

Electrical stimulation in headache treatment. For separate headache(s) or for headache generally?

Ottar Sjaastad, MD, PhD^a
Torbjørn Fredriksen, MD, PhD^b
Jan V. Jørgensen, MD^b

^a Department of Neurology, St Olavs Hospital,
Trondheim University Hospitals, Trondheim, Norway

^b Department of Neurosurgery, St Olavs Hospital,
Trondheim University Hospitals, Trondheim, Norway

Corresponding author: Ottar Sjaastad,
Gautes gate 12, 7030 Trondheim, Norway
E-mail: ellhed@online.no

Summary

Two cases of unilateral, severe headache were treated with epidural/epicranial electrical stimulation, with an immediate and most favorable response. One patient suffered from cervicogenic headache (CEH), while the headache in the other one showed some similarity to Hemicrania continua (HC): unilaterality and some localized, ipsilateral autonomic phenomena. But, there were also sufficient features to alienate this picture decisively from HC: poor response to indomethacin and, frequently, excruciatingly severe pain.

In the CEH patient, the electrode was anchored at the posterior, bony end of the foramen magnum. In case 2, it was placed above and anterior to the symptomatic side ear, i.e. at the point of maximal pain. The headache in the latter patient may be termed: “non-indomethacin-responsive chronic hemicrania”, which may not be a rare headache.

In both patients, the spectacular improvement has continued for the whole post-operative period, i.e. for around five years and 11 months, respectively.

KEY WORDS: cervicogenic headache, epidural electrical stimulation, epicranial electrical stimulation, hemicrania continua.

Introduction

The localization of pain, in particular during the initial attack phase, is a not unimportant aspect of the initial, rough headache categorization. Although “anterior” and “posterior” regions of the head are not precisely defined terms, in this context they are used to designate painful areas orbitally/frontally/temporally/anterior parietally versus a painful area occipitally.

Let us consider some examples:

In cervicogenic headache (CEH) attacks, the pain usually starts in the posterior region (and neck) and thence spreads to the frontal area (1,2). In tractor drivers' headache (3) and “persistent neck complaints with subse-

quent, transient posterior headache” (4), an awkward positioning of the neck can provoke pain in the nuchal area, which eventually spreads to the vertex, but usually not to the forehead. While CEH is unilateral (5) in its clinically recognizable form, the other two forms are bilateral.

On the other hand, cluster headache, a unilateral headache, and migraine without aura (1), frequently a unilateral headache, are initially mainly located anteriorly. The same seems to go for the rare hemicranias, like chronic paroxysmal hemicrania (CPH) and Hemicrania continua (HC). Both of these are indomethacin-responsive headaches.

The various invasive techniques used in CEH

It might seem logical to try to influence a posteriorly located headache, like CEH, through procedures directed at that area. Given the lack of optimal results, many different techniques have been tried during the last 20 years, with an occipital/cervical approach as their common denominator. Some of these invasive techniques used in Trondheim have given meaningful results in CEH:

i) Fifty patients were followed up after undergoing a “*liberation operation*” (“*neurolysis*”) on the greater occipital nerve (GON) on the symptomatic side (6). The mean observation time was 14.5 months (range, 3-43 months). The immediate, postoperative beneficial effect was “complete relief” in 46% versus “some relief” in 36%. Over time, recurrences occurred in 92% of the cases.

ii) *Radiofrequency denervation of the planum nuchale* on the symptomatic side according to Blume et al. (7). A follow up of the first seven patients after around 4.5 years showed an improvement in 71% (complete recovery in 43%). Unfortunately, one patient developed a local, sterile effusion postoperatively (8).

iii) *Foramenectomy* (n=16) (9) – and: iv) *Stabilization operations*, according to Smith-Robinson (10,11), in the cervical spine have been carried out in patients in whom CEH was also a problem (n=10+1) since the early 1990s (9,12). More recently, Smith-Robinson operations (11) were also carried out in collaboration with a German group (13-15). While the first of these studies were retrospective, the latter (13-15), involving two groups of patients with unilateral and bilateral CEH (n=32+28), were prospective and controlled, with long-term follow up. Altogether, these studies gave positive results. Our studies (13,14) are the first published ones on CEH series with surgical removal of low cervical disc herniations as a causative factor in CEH. More recently, our results have been reproduced in a prospective study by Diener's group (16).

