Fabio Antonaci Giorgio Bono Marco Mauri Monica Drottning Simona Buscone

Concepts leading to the definition of the term cervicogenic headache: a historical overview

Received: 11 October 2005 Accepted in revised form: 3 November 2005 Published online: 15 December 2005

F. Antonaci (☑) • S. Buscone Department of Neurological Sciences, University of Pavia, "C. Mondino" Foundation, Via Mondino 2, I-27100, Pavia, Italy e-mail: neuronet@libero.it

Tel.: +39-0382-3801 Fax: +39-02-700445466

G. Bono • M. Mauri University Centre for Adaptive Disorders and Headache (UCADH), Section of Varese, Varese, Italy

M. Drottning Department of Neurosurgery, Ulleval University Hospital, Oslo, Norway Abstract The idea that headache may originate from a problem at the neck or cervical spine level has fascinated and stimulated researchers for centuries. Contributions and reports seeking to clarify this issue have multiplied in the past 80 or 90 years. Bärtschi-Rochaix reported what seems to have been the first clinical description of cervicogenic headache, but it was not until 1983 that Sjaastad and his school defined diagnostic criteria for this sydrome. The current, revised International Headache Society Classification (ICHD-II) includes the term cervicogenic headache, but the diagnostic criteria it gives differ from those of the International Association for the Study of Pain

(IASP), and also from the most recent Cervicogenic Headache International Study Group (CHISG) definition (1998).

Keywords Cervicogenic headache • History of headache • Cervical spine • Anaesthetic blockade

Introduction

The term cervicogenic headache (CEH) refers to a head pain condition that has its origin in the neck. Such ideas – headache of nuchal origin – must have occupied the minds of men for centuries. A more systematic search for such headaches seems to have been made only from the early stages of last century [1]. The reason for the lack of breakthrough of such ideas was probably the vague, indistinct descriptions of the headache in the original works, and confusion with other headaches, like migraine without

aura and tension-type headache, none of which were properly defined at that time. Cardinal symptoms, beside the headache, were supposed to be dizziness, visual disturbances and tinnitus – the attacks in part lasting two to three hours. This constellation of symptoms has little to do with the CEH picture that we know today. Clinicians who conscientiously were searching for cases of headache stemming from the neck on those premises could be easily and widely misled. An overview of the early description could be the basis for a better understanding of the current terminology as far as CEH is concerned.

The early descriptions

Bärtschi-Rochaix [1] maintained that the first clinical description of a headache linked to a problem in the neck was published by Schützenberger, in 1853. Unfortunately, this description is not available in the literature. In a 1913 report – this is the first clinically significant one that we are able to access – Holmes [2] claimed that headache could originate from the neck. This author described headaches associated with the presence of painful nodules in the posterior muscles of the neck, which he attributed to fibrositis. This provided the basis for the subsequent definition of "rheumatic headache", which was described by others writing at this time [3, 4].

Barré, in 1926 (and possibly even a year earlier) [5], described a headache with greater intensity in the occipital region, associated with dizziness and with hearing and visual disturbances, and called this picture "posterior cervical sympathetic syndrome". In 1928, Lieou [6] added "pain referred to the larynx and pharynx".

In 1940, Haddon [7] described a clinical picture characterised by unilateral pressing pain, starting in the suboccipital region and radiating anteriorly to the temporal and often also to the retro-orbital region unilaterally. This clinical picture was sometimes associated with photophobia and, in severe cases, pallor, profuse sweating, hyperaesthesia in the nerve distribution area and, on occasions, also with vomiting. During an attack, the application of pressure to the greater occipital nerve (GON) or the small occipital nerve could accentuate the pain. Haddon called this pain "occipital neuralgia" and recommended, for its treatment, injections of procaine hydrochloride or alcohol at the site of the hyperaesthesia. Over the following years, there were other reports of headache possibly related to the neck, but the clinical descriptions given were less precise [8, 9]. It was to be a few more years before reports appeared that led to the inclusion, in the sphere of headache originating from the neck, of cases in which the pain was induced by the stimulation of trigger points ("mechanical headache") [10, 11].

