

## CERVICOGENIC HEADACHE

### JOSEY'S CASES REVISITED

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**ABSTRACT** - Before Sjaastad coined the term cervicogenic headache (CH) 15 years ago, neck-related headaches have been considered by different authors for many years. Even after the publication of diagnostic criteria, dispute on the clinical picture, differential diagnosis, pathophysiology and treatment of CH still persists. A paper published in 1949 by Josey reports on 6 "illustrative" cases of cervical-related headaches. Indeed, looked from a more recent perspective, those cases could eventually correspond to CH. Important topics such as the relatively high frequency, fixed unilaterality of the pain, relation to previous trauma, irradiation from the back to the forehead, normal or slightly abnormal roentgenograms, and the mechanical precipitation of attacks are some of the topics considered by Josey. The female gender was not prevalent in Josey's series. Traction and analgesics were basically the recommended treatment. CH is probably a common disorder, an idea already considered by a clinician in 1949. This syndrome was not adequately described before Sjaastad's group papers in the 80's.

**KEY WORDS:** cervicogenic headache, differential diagnosis.

#### Cefaléia cervicogênica: revisão dos casos de Josey

**RESUMO** - Muitos autores escreveram sobre cefaléias relacionadas ao pescoço antes da descrição da cefaléia cervicogênica (CH) por Sjaastad e col. Mesmo após a publicação de critérios diagnósticos, há controvérsias em relação ao quadro clínico, diagnóstico diferencial, fisiopatologia e tratamento da CH. Um artigo publicado em 1949 por Josey relata 6 casos "ilustrativos" de cefaléia relacionada ao pescoço, cujo quadro pode corresponder ao que hoje consideramos ser CH. Aspectos importantes como sua frequência relativamente elevada, unilateralidade fixa, relação a traumas prévios, irradiação pósterio-anterior, normalidade de exames radiológicos e os mecanismos de precipitação foram considerados por Josey. O sexo feminino não foi predominante na sua casuística. Tração e analgésicos foram os tratamentos recomendados. CH é provavelmente uma desordem comum, o que já havia sido considerado neste estudo de 1949. A síndrome, entretanto, não foi completa e adequadamente descrita antes de Sjaastad.

**PALAVRAS-CHAVE:** cefaléia cervicogênica, diagnóstico diferencial.

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By describing cervicogenic headache (CH), Sjaastad and co-authors renovated the discussion on neck-generated headaches, which they believe are a well-defined reaction pattern<sup>38,41</sup>. Cornerstone features according to these authors are strict unilateral pain that typically starts at the occipital-nuchal area and spreads to the ipsilateral forehead, and attack induction by neck movements and/or digital pressure over trigger points such as the greater occipital nerve (GON) or the C2 area. Additionally, there may be diffuse, vague ipsilateral arm pain or discomfort. Females tend to be more affected than males, and a trauma may precede the onset of the symptoms.

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In spite of some dispute<sup>4,11,28,35</sup>, different authors have been describing CH-like pictures over many years with surprising resemblance. Writing on his personal view of CH, Pearce cited Hilton's observations on headache related to neck abnormalities in 1860-1862<sup>29</sup>. Maigne's ideas on CH also parallel Sjaastad's concepts in many aspects<sup>24-26</sup>. Hunter and Mayfield, also in 1949, reported on successful treatment of hemicranial pain with section of sensory cervical roots and the greater occipital nerve<sup>17</sup>. Other frequently cited descriptions such as Barré-Lieou's syndrome sympathique cervicale posterieur<sup>3</sup> and Bärtschi-Rochaix' migraine cervicale<sup>9</sup> also share some characteristics with more recent reports. Bogduk and Marsland's also wrote on occipital or suboccipital headache radiating to anterior or superior parts of the head<sup>6</sup>.

