



Benito's neuralgia: the first description of the occipital neuralgia was made for Spanish doctors at the beginning of the nineteenth century

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Abstract

Background The occipital neuralgia affects 3 out of every 100,000 people and includes the neuralgia of the greater occipital nerve (GON) and the neuralgia of the minor and third occipital nerves. These nerves emerge from the posterior branches of the first cervical roots, innervate the muscles of the nape, and provide the sensitivity of the scalp. The most frequent issue is not to find causes that justify neuralgia for what is usually idiopathic. The nerve that most often causes neuralgia is the GON that is usually wrongly called Arnold's nerve, so neuralgia is also called Arnold's neuralgia.

Methods We have reviewed the first description of occipital neuralgia.

Results Two Spanish doctors, José Benito Lentijo and Mateo Martínez Ramos, had already described in detail the neuralgia of the GON before Arnold was born. The first clinical case of occipital neuralgia due to GON involvement was published by them in a Spanish medical journal in 1821, and they called it cervico-suboccipital neuralgia.

Conclusion We claim in this article the role of these two Spanish doctors in the history of Neurology.

Keywords Occipital neuralgia · Occipital nerve · Arnold's neuralgia

Introduction

Medical literature shows us that it is possible to offer new diagnoses revisiting art or historical literary documentations or reconsider the paternity of classic syndromes [1, 2].

The occipital neuralgia includes the neuralgia of the greater occipital nerve (GON) and the neuralgia of the minor and third occipital nerves, these last two being infrequent. The neuralgia of the GON is also called suboccipital neuralgia or C2 neuralgia. Medical jargon has also wrongly named it Arnold's neuralgia, though this eponymous does not appear in International

Classifications of headache or in modern medical books. The occipital neuralgia is the most common cranial neuralgia after trigeminal neuralgia with a prevalence of 3.2:100,000 people [3]. Diagnostic criteria are defined in the 3rd edition of the International Classification of Headache (ICDH3, code 13.4) [4], as we show in Table 1. It consists on a unilateral or bilateral paroxysmal pain, lancinating or stabbing, located at the back of the scalp and distributed in the territory of the GON, and, less frequently, of minor occipital nerve or third occipital nerve (Fig. 1). In cases of GON involvement, the pain may reach fronto-orbital region due to the existence of neuronal connections with the trigeminocervical nuclei in the brainstem, and may even be accompanied by conjunctival hyperemia and ipsilateral tearing. Pain can be accompanied by hypoesthesia or allodynia.

The diagnosis of occipital neuralgia is clinical and the disappearance or relief of pain with an anesthetic block of the causative nerve confirms the diagnosis. It is recommended to perform a MRI to evaluate the cervical cord (specifically C1-C2-C3) and the atlanto-occipital junction.

Occipital neuralgia is commonly considered as idiopathic. In some cases, it is related with contact of the nerve with vessels or other anatomical structures. There have been described some cases of contact between GON and aberrant branches of the posteroinferior cerebellar artery (PICA),

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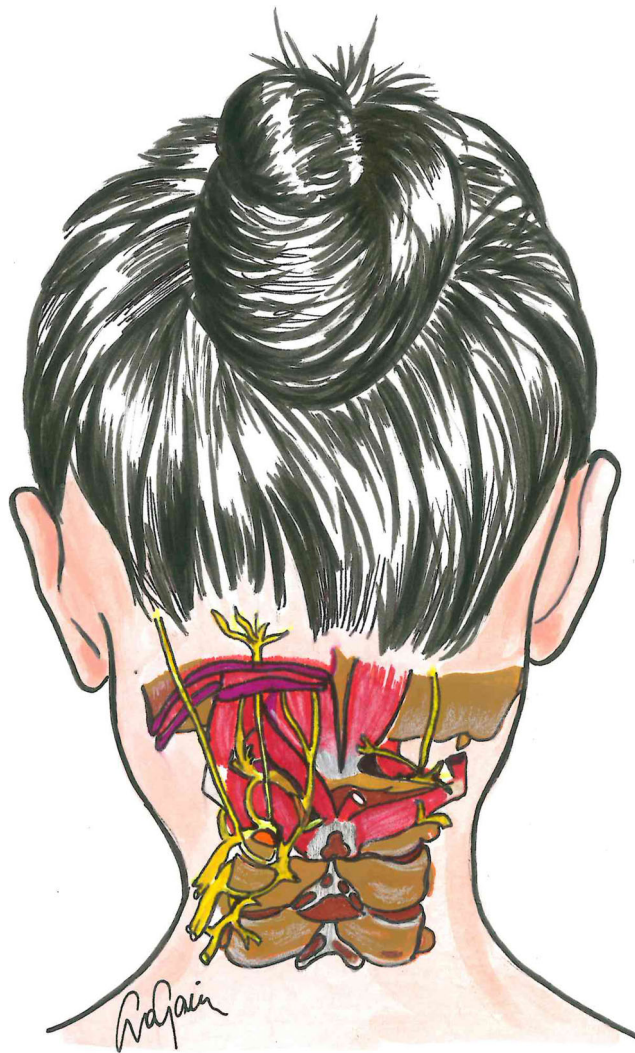
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Table 1 Occipital neuralgia diagnostic criteria (ICHD-3 classification, code 13.4) [2]