Stabilization operations are presently the preferred procedures when there are radicular phenomena in addi-

tion to headache. However, alternative methods must be available, since not all CEH patients can be assumed to show a combination with obvious radiculopathy.

v) *Epidural electrical stimulation (dorsal column stimulation)* has been used since the early 1990s in our setting for the treatment of CEH (17), diagnosed according to the criteria of the International Cervicogenic Headache Study Group (CHISG) (18) or their precursor. A summary report of the 10 first cases has been published (19). "Excellent" initial results were obtained in two patients. "Good"/ "excellent" results were found in 60% of the patients. After >8 years, 50% of them still used the stimulation on a daily basis.

Various shortcomings/complications should be envisaged with this type of therapy: failure to get the electrode high enough up in the spinal canal (preferably above C2); lead migration within the canal; rupture of the electrodes/connections. Difficulties with the electrode placement and lead migration has led to the development of alternative techniques to the spinal canal approach: *direct* positioning of the electrode at the C1 level (via small flavectomies) gave favorable results in cases where the electrode could not be brought up to the C1 level with the ordinary, intra-canal technique. In our hands, the results have, nevertheless, not equaled those obtained with stabilization operations and the Blume procedure (7) in CEH. Therefore, we recently tried to position the neurostimulator lead higher up, i.e. in the occipital area, as proposed by others (20-23), in the attempt to make this procedure as effective as the stabilization operations and the Blume procedure.

There have, in recent years, been several reports on headaches treated with GON stimulation. One may be somewhat concerned over the diagnostic accuracy in some of these reports. Some cases seem to have been classified simply as "headaches"/chronic headaches and hemicranias (23). Diagnoses such as "occipital neuralgia" and CEH have been given, without convincing diagnostic evidence. This lack of clarity currently makes the selection of cases for these operations problematic. To increase experience in this area, this treatment should preferably be tried in specific headache categories. Even though the technical problems linked to this procedure have proved considerable, at present the clinical diagnostic problems must be considered to outweigh them.

Since CEH, putatively originating in the neck (24,25), can be positively influenced by procedures targeting the neck/occipital area, could headaches mainly localized in the temporal/parietal area be improved by epicranial procedures in that area, e.g. electrical stimulation? This communication deals with this problem, illustrated by two contrasting case histories, one with an "anterior", i.e. a mainly temporal/ocular/aural headache and one with a "posterior" headache; both were treated with electrical stimulation, and both were followed prospectively, both pre- and postoperatively.

Cervicogenic headache

A male, born in 1943, had aseptic meningitis at the age of 28 years. He was a successful businessman when, at 48 years of age, he sustained a whiplash trauma when his car was hit from behind. Because the neck support

was positioned too low, his neck slid over it. The damage to his car was considerable. Immediately after the collision, he felt bewildered, disoriented and dizzy. Neck pain developed within a couple of days. After 6 weeks or so, he tried to return to work, but was unable to function properly. The pain, mainly headache, gradually increased over the next couple of months. He was again forced to stop working, partially to begin with and later completely. The headache was mainly left-sided. It started in the neck and spread to the frontal area, behind both eyes, "like a bolt", and it was chronic-fluctuating. At times, the pain could spread to the right frontal area, the maximal pain nevertheless remaining in the original area. There was also "numbness" in the arms, ulnar side, and fingers 4&5; with maximal head pain, this "numbness" was also present in the thumbs. On such occasions, there could be some mild pain in the arms; the arm symptoms could also amount to weakness. The patient had "damaged quite a number of cups". Pain radiating to the forehead could be provoked within a few minutes by bending forwards, looking upwards, and by turning the head to either side. On the other hand, the patient had to move his head slightly all the time, otherwise he would feel stiffness and tenseness developing in the neck. Physical work, e.g. shoveling snow, would spoil the rest of the day. Occasionally, he wore a neck collar.

