

Peripheral Neurostimulation in the Management of Cervicogenic Headache: Four Case Reports

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■ ABSTRACT

Introduction. Neuromodulation, mediated by invasive electric stimulation, has been shown to be effective when applied to patients with refractory and intractable neuropathic pain. Recent advances in neurostimulation have broadened the therapeutic uses of this therapy, with the placement of extraspinal electrodes for peripheral nerve stimulation.

Methods. Four patients with long-evolving, persistent, severe, uncontrolled, and localized pain in the occipital region, in whom other management options had been tried and failed, were treated with a peripheral, occipital, extraspinal electric stimulation (C1-C2-C3).

We present, as case reports, the results of this intervention in these four patients.

Results. In all cases, stimulation of the occipital region yielded good or very good global results. In all patients, continuous pain disappeared, the frequency and severity of the episodic pain decreased, function improved, and restful sleep improved. As a result of stimulation we were either able to reduce or discontinue medication usage in all of our patients. ■

KEY WORDS: cervicogenic headache, peripheral occipital stimulation, transformed migraine

INTRODUCTION

The term cervicogenic headache, referred to by many specialists as transformed headache, is characterized by head pain originating at cervical spinal

levels. The underlying causes are numerous and include degenerative processes such as osteoarthritis or inflammatory arthritis, microtraumas to the joints of the spine, joint and articular soft tissue overload, injury (cervical whiplash, chiropraxis, etc.), abnormalities of the craniocervical junction (odontoid process dislocation, synarthrosis, etc.), disc disease (rare at upper neck level), and spinal cord, nerve root or peripheral neuropathy of a structural or infectious nature. Cervicogenic headache also encompasses a large group of patients with similar symptoms in whom the origin of the pain is not always known (1,2).

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The pain of cervicogenic headache occasionally presents as extensive radiation to the skull, neck, or shoulders, and may be neuropathic and/or mechanical in nature, associated with body postures and movements and limited neck mobility. This pain may be induced by applying manual pressure to cervical trigger points, and is only slightly improved with rest and the prescription of anti-inflammatory medications. The condition may also be associated with other frequent symptoms such as instability, blurry vision, and nausea, and less frequent symptoms such as vomiting, photophobia and/or phonophobia, or a homolateral swelling sensation (particularly around the eyes).

Since some temporary pain relief is achieved by anesthetic block of the affected nerve, a positive response to blocking of the nerve is a criterion common to both cervicogenic headache and neuralgia of the occipital nerve (3,4).

Neuromodulation, mediated by invasive electrical stimulation, has been shown to be effective when applied to patients with refractory and intractable neuropathic pain. The therapeutic principle of neuromodulatory techniques involves the application of an electrical current to a series of specific and well-defined locations of the nervous system. Alo and Weiner have used occipital stimulation in 62 patients with cervicogenic headache. These authors report a 75% incidence of good to excellent results over an average duration of follow-up of 22 months. A significant number of these patients have been able to abort progression of their transformed migraine with short stimulation periods (5). In a recent study by Popeney and Alo, occipital quadripolar double electrodes and Synergy generators were implanted in 25 patients who met the criteria for transformed migraine. Over a mean follow-up period of 18.3 months, 88% of the patients noted reduction of their disability and improvement of $\geq 50\%$ in the frequency and severity of headache (6).

CASE REPORTS

We present, here, four patients with persistent and uncontrolled pain in the occipital region, treated by means of peripheral, extraspinal, electric stimulation using an implanted system with a lead positioned in the cervico-occipital zone (C1-C2-C3).

Case Report 1

A 46-year-old woman presented with a 20-year history of migraine. She suffered bilateral occipital pain extending to the parietal, frontal, periorbicular, preauricular, and supraclavicular regions. The pain was described as being continuous and of mild to moderate intensity (visual analog scale [VAS] score = 4), with frequent crises of very severe intensity (VAS = 9-10). The pain was characterized by the patient as being oppressive with a sensation of dysesthesia, paresthesia, and swelling. Initially this woman's pain was unilateral, but, in the previous 4 years, the condition had worsened and spread, proving refractory to medication management. Her painful symptoms caused her to reduce her physical activity by 25% and she suffered from poor nighttime rest and depression.