- A. Unilateral or bilateral pain in the distribution(s) of the greater, lesser, and/or third occipital nerves and fulfilling criteria B–D
- B. Pain has at least two of the following three characteristics:
1. Recurring in paroxysmal attacks lasting from a few seconds to minutes
 2. Severe in intensity
 3. Shooting, stabbing or sharp in quality
- C. Pain is associated with both of the following:
1. Dysesthesia and/or allodynia apparent during innocuous stimulation of the scalp and/or hair
 2. Either or both of the following:
 - a) Tenderness over the affected nerve branches
 - b) Trigger points at the emergence of the greater occipital nerve or in the distribution of C2
- D. Pain is eased temporarily by local anesthetic block of the affected nerve(s)
- E. Not better accounted for by another ICHD-3 diagnosis

**Fig. 1** Anatomy of the occipital nerves: greater occipital nerve (GON), lesser occipital nerve (LON), and third occipital nerve (3rdON)

fenestrations of the basilar artery, or dural fistulas. GON contacts have also been described with bone deformities of vertebrae C1 and C2 (inflammatory, fracture calluses ...). Other secundarisms infrequently described are very tight helmets on the head, lipomas, Chiari type I malformation, cervical myelitis, meningioma, multiple sclerosis, cervical surgery, neurinoma, herpes infection, or diabetes mellitus. Neuralgia of the lesser and third occipital nerve has been related to cervical radiofrequency procedures or post-surgery of C1 and C2, herniated discs, or after whiplash.

Treatment occipital neuralgia is based on drugs such as carbamazepine, oxcarbazepine, gabapentin, pregabalin, baclofen, or amitriptyline. Anesthetic blocks with or without corticosteroids are also effective and, in refractory cases, therapy with botulinum toxin type A can be proposed. If there is no response, radiofrequency of the dorsal root of C2 or GON stimulation can be performed. In refractory cases, a surgical section can be assessed. Treatment of the neuralgia of minor occipital nerve or third occipital nerves does not differ from that for GON neuralgia [5–9].

Undoubtedly, the first series of patients with occipital neuralgia, 11 in all, was reported by the well-known American neurosurgeon Frank H. Mayfield in 1949 [10]. However, more than ten citations on occipital neuralgia on the Internet explain that the first case was described in the year 1821 by doctors Beruto and Ramos with a comment that is exactly repeated, word by word, in all of them: “First described in 1821 by J. Beruto y Lentijo and M.M. Ramos, occipital neuralgia can be a debilitating disorder characterized by recurrent headaches localized in the occipital region...” and the bibliographical citation is *Beruto LJ, Ramos MM. Decades de med cirug pract. Mad 1821.*

Methods

The fact that one of the referenced authors, Ramos, had a clear Spanish surname called our attention. Anyway, it was impossible to find out who Beruto and Ramos were. It was as if they did not exist, but we had another clue, the name of the journal—*Mad* corresponded to Madrid? As it dated from 1821, we consulted the web of the on-line Newspaper Library of the National Library of Spain and we were able to find it. The publication was indeed from Madrid and its correct name was *Decadas médico-quirúrgicas y farmacéuticas*. In it, we found the article written on June 18, 1821 and our surprise was capitalized. The title was “Observation on a new species of neuralgia called cervico-suboccipital” [11] and the authors were in fact Mr. José Benito Lentijo and Mr. Mateo Martínez Ramos, doctor and surgeon holders of the town of Cevico de la Torre, Palencia, Spain.