Almost 50 years ago, Josey published a paper on patients suffering from headaches related to pathologic changes in the cervical spine (Table 1)<sup>20</sup>. Although that paper is not cited in recent articles on CH, including a comprehensive review of the literature<sup>13</sup>, the basic ideas considered there are similar to what has been considered by other authors. "This symptom complex is not a new one, but it is one which I am sure is frequently overlooked. Too often the patient is sent off on a round of investigations of eyes, sinuses and teeth, and then all too often the diagnosis is neuralgia, atypical Migraine, chronic sinusitis, constipation or psychoneurosis", said Josey quite appropriately. Before Sjaastad's group description there was great confusion about cervicogenic headache-like disorders. However, its frequency and importance have been clearly overlooked.

### Clinical picture

The headache described by Josey was frequent and usually prolonged, most often starting at the occipital region, spreading to the frontal or supraorbital area uni- or bilaterally. "The frontal pain was unilateral or bilateral", said Josey, who considered the headache to be mainly frontal: "At times the frontal headache was so predominant that only after direct questioning was any painful symptom referred to the occipital region or the neck".

The site of the pain, according to Josey, is similar to what has been considered in Sjaastad's description of cervicogenic headache<sup>15,41</sup>. The irradiation from the back to the forehead was noticed by several authors<sup>23,26,31,36,39,41</sup>. In this particular respect, according to Fredriksen et al., "All the 11 patients experienced the strongest pain in or around the ipsilateral ocular, aural, and temporal areas, and the pain also involved the neck and the nape of the neck"<sup>15</sup>, similar to Josey's cases. In our experience, it is noteworthy that the radiation pattern may eventually be overlooked by the patients since the frontal pain may be substantially more intense than the posterior headache. Maigne said in this respect that "Il n'est pas vrai qu'une céphalée cervicale soit seulement à topographie occipitale. Elles sont pour la plupart sus-orbitaires"<sup>25</sup>.

The pain unilaterality has been approached by different groups<sup>24,26</sup>. CH is considered a fixed unilateral headache in contrast to migraine, where attacks may change from one side to the other<sup>36,37,40</sup>. D'Amico et al. identified 20.8% side-locked unilaterality in migraine and 12.5% in tension-type headache<sup>10</sup>. In a series of 31 "classic migraine" patients, only one had side-locked unilaterality<sup>40</sup>. Although originally defined as a strict unilateral headache, Sjaastad himself predicted the existence of bilateral cervicogenic headache cases in the 1990 diagnostic criteria<sup>38</sup>. In fact, theoretically, the laterality criterion could be less strict, as contralateral trigeminal dysaesthesias secondary to a C2 compression have been reported<sup>33</sup>. Thus, not only bilateral cases could exist, but pain originated in one side of the neck may spread contralaterally. Maigne admitted already bilateral cases, as he wrote about palpating the posterior part of the neck: "cette manœuvre est indolore du côté opposé - sauf en cas de bilateralité de la céphalée"<sup>25</sup>.

Concerning the precipitating factors, similarities between the former and recent reports also exist. "Position of the head and neck appeared to be a definite factor. Headache frequently had its onset while the patient was in bed and in early morning. The position of the head at work, any exertion which placed a strain on the neck and driving a car were often precipitating factors. In some cases, the head was held in an obviously abnormal manner, but this was never extreme", was Josey's comment in 1949. CH patients clearly tend to suffer more headache in the morning, mostly even before getting up, which indicates that the position in bed may somehow predispose to an attack. Some of Josey's patients had attacks during the night.

As Sjaastad described, one of the main features of cervicogenic headache is the mechanical precipitation. Pressure over trigger points over the occipital-nuchal area, as the greater-occipital nerve, may originate an attack

Table 1. Patients described by Josey in 1949.

