Case Series

Management of Pain Secondary to Temporomandibular Joint Syndrome with Peripheral Nerve Stimulation

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Free full manuscript: www.painphysicianjournal.com **Background:** Temporomandibular joint syndrome, or Costen syndrome, is a clinically diagnosed disorder whose most common symptoms include joint pain and clicking, difficulty opening the mouth, and temporomandibular joint discomfort. The temporomandibular joint (TMJ) is supplied by the auriculotemporal nerve, a collateral branch of the mandibular nerve (the V3 branch of the trigeminal nerve).

Objectives: The aim of this study is to assess the effectiveness and safety of permanent peripheral nerve stimulation to relieve TMJ pain.

Study Design: This case series is a prospective study.

Setting: Pain Unit of a regional universitary hospital.

Methods: The study included 6 female patients with temporomandibular pain lasting from 2 to 8 years that did not respond to intraarticular local anesthetic and corticoid injections. After a positive diagnostic block test, the patients were implanted with quadripolar or octapolar leads in the affected preauricular region for a 2-week stimulation test phase, after which the leads were connected to a permanent implanted pulse generator. Results of the visual analog scale, SF-12 Health Survey, Brief Pain Inventory, and drug intake were recorded at baseline and at 4, 12, and 24 weeks after the permanent implant.

Results: Five out of 6 patients experienced pain relief exceeding 80% (average 72%) and received a permanent implant. The SF-12 Health Survey results were very positive for all specific questions, especially items concerning the physical component. Patients reported returning to normal physical activity and rest at night. Four patients discontinued their analgesic medication and 1 patient reduced their gabapentin dose by 50%.

Limitations: Sample size; impossibility of placebo control.

Conclusion: Patients affected with TMJ syndrome who do not respond to conservative treatments may find a solution in peripheral nerve stimulation, a simple technique with a relatively low level of complications.

Key words: Temporomandibular joint disorder, temporomandibular joint syndrome, Costen syndrome, peripheral nerve stimulation, auriculotemporal nerve stimulation, preauricular block, clinical safety and effectiveness, trigeminal neuralgia

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emporomandibular joint (TMJ) syndrome, or Costen syndrome, is a functional disorder that the most typical symptoms are articular pain and clicking, difficulty opening the mouth, and discomfort in the temporomandibular joint (1).

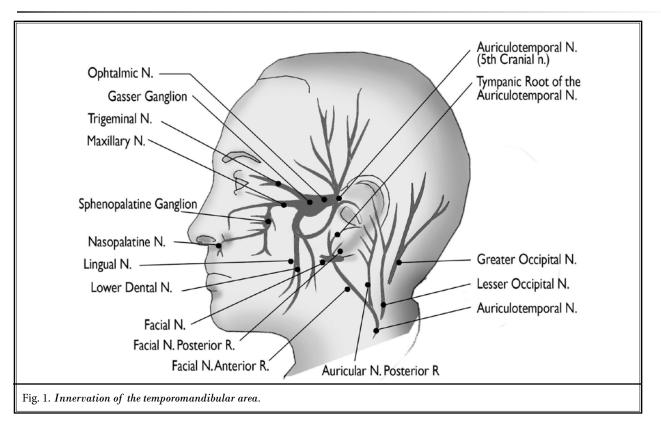
From the clinical standpoint, the usual symptom of TMJ syndrome is very intense pain in the temporomandibular joint or mandible. This pain may spread to one side of the scalp, nape, or neck, and worsen with chewing, yawning, or talking for long periods. Additionally, some patients mention temporomandibular rigidity with difficulty opening the mouth or chewing, articular clicking and cracking, and a brief feeling of closing or locking of the mandible when trying to open or close the mouth.

The diagnosis of TMJ syndrome is chiefly clinical, and diagnosis includes 1) examining for the presence of articular clicking, 2) careful inspection of the teeth and mandibular joints, 3) palpation of the mandibular joints as well as the head and face muscles, and 4) conducting radiologic studies, which may be of great diagnostic help. Today, the main diagnostic procedure is magnetic resonance imaging (MRI), which shows that in all cases of TMJ there is an anterior meniscus dislocation (2).