Beruto was really Benito, and Ramos had removed his first surname, which was Martínez. It was really impossible to find information about both in the network with these failures. Transcription errors committed by someone in the past were perpetuated from work in work, and no one read the reference repeatedly cited in the bibliography.

Data availability We obtained the historical paper cited in the manuscript from issue *Decadas médico-quirúrgicas y farmacéuticas* on the on-line Newspaper Library of the National Library of Spain.

Results

José Benito and Mateo Martínez reported the first clinical case of occipital neuralgia in a 24-page article in the Spanish journal *Decadas médico-quirúrgicas y farmacéuticas* on June 18, 1821, fully aware that they were communicating a new entity to which they decided to place the name of cervical-suboccipital neuralgia (see Fig. 2).

The article [11] tells the story of a young military man named Agustín Moratinos, who was 23 years old and was a

soldier of the 8th company of the Cavalry Regiment of the Infant King. His regiment was deployed in the provinces of Soria and Burgos on the hunt for the guerrillas of José Ariza and Cura Merino when the neuralgia began.

He was treated at the Hospital de Burgo de Osma, in the province of Soria, for the pain of occipital neuralgia, and moved after two months to Cevico de la Torre, where he was from, where he spent twelve days under the care of doctors Benito and Martínez until his death.

Until reaching Benito and Martínez, the patient had received the diagnosis of cephalalgia without fever, spasm of the head, gastric headache, sympathetic headache, and idiopathic headache. He had received repeated doses of hipecacuana, evacuating and anti-bilious, diaphoretic tisanes, leeches, cantharides on the nape of the neck, embrocations on the head, creamy milk drums, and enemas.

This is the description of the pain that Benito and Martínez made: “The patient always complained of an intense pain, alive and tearing (*el paciente se quejó siempre de un dolor intenso, vivo y desgarrante*), whose sensation referred to a tight string, according to his expression, that stretched the head and neck: its origin was between the 1st and 2nd cervical vertebra and the condyles of the occipital bone, the one that extended by irradiations by the superior part of this one until the vertex and part of the occipito-frontal muscle; on the lower part it propagated by a large portion of the splenius of the head and on the right lateral part inclining towards the muscle lateral rectus, involving the oblique muscles inferior and superior part of the complexus and, finally, the posterior recti, large and small; but examining and recognizing the parts, neither noticed swelling, tension, mutation of colour, or increased pain by pressure: invaded by more or less acute access atypical or irregular, but more sensitive at night: and complained sometimes thrown or throbbing at the time of access (*se quejaba algunas veces de lanzadas o punzadas al tiempo del acceso*) and others of stupor or numbness in remission; others spread the pain suddenly like a lightning, making him give the most terrible and disconsolate cries (*se propagaba el dolor de repente como un relámpago, haciendole dar los gritos mas terribles y desconsolados*), sometimes disturbing the senses with various and tumultuous spasmodic movements” [11].

After this description, the authors talked about the origin of the pathology of the patient defending that it was not a headache, “but a neuropathy, that is, disease of the nerves” [11]. Specifically, they used the term neuralgia in the same way of Chaussier, Bichat, Delpech, Duval, Hamel, Bailli, Puzin, Coussays, Montfalcon, Pinel, Brichetau, and Nystee, showing a great knowledge of the French medical school. They explained that “the seat of these pains always resides on the same nerves, whether on a trunk, a branch or a ganglion; but the authors hesitate about whether the neurilemmium resides in the nervous pulp, or if it consists of an alteration of their vessels” [11].

OBSERVACION
S O B R E
UNA NUEVA ESPECIE DE NEURALGIA,
DENOMINADA
CERVICO-SUBOCCIPITAL.

Por don José Benito y Lentijo, y el licenciado don Mateo Martínez Ramos, médico y cirujano titulares de la villa de Cevico de la Torre. (junio 18 de 1821.)

Noster in hoc opere scopus pertinet,
ut dilucide cognoscatur quantum
momenti in medicina offerat observatio.
Bagliv. Prae. Medic. lib. 2. pag. 164.

INTRODUCCION.

