

# Temporary resolution of hemicrania continua following ipsilateral ear piercing

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## ABSTRACT

**Background** Hemicrania continua is an uncommon subtype of trigeminal autonomic cephalgia that exhibits dramatic therapeutic response to indomethacin. Unfortunately, indomethacin is associated with a range of adverse effects, including neuropsychiatric complications, which limits its use in many patients. Although no other effective pharmacologic agents exist, there is emerging evidence for interventional treatments such as occipital nerve and vagus nerve stimulation, which may act by modulating neural activity within the trigeminovascular system.

**Case** We present a 30-year-old woman with long-standing refractory hemicrania continua who suffered adverse effects to indomethacin. She experienced temporary, but near-complete, symptom resolution following piercing of the crus of the ear helix ipsilateral to her headache, whereas contralateral piercing produced no benefit.

**Conclusions** To our knowledge, this case is the first to describe a therapeutic benefit following ear piercing in a patient with trigeminal autonomic cephalgia. We argue that symptom relief was obtained through a similar mechanism to occipital or vagus nerve stimulation.

## INTRODUCTION

Hemicrania continua (HC) is an uncommon but clinically striking headache syndrome that is classified as a trigeminal autonomic cephalgia (TAC).<sup>1</sup> HC is characterised by prolonged unilateral headache accompanied by cranial autonomic phenomena such as lacrimation and ptosis.<sup>2</sup> Crucially, HC is exquisitely sensitive to indomethacin underscoring the importance of timely diagnosis and treatment to minimise the impact on patient quality of life.

Unfortunately, many patients experience adverse effects to indomethacin, yet no established prophylactic therapies exist. In some patients, an improvement with pharmacologic agents, including topiramate and melatonin, has been reported.<sup>3</sup> More recently, interventional approaches have emerged as viable treatment options with occipital nerve stimulation and vagus nerve stimulation demonstrating benefit in small case series.<sup>4 5</sup> These treatment strategies are thought to act by modulating neural activity

within the trigeminovascular system implicated in TAC pathophysiology.<sup>6</sup>

Here, we report a case of long-standing HC in which the patient experienced symptomatic improvement following ipsilateral piercing of the crus of the ear helix: a practice known as ‘Daith’ piercing.

## CASE REPORT

A 30-year-old female patient was referred to a neurology clinic for refractory headache since the age of 16 consisting of a burning sensation over the periorbital region extending posteriorly across the scalp. The pain was exclusively left sided with sharp demarcation at the midline, and which she described as ‘splitting her head down the middle’. Her headache was constant but punctuated by severe exacerbations lasting from days to weeks with an average frequency of four exacerbations per month. There was no light or sound sensitivity, but she reported a ‘droopy’ left eyelid, periorbital swelling and rhinorrhoea during exacerbations. The patient provided written informed consent to publish her case.

The patient’s headaches did not respond to acute therapy with paracetamol, non-steroidal anti-inflammatory drugs (including ibuprofen up to 800 mg daily and celecoxib 400 mg daily) or aspirin 800 mg. She used regular propranolol 20 mg two times per day over many years for migraine prophylaxis, which was ineffective. The headaches profoundly disrupted the patient’s quality of life, and she was unemployed at the time of review. She lived in a regional setting with limited specialist access and had never previously seen a neurologist.

The patient had no other medical history and her only regular medication was propranolol. Her mother suffered from migraine without aura, but no first-degree relatives experienced headaches with similar characteristics to the patient. Cranial nerve, funduscopy and upper and lower limb neurological examinations were normal. An MRI of the brain and angiogram did not



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demonstrate any cerebral or vascular abnormalities. After researching alternative treatment options for migraine, the patient underwent Daith piercing of the left ear 12 months before neurology review. She noticed a rapid and dramatic symptom improvement: her severe exacerbations resolved and her background pain improved to the extent that she did not require analgesia for a 2-month period. Due to this therapeutic response, the patient then elected to undergo Daith piercing of the right ear, which provided no symptom benefit.

