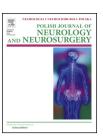


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# Case report

# Nummular headache in a patient with ipsilateral occipital neuralgia—A case report

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#### ABSTRACT

Nummular headache (NH) is a rarely recognized primary headache, the diagnostic criteria of which are contained in the appendix to the 2nd edition of the International Classification of Headache Disorders (code A13.7.1).

We present the case of a 61-year-old female who suffers, regardless of NH, from right-sided occipital neuralgia. The applied treatment – gabapentin and mianserin – had no effect. Injection of bupivacaine twice to the right occipital region resulted in neuralgia resolution up to three months, with no effect on NH.

This confirms the independence of two abovementioned head pain conditions.

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## 1. Introduction

Nummular headache (NH) is a primary, chronic head-pain condition with well-defined topographic borders covering the area of round or elliptical shape with a diameter of 2–6 cm [1–3]. Most frequently, the pain is one-sided and located on the right parietal tuber, although it can also occur in other areas of the head [1,3–5]. Nummular headache does not change its shape and size, but the intensity of pain, ranging from mild to moderate, is sometimes interrupted by periods of exacerbations and spontaneous remissions [6]. Another symptom may be a combination of sensory disorders in the area of pain including hypoesthesia, paraesthesia, dysesthesia and allodynia.

The first cases of NH were described by Pareja et al. in 2002, that made it possible to integrate this type of headache in the diagnostic criteria in addition to the 2nd edition of International Classification of Headache Disorders (ICHD II) (code A13.7.1) [2,3].

In the paper, we introduce the patient diagnosed and treated in the Department of Neurology, University of Medical Sciences in Poznan due to NH and occipital neuralgia on the right side of the head.

## 2. Case description

A 61-year-old woman was admitted to the Department of Neurology due to the constant headache in the right temporal region (Fig. 1) that has lasted for a year before admission, i.e. since March 2011. The pain of pressing character was limited to a circular field with a diameter of 6 cm. In general, the pain was ranked by the patient as 4/10 in visual analogue scale (VAS); however, in the same area the attacks of intensive pain – ranked as VAS 7/10 – did occur three times per day on average, lasting about 10 min without accompanying symptoms. These problems did not interfere with the daily activities – only skin (brush) allodynia caused pain more

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Fig. 1 - Location of the nummular headache.

intense while hair combing and waking from sleep when lying on the right side.

Since September 2011, after the fall of the left half of the face with head bending to the right, the patient has felt pain in the right occipital region, diagnosed as occipital neuralgia (neuralgia occipitalis). The pain was constant with intensity of VAS 2/10, and with bouts of acute pain exacerbations of 5/10 VAS, lasting several minutes, with the radiation to the top of the head and occurring approximately five times a month. In addition, the patient experienced regularly, once a month, the incidents of tension-type headache in the frontal region that regressed spontaneously after a few hours.

Neurological examination revealed allodynia in the right temporal region in the nummular headache and right occipital tenderness, with no other symptoms and signs observed. The pulse of the temporal and occipital arteries was easily detected with no pain of pericranial muscles.

History of the patient was remarkable for angina pectoris, gastritis and duodenitis, as well as left middle ear infection.

The results of laboratory tests (full blood count, erythrocyte sedimentation rate, C-reactive protein, international normalized ratio, activated partial thromboplastin time, D-dimer, sodium, potassium, creatinine, glucose, alanine transaminase, aspartate transaminase, total bilirubin, gamma-glutamyl transpeptidase, alkaline phosphatase, creatine kinase, and urinalysis) were within laboratory limits, with negative results of antinuclear antibodies (ANA) and RPR reaction.

Brain magnetic resonance imaging (MRI) – in PD, T2-weighted and FLAIR Images – revealed few, small hyperintense foci with maximal dimension up to 3 mm, of ischaemic character, scattered on the border of cortical–subcortical region of both frontal lobes.

MRI-angiography of the head showed no vascular defects and in cranial X-ray the thickening of the inner plates of the frontal bone was diagnosed. Radiograms of the cervical vertebral column revealed moderate reduction of cervical lordosis, reduced amount of the vertebral bodies C5 and C6, and degenerative changes of the vertebral bodies from C4 through C6. Doppler study of carotid and vertebral arteries showed an anatomical variant – the right vertebral artery bypasses transverse process of C6 vertebra and enters the C5 vertebra hole, with no other changes observed.

The current therapy based on analgesics (ketoprofen, diclofenac, ibuprofen and paracetamol) and drugs commonly used for neuropathy (carbamazepine, amitriptyline, gabapentin) did not affect the perceived pain of the head. During the stay at the department, a two-day treatment at a dose of 150 mg indomethacin daily was applied without effect. Then bupivacaine was injected into the area of right great occipital nerve with the resolution of occipital neuralgia for a period of three months. Gabapentin treatment at a dose of  $3\times 300\,\mathrm{mg}$  was applied from April 2011 to June 2012 without any pain relief.