	Sex	Age	Trauma	Pattern and site	RRM	Precipitation	Treatment
1	fem	40	Mild head trauma, onset in a week	Right, nuchal and supraorbital, constant for three weeks and frequent for 3 months	yes, laterally	Pressure to the right of the 2nd cervical body. Laying down, driving a car, exertion	Local physical therapy and a cervical collar
2	male	49	Head injury 30 years before, unconscious for 8 hours	Left eye irradiating to left occipital region. Irregular intervals (1-3 months) persisting 2-14 days. Headaches for undetermined number of years.	no	Exertion, pressure over the left of the 2nd cervical body, irradiating to the ipsilateral supraorbital region. Coughing and sneezing	Temporary relief with traction during attack. Traction recommended 3-4 times/day for 15 min.
3	male	?	no	Bifrontal, starting over either eye spreading at times to the occipital region. Almost daily for several months with remissions for weeks. Headaches and "cricks" in the neck for many years	yes, to the right	Onset in the morning with exertion or driving a car. Pressure to the right of the 2nd cervical body, irradiating to the ipsilateral supraorbital region	Halter traction for 20 min. bid. and sleep without a pillow. No effect with ergotamine, nicotinic acid, tridelenamine hydrocl., relief with codeine and ASA
4	fem	38	Injury to the coccyx at the age of 13. Fall striking her chin a week later	Acute onset of a severe throbbing headache over the right occipital region that disappeared in 30 min. Seven hs. later, severe pain in the same area radiating to ipsilateral forehead for 2 hs. Day after: same pain for 24 hs. Infrequent attacks of "stiff neck" for 10 years	no	Pressure to the right of the 2nd cervical body, irradiating to the ipsilateral supraorbital region. One time the pain was associated with coughing and sneezing	Traction for 15 min. tid: Temporary relief. Cervical collar at night
5	fem	50	Fall at the age of 5, unconscious for several hours	Daily headaches for over 25 years over the upper posterior cervical and bifrontal areas. Right shoulder and upper arm pain	yes, flexion	On awakening in the morning. Physical exertion. Pressure to the left of the second and third cervical bodies.	Traction: temporary relief.
6	male	52	No	Aching pain in the left occipital and suboccipital region. 2 days later: severe pain in the left temporofrontal region for 45 min. Pain recurred hours later	yes, all directions	Coughing and turning the head. Pressure to the left of the 2nd cervical body, irradiating to the ipsilateral frontal region	ASA and codeine. Cervical collar.

similar to the naturally occurring episodes<sup>15,38,41</sup>. According to Josey, "In practically all cases, firm pressure with the examiner's thumb just lateral to the body of the second cervical vertebra produced pain locally and also a radiating pain which was described as shooting through the head to the frontal or supraorbital region. When the symptom of headache was bifrontal, this phenomenon could often be produced on both sides. Such pressure would almost always reproduce the frontal headache complained of, but it would not be protracted". Indeed, many patients report interesting sensations in the forehead during examination. They feel for example that there is "an invisible wire" inside the head connecting the back to the forehead, so that pressure on the occipital-nuchal areas would invariably produce pain on the ipsilateral forehead. Others say the pain is "reflected" in the frontal area. They may also report that a previous physical examination provoked a long lasting attack. In Josey's paper it is not mentioned exactly where the digital pressure is exerted (Table 1). It is possible that the area where Josey palpated - lateral to the second cervical body - corresponds to what Sjaastad's group considers one of the trigger points, the C2 area, behind and just below the mastoid process<sup>15</sup>.

There has been concern on the reduction of the cervical range of motion in CH. According to Fredriksen and coworkers, "all the patients had slightly to moderately reduced range of motion in the neck, particularly on rotation. (...) During attacks, the patients tried to minimize neck movements, and they even resisted passive movements." Bärtschi-Rochaix considered that "in order of frequency, lateral flexion, rotation, neck extension and flexion are limited and/or painful on both sides. Combined neck extension and rotation are particularly unpleasant"<sup>9</sup>. Jaeger found that the range of motion was reduced in 7 out of 11 cases<sup>18</sup>. Pfaffenrath et al, using a computer assisted methodology, found hypomobility in segment C0/C1, maintained mobility in C1/C2 and hypermobility in C6/C7<sup>30</sup>. This aspect was also mentioned in Josey's article: "Manipulation of the neck usually revealed some resistance to extreme lateral motion, but there was no obvious spasm of the superficial musculature." Stiff neck posture with muscle spasm has been related to headaches of cervical origin<sup>9</sup>. Jaeger found comparatively more tender myofascial trigger points in the symptomatic side in cervicogenic headache<sup>18</sup>. We believe uncertainties in this respect still persist. Cervical mobility needs to be evaluated in larger series in order to confirm such observations.