Tinnitus, which seemed to have a slightly pulsatile quality, appeared particularly in concomitance with heavy pain. Allodynia and hyperventilation were not noted until around two years after the accident. The allodynia was pronounced, mainly on the symptomatic side, but could also be felt on the other side (the headache could also involve the contralateral side). Precipitating factors for allodynia were: combing the hair, touching the skin, and windy weather. The allodynia changed markedly after the first operation (vide infra), leaving the patient with no discomfort on touching the skin and hair; only upon the application of firm pressure to the scalp would some mild discomfort arise.

The skin-roll test was clearly positive (see comments). There was also excessive tenderness of the tendon insertions in the occipital area; palpation in this area caused pain that spread to the back, but not to the front of the head. There was tenderness over the upper facet joints on the symptomatic side (probably), and marked tenderness over the lower part of the sternocleidomastoid muscle, bilaterally. Neck mobility was gravely restricted in all directions. Range of motion could not be examined properly because he became dizzy and unwell, and hazy vision persisted after the testing. Magnetic resonance imaging (MRI) of the brain was negative. MRI of the neck demonstrated a disc herniation at C6-C7, with a relatively minor affection of the corresponding nerve roots. Anesthetic blockade of the GON at 90% pain level did not influence the pain. Cortisone injections along the tendon insertions in the occipital area were of no benefit. Acetylsalicylic acid, ibuprofen, various antidepressants in adequate dosages, and various combination analgesics, partly with codeine, were of little or no avail. In addition, he had had physiotherapy, "psychomotor therapy", lymph drainage, needle acupuncture, and traction of the neck, all without any durable effect. The pain during exacerbations was at 10+, on a scale of 0-10+, much of the time. (The au-

thors used a 0-6+ scale, Table I). He then had a strong feeling of being generally sick, with nausea, vomiting, cold sweat and unclear vision. At times, he was on the point of fainting and had to stay in bed. Life had become unbearable. He now maintains spontaneously that at the time he found life meaningless.

Given this situation and also considering the lack of effect of the various forms of therapy, dorsal column stimulation equipment was implanted when the patient was 60 years of age (December, 2003), the electrodes being positioned at the C1/C2 transition. Stimulation caused symmetrical sensation in the occipital area and arms. The improvement was immediate; this event "marked a new epoch in his life": the arm complaints disappeared and practically all the headache. The tinnitus disappeared, while the neck ache, which made up around 40% of the total pain picture, also improved. After the operation, the pain interfered very little with the patient's activities, whereas before his pain and general symptoms had precluded any normal activity. The patient himself estimated the improvement at this stage to be approximately 80%. Unfortunately, around a year after this operation, he sustained a direct trauma to the neck that left him largely back to his preoperative conditions. The electrode had migrated. It was re-positioned, with initial, good effect, but this lasted for only two weeks. The electrode had again migrated, its tip being at the C2 level. After re-operation, during which the electrode was anchored at the C0 level (at the bony part of the posterior end of the foramen magnum), under direct vision, the full effect of the stimulation was restored. Close to two years later, i.e. in February 2008, pain attacks recurred when the neurostimulator stopped functioning. It was replaced. Ever since, the effect of the stimulation has been good. In January 2009, the patient reported that he could still experience some pain in the neck and sometimes in the posterior part of the head, up to 2-3+, but mostly on provocation. Provoked headache would either vanish spontaneously, or ibuprofen would abate it. But he maintained that, for all practical purposes, his headache had disappeared. His memory and concentration had improved so much that he was once again able function adequately as a board member.

Non-indomethacin-responsive chronic hemicrania

A female, born in 1977, had from the age of around 23 years experienced yearly, several-weeks-long episodes of so-called sinusitis, all of which appeared on the right side. Symptomatic side lacrimation accompanied these episodes, as did nasal secretion, which only once may have had a yellowish tinge to it. Each time, antibiotics were felt to have abolished the complaints. Then, at the age of 28 years, these right-sided symptoms suddenly recurred and this time antibiotics were ineffective. The pain was chronic-fluctuating from the onset. Exacerbations could last from one or two days to over a week. Headache clearly impacting on daily life was present on around 75% of the days during periods of registration. At its maximum, the pain could, in our estimation, be rated at 6+ on a 0-6+ scale, and it severely affected the patient's sleep; she was unable to lie quietly, and would lie on the floor and bang her head (Table I). At times, she had destructive thoughts. At best, she experienced an

annoying feeling of heaviness; this feeling could last up to a week. Pain was most marked in the anterior temporal/frontal/aural areas, behind the eye, and in half of the nose. Exacerbations could start as ocular "irritation": itching/burning. At its maximum, the pain would increase each time the symptomatic side foot touched the ground during walking. The pain was "deep" and might be pounding when severe; the pounding was exacerbated during the Valsalva maneuver. There was occasional numbness in the symptomatic side hand and foot (!), but not simultaneously. Phono- and photophobia were not present initially; more recently they could occur, but rarely and to a mild degree.

