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Migraine presenting as chronic facial pain

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Abstract

We report the case of a 44 year old woman with chronic facial pain. She was treated with several analgesics, prophylactic medications and infiltrations, but all treatment modalities were ineffective. Finally, the diagnosis of medication-overuse headache complicating migraine without aura was made and an appropriate treatment was initiated. Migraine is a very common primary headache and rarely presents as isolated facial pain. Stimulation of the dura with activation of the trigeminovascular system can result in pain in any of the three divisions of the trigeminal nerve. This is the anatomic basis of migraine pain presenting as referred pain to the second division of the trigeminal nerve. The atypical presentation of migraine pain can easily lead to inappropriate treatment regimens.

Key words: Migraine; facial pain; trigeminovascular system; medication-overuse headache.

Introduction

We report the case of a patient with chronic facial pain. She was treated in several ways, but all were ineffective. Finally, the diagnosis of medication-overuse headache complicating facial migraine was made and an appropriate treatment was initiated.

This case report illustrates three issues. First we review the concept of migraine presenting as facial pain. Second, we show that medication-overuse headache may have a facial presentation. Third, we highlight the fact that infiltrations are easily applied, but not always necessary to treat facial pain.

Case report

The patient is a 44 year old woman. Her medical history comprises an appendectomy and an ectopic pregnancy. At the time of consultation she was taking daily venlafaxine and a suppository containing ergotamine tartrate.

She was referred to our headache clinic for a problem of left-sided facial pain. She had been suffering from this pain since the age of 21. The pain was located in the left maxillar region. Initially she felt this pain episodically lasting for 24 hours. It was accompanied by nausea, vomiting and anorexia. There was no phonophobia, nor photophobia. During these painful episodes, she had an urge to move rather than to lie down or to sit still. The pain was very severe but not electric shock-like. There were never any associated autonomic phenomena nor facial trigger points. A preceding aura was absent.

The following years, the frequency of the painful episodes increased evolving to a daily pain. She started using ergotamine tartrate - caffein (Cafergot®) suppositories, which seemed to control the pain.

At the time of consultation, the patient had been using daily the suppository for more than 12 years. When she didn't use it, she would suffer from a severe burning pain localised in the left maxillar region. For this facial pain,. several dental treatments were done. Later a neurosurgeon injected the cavum of Meckel with glycerol, resulting in absence of pain during a week. Later on, a stabbing pain reoccurred.

She tried treatments with maprotiline, sodium naproxen, haloperidol, orfenadrine hydrochlorine, flupentixol, clomipramine hydrochloride, pimozide and melitracen without success. An infiltration with lidocaine and repeated infiltrations with a steroid (Depo-medrol®) controlled the pain only temporarily.

Five years later, four infiltrations of the stellate ganglion were performed and treatments with melitracen, flupentixol, passiflora extract, verapamil and amitriptyline did not show any effect. About six years later, she was referred to us by her family physician. The characteristics of the pain were as described above. The neurological examination was normal. Keeping in mind the associated features of the facial pain, and the duration of the attacks, the

diagnosis of facial migraine was considered. The diagnosis of medication overuse headache was also made. To abort this overuse, the ergotamine was withdrawn. She was admitted for five days and treated with dihydroergotamine 1 mg given intramuscularly every 8 hours, preceded by an injection of metoclopramide hydrochloride 10 mg given intravenously. The pain did not resolve completely, but after 5 days she was satisfied with the result of this treatment. A preventative treatment with bisoprolol fumarate 5 mg was initiated. Further use of the ergotamine suppository was prohibited. To control for severe pain, a treatment with domperidone 20 mg and sodium naproxen 550 mg was prescribed, with the restriction not to use these medications for more than 3 days a week. At follow-up, she was still satisfied with her condition. The attacks of stabbing or burning pain in the left maxilla hadn't subsided, but they appeared episodically instead of daily. She was using the same suppositories again, but only once a week, when the pain was too severe.

Discussion

In patients with facial pain without dental, sinus or ear disease, the differential diagnosis mainly includes cluster headache, trigeminal neuralgia and migraine (Feinmann *et al.*, 1993).