The analgesic response to occipital neuronal block with local anesthetic was satisfactory, with improvement in excess of 50%, although lacking persistence over time. With the diagnosis of cervicogenic headache, electrical stimulation was carried out using an external stimulator with a C1-C2-C3, left, extraspinal, subcutaneous lead placement, for a test period of 1 month. Over this one month the patient derived 50% improvement of pain on the left side, with persistent symptoms on the right side. At the end of the trial, her test lead was removed. Three months later, after removal of this trial lead, two Pisces Quad®, quadripolar, C1-C2-C3, left and right double leads (Medtronic, Inc, Minneapolis, Minnesota, USA), with double-loop anchorage and fixation of the electrodes, were implanted at the medial cervical vertebral level (Fig. 1A,B) These leads were subcutaneously tunneled and connected to a Synergy® neuropulse generator (Medtronic, Inc, Minneapolis, Minnesota, USA), housed in a left gluteal pocket. The lead configuration and electric current parameters that we used in this patient are as follows:

- amplitude, 2.7 V;
- pulse width, 330 msec;
- frequency, 60 Hz; and
- the programmed polarity of the electrodes was as follows: left lead with poles 0 (distal) (-), 1 (+), 2 (+), 3 (+) and right lead with poles 4 (distal) (-), 5 (+), 6 (+), 7 (+).

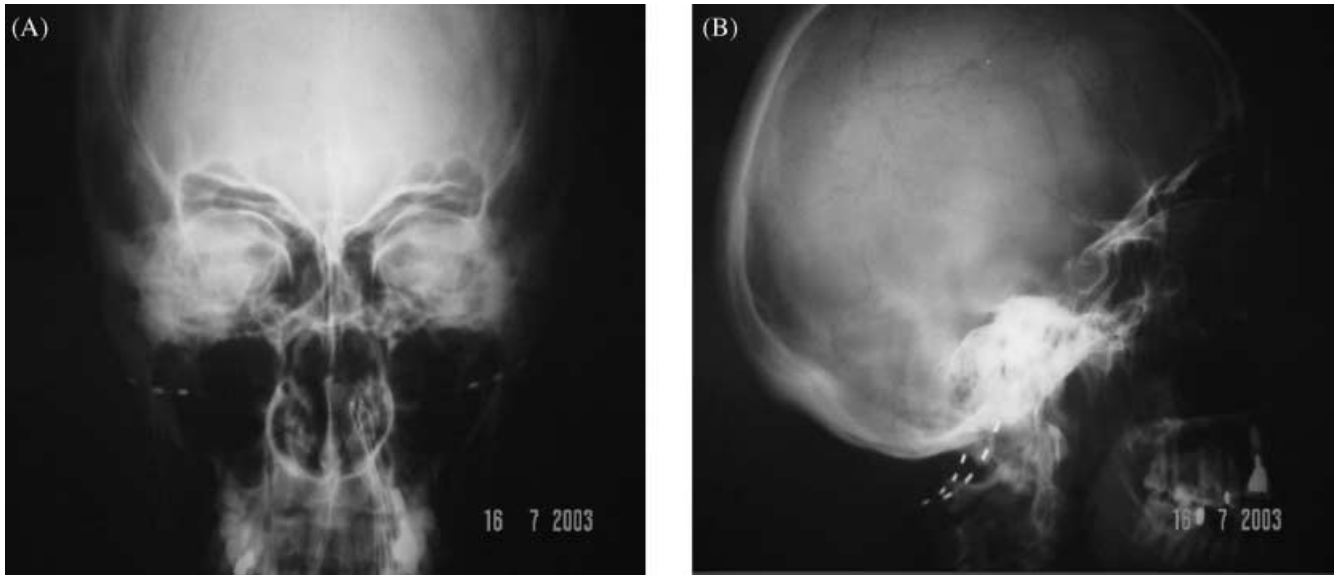


Figure 1. Case 1. A 46-year-old woman with cervicogenic headache. X-ray image showing positioning of the electrodes for C1-C2-C3 bilateral extraspinal stimulation. A: anteroposterior projection showing the right and left quadripolar electrode. B: lateral projection showing the double electrode.