Based on the clinical characteristics, the patient was commenced on an up-titrating dose of indomethacin for treatment of HC. At a dose of 50mg three times per day, she experienced complete resolution of headache approximately 30min after her third dose. Medication withdrawal several days later led to a recrudescence of her symptoms. She experimented with her indomethacin regimen and found that maintaining a daily dose of 25–50mg reliably prevented headache recurrence. After approximately 1month of therapy, she developed light headedness, anxiety and a distressing sensation of depersonalisation that resolved after ceasing indomethacin. She trialled topiramate 50mg daily, which was self-ceased after 2 months due to lack of perceived benefit, and then underwent left-sided greater occipital nerve (GON) block. She reported a partial response to GON block over the following month with persistent background pain but only two mild exacerbations lasting under 24 hours, after which her headaches returned to their previous frequency and severity.

## DISCUSSION

HC is an uncommon subtype of TAC that is often incorrectly diagnosed and treated. HC is under-recognised by clinicians, including neurologists, and symptoms may be confused for unrelated conditions such as dental or temporomandibular disorders.<sup>7</sup> TACs are associated with high rates of disability, occupational absenteeism and reduced social engagement.<sup>8</sup>

Due to the remarkable therapeutic response of HC to indomethacin, a misdiagnosis has profound consequence on patient quality of life. Unfortunately, indomethacin is poorly tolerated in over 30% of patients and associated with a range of adverse effects.<sup>9</sup> Gastrointestinal disturbance is most commonly recognised but neuropsychiatric adverse effects, evident in this case report, occur in up to 3% of patients.<sup>9</sup> Therefore, there exists an urgent need for novel treatment strategies.

The pathophysiology of TACs is complex. Pain may arise through activation of the trigeminovascular system, which consists of nociceptive fibres terminating in the trigeminocervical complex, and ascending pathways to thalamic nuclei, which project to the pain neuromatrix.<sup>10</sup> Cranial autonomic symptoms are thought to arise from a reflex arc involving the pontine superior salivatory nucleus, and dysregulation of these pathways may be driven by the posterior hypothalamus.<sup>11</sup> This framework provides a

rationale for the use of interventional therapies in HC.<sup>12</sup> Symptomatic improvement has been observed after sphenopalatine ganglion blockade and non-invasive vagus nerve stimulation, perhaps via modulation of the trigeminal–autonomic reflex.<sup>5 6 13</sup> In a cross-over study of six patients receiving occipital nerve stimulation, a branch of the C2 nerve root, five patients reported significant symptom improvement.<sup>4</sup> Here, C2 stimulation may modulate ascending trigeminal pain pathways due to afferent inputs converging within the trigeminocervical complex, perhaps through an inhibitory gate control mechanism.<sup>10</sup>

Daith piercing has been reported to provide symptom benefit in migraine, but, to the best of our knowledge, this is the first case to document clinical improvement in a TAC.<sup>14</sup> Although placebo effects are common following interventional treatment of headache, we feel this is unlikely to account for the response in this patient. Not only did contralateral piercing provided no benefit, but the patient experienced only mild improvement with GON block, which would also be expected to carry a high likelihood of placebo effect.<sup>15</sup> The crus of the ear helix receives sensory supply from the vagus nerve and C2/C3 nerve roots via the auricular nerve, which project to the superior salivatory nucleus and spinal trigeminocervical complex, respectively. We hypothesise that symptomatic benefit was obtained through modulation of ascending pain pathways within the trigeminocervical complex, through either overlapping cervical sensory representation or via the trigeminal–autonomic reflex.<sup>6 12</sup> It is interesting to note that the response to GON blockade was less pronounced than ear piercing in this patient. This could reflect differences in either the nature of the peripheral stimulation, or the preferential contribution of vagal afferents to the analgesic effect.

## CONCLUSION

This case is the first to report a symptomatic benefit with Daith ear piercing in a patient with HC, and we propose a similar therapeutic mechanism to occipital nerve or vagus nerve stimulation. Although this case highlights the potential therapeutic mechanisms of HC, Daith piercing is not a recommended treatment modality as it is unlikely to provide persisting benefit and is associated with a risk of infection. This case also demonstrates the range of adverse effects associated with indomethacin, including neuropsychiatric complications, which are often under-recognised.

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