INNERVATION

The TMJ is innervated by the auriculotemporal nerve (a collateral branch of the mandibular nerve), which in turn is a ramification of the trigeminal nerve. It emerges from 2 roots coming from the mandibular nerve's posterior division. These branches form a buttonhole through which the meningeal artery passes, and then they join in a single nerve. The auriculotemporal nerve runs laterally to the mandible's neck, where it gives birth to the parotid branches, and continues upward to innervate the ear, the external auditory conduct, the external side of the tympanic membrane, and the skin of the temporal region. It also provides a couple of articular branches for the temporomandibular joint. The auriculotemporal nerve is highly sensitive to pain, and its irritation causes the abundant and strong symptoms experienced by patients with TMJ syndrome.

Both sensitive and vegetative nerve fibers form the auriculotemporal nerve. It supplies sensitive information, among others, from the temporomandibular joint, outer ear, and skin of the temporal, pterional, and outer ear regions. Its vegetative role comprises the parasympathetic and sympathetic innervations of the TMJ and temporal, pterional, and outer ear regions (Fig. 1).



METHODS

The hospital's Ethics Committee approved this prospective study, and enrolled patients signed a written consent prior to their inclusion in the study. We included 6 consecutive patients, all of them female, aged 24 - 49 (average 32.66) years. All patients presented with temporomandibular pain, with unilateral pain in 5 patients and bilateral pain in one patient. The pain had neuropathic characteristics that were perceived as electric, lancinating, and oppressive, with dysesthesia/paresthesia. All patients suffered from poor sleep, associated depressive symptoms, and limited mouth opening. All had undergone one or more maxillofacial surgeries without satisfactory results in terms of pain relief and improved mouth opening, as well as various pharmacological and orthotic treatments, none of which provided relief. In all patients, the pain evolved over a period longer than 2 years, reaching 8 years in 2 patients.

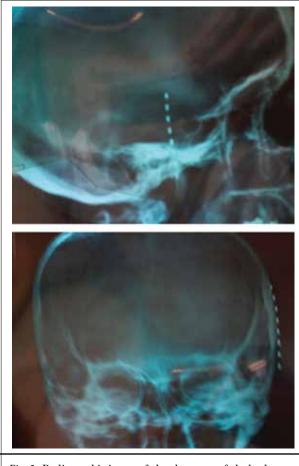
All patients had been treated, without success, with intraarticular infiltrations of local anesthetics and corticoids, after which they underwent a pre-auricular block of the affected temporomandibular joint with 5 mL of lidocaine 2%. The block provided immediate pain relief in all patients and increased their ability to

open the mouth; however, the analgesia was temporary, and both the pain and lack of functionality reappeared at the end of the lidocaine's effect duration. Nerve blocks are considered standard tools to confirm the presence of peripheral neuralgia; therefore, we considered that pain suppression after a pre-auricular block was a prerequisite for candidacy for implantation of a peripheral nerve stimulation (PNS) system. In all 6 patients, after the pre-auricular block with local anesthetic (Fig. 2), pain decreased by 80 – 100% from its initial intensity with an effect duration of more than 6 hours. Therefore, all 6 patients were accepted for temporomandibular trial stimulation by implantation of a pre-auricular lead.

The main inclusion criteria for PNS (3) were 1) severe pain in the temporomandibular joint (uni- or bilateral) with a duration of more than 6 months; 2) failed or inadequate relief from other treatments, including intraarticular infiltrations and surgical procedures; 3) durable relief and increase in functionality after local anesthetic infiltration in the ipsilateral pre-auricular region; and 4) being declared fit for neurostimulation treatment by a psychologist. Exclusion criteria were common to all other candidates for neurostimulation.



Fig. 2. Auriculotemporal nerve puncture technique using the preauricular approach.



Fig, 3. Radiographic image of the placement of the lead.

All patients signed a written consent. The implanted system (Advanced Neuromodulation Systems, Inc., Plano, TX, USA), had 6 leads, including 3 quadripolar leads (Axxess, model 4143) and 3 octapolar lead (Octrode, model 3183). After infiltrating the needle's entry point with local anesthetic, the surgeon located the mandibular angle and performed a small incision. A 14-gauge or 18-gauge needle was tunneled subcutaneously in a caudocephalic direction adjacent to the pinna, targeting the tip toward a point located 1 cm anterior to the tragus in order to avoid damage to the vasculonervous structures, until electric paresthesia overlapping the painful area was obtained. Once the desired paresthesia was obtained, the lead was tunneled to the patient's back and connected to an external test stimulator. Stimulation parameters were programmed according to conventional spinal cord stimulation procedures, seeking the stimulation program that provided maximum relief to each patient (Fig. 3).

