

Gregory S. Antonarakis  
Mike Hughes

## A case of unclassified headache involving bilateral nuchal to frontal pain associated with burning eyes and lacrimation

Received: 17 April 2006  
Accepted in revised form: 12 May 2006  
Published online: 15 June 2006

G.S. Antonarakis (✉)  
28 Philosophes,  
CH-1205 Geneva, Switzerland  
e-mail: gregory.antonarakis@zoo.unige.ch  
Tel.: +41-22-7891007  
Fax: +41-22-3796795

G.S. Antonarakis  
Clinique Dentaire de la Jeunesse,  
Geneva, Switzerland

M. Hughes  
General Dental Practice,  
Gwynedd, Wales, UK

**Abstract** A 21-year-old Caucasian male presented to an oral medicine clinic with bilateral nuchal to frontal headache that was associated with burning eyes and lacrimation. Following a string of previous consultations with a variety of specialists, no diagnosis had been made. Treatment was carried out empirically, with the best improvement experienced when the patient was put on a combination of gabapentin and dothiepin treatment. This case highlights the importance of multidisciplinary management of headaches, as well as constant re-

sion of the International Classification of Headache Disorders.

**Key words** Headache · Multidisciplinary treatment · Headache classification

### Case history

A 21-year-old Caucasian male fitness instructor presented following a long string of referrals to an oral medicine clinic complaining of chronic headache. According to the patient, these headaches were significantly affecting his quality of life and ability to perform his job properly. It had been about three years since the start of the headaches. His initial complaint was of pain in the nuchal region bilaterally. This pain radiated to the top of his head into his frontal region, again bilaterally. There was no history of flashing lights, photophobia, phonophobia, nausea or vomiting associated with the pain. The patient did however experience associated burning of the eyes and lacrimation, but without the presence of either electric

shock or allodynia. Another occasional related symptom was shoulder tension.

Since its initial onset noticed by the patient, the pain had changed in its distribution and character throughout the day. Originally the pain had occurred daily and was constant. With time this changed to a more occasional pain, however still occurring daily. The pain seemed to peak on waking in the morning, as well as in the late evening. Infrequently, the patient was woken up by the pain. In the morning, it was more sharp in nature, eventually fading and developing into a more dull but burning sensation. The duration of each individual headache was from waking until the patient went to bed, therefore lasting throughout the day, with a variation only in intensity.

Certain aggravating and relieving factors were mentioned by the patient during history taking. If the patient

pushed on top of the head then the pain worsened. When the patient closed his eyes and applied pressure over his eyelids, the burning sensation in the eyes would be eased.

The past medical history of the patient included nephritis, which also led him to being hypertensive. He was under the regular care of his nephrologist. His medication included ramipril, bendrofluzide and penicillin. There was no history of anxiety or depression.

Prior to arriving at the oral medicine clinic, he had been examined by a wide range of specialists. Consultations with a general practitioner, neurologist, anaesthesiologist, otolaryngologist, ophthalmologist, psychiatrist and physiotherapist including investigations such as radiography, computed tomography and magnetic resonance imaging led to the exclusion of any underlying problem and any headache disorders, but a diagnosis remained elusive. Treatments had however been carried out on an empirical and palliative basis to alleviate the pain. These had proved unhelpful as regards long-term pain control. The pain failed to respond to analgesics (including indomethacin), amitriptyline, physiotherapy, traction, osteopathy, reflexology, acupuncture, botulinum toxin (botox) and a transcutaneous electrical nerve stimulation (TENS) machine. The only short-term relief had been achieved by an occipital nerve block with local anaesthetic and 40 mg depomedrone performed by the anaesthetist. The pain relief in this case had been a marked decrease in intensity but only lasted a couple of weeks. In spite of this, when repeated, subsequent injections did not give the same benefit.

At the oral medicine clinic, following a thorough history-taking, an examination was carried out only to lead to a lack of indicative clinical findings once more. No signs of bruxism, temporomandibular joint dysfunction or pathology were noted. It was decided nevertheless to construct a lower hard occlusal splint. When worn for two months however, still no improvements were seen. Following this, treatment went down the line of medication commonly used for facial pain. First, gabapentin 300 mg three times daily was prescribed. This was later increased to 600 mg three times daily. This seemed to mildly ease the pain, which was noticed more by the patient as a result of the sudden increase in pain intensity if the medication was stopped. Dothiepin 50 mg nocte was later added. Pain intensity with dothiepin was the same but the duration of each individual headache seemed to be less, not always lasting throughout the day but only until the early evening. Dothiepin 25mg was added to be taken in the morning, which did not seem to offer any significant improvement in symptoms. Overall, the headache was still present daily, but the symptoms were better when the patient was on the medication than without it. The intensity of pain was diminished, the duration decreased by a few hours and the eye symptoms showed an improvement.