Since July 2012, the patient has been taking omeprazole and the analgesics temporarily only.

In mid-January 2013, bupivacaine injection was re-applied around the right great occipital nerve giving a substantial relief of the pain for three months. The treatment with mianserin at an initial dose of 10 mg once daily at night, titrated every week by 10 mg, was started. After six weeks, no pain reduction was noticed in the area of NH. Because of a sleep disorder (excessive daytime sleepiness with frequent arousals during the night), and non-vertiginous dizziness with mianserin dose of 60 mg daily, it was decided to stop the treatment.

Nummular headache and occipital neuralgia occurred unilaterally, but the painful areas did not overlap. The applied oral treatment did not affect both types of pain, only injections to suboccipital neuralgia resulted in resolution of disorders for three months. In the patient's opinion nummular headache does not make daily activities difficult, and the only problem is the skin (brush) allodynia in the painful area.

#### 3. Discussion

Nummular headache is rarely described primary headache occurring more frequently in women, usually in the fourth and fifth decade of life (range 4–79 years) [1,3].

The pathogenesis has not been definitively determined, although the entity is widely recognized as epicrania, i.e. the pain that comes from the inner or outer cranial and scalp layers, along with vessels and nerves [3,4]. Periosteum is sensitive and the shifting of inner layers by the dura seems to cause the main problem of the disorder [1]. Due to the location, NH was classified to neuralgia of the trigeminal nerve ending branches [2]. Furthermore, lower pain threshold in the affected area of dysestesia triggered by touch [5], cases with trophic changes of the skin such as hair loss, topical warming, flushing [7] and casuistic reports of multifocal presence of pain [8,9] all seem to advocate peripheral origin of the pain. The other mechanism of NH appearance might be based on central sensitization of pain that is justified by the lack of effectiveness of the local lidocaine block [10,11] and the case of pain relief after surgical removal of tentorial meningioma [12].

The differential diagnosis of NH requires exclusion of somatic and psychogenic diseases as well as primary headaches including chronic migraine, chronic tension-type headache, new daily headache and hemicrania continua [13]. Nummular headache rarely occurs with other idiopathic headaches and has, in general, an independent course [4,13]. Among other neuralgias, it is distinguished with clearly restricted painful area without radiation [1]. In addition,

patients with NH did not reveal increased sensitivity of pericranial tenderness which may be clinically useful in the differentiation from tension-type headache or migraine [14].

The secondary nature of NH may be due to local skin lesions (fusiform aneurysm of the superficial temporal artery branch), damage to the bones of the cranial metastases, osteomyelitis, Paget disease, multiple myeloma [1], or intracranial changes, i. e. meningioma [12] and arachnoid cyst [15]. Basically, NH is not associated with head injury or anxiety disorders and depression [1,6]. The increased incidence of Sjögren syndrome among subjects with NH is probably associated with neuropathy of sensory fibres of trigeminal nerve [16]. It is believed that the distal damage to the branch of the trigeminal nerve by the varicella-zoster virus (VZV) herpetic changes may also be the cause of pain [17].

Except from rare cases of secondary NH, the pathogenesis is not uniquely recognized that results in the multidirectional mode of treatment including drugs, applied in neuropathic pain [1,10,18]. Currently, it is recommended to apply non-steroidal anti-inflammatory for mild pain [1]. In the pain of moderate intensity, gabapentin at a dose of 300–900 mg daily [19] or tricyclic antidepressants [3] are sometimes effective. In cases resistant to the above treatment, an injection of botulinum toxin type A [20,21] or transcutaneous electrical nerve stimulation [22] should be considered. It has been shown that topical anaesthetics do not improve the pain status [10,11]. In vast majority of the cases, there is a resistance to any treatment. In these circumstances, the exclusion of symptomatic nature of the pain is both advisable and sufficient attitude.

The presented description of the history of the patient confirms the independence of the NH and occipital neuralgia. The applied treatment had no effect on pain, while confirming the effectiveness of bupivacaine injections for occipital neuralgia. Recent reports indicate that NH is usually mild pain, resistant to treatment, which may resolve spontaneously in a different period of time, lasting for up to a decade. In most cases, the physical examination did not show any deviation with no family history of the disorder. Due to the ten-year NH recognition time, further, long-term observation of patients is required to seek more effective methods of treatment and updated criteria for diagnosis.

#### Conflict of interest

None declared.

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## **Ethics**

The work described in this article has been carried out in accordance with The Code of Ethics of the World Medical Association (Declaration of Helsinki) for experiments involving

humans; Uniform Requirements for manuscripts submitted to Biomedical journals.

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