In all cases, the pain intensity and the patient's general condition were assessed before the beginning of the trial stimulation and at 2 weeks after the lead implantation, after which the test phase was considered to be over and a permanent pulse generator (Genesis model 3608; Advanced Neuromodulation Systems, Inc.) was implanted. All patients who passed the trial phase were followed up at 4, 12, and 24 weeks after receiving the permanent implant.

The tools used to evaluate each patient's general condition were the SF-12 Health Survey and the Brief Pain Inventory (BPI). All surgical procedures and programming sessions were recorded on special forms, along with the results of a patient satisfaction questionnaire and reports of any complications that occurred during the treatment.

RESULTS

All patients answered several questionnaires before being treated with electric neurostimulation. The results showed that the mean pain score before treatment was 7 on a 0 - 10 visual analog scale (4-8), with average relief of 30% obtained from previous medications. All patients reported pain in the temporomandibular region that limited their ability to open their mouth wide.

Five out of 6 patients experienced relief greater than 80% compared to their basal pain intensity. Of the 6 patients, 5 continued with neurostimulation therapy, with a level of analgesia that allowed them to open their mouth, yawn, and chew without being prevented by pain. One patient stopped the treatment because of facial stimulation that provoked lip retraction and uncomfortable paresthesias in the ipsilateral eye.

Because of the relative simplicity of the surgical procedure, no surgical or postsurgical complications were observed. Consequently, all patients were able to complete the 2-week trial stimulation phase before the implantation of the permanent pulse generator. Parameter adjustments were carried out without complications in the postoperative room, and all systems were programmed with a single set of parameters (without subprograms or stim-sets).

Regarding the only patient with bilateral pain, after a successful 3-month stimulation therapy in her left TMJ a second octapolar lead was implanted in the right side with similar results in terms of pain relief, functionality and satisfaction. Results of the SF-12 Health Survey regarding patients' perceived general health were very positive for all specific questions, especially

Question	Scale	Baseline	4 wk	12 wk	24 wk	Improvement
In general, would you say your health is:		2.5	2.4	2.4	2.3	- 7%
Does your health limit you in moderate activities?		1.1	2.0	2.1	2.2	100%
Does your health limit you in climbing stairs?		1.1	2.0	2.0	2.1	92%
Accomplished less than you would like?		1.1	1.4	1.5	1.5	38%
Were limited in the kind of work or other activities?		1.1	1.3	1.4	1.4	31%
Accomplished less than you would like due to emotional problems?	1 – 2	1.1	1.5	1.4	1.7	54%
Didn't do work or other activities as carefully as usual due to emotional problems?		1.1	1.5	1.4	1.7	54%
Did pain interfere with your normal work?	1 – 5	1.8	3.3	3.3	3.4	95%
Have you felt calm and peaceful?	1 – 6	3.1	3.5	3.6	3.9	27%
Did you have a lot of energy?	1 – 6	2.7	3.3	3.4	3.6	34%
Have you felt downhearted and blue?		3.5	4.3	4.3	4.6	31%
Has your physical health or emotional problems interfered with your social activities?	1 – 5	2.3	2.5	2.5	2.6	11%

Table 1. Patients' quality of life assessed over the study period according to the results of the SF-12 Health Survey.

those concerning the physical component (Table 1). It is necessary to mention that responses to the generic question, "In general, would you say your health is...," did not show an improvement, and the mean score for this item decreased 7% from baseline. Paradoxically, this result does not correspond with results for the rest of the questions, all of which showed a significant improvement.

All patients reported dramatic improvement in pain according to the results of the BPI, with an average improvement of 72% at week 2, 70% at week 12, and 73% at week 24 (Table 2). All patients reported returning to normal physical activity and restful sleep. At the end of the trial, 4 patients had discontinued their analgesic medications, and one patient had reduced their gabapentin dose by 50%. Five patients continued with the treatment and used their stimulation system regularly at the end of the trial. All 5 patients made intensive use of their stimulators for 13 – 18 hours per day.

COMPLICATIONS

As explained above, one patient suffered uncomfortable facial nerve stimulation provoking ipsilateral lip and eye retraction. The lead was repositioned 2 weeks after the initial implantation, but coverage was partly lost and the stimulation system was finally removed.

In one patient, the lead/extension system had to be explanted due to granuloma formation in the lead's anchoring area 15 days after the initial implant. As the achieved relief had been significant, another lead was implanted 5 months later and, after a positive 20 day

Table 2. Pain intensity and ADL interference assessed overthe study period according to the results of the BPI.