Migrainous traits such as nausea, vomiting, photo- and phonophobia may occur in CH, but to a comparatively lesser extent. In 11 cases, Fredriksen found nausea in 7, vomiting in 6, photophobia in 5 and phonophobia in 10<sup>15</sup>. In Sjaastad's series, nausea/vomiting occurred in 70-85% of the migraine patients and in 55% of the CH group<sup>36</sup>. One of Maigne's patients with "céphalée cervicale" had, according to his description, "le caractère nettement migraineux. (...) tous les neurologues ou allergologues consultés avait posé le diagnostic de migraine vraie"<sup>25</sup>. In a series of 15 patients, nausea, vomiting and phono-/photophobia were present in 5, 2 and 5 cases, respectively<sup>31</sup>. Pikus and Phillips reported on 31 CH cases. Nausea and/or vomiting were present in 7 and even fewer had photophobia<sup>32</sup>. In our material, nausea (52.7% in CH, 89.2% in migraine) vomiting, (22.2% in CH, 60% in migraine) photo- and phonophobia (52.5% in CH, 86.1% in migraine) were significantly lower in CH as compared to migraine (Vincent M & Luna R, unpublished results). According to Josey, "nausea of slight degree was present only if the headache was particularly severe. There was no vomiting".

Josey did not consider the quality of the pain in detail: "In almost all cases there is associated aching in the posterior cervical region or a history of attacks of "cricks" in the neck." One of the patients was described as having an aching pain in the back and a sharp forehead pain. Patient 2 was said to have a dull to sharp ache across the frontal region. Sjaastad considers the pain to be moderate, non-excruciating and usually non-throbbing<sup>36</sup>. Case 4 had acute severe throbbing pain at 6:00 AM.

As shown in Table 2, female sex tend to predominate in most series. Josey did not have a clear-cut female preponderance, but this may be due to the small size of that material. Only one series had more males than females<sup>19</sup>. In general, CH patients tend to be older than migraine and tension-type headache patients. Ages were similar in most reports (Table 2).

Josey stressed that, although the cervical spine could be the source of pain in many patients, the existence of a disease perhaps was not an obligatory finding: "It will be shown that demonstrable arthritis of the cervical part of the spine is not necessarily present". Even with the technology available today, imaging of cervical structures may be normal in CH<sup>14,31</sup> and patients with substantial degenerative cervical disorders do not necessarily have pain. This was also Maigne's opinion: "l'examen clinique et radiologique classique du rachis cervical est généralement normal. C'est sur une sémiologie plus fine que se fait le diagnostic"<sup>25</sup>. This suggests that the

Table 2. Female preponderance and age in cervical-related headaches. Josey's cases compared to other series.

Author	n	Females	Age
Josey <sup>20</sup>	6	50%	45.8±6.3*
Fredriksen et al. <sup>15</sup>	11	100%	43±?
Bogduk and Marsland <sup>6</sup>	10	60%	39.8±10.4
Pfaltenrath et al. <sup>31</sup>	15	53.3%	47.4±?
Jansen et al. <sup>19</sup>	16	31.2%	37.5±?
Jaeger <sup>18</sup>	11	90.9%	33.8±6.3
Pikus and Phillips <sup>32</sup>	31	51.6%	41.1±11.8
Vincent and Luna <sup>44</sup>	36	88.8%	43.9±13.9

\*n=5 for age; \*\* (Vincent M, Luna R. Cervicogenic headache: a comparison with migraine and tension type headache: unpublished results).

Ages are shown as mean ± SD, taken or calculated from the original data when available or possible.

current imaging techniques may be sometimes insufficient for detecting the abnormalities related to CH; that visible cervical lesions are not necessarily the source of all CH, or that CH abnormalities are not anatomically determined and occur at the functional or biochemical level. Again, if a cervical abnormality is found in a patient suffering from headache this is not a definite proof that the pain is caused by that abnormality. Further studies are required before we can subdivide CH into various forms according to different putative cervical lesions.