Jabs (idiopathic stabbing headache) had been present almost daily since onset and felt like needle pricks, mostly in the symptomatic side vertex area, but the localization could vary, and occasionally jabs were felt in the external meatus. They usually lasted 3-5 seconds, but they could, on rare occasions, last up to a couple of minutes; they were intense and could appear in short-lasting series. These jabs could be associated with "jolts", but not with "vocalization". Tinnitus, apparently on the symptomatic side, increased *pari passu* with the pain, but could also appear independently of it. Attack-related allodynia appeared around two years after onset and was located within the headache zone. Episodes with hyperventilation during severe headaches started approximately two years after onset of headache, which she ascribes to nervousness.

The following drugs had been tried: gabapentin, 1200 mg per day; verapamil 480 mg per day; topiramate and kandesartan (100 mg per day), all without any appreciable effect. Moreover, trimipramin (100 mg per day) and amitriptylin (25-50 mg per day) as well as diazepam were tried, also without any appreciable effect. Sumatriptan, on the other hand, could more or less remove the pain for 4-6 hours. However, because of the daily frequency of the pain, she soon developed overuse problems, and the drug had to be discontinued. Indomethacin was tried as atypical HC was suspected. Initially, on 75-150 mg per day, there usually seemed to be only minimal pain, but after a while – even on 150 mg per day – there could be attacks of 50-60% intensity. Increasing the dosage to 200-250 mg per day did not essentially change the situation; there were no pain-free days. At one time, the patient probably had an in-

Table I - Headache scores in both cases, compared with cervicogenic headache.

	Patient 1	Patient 2	CEH*
Cervicogenic factor (score 0-5+)	4.5	1.-1.5**	2.37
CEH criteria, no. (scale 0-6+)	6	1-2 **	6.0
Mean intensity (scale 0-6+ or: %)	5-6	45%***	3.8

Abbreviations and symbols: CEH=cervicogenic headache; *=Data regarding "core cases" [n=41,Vågâ series (2)]; ** Mean entire Vågâ series (2): 0.79+; *** See text.

domethacin headache. When on adequate dosages of indomethacin, the patient did not notice any pain accentuation when taking steps; the jabs vanished, as did the destructive thoughts and the nightly awakening due to pain.

Clinical examination demonstrated that the range of motion in the neck was probably within normal limits. Tenderness along the tendon insertions in the occipital area may have been slightly increased.

MRI scans of the brain and cervical spine were negative, and so was MRI angiography, including "sections" through the vessel wall of carotid and vertebral vessels, bilaterally. CSF was normal, including Borrelia antibodies; the latter test was also negative in serum samples. CT examinations of the sinuses were normal.

Due to the lack of effect of drugs, electrical stimulation was implemented at the age of 31 years. The electrodes were placed around 2 cm above and 4-5 cm in front of the symptomatic side ear, and were anchored in the galea. The pain picture started to improve immediately, and the favorable situation has lasted ever since (approximately 11 months). Daily recording of pain was carried out periodically during the last two years prior to the operation (no. of days: 132) and periodically since the operation (no. of days: 141) (Table II).

In the patient's estimation, the lowest level of headache she experiences is 10%. At this level, her complaints are a nuisance more than anything; at 15% intensity the headache influences her daily life, whereas at 20%, she would take an analgesic pill. It is worth noting that her headache scale ratings end at around 60-70%. This is because she feels that the headache that she has heard of from other patients sounds worse than her own (even when hers is so bad that she tumbles around on the floor and wants to bang her head). The mean preoperative headache pain level, i.e. 45%, must be viewed in this light and should accordingly probably be rated 10-20% higher, or more (Table I). Our reasoning is this: during a >2 month-long discontinuous registration, there were 1/3-1/2 days with pain >=60%, all of which may, in reality, have corresponded to an 80-100% pain level.

