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Case report

Implantable peripheral subcutaneous occipital neurostimulation for the treatment of refractory Arnold's neuralgia: Cases report☆

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ABSTRACT

Introduction: Peripheral nerve stimulation has yielded promising long-term results for managing occipital neuralgia refractory to pharmacological and primary interventional management.

Objective: To report the efficacy obtained in two cases of intractable occipital neuralgia that underwent implantable peripheral subcutaneous neurostimulation.

Material and methods: This is a descriptive and retrospective case report of severe Arnold's neuralgia refractory to pharmacological and interventional management, undergoing implantable peripheral subcutaneous neurostimulation performed during the last 10 years from 2006 to 2016 at the Pain Clinic Service of the National Medical Center Hospital "20 de Noviembre".

Results: Two cases of severe occipital neuralgia of traumatic etiology, refractory to pharmacological management, with over 9 years of evolution, were selected. Following management with implantable peripheral subcutaneous neurostimulation, the patients experienced 80–100% pain relief.

Conclusions: Peripheral subcutaneous neurostimulation seems promising as a short- and long-term therapy for the management of severe intractable occipital neuralgia.

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Neuroestimulación subcutánea periférica implantable de nervios occipitales para tratamiento de la neuralgia de arnold refractaria: reporte de casos

Palabras clave:
Estimulación eléctrica
Cefalea
Dolor
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Reporte de caso

RESUMEN

Introducción: La estimulación de nervio periférico ha obtenido resultados promisorios a largo plazo en el manejo de la neuralgia occipital refractaria a manejo farmacológico e intervencionista elemental.
Objetivo: Reportar la eficacia obtenida en dos casos de neuralgia occipital refractaria que fueron sometidos a neuroestimulacion subcutánea periférica implantable.
Material y Métodos: Es un informe de casos, descriptivo y retrospectivo, con diagnóstico de neuralgia de Arnold de intensidad severa refractario al manejo farmacológico e intervencionista, que hayan sido manejadas con neuroestimulacion subcutánea periférica implantable, realizada durante los últimos 10 años en el periodo comprendido entre los años 2006-2016 en el Servicio de Clínica del Dolor del Hospital Centro Médico Nacional "20 de Noviembre".
Resultados: Se obtuvieron dos casos de neuralgia occipital de intensidad severa y etiología traumática, refractario a manejo farmacológico de más de 9 años de evolución, quienes presentaron alivio del dolor 80-100% luego del manejo con neuroestimulacion subcutánea periférica implantable.
Conclusiones: La neuroestimulacion subcutánea periférica parece ser una terapia prometedora a corto y largo plazo en el manejo de la neuralgia occipital de intensidad severa intratable.

Introduction

Occipital neuralgia (ON) – or Arnold's neuralgia – is an acute, electrical, paroxysmal, and occasionally throbbing disorder originating from the occiput and extending along the posterior scalp at the distribution of the occipital nerves.[1] Peripheral nerve stimulation (PNS) in refractory cases is a promising strategy, with minimal adverse effects and long-term efficacy.[2]

NO is a primary and recurrent headache in the occipital region.[3] The etiologies associated with the development of this condition are: (1) Trauma, (2) Anatomical, (3) Tumors, (4) Infections, (5) Degenerative changes, and (6) Idiopathic. The most frequent etiology however is trauma.[4,5]

Materials and methods

Two cases of ON, refractory to pharmacological and interventional therapies, with severe neuropathic pain characteristics, previously managed with Implantable Peripheral Subcutaneous Neurostimulation devices in the last ten years from 2006 to 2016 at the Pain Clinic Service of "20 de Noviembre Hospital." A complete medical history was taken from each patient, emphasizing the algological assessment to establish the adequate interventional therapy based on the patient's evaluation. Once the interventional procedure was performed in each patient, the pain intensity has been monitored on a monthly basis until the present.

Results

Case 1

63-year old female evaluated at the pain clinic for the first time in 2014, with no relevant pathological history. The patient experienced a traumatic brain injury upon falling from a theestory building. She began experiencing pain at 17 years of age, with a 46-year evolution characterized by burning, shooting headache initiating from the occipital region and radiating to the right hemicranial and posterior cervical regions. The pain was constant with baseline intensity according to VAS of 5/10 and exacerbations of up to VAS 8/10, intensifying with palpation of the occipital region (Arnold's points) and sporadically at neck extension and flexion, with associated allodynia. The patient failed to respond to multiple medications. A test neurostimulation electrode was placed for one week, resulting in 100% improvement of pain, so therapy was considered effective. An Implantable Peripheral Subcutaneous Neurostimulation device was placed in September 2014. The implant procedure basically comprises two stages: (1) placement of the peripheral nerve stimulation electrodes, and (2) subcutaneous placement of the impulse generator. During the trial, only phase 1 of the procedure was implemented (placement of the stimulation electrodes). At this stage, the electrodes are connected to an external impulse generator, thus avoiding the need to do a subcutaneous placement of the impulse generator. The procedure took place in the sterile zone of the fluoroscopy suite, with the patient placed in left

lateral decubitus under non-invasive monitoring, conscious sedation (Midazolam 0.15 mg/kg and Fentanyl 1 mcg/kg), oxygen supplied trough nasal cannula at 3 L/min. Povidone-iodine was used to wash the back of the neck and the front of the chest and sterile drapes were placed.