Despite the absence of a specific diagnosis, the headache was managed empirically and some improvement, albeit not remarkable, was attained. This would possibly suggest a neuropathic pain, as the medication used in this case, eventually showing an improvement in symptoms, is normally indicated for neuropathic pain. Its appearance, however, does not fit into any specific diagnosis.

---

## Comments

The headache described here seems to be associated with unique symptoms, not permitting a diagnosis to be made. According to the 2nd edition of The International Classification of Headache Disorders (ICHD-II), it would therefore fall under the category of 14.1: Headache not elsewhere classified [1]. A small minority of headaches do not meet recognised criteria, and even after the keeping of a diary cannot be reliably diagnosed. The most important requirement in such cases is to exclude serious causes.

For presenting complaints of headaches, once serious causes have been excluded, there are no real diagnostic tests available. The history is thus all-important. Only symptoms, the patient's subjective experience of changes in his or her body, are available, and difficulties arise when the practitioner can find no objective changes to explain the patient's subjective experiences [2]. Sometimes, despite the absence of a definitive diagnosis, treatment can be begun nevertheless, on an empirical basis, as was the case here.

When faced with symptoms of headache, the patient (as well as the general practitioner at times) is unsure as to which specialist to consult. Artificial distinctions in clinical presentation lead patients to different specialists providing different treatments, including dentists, neurologists, otolaryngologists, osteopaths, chiropractors and psychiatrists, with little collaboration [2]. Different specialists then tend to refer patients to one another when they consider that diagnosis and management is not under their competence. In this way, patients lose time within referral systems and waiting lists, without resolution of the headache. What would benefit both patient and specialist in these cases would be the existence and use of multidisciplinary headache clinics where many specialists and their expertise come together and collaborate in the diagnosis and treatment of headache disorders.

Chronic symptoms in headache cases are common, often persistent, and associated with significant distress, disability and unnecessary expenditure of medical resources. Patients want to know what is wrong with them rather than what is not wrong. Enormous benefit is gained from having their symptoms acknowledged as part of a

group of conditions with which clinicians are familiar. Reassurance about the non-malignant nature of the chronic pain is also important [2].

Regarding treatment, it may be more helpful to assess patients in terms of disability and coping strategies rather than pain intensity itself [3]. This provides the treating practitioner with useful information concerning both the possible cause of headache as well as possible responsiveness to treatment. Having said this however, depression is a common feature of headache cases but is suggested to be more likely a consequence of living with pain rather than a precursor to it [4]. Pain relief with gabapentin and dothiepin, a tricyclic antidepressant (TCA), was chosen in this case as gabapentin targets neuropathic pain, and TCAs are considered to act by altering the sensory discriminative component of pain [5]. Drugs such as dothiepin are of

proven, if modest, benefit in the management of head pain [2].

There is a growing body of evidence showing that different headaches require quite different treatments. Detailed and accurate diagnoses need to be made and treatments tailored accordingly [6]. Treatment without an accurate diagnosis may be problematic, and thus multidisciplinary management is recommended, increasing the likelihood of accurate diagnosis and treatment. A high-quality classification of headache disorders (ICHD-II) enables better headache research, understanding of headache, communication and, ultimately, management of a set of disabling neurological disorders [7, 8]. However, regular revisions are still necessary to keep the classification up-to-date, and reporting of cases not fitting into the classification should aid in this.

---

## References

1. Headache Classification Subcommittee of the International Headache Society (2004) The International Classification of Headache Disorders, 2nd edition. *Cephalalgia* 24[Suppl 1]:9–160
2. Madland G, Feinmann C (2001) Chronic facial pain: a multidisciplinary problem. *J Neurol Neurosurg Psychiatry* 71:716–719
3. Madland G, Feinmann C, Newman S (2000) Factors associated with anxiety and depression in facial arthromyalgia. *Pain* 84:225–232
4. Dohrenwend BP, Raphael KG, Marbach JJ, Gallagher RM (1999) Why is depression comorbid with chronic myofascial face pain? A family study test of alternative hypotheses. *Pain* 83:183–192
5. Pettengill CA, Reisner-Keller L (1997) The use of tricyclic antidepressants for the control of chronic orofacial pain. *Cranio* 15:53–56
6. Olesen J (2004) The International Classification of Headache Disorders, 2nd edition: application to practice. *Funct Neurol* 20:61–68
7. Olesen J, Steiner TJ (2004) The International classification of headache disorders, 2nd edn (ICHD-II). *J Neurol Neurosurg Psychiatry* 75:808–811
8. Peatfield R (2004) A revised classification of headache disorders. *BMJ* 328:119–120