Postoperatively, headaches of any significance on the symptomatic side have only occasionally occurred (Table II), at times requiring paracetamol, 500 mg. The patient could go two weeks without any headache complaints. There could, on rare occasions, be headache of

40-60% intensity. The headaches were usually short-lasting. The highest percentage noted on any given day was invariably included in the calculations (Table II). Pure non-symptomatic-side headache also appeared, but much more infrequently than that on the symptomatic side. Left-sided headache was generally mild, but pain of up to 40-60% intensity has been reported, and one attack lasted six hours. The pain on the non-symptomatic side seemed to appear in the same area as that on the symptomatic side, even including the one side of the nose. The patient herself declared herself "99% improved", although she later adjusted this to ">90% improved."

Autonomic symptoms and signs preoperatively. In the preoperative period, the patient naturally was particularly concerned about her head pain. Autonomic and other particular phenomena cannot be assumed to have been recorded with an exactness that makes them suitable for any quantitative estimates or direct comparison with such phenomena from the post-operative period. However, with these reservations, it is probably fair to state the following. Lacrimation on the symptomatic side was sometimes massive, also occurring in the presence relatively strong supraorbital pain on the non-symptomatic side. Occasionally, non-symptomatic-side lacrimation was present. Otherwise, the following phenomena were occasionally present on the symptomatic side: conjunctival injection, nasal stuffiness and a feeling of pulsation in the eye. These phenomena could antedate pain exacerbations; they were regularly more short-lasting than the pain attack per se. The nostril on the symptomatic side was constantly blocked, whereas it was open on the opposite side. During an attack, rhinorrhea was an additional feature, but only on the symptomatic side. A clear miosis (asymmetry of up to 4%) and a sagging upper eyelid came and went on the symptomatic side. The latter phenomena could last the entire day, independent of headache intensity; once, they seemed to be present more or less continuously for a whole week. These features were documented on photos and were observed by one of the authors (OS). Allodynia was frequent and was most marked during periods with strong pain and always seemed to be within the area of the pain.

Postoperative autonomic phenomena. Of the local autonomic features, lacrimation was clearly the most frequently occurring one. On one occasion, around 4 months postoperatively, heavy lacrimation occurred on the non-symptomatic side (i.e. left side) during a bout of rather severe supraorbital pain. At another time, relatively heavy, non-symptomatic-side headache was accompanied by nausea and vomiting. Rhinorrhea, conjunctival injection, and miosis – in that order of frequency – all occurred only in conjunction with lacrimation. Miosis was observed only during the first two months postoperatively. Lacrimation largely occurred without headache; headache also largely appeared without autonomic phenomena; thus, headache and autonomic phenomena usually appeared independently. It seems as though the miosis may have been linked to periods with strong complaints. Allodynia, jabs, and hyperventilation were not reported postoperatively.

Table II - Case 2: Pre- and postoperative headache complaints.

	Pre-operative	Postoperative
No. of registered days	132	141
Severity (in %, mean)	45*	5
Severity variation (%)	10-70	0-60
No. of headache days (>10%)	**	37

* This figure could be 10-20% higher, see text;

** Two examples: 2 weeks, continuous registration: daily headache; 31 days of continuous registration: 7 days with just discernible headache (around 10% intensity); the remaining days: intense-excruciating headache.

Technical data

The following equipment was implanted: Pisces Quad electrodes; a standard, subcutaneous extension and an Itrel 3 pulse-generator (Medtronic Inc., Minneapolis). The stimulation parameters were: Patient 1: electrodes 0 & 1 were negative and the neurostimulator case was positive. The frequency was 90 Hz; pulse width 240 μ s; amplitude 2.5 (0-6); 4 min "on" and 6 min "off", with a soft start; Patient 2: electrode 2 was negative, electrodes 1 & 3 positive. The frequency was 65 Hz; pulse width 210 μ s; amplitude 1.95 (0-4), with continuous stimulation.