The intended purpose of our stimulation parameters was to induce pleasant paresthesia within the patient's painful area. With preestablished, physician-programmed ranges, the patient had the option of self programming her stimulation. Stimulation time (continuous or intermittent) was activated by the patient in response to the time sequence of her symptoms.

This patient's continuous pain was cured by her stimulation (VAS = 0); moreover, her intermittent crises of pain became less frequent and of lesser intensity, and are aborted completely by maintaining continuous stimulation. All medications have been discontinued and the patient is now able to go about her activities of daily living in a normal fashion. This patient now has a normal family and social life and her nighttime rest is good. This patient has been followed up for 16 months, and her initial excellent analgesic response still persists. In the first 2 months the patient required continuous stimulation during the day, although at present the duration of stimulation has been reduced to 2 hours, three times a day.

Case Report 2

A 43-year-old woman presented with a history of C2-T1 syringomyelia with Arnold-Chiari malfor-

mation. Surgery, in the form of a suboccipital craniotomy with resection of the C1 posterior arch, dural sac aperture and plasty, and decompression of the cerebellar amygdala, had been performed to treat this condition, 11 years previously. The patient developed progressive localized occipital, cervicospinal and upper right limb pain after the operation.

During the first 4 years, she remained practically asymptomatic with analgesic medication and the use of occasional muscle relaxants. However, her clinical complaints gradually worsened with deep continuous pain that increased at night, requiring medication treatment everyday. She, moreover, suffered crises once or twice a year lasting about 1 month. The pain in these periods was described as very intense (VAS = 9) and was accompanied by sensation of numbness, itching, loss of strength, reduced sensitivity and vasomotor alterations of the upper limb with global edema, cold and blue skin, and functional limitations.

Magnetic resonance imaging revealed moderate to severe cervical spondylodiscarthrosis, with disc protrusions at multiple levels, cervical spinal canal stenosis, and a syrinx cavity from C2 to C6. With the diagnosis of complex regional pain syndrome, type 2, sympathetic block was performed and pharmacologic treatment and rehabilitation was provided without satisfactory response.