Josey considered that the loss of the normal cervical lordotic curvature was a common factor in this sort of headache, but did not state whether this was the cause of the pain. The abnormalities reported in 1949 were fusion of fifth and sixth cervical bodies, arthritic changes, and straightening of parts or the entire cervical spine. The observations, however, lack objective measurements. The reported finding may be non-specific<sup>27</sup>. Controlled studies would be needed to better address roentgenologic abnormalities in cervicogenic headache. In this respect, Watson and Trott's material on "cervical headache" suggests that patients may have forward head posture as compared to controls<sup>44</sup>. It would be interesting to use current diagnostic criteria to select patients for studying head posture and abnormal cervical curvatures.

Trauma, a minor criteria according to Sjaastad, has been related to cervicogenic headache, but this subject is debatable. Josey did not consider trauma as an important cause of headache: "A history of trauma was usually not obtained except on direct and insistent questioning and often was of such slight degree, or so far in the past, that the patient was not impressed with its relationship to headache". Bärtschi-Rochaix considered that trauma is related to unilateral cervicogenic headaches whereas bilateral or alternating pain was caused by arteriosclerosis or degenerative changes<sup>9</sup>. In other series trauma was also related to cervicogenic headache<sup>10,32</sup>. According to Maigne, trauma is "frequent", but may be mild and occur far before the onset<sup>24</sup>. We believe trauma is not an obligatory predisposing event, present in only 20% of the CH patients (Vincent M & Luna R, unpublished results). Whiplash trauma and its importance in headache is an unsolved issue. Although it has traditionally been considered as a possible source of head pain<sup>2,15</sup>, existing data doubt this aetiological factor<sup>1,34</sup>.

### Pathophysiology

Josey considered that the model of head innervation - pain in frontal and supra-tentorial areas being transmitted by the trigeminal nerve and posterior/infratentorial pain carried by the ninth and tenth cranial nerves - was inadequate, although Josey did not point an alternative possibility: "This explanation fails to account for certain headache phenomena, particularly with the headaches associated with pathologic changes in the cervical part of the spine". In fact, the ultimate explanation for certain cervical abnormalities producing pain in trigeminal areas remains speculative. According to the so-called Kerr principle, connections between cervical sensory fibres and the trigeminal nucleus could explain why neck-generated pain may be felt in the forehead<sup>21,22</sup>. However,

sturdy evidence supporting this mechanism is yet not available, neither is there an explanation for the sometimes prolonged relief of frontal pain following cervical sensory nerves blockades<sup>43</sup>. It was shown recently that stimulation of the feline greater occipital nerve increased metabolic activity in the dorsal horn at the level of C1 and C2 ipsilaterally, and by a lesser amount contralaterally. In this study, neuronal activation appeared contiguous with the trigeminal nucleus caudalis and was in the same distribution as has been seen when trigeminally-innervated structures are excited<sup>16</sup>. This may be important for explaining the pathophysiology of cervicogenic headache in the future.

Still on the pathophysiology of CH Josey said: "The exact nature of the anatomic or physiologic pathologic change cannot be definitely stated. From what is known of the innervation of the fore part of the head and the structures within the cranium above the tentorium by the fifth cranial nerve and the posterior aspect of the head and the subtentorial intracranial structures by the upper three cervical and the ninth and tenth cranial nerves, it is difficult to explain the transmission of the headache through these pathways." The possibility of connections between cervical nerves and the sensory nucleus of the fifth cranial nerve was also considered by Josey, but "it is difficult satisfactorily to conceive any direct connection of the nucleus playing a direct part in the pain involved in these cases".

### Do Josey's patients meet Sjaastad et al's criteria for cervicogenic headache?

According to the diagnostic criteria by Sjaastad et al.<sup>38</sup>, two topics are obligatory in cervicogenic headache: (I): Unilaterality of the pain without sideshift and (II): Symptoms and signs of neck involvement (at least one of the following points: similar pain triggered by neck movement and/or sustained awkward positions; similar pain elicited by external pressure over the ipsilateral upper, posterior neck region or occipital region; ipsilateral neck, shoulder and arm pain; reduced range of motion in the cervical spine). Other items rather characterize the pain and are not necessarily required for the diagnosis.