Entre las infinitas y malogradas víctimas, que ha sepultado en la ruina y desolacion la guerra fratricida, que provoca en nuestros dias la desmedida ambicion de una clase privilegiada; que aplauden la ignorancia, el egoismo y la mala entendida religion, y que sostiene con osadía el fanatismo de un indigno ministro del sacerdocio acompañado de un corto número de foragidos; los anales médicos reclaman
Tom. III, N. IV. 10

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Fig. 2 Cover of the original article in the Spanish journal *Decadas médico-quirúrgicas y farmacéuticas* [9] that describes the first case of occipital neuralgia published by José Benito and Lentijo and Mateo Martínez Ramos in Madrid on June 18, 1821. Newspaper Library of the National Library of Spain

They continued their discussion explaining opinions of Boerhave, Tisot, Barthet, Ackerman, Stoll, or Fothergill. In fact, they mention the criteria of neuralgia proposed by Chaussier: “1. Nature of pain; 2. The seat and spread of it; and 3. Secondary phenomena according to the point where it is fixed” [11]. Based on these criteria, Benito and Martínez decided to call it cervical-suboccipital neuralgia. On the other hand, they explained that the patient did not present clinical signs of scrofulous, rheumatic, or syphilitic viruses, ruling out organic causes.

However, both showed great humility by stating that “When we say that this is a new species, we do not try to have the glory of being the only ones who have observed it but without dispute it belongs to us to be the first to call attention to this point. We invite our readers to do new research on neuralgia of the head, as we suspect are many hemicranias and headaches that occur frequently, and deceive the practical” [11]. Its honesty was maximum when explaining that perhaps a pain of a clergyman described by André or the occipital lockjaw of Sauvage were already cases of neuralgia like the one they described.

Both authors excused the Spanish Medicine for its ignorance regarding neuralgias, as they explained that at that moment, the only work translated into Spanish language (and no broadly known) that mentioned the neuralgias was the Medical Manual of Nysten.

On the other hand, Benito and Martínez considered an absurd error that they were prevented from performing the autopsy on the body of the young soldier based on a false piety and veneration of the remained materials of man.

They also regretted that they were not allowed to perform a cauterization. The cauterization was a surgical instrument in the form of a metal rod with a handle at one end and different shapes at the other end according to what one wanted to cauterize. This end was burning and applying it, and an eschar was induced. In a certain way, cauterization could be considered as the ancestor of thermic ablative radiofrequency by percutaneous approach performed still today by neurosurgeons in neuralgias. The authors had seen Dr. Antonio Hernández Morejón, one of the more famous Spanish doctors of that period, performing this therapy with success on a neuralgia of the femoropopliteal nerve in Madrid.

They attributed both prohibitions, autopsy and cauterization, to the ignorance of the vulgar and the populace, to the prevailing religious fanaticism and to the lack of protection of the medical professions by the Government. Finally, the authors made a liberal plea in defense of the Constitution and ended by exclaiming: Hopefully this time and the last time they raise their just complaints the children of Aesculapius in the midst of a country eminently heroic and liberal!

Discussion

How is the eponymous Arnold associated with the occipital neuralgia of the GON? Julius Arnold (1835–1915), a Swiss

pathologist of German descent, is known for his contributions to the Chiari malformation. He participated in more than 180 publications that are difficult to access. However, in none of the articles on occipital neuralgia existing in the Pubmed base is any work by Julius Arnold cited.

Interestingly, the father of Julius Arnold, the German pathologist Friedrich Arnold (1803–1890), did describe a neuralgia, that of the superior laryngeal nerve. Moreover, Friedrich Arnold was the first physician to describe the atrial branch of the vagus nerve, called Arnold’s nerve in Anatomy in his honor, as well as the channel in the petrous portion of the temporal bone that contains the nerve (Arnold’s canal). He also described the otic ganglion (Arnold’s ganglion) and the cough reflex (Arnold’s reflex). Therefore, referring to GON as Arnold’s nerve is incorrect because Arnold’s true nerve is the atrial branch of the vagus nerve. Some authors had already noted this incongruence [12, 13]. However, there is a picture of peripheral vessels and nerves of the head that shows the GON in the book *Icones nervorum capitis*, but his book (one of the most brilliant works of century anatomical literature) was published in Heidelberg in 1984 [14–16].

It is noteworthy that José Benito and Mateo Martínez published their work in 1821, and Julius Arnold was born in Zurich on August 19, 1835, and his father Friedrich Arnold was only 18 years old then. So, the neuralgia of the GON would have to be called Benito’s neuralgia (or Benito’s and Martínez’s), and not Arnold’s.

