# Conjugal Hemicrania Continua – A Chance Occurrence or a New Entity?

Dear Editor,

Hemicrania continua (HC) represents approximately 2% of all headaches in clinical settings and is probably the second most common trigeminal autonomic cephalalgias (TACs).[1,2] There are two reports of familial HC in the literature.[3,4] We report a non-consanguineous couple, married for nearly 15 years, who developed HC one after the other (conjugal HC).

## Case 1 (Husband)

A 43-year-old man presented with a 7-year history of continuous left-sided headaches with superimposed exacerbations. He never experienced remission. The background pain was mild-to-moderate, non-pulsatile, and felt on the left side of the head, including the orbit. The exacerbations, throbbing in quality, were mainly localized in the orbital and supraorbital regions. The intensity of exacerbations varied from 8/10 to 10/10 on the visual analog scale (VAS). The frequency of exacerbations ranged 2–3 per day to once per week. The superimposed attacks used to last between 30 min and 12 h. The exacerbations were associated with ipsilateral conjunctival injection, lacrimation, and rhinorrhea. During exacerbations, the patient would pace, rock in a chair, or hold the aching part. Exacerbations were sometimes accompanied by nausea, vomiting, and phonophobia. Physical examination was normal. Investigations, including a complete blood count, ESR, thyroid profiles, liver function tests, kidney function tests, antinuclear antibody, an MRI of the brain, an MRA for both intracranial and extracranial vessels, CSF analysis, and serological testing for HIV and HBV, did not reveal any abnormalities.

He received a number of drugs (including antidepressants, valproate, propranolol, lithium, ibuprofen, and paracetamol). However, none of these treatments had any notable effects on headache. We suspected a possibility of HC, and indomethacin was started. There was a marked response at the dose of 50 mg tid. The background headache and exacerbations subsided completely within 72 h. We followed the patient for more than 18 months. Skipping the dosage of indomethacin always resulted in a reappearance of the symptoms. The reinstitution of indomethacin always provided a complete response.

## Case 2 (Wife)

A 37-year-old lady, patient 1's wife, described a 2-year history of right-sided headaches that were similar to her husband's. The headache began roughly 5 years after the husband's headaches began. There was a continuous, mild-to-moderate, dull ache on the right side of head. The headache has been constant since its onset. There were 1–2 exacerbations each day over background headaches, lasting for 30 min to 12 h. Most exacerbations were associated with ipsilateral lacrimation, conjunctival injection, and rhinorrhea. There was agitation and pacing activities during severe attacks. General and neurological examinations were normal. She had been subjected to a number of investigations, all of which were normal. It included a complete blood count, ESR, plasma glucose, thyroid profiles, liver function tests, kidney function tests, antinuclear antibody, MRI of the brain, MRA for both intracranial and extracranial vessels, and serological testing for HIV and HBV.

Prior treatments with propranolol, valproate, topiramate, amitriptyline, naprosyn, ibuprofen, and paracetamol were without benefit. Treatment with 25 mg tid indomethacin produced an immediate and complete response. In the 18-month follow-up, she responded well to indomethacin. Skipping indomethacin (planned or due to poor compliance) always resulted in headache recurrence.

Both patients fulfilled the ICHD-3 criteria for HC.[1] Immediate reappearance of headaches on discontinuation or skipping of indomethacin is a strong feature of HC.[5] There are two documented cases of HC in the same family. Weatheral and Bahara reported the first case of familial HC.[3] They reported HC in a mother and daughter at the same time of life. Huang and Newman recently reported another case of familial HC in a mother and daughter.[4] Any disease discovered in a married couple is referred to as conjugal disease. Thus, we can label it as conjugal HC. To the best of our knowledge, conjugal HC has not been reported in the literature before.

Any disease that develops in families frequently has a hereditary cause. However, conjugal cases do not favor genetic etiology. There are at least four possible explanations for the HC in this couple: (1) transmission by sexual or close contact, (2) shared environments, (3) shared pain due to empathy and synesthesia, and (4) incidental HC.