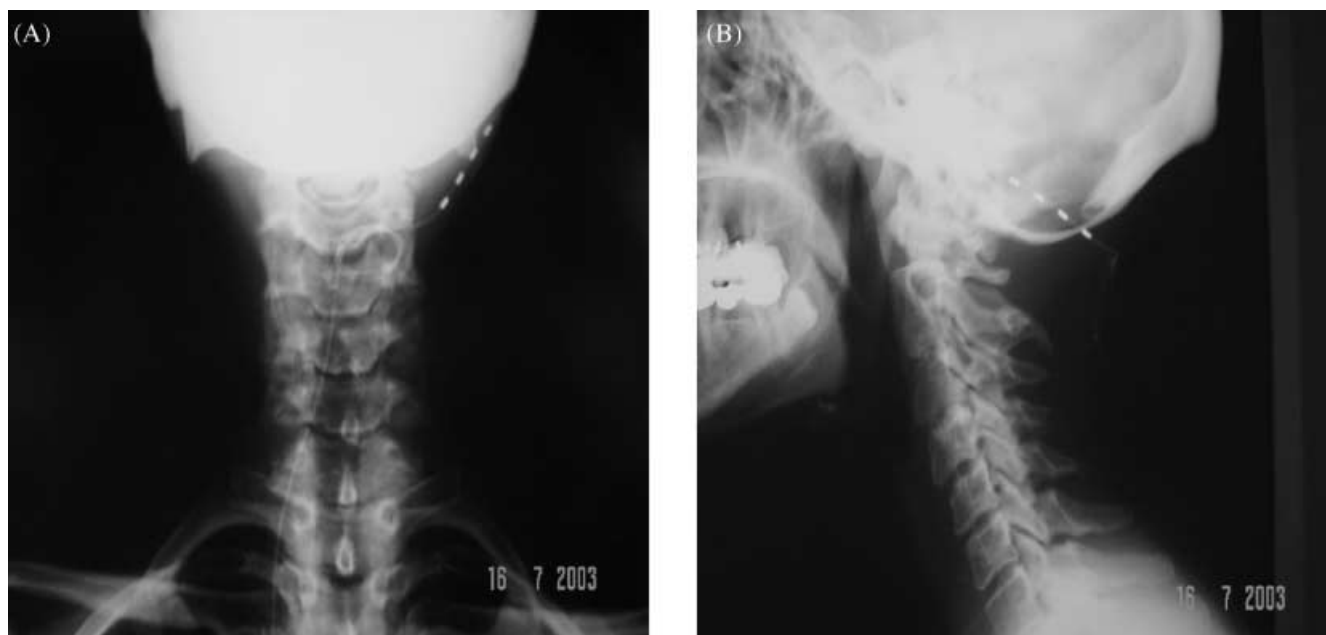


Figure 2. Case 2. A 43-year-old woman with complex regional type 2 cervical pain syndrome. X-ray image showing positioning of the lead for C1-C2-C3 right extraspinal stimulation. A: anteroposterior projection showing the right subcutaneous quadripolar lead with double-loop anchoring and fixation at medial cervical vertebral level. B: lateral projection of A.

The anatomical alterations in this case were considered by us to be a contraindication to cervical spinal cord stimulation placement. Right occipital neuronal block was performed on two occasions with local anesthetic, affording over 50% improvement that nevertheless subsided over subsequent days.

A Pisces Quad, quadripolar, C1-C2-C3, right, extraspinal and subcutaneous lead, with double-loop anchorage and fixation at the medial cervical vertebral level (Fig. 2A,B), was implanted for peripheral stimulation. This trial, providing this patient with at least 50% reduction in her pain, lasted 7 days. After the trial, the implanted lead was subcutaneously tunneled and connected to an Itriel-3® neuropulse generator (Medtronic, Inc, Minneapolis, Minnesota, USA), housed in a left gluteal pocket. Lead configuration and electric current parameters used in this case are as follows:

- frequency of 55 Hz;
- pulse duration, 210 μ sec;
- intensity amplitude 2.0 V; and
- the electrodes were programmed as follows: right lead with poles 0 (proximal) (+), 1 (+), 2 (-), 3 (-).

The results of occipital stimulation in this patient have been very good with an overall, global symptom improvement of 75–80%. Her pain intensity had decreased to a VAS of 0 and her reliance on medication was completely abolished by stimulation. The patient has returned to work. After a follow-up of 9 months, the patient uses stimulation on an intermittent basis for short time periods of 15–20 min, 2–3 times a day.

Case Report 3

This is an 80-year-old woman with a 4-month history of acute, C1-C2-C3, left herpes infection developing into chronic left occipital pain extending to the parietal and auricular regions. The pain was described by the patient as burning and itching with very frequent and recurrent crises of brief pain described as a severe darting sensation. She rated this pain on a VAS as a 9/10. Tactile sensitivity alterations and allodynia were noted.