It is worth mentioning the important studies conducted by Ray and Wolff [12]. These authors showed that stimulation of the sensory nerve endings above or below the upper surface of the tentorium cerebelli produced head pain that could be felt centrally (at the vertex) and frontally. Over the next few years, the afferent connections of the upper cervical nerves were also investigated [13]. In 1949, Hunter and Mayfield [14] made a major contribution to clarifying and developing the concept of CEH, describing 11 patients presenting with recurrent attacks of severe migraine-like pain which, at its height, could become holocranial; the pain started in the suboccipital region but radiated to the vertex, to the temporal

region and to the periorbital region, often bilaterally. The attacks would be accompanied by symptoms such as lacrimation, facial flushing, profuse sweating and, on occasions, nasal congestion on the more severely affected side. In a high percentage of cases, the patients had sustained a direct trauma to the neck. The clinical picture also included (in just a few patients) postural instability and dizziness, and a couple of patients also complained of vomiting during particularly severe attacks. This pain could be interrupted by anaesthetic blockade of C2. Blockade of the GON also modified the pain of an attack, but less rapidly and less completely. The patients described in this report underwent avulsion of the GON, either with intraspinal section of the sensory root of C_2 , or with intraspinal sections of the sensory roots of C₂ and C₃. Raney and Raney [15], meanwhile, investigated the intervertebral cervical disc as a possible trigger factor for headache.

The next important contribution was Bärtschi-Rochaix's 1949 monograph on "cervical migraine" [16]. Practically all the patients described (32 out of 33) had headache or "paraesthesia of the head". The pain would start at the back of the head and extend to the central, parietal or frontal regions. All the patients but one showed signs and symptoms linked to the neck, mainly nuchal pain on the same side as the headache, and many of them had reduced mobility of the cervical spine. Four patients had a history of drop attacks. Around half the patients suffered brief scintillating scotoma episodes (n=10) or visual fogging (n=5) coinciding with the headache attack. In a high number of cases (n=18) the pain/paraesthesia - mainly pain but sometimes paraesthesia (n=6) – spread to the upper extremities, usually unilaterally, but occasionally bilaterally. This description was the first to include the finding of headache following cervical trauma [11], and in his 1968 series Bärtschi-Rochaix [1] also included patients with degenerative alterations at the cervical level. He described radiological alterations in the uncovertebral facet joints at levels C₃-C₄, C₄-C₅, C₅-C₆ and C₆-C₇, the most characteristic being those at levels C_3 - C_4 and C_5 - C_6 . The author called these radiological findings "casseroles entassées" and "assiettes empilées"; the first of these expressions described exostoses of the uncovertebral joints of both distal and proximal vertebra, whereas in the second case only distal exostoses (affecting the uncinate process) were clearly visible. The patients studied presented morphological alterations of the uncovertebral facet joints and Bärtschi-Rochaix divided these alterations into four stages. The author used the term "cervical migraine" because he considered this migraine-like picture (unlike classic or "carotid" migraine) to be attributable to an alteration of the posterior circle.

The surgical evidence and the blockade effect

Cloward [17] studied projection of pain following stimulation of the anterior and anterolateral part of the disc during 114 cervical discographies. Pain following stimulation of C₃-C₄ was referred to the scapular region, whereas stimulation of C₄-C₅, C₅-C₆ and C₆-C₇ produced pain at the superiormedial, medial and inferior border of the homolateral scapula. Stimulation at the level of the median line, anteriorly, induced pain in the middle part of the thoracic area. Most of the patients perceived pain referred from the posterior surface of the lower cervical discs in one or more of the following areas: (1) the upper part of the scapula, (2) the base of the neck, (3) the upper part of the shoulders, (4) the inferior border of the scapulae, (5) the upper arm, radiating down as far as the elbow. This last point is particularly interesting, as pain radiating to this area is one of the characteristics of CEH.

Pentecost and Adriani [18] described the effects of cervical blockade in patients presenting with unilateral headache, with pain starting in the suboccipital region and radiating upwards to the occiput of the cranium. The authors submitted 63 patients to anaesthetic blockades of C₂ and C₃. Six patients did not derive any benefit; in 10 the pain improved partially; and in the remaining patients the pain was, temporarily, eliminated completely. In some patients, GON and small occipital nerve blockades were tried as an alternative, but these produced only partial resolution of the pain.

The anatomical basis

Kerr and Olafson [19] demonstrated, in the cat, convergence of trigeminal and cervical afferents in the intermedial and ventral dorsal horn at the level of the upper cervical cord. On the basis of this anatomical pathway it is possible to hypothesise a spreading of the hemicranial pain from cervical to trigeminal areas. According to these authors, this convergence could also explain a reflex pathway, probably related to head turning in response to trigeminal stimuli. Kerr [19–21] believed that atypical facial neuralgias [19] and other facial and cranial pain syndromes could be explained on this basis. These authors then described a recurrent clinical picture characterised by severe, periodic headache, aggravated by head and neck movements. Typically, these patients presented pain that began with a deep sensation of pressure in the suboccipital region unilaterally, which could extend to the occipital, parietal and fronto-orbital regions and down to the shoulder and arm; sometimes, it could even radiate to the mandibular and maxillary regions.