Comments

The similarities and differences between these two patients will be discussed. As we judge the situation, the chances that these two patients suffer from the same basic disorder are close to zero. Case 1 seems to show a CEH picture, given that there is *fulfillment of each of the CEH criteria* (18) (Table I). These criteria are: unilaterality; reduced range of motion in the neck; shoulder pain and arm pain; mechanical provocation of attack/exacerbation by awkward neck positions, and by external pressure against circumscribed, sensitive areas in the neck. Also, the recently proposed, extra criterion: initial pain of attack in the neck (2) was met; in short, a total of 7 CEH criteria. Moreover, his cervical abnormality index, or cervicogenic factor (CF) (26), was 4.5+, a clearly abnormal value (scale:0-5+); mean, entire Vågå material (n=1834): 0.79+ (Table I). The skin roll test (on the "arch" of the shoulder area) was positive, with 35 mm skin-fold width on the symptomatic versus 25 mm on the non-symptomatic side, the skin in this area being extraordinarily tender, in particular on the symptomatic side. The skin-roll test thus showed three clear abnormalities: i) an asymmetry of 10 mm (>3 mm is considered abnormal (27)); ii) an apparently clearly abnormal value (35 mm) on the symptomatic side, given that >25 mm is considered abnormal [the mean for the headache-free individuals in the Vågå study n=246: 14.3mm \pm 5.7 (27)]; iii) the extraordinary tenderness.

Despite the fact that drug responses were not tested systematically, it is felt that this case is clearly different from our female case, because of: the onset after trauma; the severely decreased range of motion of the neck; the mechanical pain provocation, etc. There were traits out of proportion with the ordinary CEH picture: the excruciating pain of the attacks/near-fainting episodes/general feeling of malaise. These latter phenomena attest to the gravity of the picture and they all disappeared upon electrical stimulation.

The constellation of symptoms and signs in case 2 was quite different.

The CF (26) was: 1-1.5+; there was no spreading of pain to the head. The CEH criteria were relatively poorly met (Table I). It is noteworthy that 1+ is due to the unilaterality of the pain, which happens to be a trait common to both CEH and case 2.

On the other hand, the clinical picture in case 2 was reminiscent of HC, i.e. unilaterality and autonomic phenomena (28). However, two essential features did not quite fit into the overall picture of HC: i) the sometimes

excruciating pain intensity, i.e. 6+, on a 0-6+scale (intensity is not known to be excruciating in HC); ii) the far from absolute indomethacin response, despite a daily dosage of up to 250 mg. Lack of indomethacin response – or an only partial response – is not consistent with a diagnosis of HC.

From the differential diagnosis point of view, the sumatriptan effect in case 2 should be heeded: sumatriptan is not known to influence HC highly positively (29), although the effect in HC seems to be marginally better than that in CPH, where it is totally lacking. Although the clinical picture in case 2 bears some resemblance to HC, the arguments against HC seem to be considerable. We are probably faced with a case of headache similar to, but clearly distinct from HC. Such cases may not be rare and may have contributed to the belief that HC *per se* "is not that rare" (30).

Therefore, the observed good stimulation response in this case cannot be extrapolated to HC, even though, of course, the present observations do not allow it to be concluded that electrical stimulation will not be helpful in HC.

So, has HC as such ever been successfully treated with electrical stimulation? According to one report of a solitary case of unilateral headache, initially published under the heading HC (31), the pain was localized in the occipital area; HC is generally an anteriorly located headache. Another argument against a diagnosis of HC in this case is that even an indomethacin dosage of 225 mg did not make the patient pain-free (remaining pain: 2/10). Another disturbing fact is that the pain apparently started immediately after a head trauma with loss of consciousness. Thus, in spite of the presence of some traits showing some similarity to HC, there is no convincing evidence that this is a case of HC. This case, in other words, does not prove that HC is remediable by electrical stimulation. Whether our case and this female case (31) belong to the same category is an open question. A lot depends upon the role of the head trauma in the reported case, which is – and will remain – uncertain.