In our patient, the diagnosis of cluster headache was excluded mainly because pain attacks lasted longer than 180 minutes, and because of the lack of autonomic features (Lipton *et al.*, 2004). Although the latter is no longer required in the 2004 IHS classification when a sense of restlessness or agitation is present. The facial pain of our patient was not short-lasting nor electric shock-like; trigger points were absent, making the diagnosis of trigeminal neuralgia unlikely.

We considered the possibility that her facial pain was in fact facial migraine. Recently, Gaul *et al.* reported two patients with attacks of unilateral dental pain as their main complaint (Gaul *et al.*, 2007). Except for pain localisation, the pain attacks fulfilled IHS criteria for migraine with and without aura (ICHD-II 2004). Based on these criteria, the initial pain of our patient was considered to be migraine, as she had more than 5 attacks of pain lasting about 24 hours. The pain was located unilaterally, was moderate to severe in intensity and associated with nausea or vomiting. At the initial consultation the diagnosis of probable ergotamine-overuse headache was made, based on the IHS criteria. These criteria have been revised (Olesen *et al.*, 2006), and field-tested by Zeeberg *et al.* (2009). Using the appendix criteria, our patient would fulfil the criteria for medication overuse headache.

Facial migraine is a migraine subtype which is sometimes called lower-half migraine by a number of authors (Raskin, 1988; Okeson, 1996). Lower-half migraine comprises pain reported in the lower half of the face, mainly the lower jaw and cheek. Yet few attempts have been made to characterize or categorize patients presenting with such pain (Penarorrocha *et al.*, 2004). Facial migraine usually develops in the first 4 decades of life, followed by a gradual decrease in the frequency of pain crises (Raskin, 1988). The condition predominates among women, as is classically the case with migraine (Okeson, 1996). Our patient was female and developed the pain at the age of 21.

The exact pathogenesis of migraine remains to be determined. Our current understanding of migraine pain is that it is caused by inflammation and dilation of the meningeal blood vessels. The inflammation results from the actions of neuropeptides CGRP and substance P, which are released from primary sensory nerve terminals innervating the dural vessels (Bussone, 2004). The trigeminovascular system is comprised of sensory fibres that densely innervate the cerebral blood vessels and dura mater (Del Rio *et al.*, 2004). The ophthalmic division of the trigeminal nerve innervates the vast majority of the intracranial structures (Alonso and Nixdorf, 2006). This extensive intracranial innervation by the ophthalmic nerve explains why headache is perceived by most patients as periorbital pain. The maxillary division of the trigeminal nerve gives off a dural branch, the nervus meningeus medius, which supplies innervation to the dura of the anterior floor of the middle fossa (Larrier and Lee, 2003).

Migraine pain can be perceived as a referred pain because nociceptive fibres coming from the intracranial structures converge on the same pool of second-order sensory neurons within the trigeminal nucleus caudalis or upper cervical dorsal horns together with nociceptive inputs from cutaneuous tissues (Dalkara *et al.*, 2006). It is recognized that stimulation of the dura with activation of the trigeminovascular system can result in pain in any of the three divisions of the trigeminal nerve (Daudia and Jones, 2002). In an identical way, pain occurring in migraine may be referred to the second division of the trigeminal nerve and present as facial pain. In the series of facial migraine reported by Benoliel et al, all patients referred to oral pain (Benoliel *et al.*, 1997). In a similar way, patients with migraine frequently may complain of neck discomfort without pathology at the cervical spine (Bartsch and Goadsby, 2003).

Neither the International Headache Society (IHS) nor the International Association for the Study of

Pain (IASP) specifically mention the condition of facial migraine (Mersky, 1994; Benoliel *et al.*, 1997; Bartsch and Goadsby, 2003). We believe that the pain of migraine might be located in the face, as do other authors (Penarorrocha *et al.*, 2004; Daudia and Jones, 2002; Obermann *et al.*, 2007). The terminology in the literature to describe the distribution of pain is that the area below the eyebrows/supraorbital rims is regarded as facial pain, whereas the forehead and temporal region are included in the term headache (Daudia and Jones, 2002). Patients with episodic pain in the distribution of the first and second division of the trigeminal nerve and whose other diagnostic criteria are consistent with migraine would often be given the diagnosis of migraine by a neurologist. However, those with pain restricted to the second division of the trigeminal nerve would often be misclassified. The facial pain in our patient was mainly located in the V2-territory of the trigeminal nerve. Migraine confined to the second division of the trigeminal nerve has rarely been described, but is mentioned by some authors (Campbell, 1990; Bradley, 2000; Silberstein, 2001; Daudia and Jones, 2002; Obermann *et al.*, 2007). Some patients have pain confined only to the V3 territory (Silberstein, 2001).