Secondary causes of HC include leprosy and HIV.[2] Both may spread through close or sexual contact. However, we did not get any evidence of either leprosy or HIV infection. Therefore, the possibility of a contagious agent was less likely. Any disease in family members raises the likelihood of a shared environmental factor. The main point in favor of shared environmental factors is that disease would start in all members at the same time.[6] The symptoms in the wife started roughly 5 years after the onset of HC in her husband. This point refutes the possibility of shared environmental factors in the development of conjugal HC.

Feeling pain while witnessing someone else's pain is not rare. Such pains are described during pregnancy, when a

male partner experiences the same discomfort or pain as his pregnant partner (couvade syndrome). Empathy has been suggested for such emotional pain. Bradshaw and Mattingley[7] reported the first clinical case of feeling mirrored pain when observing another person in pain. Several studies were published in which normal controls developed pain after witnessing pain in others. Giummarra *et al*.[8] described eight patients who experienced phantom pain as a result of observing, thinking about, or inferring that another person was in pain. Prakash *et al*.[9] reported two patients with HC whose background continuous pain worsened or reappeared when they thought about their pain. Such a phenomenon is called "mirror pain synesthesia" or "synesthesia for pain."[8,10] It could be one of the mechanisms for the development of HC in the wife. HC is a very disabling headache disorder as pain persists throughout the day. Severe exacerbations that could persist for many days could make the situation even worse. It might have caused an HC-like headache in the wife by inducing empathy or mirror pain synesthesia.

The fourth and most likely possibility is that each patient is sporadic, and conjugal HC in this couple is a chance occurrence. This is just a single-case port. Therefore, no definitive conclusion can be drawn here. We need more cases to determine the causes behind conjugal HC.

## Financial support and sponsorship

Nil.

## Conflicts of interest

There are no conflicts of interest.

**Sanjay Prakash, Anurag S. Prakash**[1]

Department of Neurology, Smt. B. K. Shah Medical Institute and Research Centre, Sumandeep Vidyapeeth, Piparia, Waghodia, Vadodara Gujarat, [1]Department of Medicine, Parul Institute of Medical Sciences and Research Centre, Parul University Waghodia, Vadodara, Gujarat, India

**Address for correspondence:** Dr. Sanjay Prakash, Department of Neurology, Smt. B. K. Shah Medical Institute and Research Centre, Piperia, Waghodia, Vadodara, Gujarat - 391 760, India.
E-mail: drprakashs@yahoo.co.in

## References

1. Headache Classification Committee of the International Headache Society (IHS). The International Classification of Headache Disorders, 3rd edition. Cephalalgia 2018;38:1–211.
2. Prakash S, Patel P. Hemicrania continua: Clinical review, diagnosis and management. J Pain Res 2017;10:1493-509.
3. Weatherall MW, Bahra A. Familial hemicrania continua. Cephalalgia 2011;31:245–9.
4. Huang H, Newman LC. Hemicrania continua in a family: A report of two cases. Headache 2021;61:1132-5.
5. Prakash S, Rathore C, Rana K, Joshi H, Patel J, Rawat KS, *et al*. A long-term prospective observational study in 31 patients with hemicrania continua. Cephalalgia Rep 2019;2:2515816318824694. doi: 10.1177/2515816318824694.
6. Rajput AH, Rajput A. Parkinsonism in both members of married couples. J Neurol Neuromedicine 2016;1:7-17.
7. Bradshaw JL, Mattingley JB. Allodynia: A sensory analogue of motor mirror neurons in a hyperaesthetic patient reporting instantaneous discomfort to another's perceived sudden minor injury? J Neurol Neurosurg Psychiatry 2001;70:135–40.
8. Giummarra MJ, Gibson SJ, Georgiou-Karistianis N, Bradshaw JL. Central mechanisms in phantom limb perception: The past, present and future. Brain Res Rev 2007;54:219-32.
9. Prakash S, Shah ND. Delayed response of indomethacin in patients with hemicrania continua: Real or phantom headache? Cephalalgia 2010;30:375–9.
10. Fitzgibbon BM, Giummarra MJ, Georgiou-Karistianis N, Enticott PG, Bradshaw JL. Shared pain: From empathy to synaesthesia. Neurosci Biobehav Rev 2010;34:500-12.

**Submitted:** 10-Sep-2023 **Revised:** 29-Sep-2023 **Accepted:** 15-Oct-2023
**Published:** 29-Nov-2023



**DOI:** 10.4103/aian.aian_805_23