José Benito Lentijo and Mateo Martínez Ramos were the titular doctors of Cevico de la Torre in the year 1821. Cevico is a municipality of the region of the Cerrato, in the Province of Palencia, autonomous community of Castile and Leon, Spain. The creation of this villa dates back to Celtiberian times and persisted during Roman, Visigothic, and Arab times. It currently has about 600 inhabitants although in the nineteenth century, it reached more than 2,000.

José Benito and Mateo Martínez had to live through a politically very convulsed and violent Spanish era, for in just 50 years, there followed the French invasion of Napoleon and the war of independence, two rebellions, a constitution, an absolutist monarchy, and the establishment of the parliamentary monarchy. In between, the loss of the colonies of the Spanish empire and three internal wars followed one another. Both, José Benito and Mateo Martínez, were clearly liberal, as is shown from the political position they proclaim in the article, something that today would be unprecedented in an article in a medical journal. It is necessary to emphasize that the absolutist king of Spain, Fernando VII, had been defeated after a revolution and a liberal regime had been established in 1820 that would last only 3 years.

José Benito Lentijo was born in Valladolid on November 25, 1796 and was a Spanish doctor known in his time. He studied Medicine and Philosophy at the University of Valladolid and obtained a bachelor’s degree in Medicine *nemine discrepante* on November 2, 1815. Finally, he obtained a license in

Medicine on October 30, 1819. He studied on *tabes dorsalis* and leprosy during this academic period.

Later, he was the titular doctor of Villacastín 1819–1820 (province of Segovia), of Cevico de la Torre 1820–1822 (province of Palencia), and finally of Valladolid and Salamanca.

On December 12, 1812, he was admitted to the Royal Academy of Medicine of Valladolid, and on February 12, 1831, she was appointed secretary of the Royal Academy of Medicine and Surgery of Castile. Later, he was admitted to the Royal Academy of Medicine and Natural Sciences of Madrid, Royal Academy of Medicine practice of Barcelona, Royal Medical-Surgical Academy of Seville, Cordoba, Extremadura, and Cádiz. He performed multiple tasks and administrative positions: examinations of doctors, translator and censor of scientific works, inspector of mineral waters, commissioned of epidemics, and vaccination.

He translated the *Treatise of Modern Practical Medicine* of R Thomas in 1824, the *History of the Plegmasias or Chronic Inflammations* of FJV Broussais in 1828, and the *Elementary Compendium of Chemistry applied to the Medicine* of E Julia-Fontenelle in 1831. Moreover, José Benito wrote the *New Manual of Chemical-medical Hydrology* or analytical treatise of the mineral waters considered according to their diverse species and applications to the arts, to the domestic economy and to the Medicine. Finally, he collaborated with the medical Spanish journals *Gaceta Médica de Madrid* and *Décadas médico-quirúrgicas y farmacéuticas*.

He died of cholera at only 38 years of age, on August 12, 1834. That epidemic ravaged Valladolid causing more than 2000 deaths and apparently, José Benito had an active role in the fight against this disease, because the Governor of Salamanca wrote to his death: “he has died, the victim of his zeal, at the head of the choleric of Valladolid.”

Unfortunately, we have not found any biography of the second author, the surgeon, Mateo Martínez Ramos, although there are notices placing him as a surgeon from Vitoria, in the north of Spain.

Conclusion

Thus, it seems unquestionable to defend that the first case described and argued of occipital neuralgia in the history of Medicine, produced by neuralgia of the greater occipital nerve, has to be attributed to two Spanish doctors, José Benito and Mateo Martínez. In fact, they already called it cervico-suboccipital neuralgia.

We so propose that occipital neuralgia, when it is produced by the involvement of the greater occipital nerve, ceases to be called Arnold and is renamed Benito’s neuralgia.

Authors’ contributions Both authors have equally contributed to this work.

Compliance with ethical standards Ethical approval and informed consent to participate is not applicable in this paper.

Consent for publication Not required.

Competing interests The authors declare that they have no conflicts of interest.

Abbreviations *GON*, greater occipital nerve; *ICHD*, International Classifications of Headache Disorders; *MRI*, magnetic resonance imaging; *PICA*, posterior inferior cerebellar artery; *LON*, lesser occipital nerve

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