The fact that the pain of migraine is located in the face, easily leads to iatrogenic damage and inappropriate dental treatments (Vickers *et al.*, 1998). In the series of Obermann *et al.* 5 patients were wrongly diagnosed with trigeminal neuralgia and treated with carbamazepine and gabapentin (Obermann *et al.*, 2007).

We conclude that facial pain might be an expression of migraine and medication-overuse headache. Recognition of this migraine subtype is essential not only to instaure proper treatment, but also to avoid unnecessary therapeutic procedures.

REFERENCES

Alonso A, Nixdorf D. Case series of four different headache types presenting as tooth pain. JOE. 2006;32:1110-1113.

Bartsch T, Goadsby P. The trigeminocervical complex and migraine:current concepts and synthesis. Current pain and headache reports. 2003;7:371-376.

Benoliel R, Elishoov H, Sharav Y. Orofacial pain with vascular-type features. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 1997;84:506.

Bradley WG. Neurology in clinical practice. Vol 1. Boston, Butterworth Heinemann. 2000:267.

Bussone G. Pathophysiology of migraine. Neurol Sci. 2004;25:239-241.

Campbell K J. Manifestations of migraine. Neurol Clinics Headache. 1990;8:847.

Commitee on classification of headache of the national institute of neurological diseases and blindness. Classification of headache. J Am Med As. 1962; 179:717-718.

Dalkara T, Zervas Nt, Moskowitz MA. From spreading depression to the trigeminovascular system. Neurol Sci. 2006;27 Suppl:86-90.

Daudia AT, Jones NS. Facial migraine in a rhinological setting. Clin Otolaryngol. 2002;27:521-525.

Del Rio M, Linera J. Functional neuroimaging of headaches. Lancet neurol. 2004;3:645-651.

Feinmann C, Peatfield R. Orofacial neuralgia. Drugs. 1993;46 (2):263-268.

Gaul C, Sandor P S, Galli U, Palla S, Ettlin D A. Orofacial migraine. Cephalalgia. 2007;27:950-952.

Headache Classification Subcommittee of the International Headache Society. The International Classification of Headache Disorders 2nd Edition. Cephalalgia 2004;24 (Suppl 1).

Headache Classification Committee: Olesen J, Bousser M-G, Diener H-C, Dodick D, First M *et al.* New appendix criteria open for a broader concept of chronic migraine. Cephalalgia. 2006; 26:742-746.

Larrier D, Lee A. Anatomy of headache and facial pain. Otolaryngol Clin N Am. 2003;36:1041-1053.

Mersky. International association for the study of pain. Classification of chronic pain: descriptions of chronic pain syndromes and definition of pain terms. 2nd ed. Seattle, IASP Press. 1994:68.

Obermann M, Mueller D, Yoon M-S, Pageler L, Diener HC et al. Migraine with isolated facial pain:a diagnostic challenge. Cephalalgia. 2007;27: 1278-1282.

Okeson JP. Differential diagnosis and management considerations of primary headache. Orofacial Pain. Guidelines for assessment, diagnosis and management. The American academy of orofacial pain. Chicago, Quintessence book. 1996:61-66.

Penarorrocha M, Bandres A, Penarrocha M, Bagan J. Lower-half facial migraine:a report of 11 cases. J Oral Maxillofac Surg. 2004;62:1453-1456.

Raskin NH. Headache. 2nd ed. New York, Churchill Livingstone. 1988:215-227.

Silberstein. Headache and other head pain. 7th ed. New York, Oxford university press. 2001:478-479.

Vickers E, Cousins M, Walker S, Chisholm K. Analysis of 50 patients with atypical odontalgia. Oral Surg Oral Med Oral Pathol. 1998;85:24-32.

Zeeberg P, Olesen J, Jensen R. Medication overuse headache and chronic migraine in a specialised headache centre:field-testing proposed new appendix criteria. Cephalalgia. 2009;29:214-220.

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