine are the preferred TCAs compared to amitriptyline, as these medications cause less sedation, cognitive impairment, orthostatic hypotension, and constipation.[24,25]

Clonazepam, like other benzodiazepinic agents, requires close monitoring due to its well-established potential for producing physical and psychological dependence. Dependence on benzodiazepine therapy, however, varies depending on the dosage, duration of therapy, and pharmacological properties. Short-acting, highly potent agents such as alprazolam are more likely to cause dependence than longer-acting agents such as clonazepam.[26] Physical dependence should be expected with long-term treatment and should not be confused with psychological dependence ("addiction"), manifest as drug abuse behavior. Long-term benzodiazepine therapy, therefore, should never be abruptly discontinued to avoid the development of an abstinence syndrome.

This case report presents PIFP as a possible complication of dental implant surgery. It also suggests that a combination of nortriptyline, clonazepam, and relaxation training procedures may be helpful in the management of PIFP. Future studies are needed to determine the exact prevalence of PIFP following dental implant surgery.

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