During the first phase, 60 mg of 1% lidocaine were infiltrated into the skin and the subcutaneous tissue at the level of C1–C2 under fluoroscopy with a posterior-anterior approach, making a 1 cm craniocaudal incision at this level on the midline using the St Jude Medical™ implant kit. The 2.032 mm gauge introducer needle (Model 1114) was inserted with a right medial–lateral approach, following the transverse occipital curvature and avoiding overshooting the superficial muscular fascia, for safe placement of the electrodes over the occipital nerves. Through the introducer needle in place and under fluoroscopic visualization, the ST Jude Medical™ four-pole 60 cm Quattrode St Jude Medical™ ¾ electrode series No 3146 was advanced, following the same approach to place the left occipital electrode. The programmer engineer triggers a stimulation to confirm the correct positioning of the electrodes. The introducer needles are then removed and then the electrodes are fixed over the superficial muscular fascia using the St Jude Medical™ fixator kit model 110. During the second phase, the skin and the subcutaneous cell tissue is infiltrated with 100 mg of 1% lidocaine at the level of the middle third of the right clavicle, 2–3 cm infraclavicular in the medial-lateral direction. A 3 cm incision in the medial-lateral direction is made and then a plane by plane dissection is performed in a "hockey stick" pocket design, not exceeding the muscular superficial fascia, so as to properly insert the impulse generator. The St Jude Medical™ implant kit – model 1112 – tunneling device is used to subcutaneously place the stimulation electrodes all the way to the pocket and are then connected to the impulse generator using the St Jude Medical™ implant kit screwdriver. The programmer engineer tests the stimulation once again, to confirm the correct positioning of the electrodes and the proper functioning of the impulse generator. The wound is closed in layers and posterior-anterior X-rays of the impulse generator and the stimulation electrodes are taken as a final reference. The interventional procedure is thus completed uneventfully. Follow-up to this date shows 100% pain relief without the need to use any adjuvant pharmacological treatment.

Case 2

66-year old female with a history of a car accident in 1999 in which the patient sustained forced neck hyperextension. The patient was evaluated at the service in 2005, when she complained of constantly evolving blazing burning pain in the occipital region that irradiated into the right hemicrania with baseline VAS intensity of 6/10, exacerbations of up to 10/10 under palpation of the occipital region (Arnold's points), and during neck flexion and extension movements. Partial improvement was obtained (30%) with multiple therapies. A test neruostimulation electrode was placed for one week using the above-mentioned technique, achieving 80% pain improvement, so the intervention was considered effective. Also in 2008 an implantable peripheral subcutaneous device was placed according to the technique previously described, achieving 80% pain reduction. To this date, the average VAS has been 1–2 together with adjuvant pharmacological therapy with oral pregabalin 75 mg every 24 h.

Discussion

The first reference in the published literature about this technique was described by Picaza in 1977.[6] However, the technique became popular in the United States following a series of cases reported by Weiner in 1999.[7]

Physiologically, the analgesic effect of electrical stimulation in based on the gate control theory described by Melzack and Wall,[8] through a "closing gate system", whereby the electrical stimulation of large diameter fibers (A beta), inhibits the nociceptive transmission of the small diameter fibers (A delta and C). However, further research has suggested alterations in the axonal transmission of broad dynamic range and the facilitation of the descending inhibitory pathways and potential changes in neurotransmitters, particularly GABA (aminobutiric acid), but also glutamate, adenosine, acetylcholine, substance P, brain-derived neurotrophic factor, bradykinin, inter alia.[9] Likewise, electrical stimulation enables the orthodromic activation of the serotonin descending pathways, in addition to the effects on the processing of nociception in the brain centers.[10]

Stimulation electrodes are placed percutaneously at the subcutaneous level and positioned in the parasagittal direction along the path of the occipital nerves at the level of C1. A test should be done by placing a peripheral nerve stimulation electrode to evaluate the efficacy of the implantable device to relief pain. Such test is also critical for the prognostic efficacy of electrical nerve stimulation to relief pain at the spinal cord and brain level. Weiner published a number of retrospective trials in 150 patients with long-term 70–75% pain relief.[11] Systemic reviews have reported grade III evidence recommendations since most of those reviews comprise retrospective trials and case series.[12] In terms of primary interventional management, the use of botulinum toxin has reported a high level of treatment-refractory short term response.[13]

Two cases with severe intractable pain with over 9 years of evolution are identified, both due to trauma, which is the most frequent etiology in the development of Arnold's neuralgia.[14]

The percutaneous technique for implanting electrodes uses a minimally invasive procedure, with a low probability of experiencing complications.[15] Quadripolar percutaneous electrodes and blades are currently available, resulting in over 50% pain relief in long term reports.[16,17] The most frequently reported complication is migration of the stimulation electrode following implantation, with an incidence ranging between 13.9 and 24%.[18]

The efficacy of this implantable interventional therapy is based on a timely evaluation of a test neurostimulation electrode for 5 days. If during this period of time the patient experiences over 50% pain relief, he/she will be considered a good candidate for implantable therapy. On patients who have received implantable therapy, the cost-effectiveness outcome and long-term pain relief have proven to be superior to pharmacological therapy and basic interventional management, with a considerable reduction in the daily dose of drug therapy and its associated side effects. The half-life of the batteries

in the implanted neurostimulator is around 10 years and the battery may then be replaced.[19]

Conclusions

Peripheral Nerve Stimulation (PNS) for managing Arnold's neuralgia looks promising, with long-term 80–100% pain relief and minimally invasive approach free from any reported complications.

Randomized, systematic trials with sound statistical design are required to provide stronger scientific evidence to support the use of the Implantable Peripheral Subcutaneous Neurostimulation device for the management of this condition.

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Ethical disclosures

Protection of human and animal subjects. The authors declare that the procedures followed were in accordance with the regulations of the relevant clinical research ethics committee and with those of the Code of Ethics of the World Medical Association (Declaration of Helsinki).

Confidentiality of data. The authors declare that they have followed the protocols of their work center on the publication of patient data.

Right to privacy and informed consent. The authors declare that no patient data appear in this article.

Conflict of interest

The authors have no conflict of interest to declare.

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