In a more recent communication, the same group dealt with a series of 15 patients, two of whom were diagnosed as HC (32). The clinical picture was summarily described. Indomethacin was apparently effective, but was discontinued, reportedly due to complications. Dosage, duration and degree of effect of the indomethacin trial are not given. One piece of information may seem important: "Each patient suffered from head pain that involved the C2 distribution (with or without pain in the other regions of the head)". This should thus include the two HC patients. At face value, then, it seems that these two cases are not readily acceptable as HC cases.

In a recent study, a mini-stimulator was implanted over the symptomatic-side occipital nerve in six HC patients (33). The indomethacin test was reportedly positive in all of them, at an indomethacin dosage of 100-200 mg. It is not stated by which route indomethacin was administered. In another recent article by the same group, it is reported that indomethacin at this dosage was administered intramuscularly as a test dosage (34). The recommended dosage for parenteral indomethacin as a diagnostic test is 50 mg (35), since 100 mg does not add any information of diagnostic value. By doubling, or even quadrupling this test dosage, a factor of unknown con-

sequences is introduced. Disorders essentially different from HC may respond in a positive way. The recommended dosage for the acute treatment of the colicky pain of urolithiasis (!) is also 50 mg intramuscularly. There must be a clear correspondence between the test dosage and the presumed long-term dosage in treatment; otherwise, the prognosticating role of the "in-dotest" will be fictitious.

It is hard to understand why the authors chose this policy. In spite of the positivity of the indomethacin test, the long-term efficacy of indomethacin proved to be mediocre: it was not absolutely effective in any subject; it "improved" the situation in three, and the dosage was 150-225 mg per day. A diagnosis of HC can hardly be sustained in this group. In four of the patients, there seemed to be good response to the mini-neurostimulator, but not in two. According to the authors, this is an "effective treatment for HC". In our estimation, this treatment seemed to provide ample/good response in some cases of a unilateral headache that, however, was not proven to correspond to HC.

In our second case, electrical stimulation influenced very favorably the mainly temporal/anterior headache, which was apparently different from HC and CEH. This leads us to the conclusion that extracranial electrical stimulation may have a beneficial influence on more than one type of headache and also on headaches not primarily or mainly located posteriorly. It would be extremely interesting to identify characteristic traits able to predict suitability for such electrical stimulation.

Headaches remediable with electrical stimulation may, on the basis of our present insight, preferably have the following qualities: unilaterality, severe intensity, chronic-fluctuating headache; they may either be CEH-like, or have an anterior/temporal/aural location and be combined with variegated, marked, ipsilateral, autonomic phenomena. The headache has, in both cases, been characterized as "deep", and co-existing allodynia may be marked. In our female patient, the indomethacin effect was far from absolute. On the other hand, the sumatriptan effect was satisfactory. Whether this latter feature would also apply in a larger series is uncertain.

The question of electrode placement in headaches that are not primarily occipitally located is a burning one. The placement seems to have been near optimal in our case. At this stage, one must be willing to accept some trial and error, even intra-individually, to find the ideal site. We are skeptical about improvements that appear weeks/months after the operation with this kind of treatment.

We are clearly faced with a nosological problem: a non-posterior, unilateral headache that is non-CEH and non-HC and does not have obvious neuralgiform traits, but is most favorably influenced by electrical stimulation, cannot be readily categorized. Over recent years, we have observed five other patients with traits resembling those in our case 2. During this period we have seen no new cases of HC. The present patient is the first one of this group operated on in this way. Due to the positive outcome of the invasive procedure in this patient, she is reported separately. These cases seem to have little more than the unilaterality of pain and some autonomic features in common with HC. Indomethacin responsiveness is a fundamental quality of a headache, and it creates a wall between responders and non-responders.

As we have claimed for years, patients on the two sides of the wall are not akin and must not be mixed up. As we see it, a totally neutral term vis-à-vis HC must therefore be sought for these cases. "Non-indomethacin-responsive chronic hemicrania" would be such a term: this headache is a hemicrania, and it does not respond to indomethacin in a clinically useful way. "Continuous" is too loaded a term that immediately causes thoughts of HC to cross one's mind. "Continuous" is therefore substituted by "chronic". This group – it may be abbreviated as "Nirch" – is hardly a homogeneous group; it may include several minor, unilateral groups. A sublime task will be to divide the group correctly. HC as such will probably remain as a group in its own right: an entity sui generis.

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