Patient	Baseline	4 wk	12 wk	24 wk	Improvement		
1	7.3	0.3	0.3	0.2	97%		
2	7.9	4.0	4.6	1.7	78%		
3	7.4	1.2	1.3	1.2	84%		
4	7.8	0.4	0.4	0.4	95%		
5	7.4	System removal					
6	7.1	0.3	0.3	0.3	96%		

ADL, activities of daily living; BPI, Brief Pain Inventory.

test-phase, she received the permanent stimulation system.

DISCUSSION

TMJ disorders were described as early as the time of Hippocrates, but it was James Costen in 1934 who first gathered a group of symptoms related to TMJ functional disorders, which would later be known as Costen syndrome. Among the most frequent symptoms of Costen syndrome, we find: 1) excessive tension of the mandible's muscles, limiting the joint's movement; 2) defective alignment of the upper and lower teeth, leading to unbalanced movement of the mandibular joint; 3) abnormal position or dislocation of the TMJ or its inner cartilage; and 4) evolving anomalies such as condylar alteration, congenital defects, acromegaly, traumatisms or luxations, joint inflammation or infection, and bone tumors (sarcomas, metastases, osteomas) (9). TMJ syndrome is associated with a series of risk factors, with some of the most frequent being 1) female, 2) age between 30 and 50 years, 3) clenching or grinding the teeth, 4) use of badly adjusted dentures, and 5) the presence of associated pathologies, such as fibromyalgia, stress, and arthritis. Because a high number of patients with TMJ syndrome have associated components of anxiety and psychophysiological disorders, the initial step with these patients is to eliminate the stress and try to relax the joint, which sometimes makes it possible to control both bruxism and articular clicking. The management of these patients includes (10):

Administration of a bland diet for obviating the articular stress. Chewing gum and stimulating drinks, such as coffee, tea, or alcohol, must be avoided. These measures are intended for attaining pain relief, which sometimes can also be achieved by the application of cold or tepid towels, restriction of mandibular movement, or by medical treatment, usually with paracetamol, nonsteroidal anti-inflammatory drugs, muscle relaxants, or antidepressants. In some patients, anxiolytics may also be administered for 2 weeks. At times, it is advisable to combine the pharmacological treatment with TMJ infiltrations of mixed local anesthetics and corticoids.

Use of a dental protector is recommended, with the aim of relaxing the mandibular muscles and avoiding clenching or grinding of the teeth.

Physiotherapy has been proven as a useful method for management of TMJ syndrome. The patient learns to perform small exercises aimed at reinforcing the local muscles and improving the joint's stability. For example, patients may softly massage the masseter muscle for 5 minutes, 2 or 3 times a day, followed by compressing a thick pencil with the teeth for approximately 5 minutes.

Surgical procedures are considered the last resort when all the described treatments have failed to produce pain relief.

In the past few years, peripheral electrical nerve stimulation has become a common practice, producing good medium- and long-term results in the management of chronic pain. Its use for peripheral nerve disorders is not well documented, and its application in patients with chronic pain secondary to Costen syndrome is not currently confirmed by clinical trials reported in the medical literature. The aim of this study was to assess the effectiveness and safety of peripheral electrical nerve stimulation in a series of 6 patients with pain secondary to TMJ syndrome in a prospective study. Chronic pain secondary to TMJ is a condition that may affect a much larger population than commonly thought. The number of patients who may be candidates for neurostimulation because of intense pain and lack of success with other treatments is unknown, but these patients could constitute an important population group.

Invasive procedures have proved very effective in the management of various painful syndromes refractory to medical treatment, but their use is controversial because they destroy nerve structures that cannot be easily rebuilt. On the other hand, neurostimulation is completely reversible and, in the case of the patient who decides not to continue with the treatment, the system can be removed without causing problems.