Kehr et al. [22] described two surgical methods: uncosectomy and uncoforaminotomy according to Jung (anterolateral approach), indicated in cases of "cervicocephalic syndrome, Barré-Lieou syndrome and cervicobrachial syndrome". The first of these three syndromes included, in addition to the headache: cochlear, vestibular and ocular symptoms, a sensation of postural instability, lower limb weakness, or drop attacks, and psychological symptoms, such as generalised weakness and depression. The symptoms occurred episodically and were triggered or worsened by neck movements, particularly rotation and extension. Pasztor [23] emphasised the need, in these cases, to remove any fibrotic tissue around the vertebral artery.

Knox and Mustonen [24], investigating a sample of 30 patients (27 women and three men, aged 17–72 years), described greater occipital neuralgia as an ocular pain syndrome in which the eye, the eye socket and the temple could be affected; hyperaesthesia was found on application of pressure to one or both the occipital nerves, and the authors considered this a key criterion for diagnosis. The therapy proposed was local injection of an anaesthetic drug into the most affected area.

Chouret [25] affirmed that "greater occipital neuralgia headache" had an occipital, temporal and frontal distribution and was usually bilateral. The pain was described as dull and constant.

Definition of occipital neuralgia, on the other hand, proved more problematical. According to the most widely accepted description, it is a stabbing pain in the nerve distribution area, similar to that of trigeminal neuralgia.

As we have shown, the literature reports many cases of headache that can be linked to cervical spine disorders, even though the majority of these, because of a lack of evidence, had fallen into oblivion. The various descriptions that, over the decades, have appeared in the literature present very diverse features, even though, in retrospect, it may seem that they also have many features in common. In general, it can be remarked that even though the idea that headache can be related to disorders of the neck is accepted to differing degrees by the different schools of thought, there is still no consensus on the question of whether the neck can play a key role in the genesis of a headache. Up until the start of the 1980s, headache originating from the neck had no place in the International Headache Classification [26].

The term "cervicogenic headache"

The term cervicogenic headache was introduced by Sjaastad and co-workers in an article published in 1983 [27]. The first description of CEH was greeted somewhat sceptically in scientific circles. In the years that followed, there continued to be considerable opposition to these clinical criteria and headache scientists only gradually came to accept the con-

cept. This real reluctance – even after Sjaastad's 1983 contribution - to entertain the idea that headache can derive from the neck probably constitutes the best evidence that the problem had not, until that point, been properly tackled. The whole CEH story – and unfortunately it is not the only one of its kind in the history of medicine - reveals the conservative outlook of the medical community and its unwillingness to embrace new theories, even when these are based on logical arguments and supported by evidence. The term cervicogenic headache by definition describes an out-and-out headache in which there is clear involvement of the neck and possibly a neck-related aetiopathogenesis: "cervicogenic" meaning that which originates in the neck. From this perspective, the term "cervical" is misleading, while "cervical headache" is a contradiction in terms. Nevertheless, this was the term used in the diagnostic criteria published by the International Headache Society (IHS) in 1988 [28].

A crucial step in the evaluation of these nosographic aspects came in 1987 when Ottar Sjaastad, in Florence, set up the Cervicogenic Headache International Study Group (CHISG). This group has met annually ever since. Its diagnostic criteria were first published in 1990 [29] and revised in 1998 [30].

International coding of cervicogenic headache

The International Association for the Study of Pain [30] accepted cervicogenic headache a decade ago as a dis-

tinct headache syndrome, utilising principally the Sjaastad criteria [31].

The IHS recently published a new version of its classification [32] in which the term cervicogenic headache appears, but with diagnostic criteria different from those of the CHISG. Moreover, the present criteria, as the previous in 1988, may be somewhat unsuited for clinical headache work. In the IHS criteria the pain location is not properly specified, the causes of pain are only vaguely known, neuroimaging would suffice for diagnosis, there is no mention of precipitability of pain and, last but not least, the ipsilateral shoulder/arm symptom radiation is not reported.

The definition of "cervicogenic headache" marked a turning point. Indeed, the state of our knowledge now is entirely different from what it was when this term was first introduced. Since that time, many cases of CEH have been reported and in many scientific circles the definition and the diagnostic criteria are rapidly gaining ground, and support for these nosographic criteria is increasing almost exponentially. Nevertheless, the fact remains that neither the term nor the concept are yet accepted by the entire scientific community. One major obstacle to full acceptance of CEH as a separate entity is the fact that both the definition and the diagnostic criteria are regarded as "homespun". We believe that the current nosographic criteria, established by the CHISG, constitute an important starting point for conducting clinical studies in large series of patients and for a rigorous assessment of the whole question of CEH.

