A New Variant Nummular Headache: Large Diameter Accompanied with Bitrigeminal Hyperalgesia and Successful Treatment with Carbamazepine

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ABSTRACT

Nummular headache (NH) is defined as a focal head pain that is exclusively felt in a small area, which is typically 1-6 cm in diameter. Neurological examinations are normal in all patients, but this report describes a new variant. A patient was identified who presented with focal head pain that was approximately 8 cm in diameter and bitrigeminal hyperalgesia on neurological examination. Treatment with carbamazepine provided significant analgesic relief in terms of both the frequency and intensity of pain. The findings enlarge the clinical diversity of this headache disorder. The pathogenic mechanisms of NH may be similar to trigeminal neuralgia in particular patients.

KEYWORDS: Nummular headache, Hyperalgesia, Analgesia, Carbamazepine

INTRODUCTION

Nummular headache (NH) is a well-defined clinical condition characterized by local pain that is in a small (typically 1-6 cm diameter), rounded or elliptical area of the head. The pain remains confined to this coin-shaped area and does not change in size or shape over time. It is of mild to moderate intensity and can be either continuous or intermittent. After the first description of this condition in 13 patients in 2002 (23), it has been included in the classification of the research diagnostic criteria of the International Classification of Headache Disorders, 2nd Ed. (Table I) (18). At present, more than 100 additional cases have been reported across many different countries (1, 3-6, 8, 10-17, 25, 26, 28-32), of which some have shown different features. In this paper, another new clinical feature is described, which was identified in a patient who was admitted to the Department of Neurology at the Second Hospital of Jilin University in April 2010. This report will highlight the features of this case that increase the clinical diversity of NH.

CASE REPORT

A 59-year-old man presented with 1 month history of headache that consisted of lancinating pain attacks that lasted for several seconds. These attacks were described as paroxysmal, electric shock-like pain that were similar to those described in trigeminal or occipital neuralgia. The pain was non-radiating and could be evoked by touching the area in question. This area covered the area of an 8 cm diameter circle in the right temporal-parietal region, without side shift (Figure 1). The pain was of a severe intensity, fluctuating around eight out of ten, and was not associated with any localized or generalized autonomic symptoms. The patient had a past medical history of significant coronary artery disease and hypertension. He had no history of headaches.

Tender pressure points were found in the symptomatic area. Bitrigeminal hyperalgesia was present, but otherwise neurological and physical examinations were normal. Blood tests, X-rays and ECG were also normal. The basal ganglia...
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and corona radiata had multiple bilateral punctiform and lamellar low density areas on the brain CT scan. Right- and left-sided carotid artery stenoses were present (10% and 20%, respectively) on the head and neck computed tomographic angiography (CTA) scan. The patient had not taken any symptomatic or preventive medications to alleviate the pain. The changes on the brain CT scan and the head and neck CTA suggested a diagnosis of lacunar infarction. However, there were some differences between this case and the diagnostic criteria for NH. Firstly, the diameter of the circular region of pain measured 8 cm, which exceeded the typical diameter of 2-6 cm in NH. Secondly, there were signs of bitrigeminal hyperalgesia, yet most patients with NH have normal neurological examinations, apart from their symptomatic area. However, we believe the features of this case overlapped with the diagnostic criteria of NH, as the area of pain was limited to a circular area over the right temporal-parietal region, which did not change in shape and size in one month. Lacunar infarction is not usually associated with such headaches. Therefore, the diagnosis of NH was made.

The patient was treated with oral carbamazepine at a dose of 100 mg twice daily. The dose was increased to three times daily after seven days, since the pain remained unchanged. It was then changed again to 200 mg in the morning and 100 mg at noon and night after nine days. The pain reduced over this time, as well as the intensity and incidence of the headaches. There were no significant signs on neurological examination and the bitrigeminal hyperalgesia resolved after 14 days. The same dosages were given for a further 10 days and then stopped. The patient was discharged the next day and remained pain free over six-month follow-up period.

DISCUSSION

Since the original description by Pareja, many reports of cases have enlarged the clinical knowledge of NH. It is now known that the lancinating pain attacks may be the only manifestation of NH (27). NH may appear in two distinct areas in the same patient. The symptomatic areas may be located on one side or both sides of the head. The pain may emerge at the same time or follow a different course in each of the affected areas (7). Some patients have shown trophic changes within the painful area (25). These descriptions might be considered to be the symptomatic form of NH. In the present case, a new clinical expression of NH is reported, with features of a short duration, a large diameter in the right temporal-parietal region, lancinating pain attacks that only lasted for seconds in that area, and bitrigeminal hyperalgesia on the neurological examination.

As a primary headache form, the painful area may be localized in any part of the head, but is usually located in the parietal region; the pain does not change in shape or size over time. However, the affected area in this patient was limited to the right temporal-parietal region. The shape of the area of NH pain in this patient was circular, did not change in form and had a diameter that measured approximately 8 cm. The intensity of pain was severe, which was similar to the cases already reported (3, 4, 8, 13, 14, 19, 25, 26), although it is worth noting that NH pain is typically described as mild to moderate (18, 23). The majority of the pain quality of NH has been described as pressing, burning or dull (8, 11, 19, 23, 24, 26, 29, 32). Lancinating pain attacks, without baseline pain, as the only manifestation (2 patients) have only previously been reported by Ruscheweyh et al (27); the present case brings this total to three. Neurological examination of patients with NH is usually normal, except inside the painful area, which typically exhibit tenderness, paresthesia, hyperesthesia and allodynia. This present case is the first in which bitrigeminal hyperalgesia was found as the only sign on the neurological examination. After treatment with instigated with carbamazepine, the pain gradually resolved and had disappeared midway through the course of treatment.
The treatment of NH is difficult. Different drugs and methods have been used for some cases, such as paracetamol, non-steroidal anti-inflammatory drugs, indomethacin, gabapentin, duloxetine, topiramate, amitriptyline, nortriptyline, clomipramine, local botulinum toxin, local subcutaneous lidocaine application, local infiltrations and auriculotemporal nerve blocks, and local transcunetaneous electric nerve stimulation. Some may reduce pain, but the most effective treatments do not require repeated applications. The new treatment option for NH is the use of local botulinum toxin (2); however, there are some different opinions with regards to its efficacy (9).

This case study highlights that carbamazepine may be a successful treatment option for NH. The reason may be due to the type of pain quality. The patient described in this case presented with lancinating pain attacks, such as those encountered in trigeminal or occipital neuralgia. This new finding is interesting as the etiology of NH has yet to be fully described. Possible mechanisms might include neuropathy of a terminal branch of a cutaneous scalp nerve and focal, nociceptive-type pain that stems from epicranial tissues (11, 26). The manifestation of electric-shock-like pain paroxysms without continuous pain, in this case, is termed as the terminal branch neuralgia hypothesis. The peripheral nerve lesion should be along the peripheral nerve in either its extracranial or intracranial course but does not have to be restricted to within the painful area. This hypothesis has been challenged because, in some cases, the painful area extends across the midline and subcutaneous infiltration of the painful area with local anesthesia is unsuccessful in reducing pain in the majority of cases. However, the successful treatment of the pain in the present case does support this hypothesis. The pathogenic mechanisms of NH may be similar to that of trigeminal neuralgia in some patients.

The case presented in this paper is a new variation of the clinical presentation of NH. It was a large diameter (8 cm), accompanied by lancinating pain attacks and presented with bifocal hyperalgesia on neurological examination. The successful treatment with carbamazepine alone offers an insight into the pathological mechanisms that underlie NH and gives some guidance as to its therapeutic treatment.

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