With a diagnosis of postherpetic neuralgia, drug treatment was provided via the oral route and therapeutic occipital nerve blocks with anesthetic were performed. The patient did not tolerate the medications prescribed and the nerve block procedures performed were satisfactory, although

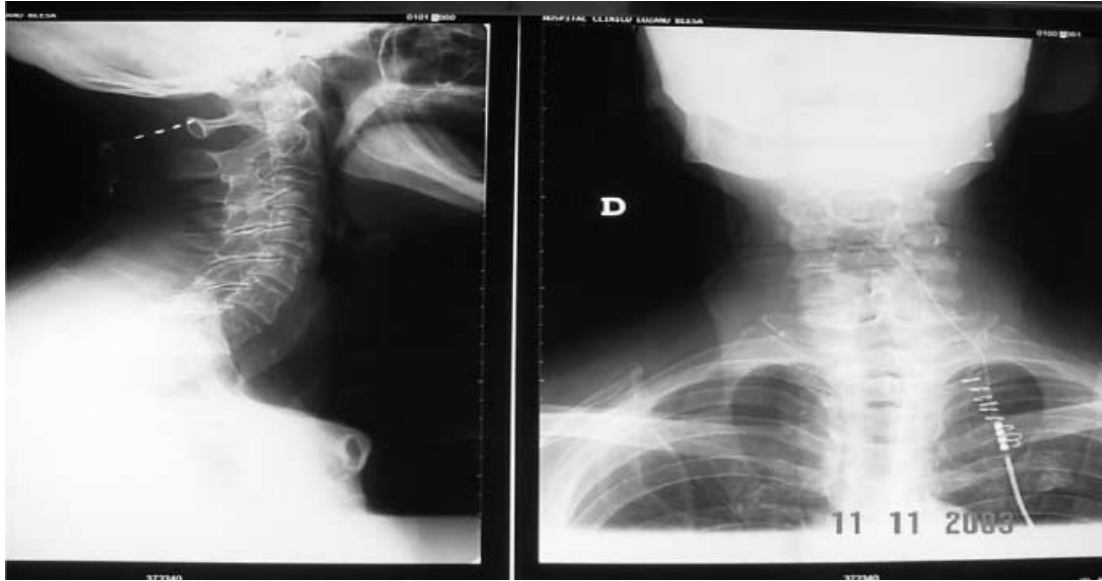


Figure 3. Case 3. An 80-year-old woman with C1-C2-C3 left postherpes neuralgia. X-ray image showing positioning of the lead for left occipital extraspinal electrical stimulation. Lateral and anteroposterior projections showing the left subcutaneous quadripolar lead with double-loop anchoring and fixation at medial cervical vertebral level, and left cervicodorsal paravertebral subcutaneous tunneling.

short lasting. After 6 months of conservative therapies, we considered the option of C1-C2-C3, left, extraspinal, subcutaneous, electrical stimulation.

A trial was performed with a Medtronic Pisces Quad quadripolar lead (Fig. 3) over a period of 9 days, with over 50% improvement. After this test period, a permanent system was implanted, with a Medtronic Itrel-3 neuropulse generator in a left gluteal pocket. This patient's stimulation system was programmed to the following parameters:

- electric current with a frequency of 60 Hz;
- pulse duration of 450 μ sec;
- intensity amplitude at 0.75 V;
- the patient had the option of self-regulation as need required; and
- activated electrodes of this left implanted lead were 0 (distal) (+), 1 (-), 2 (-), 3 (-).

After a follow-up period of 6 months, the patient states that she experienced 90% improvement in her pain, which now was rated to be 0 without drug treatment. Presently, she applies stimulation for 15 min, three times a day.

Case Report 4

This patient is a 33-year-old woman with a history of asthmatic bronchitis, ulcerative colitis, and

surgery at 20 years of age for a left internal carotid artery aneurysm. The patient began to suffer pain described as being of a neuropathic nature located in the occipital, temporal, and upper left limb regions 2 years after the aneurysmal surgery. Associated vascular reactive alterations were also observed. This condition of continuous mild pain (VAS = 3) and intermittent crises, usually coinciding with changes in climate, gradually worsened. Her crises were described as consisting of very severe and disabling pain (VAS = 8) with temporomandibular joint dysfunction, conjunctival injection, lacrimation, and functional impairment of the upper left limb with global lymphedema. This pain limited her daily family, social, and occupational life. Postoperative complex regional pain syndrome, affecting the side of the face, neck and occipital regions, and upper left limb, was diagnosed. Medication treatment and sympathetic block proved unsuccessful. Occipital neuronal block with local anesthetic afforded temporary improvement of over 50%.