References

- Bärtschi-Rochaix W (1968) Headaches of cervical origin. In: Vinken PJ, Bruyn W (eds) Handbook of clinical neurology. Vol. 5: Headache and cranial neuralgia. North Holland, Amsterdam, pp 192–203
- 2. Holmes G (1913) Headaches of organic origin. Practitioner 1:968–984
- 3. Patrick HT (1913) Indurative or rheumatic headache. JAMA 7:81–86
- Luff AP (1913) The various forms of fibrositis and their treatment. BMJ 1:756–760
- Barré M (1926) Sur un syndrome sympathique cervical postérieur et sa cause fréquente: l'arthrite cervicale. Rev Neurol 33:1246–1248

- Lieou YC (1929) Syndrome sympathique cervical postérieur et arthrite chronique de la colonne vertébrale cervicale. Schuler Mink, Strasbourg
- Haddon SB (1940) Neuralgic headache and facial pain. Arch Neurol 43:405–408
- 8. Perelson HN (1947) Occipital nerve tenderness: a sign of headache. South Med J 40:653–656
- Campbell D, Parsons C (1944)
 Referred head pain and its concomitants. Report of preliminary experimental investigation with implications for the post-traumatic head syndrome.

 J Nerv Ment Dis 99:544–551
- Travell J, Rinzler SH (1952) The myofascial genesis of pain. Postgrad Med 11:425–434

- 11. Travell J (1962) Mechanical headache. Headache 7:23–29
- 12. Ray BS, Wolff HG (1940) Experimental study of headache. Arch Surg 41:813–856
- Escolar J (1948) The afferent connection of the 1st, 2nd, and 3rd cervical nerves in the cat. J Comp Neurol 89:79–92
- 14. Hunter CR, Mayfield FH (1949) Role of the upper cervical roots in the production of pain in the head. Am J Surg 48:743–751
- 15. Raney AA, Raney RB (1948) Headache: a common symptom of cervical disc lesions. Report of cases. Arch Neurol Psychiatr 59:603–621
- Bärtschi-Rochaix W (1949) Migraine cervicale, das encephale syndrome nach Halswirbeltrauma. Huber, Bern

- Cloward RB (1959) Cervical discography. A contribution to the etiology and mechanism of neck, shoulder, and arm pain. Ann Surg 150:1052–1064
- Pentecost PS, Adriani J (1955) The use of cervical plexus block in the diagnosis and management of atypical cephalalgia of cervical origin. Anesthesiology 16:726–732
- Kerr FW, Olafson RA (1961)
 Trigeminal and cervical volleys.
 Convergence on single units in the spinal gray at C1 and C2. Arch Neurol 5:171–178
- Kerr FW (1961) Atypical facial neuralgias: their mechanism as inferred from anatomic and physiologic data. Mayo Clin Proc 36:254–260
- Kerr FW (1963) Mechanisms, diagnosis and management of some cranial and facial pain syndromes. Surg Clin North Am 43:951–961

- Kehr P, Lang G, Jung FM (1976)
 Uncusektomie und
 Uncoforaminektomie nach Jung.
 Techniq, Indikatione, Resultate,
 Langenbecks. Arch Chir 341:111–125
- Pasztor E (1978) Decompression of the vertebral artery in cases of cervical spondylosis. Surg Neurol 9:371–377
- Knox DL, Mustonen E (1975) Greater occipital neuralgia. An ocular pain syndrome with multiple etiologies. Trans Sect Ophthalmol Am Acad Ophthalmol Otolaryngol 79:513–519
- Chouret EE (1967) The greater occipital neuralgia headache. Headache 7:33–34
- (1962) Ad hoc committee on the Classification of Headache of NIH. JAMA 179:717–718
- Sjaastad O, Saunte C, Hovdal H, Breivik H, Grømbaek E (1983)
 "Cervicogenic" headache. An hypothesis. Cephalalgia 3:249–256

- 28. Hedache Classification Committee of the International Headache Society (1988) Classification and diagnostic criteria for headache disorders, cranial neuralgias and facial pain. Cephalalgia 8[Suppl 7]:1–96
- Sjaastad O, Fredriksen TA, Pfaffenrath V (1990) Cervicogenic headache: diagnostic criteria. Headache 30:725–726
- Merskey H, Bogduk N (eds) (1994)
 Classification of chronic pain: description of chronic pain syndromes and definition of pain terms. IASP Press, Seattle, WA
- Sjaastad O, Fredriksen TA, Pfaffenrath V (1998) Cervicogenic headache: diagnostic criteria. The Cervicogenic Headache International Study Group Headache. 38:442–445
- 32. Headache Classification Subcommittee of the International Headache Society (2004) The International Classification of Headache Disorders, 2nd edn. Cephalalgia 24[Suppl 1]:1–155