At least some of Josey's cases seem to fulfill the criteria for cervicogenic headache (Table 1). Probably many would do so if we had the opportunity to re-examine them according to the present diagnostic criteria and use diagnostic blockades. All but 2 had fixed unilateral headaches, none had alternating pain. Mechanical precipitation was seen all patients. Sustained neck awkward positioning was clearly reported in at least 1 patient, but 2 more had the onset in the morning, suggesting that the position in bed could favor an attack on awakening. Josey's case 1 illustrates the importance of movement and positioning: "headaches would reappear after she lay down at night and also on any excess exertion. Driving an automobile, with the necessary frequent movements of the head, would always produce an aching sensation in the neck and the right supraorbital headache. (...) the patient would frequently awaken at night with headache and would apply the cervical collar for relief." Ipsilateral neck, shoulder and arm pain was reported in one case. The remainder criteria such as female sex (3 cases) and pain starting in the neck, eventually spreading to oculo-fronto-temporal areas where the maximum pain is located (at least 2 cases) were also more or less fulfilled by Josey's patients. The patients seen in 1949, at least partially, certainly suffered from a headache similar to what Sjaastad considered as cervicogenic headache later on.

### Treatment

Josey used traction in 4 of the 6 described cases, with apparently good effect. Cervical traction has been proposed as an effective treatment for headache<sup>12</sup> but our practice do not support this procedure, specially in cervicogenic headache. However, this matter will have to be comprehensively studied using adequate diagnostic criteria in carefully selected materials for a definite conclusion. The analgesics used in Josey's cases may be useful. This effect should be regarded as non-specific.

Cervical manipulations (rotation and lateral flexion) was the treatment of choice for cervical headaches according to Maigne<sup>26</sup>, but this approach may be less effective than he suspected<sup>42</sup>. Cervical collars may just provide temporary relief in our experience. Neurolysis of the greater occipital nerve may reduce the pain but for a short period<sup>4</sup>. Following blockades of different cervical structures and nerves<sup>6,7,26,31,41</sup>, sometimes important procedures for CH diagnosis<sup>38</sup>, the pain tends to recur sooner or later. Blume proposed percutaneous radio-frequency denervation in the occipital nerve territory for what he called "persistent occipital myalgia-neuralgia syndrome"<sup>15</sup>. Freeing upper cervical roots compressed by vessels reduced the pain in patients with permanent hemicrania<sup>19</sup>.

In brief, a definite and unique treatment for CH is still not available. Probably this syndrome may require different approaches according to the underlying pathology.

## FINAL COMMENTS

The term cervicogenic headache, introduced by Sjaastad's group in the beginning of the 80's, finally organized confusing concepts of neck-related headaches in vogue for many years. We consider that as the first comprehensive description of cervicogenic headache. A paper on headache related to cervical abnormalities written in 1949<sup>20</sup> is similar in many respects to other reports from different authors. This illustrates how frequent such disorders may be and how confusing the diagnosis might have been.

The cases described in 1949 could partially correspond to cervicogenic headache as we know it today. The coincidental traits observed by different authors strength the existence of a distinct cervicogenic headache syndrome, an aspect that should be considered by sceptical clinicians. Josey said this syndrome was common as 20 patients were seen in two years. The main aspects of this headache are fixed unilaterality; irradiation from the back of the head to frontal areas; relative paucity of migrainous traits; and provocation of attacks by digital pressure over trigger points in the back of the head and neck or cervical awkward positions. Even having a cervical origin, the pain is not occipital or cervical, but rather locates to the forehead, ocular end/or temporal regions. The pain may typically irradiate from the back to the front.

In spite of the fact that neck-related headaches have been recognized for a long time, there is still confusion and difficulties concerning its identification and classification. The pathophysiology and treatment of CH remain partially unknown. Systematic studies may help in distinguishing cervicogenic headache, an apparently frequent syndrome, from other similar headaches.

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