A Pisces Quad, quadripolar, C1-C2-C3, left, extraspinal and subcutaneous lead with double-loop anchorage and fixation at the medial cervical vertebral level was implanted (Fig. 4) for a 7-day test stimulation period. A permanent system consisting of a Medtronic Itrel-3 neuropulse generator



Figure 4. Case 4. A 33-year-old woman with postoperative complex regional pain syndrome affecting the side of the face, neck and occipital regions, and upper left limb. X-ray image showing positioning of the lead for C1-C2-C3 left extraspinal stimulation. Lateral and anteroposterior projections showing the left subcutaneous quadripolar lead with double-loop anchoring and fixation at cervical vertebral level.

was implanted in a left gluteal pocket, resulting in over 50% pain improvement. The programmed lead configuration and electric current parameters were as follows:

- electric current with a frequency of 40 Hz;
- pulse duration of 450 μ sec;
- intensity amplitude of 0.3 V with the option of self-regulation as required by the patient; and
- the left lead was programmed with poles 0 (distal) (+), 1 (+), 2 (-), 3 (disconnected).

At 4 months, the patient stated that her global improvement was over 50%. Her continuous pain score was VAS = 0 with 60–70% resolution of the vascular reactive disorder and improvement in upper left limb function. She has been able to resume her daily family and social life. At present, the patient applies stimulation for 2 hours per day, and on a continuous basis during her pain crises.

DISCUSSION

Epidural, spinal cord neurostimulation with an implanted system was first used in clinical practice, in 1967, by Norman Shealy for the treatment of pain in terminal cancer patients. During the 1970s other investigators studied this technique

and extended its indication to the treatment of chronic pain associated with multiple nonmalignant diseases, and to “reeducation” of the nervous system in neurogenic bladder patients or individuals with motor and paretic spastic alterations. In recent decades, this treatment mode has been further extended to other diseases, with a progressive increase in both the number of treated patients and in the range of indications. These newer indications for spinal cord stimulation include intense back pain with surgical failure; nerve root condition secondary to postsurgical epidural fibrosis and arachnoiditis; painful neuropathy of different origins and affecting the limbs; critical reflex sympathetic algodystrophy, now known as complex regional pain syndrome; ischemic vascular disease of the limbs, comprising severe situations in which revascularizing treatment has either failed or proves nonviable (Leriche and Fontaine grades III, IV, and also some grade IIb cases); and coronary disease with unstable angina in the absence of other treatment options (affording reductions in anginal crises). The method has also been applied as symptomatic treatment in urinary incontinence associated with uninhibited reflex neurogenic bladder in multiple sclerosis patients (7–11).

Recent advances in neurostimulation have extended the therapeutic potential of neurostimulation to placement of extraspinal electrodes for stimulating peripheral nerves (occipital, supra-orbital, median, ulnar, radial, genitofemoral, peroneal, saphenous, and posterior tibial) (12). The indications for occipital stimulation include selected patients diagnosed with cervicogenic headache or occipital neuralgia in the form of C1-C2-C3 spinal transformed migraine refractory to treatment. Patients with this disorder must have diagnostic confirmation through temporary improvement or remission of symptoms in response to occipital nerve block and all patients must have no contraindications to the procedure including overriding psychiatric disorder, bleeding disorder, or systemic or local infections. Patients with cervicogenic headache or occipital neuralgia clinically present with muscle tension at the occipital base, and at the lower nape of the neck level, affecting the semispinal muscles of the head and neck vertebra. Such rigidity can be followed by discomfort with proximal radiation into the territories of one or both C1-C2 (greater occipital nerve) and C3 branches (lesser occipital and auricular nerves), i.e., commonly exhibiting a cyclic and recurrent pattern in spinal transformed migraine. Approximately 80% of these patients develop tactile allodynia in the territories of C1-C2-C3.