The results obtained in the patients in this study support, in our opinion, the proposed technique. To validate changes in the quality of life, we used the Spanish version of the SF-12 Health Survey, adapted to the Spanish population by Alonso et al (4,5). Results are expressed in percentiles, and the value scale is directly proportional to the patient's degree of health. It was easy for the patients to understand, and it took 2 minutes, on average, to fill out the questionnaire. The mean scores improved for 11 out of 12 items, leading us to think that the technique improved the quality of life for our patients. Beneficial results were also found in terms of pain relief according to results from the BPI. The BPI is a multidimensional pain assessment survey that provides information not only about pain intensity, but also about its interference in daily activities, thus making it a suitable tool for assessing the effectiveness of analgesic treatment. The BPI was developed by Daut in 1983 (6), and the Spanish version was validated by Badía et al in 2002 (7). It consists of 2 sections, namely "pain intensity" (4 items) and "interference in activities of daily life (ADL)" (7 items). Each question is scored from 0 (absence of pain/absence of interference in ADL) to 10 (worst imaginable pain/highest interference in ADL), and a summarized score from the individual results is calculated for each section. It is remarkable that all 5 patients reported overall improvements of more than 50%, with 3 patients reporting improvements of more than 90%. The results corroborate the technique's effectiveness in this group of patients.

Electrical neurostimulation systems are classified as intraspinal or extraspinal, depending on the stimulated elements in each case. Aló and Holsheimer (8) established the intra- and extraspinal classifications and considered that intraspinal systems always targeted a specific spinal nerve, while extraspinal stimulation's target was always an extraspinal nerve. Traditionally, the term "peripheral" has referred to stimulation with the lead implanted on a peripheral nerve (tibialis posterior, median, sciatic, or radial), leaving the term "subcutaneous" for leads implanted without the presence of a specific peripheral nerve. However, on some occasions these concepts have been mixed up (11). Today, the main problem we encounter is the combination of both denominations, because in some cases the classification is built on the surgical technique and in other cases on the type of stimulation applied. Although both PNS and subcutaneous stimulation (PNfS) are carried out in a similar way from the technical point of view, the targeted and achieved results are different. PNS seeks, in all cases, to achieve paresthesia in the territory innervated by the stimulated nerve; however, when performing PNfS, paresthesia distribution is an electric field around the active poles, without obtaining a defined nervous distribution (12).

PNS was first performed in 1967 and was described by Wall and Sweet (13), who demonstrated that the electrical stimulation of a nerve provoked hypoesthesia and analgesia distally to the stimulated spot. This kind of stimulation reached its peak of popularity in the 1980s (14,15). The main indication for PNS is the existence of neuropathic pain in the territory corresponding to only one or 2 nerves, so that stimulation can be performed specifically on the affected nerves. PNS is based on the same principles as spinal cord stimulation. One of the main difficulties of this technique is precisely locating the electrodes to provide adequate stimulation to the targeted territory, since previous surgeries and traumatisms may make the lead placement in the selected area difficult. Well-innervated areas, such as the trunk, neck, head, and face, are sites that are more favorable for PNS, and this kind of stimulation has been used in patients with migraine, facial neuralgia, inguinal and other peripheral neuropathies, as well as chronic low back, pelvic, and perineal pain (16). There are studies supporting the effectiveness of PNS, to a greater or lesser degree, for migraine (17), cluster headache (18), trigeminal neuralgia (19), postherpetic neuralgia (20), and post-groin surgery pain (21). There are even works referring to transcutaneous stimulation in orofacial disorders (22). However, we failed to find any studies assessing the effectiveness of PNS for treating pain in TMJ syndrome.

The surgical technique we used is relatively simple, especially in connection with the test phase. The leads

were introduced percutaneously under local anesthesia, so that the patient could describe the area of paresthesia. Percutaneous leads were connected, directly or through extension cables, to an external trial stimulator. The leads may be located around the painful area or right on the most painful spot. The implantation depth is a critical factor because, if the leads are too superficial, the provoked paresthesia may produce pain and, if they are too deep, it may not be perceived, may be distorted, or may even cause muscular contractions (23). The basic aim is to achieve paresthesia coverage in the entire painful area with maximum patient comfort. The test phase may last 2 or 3 weeks and, once its effectiveness is assessed, the implanted pulse generator is implanted in the buttock or abdominal region during the second, or permanent, surgical phase.

CONCLUSIONS

Pain secondary to TMJ syndrome is, in certain patients, a clinical problem that is hard to solve. TMJ pain can lead to a severe decline in the quality of life, and can seriously affect the person's family, social, and work environments. Patients with chronic pain that has proved refractory to previous treatments (pharmacological or rehabilitative) may find a solution in PNS. The technique is quite simple, bearing in mind that the electrodes must be placed neither too deeply nor too superficially, and it has a relatively low level of pre- and postoperative complications. The limitations of this study are the small number of patients, and the impossibility of utilizing a double-blind, placebo-controlled approach, owing to the patient's ability to feel the stimulation and, therefore, be aware when the treatment is active.

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