In all of our patients implantation was performed in two stages. The first stage was comprised of the technique described by Alo (5). During this stage, the lead(s) is(are) placed for an external trial of efficacy, preferably conducted by the patient on an outpatient basis in the home. In this first stage, the patient is required to keep a daily pain diary of the evolution of his or her pain and the actual time or times of stimulation. If the results are good, with at least 50% pain reduction, connection and subcutaneous housing of the generator is carried out during the second stage.

Technically, in our clinic, the first surgical stage is performed with the patient lying prone. A small longitudinal incision is made at the C1, medial, cervical vertebral level for placement of both a single lead for unilateral pain or two leads for bilateral pain. A 1-cm subcutaneous pocket is prepared on either side of this incision. Following dissection of this pocket, a 15G Tuohy needle, curved to follow the curvature of the occiput, is

inserted and advanced with the needle bevel facing downward in the subcutaneous plane and directed toward the lower portion of the ear. Superficial insertion traversing the dermis, or too deep an insertion through the fascia, could cause ineffective or painful stimulation. Either one electrode for unilateral pain or two electrodes for bilateral pain are positioned under fluoroscopic guidance. After the needle is removed, testing for concordant paresthesia to the cervico-occipital-auricular regions is performed intraoperatively. This portion of the procedure requires that the patient be awake and able to give positive feedback of the distribution of paresthesia felt. Once the electrode is deemed in the proper place, the proximal portion of the electrode(s) is fixed in a circular position with a silk suture to prevent displacement. In our patients, external stimulation was maintained for the duration of a test period lasting 1 month in the first case and about 7 days in the remaining three cases. After this test period, the definitive and permanent system is implanted by preparing a gluteal, subcutaneous pocket to house the neuropulse generator. Final x-rays are performed to confirm lead placement. Stimulation is carried out on a continuous or intermittent basis, depending on the severity of the patient's symptoms, and pulse intensity is variable and can be regulated by the patient within a predefined range.

Overall, we are encouraged by our results. After 2 years, case #1, the patient with bilateral cervicogenic headache, still has good analgesic response to occipital stimulation. As stated previously, this technique was selected in case #2, the patient with cervical complex regional pain, because cervical medullary electrical stimulation was contraindicated due to the presence of a syrinx at that level. This patient has approximately 50% reduction overall in her symptoms. This technique was indicated in our third case of left C1-C2-C3 postherpetic neuralgia because the condition could not be controlled with medication due to intolerance to all medications tried. It should also be stressed that the advanced age of the patient did not prevent understanding of the technique or adequate use of the method. Our fourth case, a patient with a vascular dysreactive condition, had an overall 60-70% improvement in symptoms including resolution of her vascular symptomology.

Unlike the technique used by Alo et al. where electrodes were placed away from the cervical midline in a rostrocaudal projection (5), all electrodes, in all of our patients, were generally placed at the site reproducing paresthesias within the painful area.

CONCLUSIONS

Severe cervicogenic headache is usually difficult to treat, and leads to situations of severe pain and impaired patient daily, family, social, and occupational life. These situations are typically refractory to drug treatment, rehabilitation therapy, nerve block, denervation, and other management approaches. Peripheral, extraspinal, cervico-occipital (C1-C2-C3), electrical stimulation, carried out with an implanted system, requires a minimally invasive surgical procedure that offers good to very good results in many cases refractory to more conservative interventions.

In all of our cases, reported in this paper, occipital stimulation offered good or very good global results with complete resolution of the patient's continuous pain complaint, a reduction in the frequency and intensity of pain crises, improved function, improvement in nighttime rest, and increase in daily life activities. We also found that medication use was either reduced or discontinued entirely. Based on these clinical observations, we believe that cervico-occipital stimulation offers hope of pain and symptom resolution to a group of patients that, heretofore, had little hope of pain and symptom relief.

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