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Case report

Continuous hemicrania responsive to early testing with indomethacin: case report

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Abstract

Introduction
Hemicrania continua (HC) is a primary headache characterized by persistent unilateral pain of moderate intensity, associated with autonomic signs and responsive to indomethacin. Early diagnosis is important because it is a highly disabling condition, and appropriate and immediate treatment can lead to symptom remission.

Case Report
A 63-year-old woman presented with a 5-day history of intense and continuous left temporal headache without remission, associated with conjunctival hyperemia, tearing, and semiptosis. Pain intensity was rated 9/10, with no response to common analgesics. Neurological examination confirmed ipsilateral autonomic signs, without other focal deficits. Brain MRI with contrast showed no abnormalities. Although she did not meet the minimum duration criterion for HC, early treatment with indomethacin (50 mg twice daily) was initiated, resulting in significant clinical improvement within six days, with remission of pain and autonomic signs.

Discussion
The diagnosis of hemicrania continua is based on continuous unilateral headache, presence of ipsilateral autonomic symptoms, and complete response to indomethacin. However, many patients may not meet the duration criterion at initial presentation.

Conclusion
Early therapeutic evaluation with indomethacin should be considered in patients presenting with continuous unilateral headache and autonomic symptoms suggestive of hemicrania continua, even when the minimum duration criterion has not yet been reached. Early diagnosis may significantly reduce disease impact and improve clinical outcomes.

Keywords:
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Introduction

Trigeminal autonomic cephalalgias (TACs) are rare disorders characterized by unilateral attacks of throbbing, severe pain, usually in the orbital region, associated with unilateral cranial autonomic changes such as tearing, conjunctival hyperemia, and eyelid ptosis. Four subtypes of trigeminal autonomic headaches (TACs) are defined: cluster headache (CH), hemicrania continua (HC), paroxysmal hemicrania (PH), and short-lasting unilateral neuralgiform headache attacks (SUNCT and SUNA) (1).

HC presents with continuous headache of moderate intensity, unilateral, with episodes of periodic exacerbations. Most patients experience pain on only one side of the skull; however, some may have pain attacks that alternate sides (2,3). The duration and frequency of exacerbations are highly variable and are usually associated with ipsilateral autonomic disturbances in the painful region (5). A large number of patients have migrainous features during the worsening phases (5). Although it is not as common as migraine and tension-type headache, HC may not be as uncommon as proposed, since it is still a neglected syndrome (1–7).

Brainstem connections between the trigeminal system and the parasympathetic nervous system help explain how a trigeminal-based headache presents with autonomic symptomatology (8). Pathophysiological mechanisms similar to other TACs have been proposed for HC, and involvement of the trigeminal-autonomic reflex, central or peripheral, appears to be important (9). Furthermore, functional neuroimaging studies show notable activation of the posterior hypothalamic region during pain exacerbations, and there is no evidence of an associated genetic component, which emphasizes its inclusion in this group (10).

Brain Magnetic Resonance Imaging (MRI) is recommended, with study of the pituitary region and cavernous sinus. In cases of TACs refractory to three treatments, additional MR angiography of the carotid and vertebral arteries is requested in order to identify possible symptomatic causes (8).

Patients with HC show complete and dramatic response to therapeutic doses of indomethacin (1–3,6), and this response is a mandatory diagnostic criterion, although there are exceptions in the literature (2).

Thus, this case report aims to analyze the clinical presentation of a 63-year-old patient with a history of headache in the left temporal region, in order to stimulate discussion about the importance of early diagnosis and testing for Hemicrania Continua, even when the patient does not fulfill the clinical criteria regarding pain duration, but already presents a suggestive clinical picture.

Case report

A 63-year-old female patient, previously obese and hypertensive, using captopril 25 mg twice daily, with no prior history of headache, presented with left temporal pain without lateralization change, severe intensity (reported as 9 on a 0–10 scale), pulsatile, associated with restlessness, for 10 days, without remission periods, only with fluctuations in intensity, and no identified aggravating or relieving factors.

Due to these complaints, she used common analgesics such as dipyrone 2 g/day for two days without medical guidance, with no improvement. She sought emergency care twice within five days and received intravenous dipyrone (500 mg/mL) at both visits, without symptom relief.

Physical examination revealed left semiptosis, conjunctival hyperemia, and tearing, with no extraocular motility abnormalities. Tactile and pain sensations were preserved in the trigeminal nerve territory. Other cranial nerves were normal. Pupils were isochoric and reactive to light. Fundoscopic examination was normal. No other relevant neurological findings were observed.

Contrast-enhanced MRI showed no abnormalities. Due to the possibility of trigeminal autonomic cephalalgias, a therapeutic trial with indomethacin 50 mg every 12 hours was initiated, with gastric protection (omeprazole 20 mg). After 48 hours, there was significant regression of symptoms, with complete remission after approximately five days.

Follow-up was scheduled eight days later. The patient returned seven days after the first consultation, with recurrence of pain and autonomic signs. Indomethacin was maintained at the same dose for eight months, with serial consultations initially monthly, then every two months. Afterwards, dose tapering from 100 mg/day was attempted, reducing 25% each month. The patient remained symptom-free six months after complete withdrawal.

Discussion

The International Classification of Headache Disorders (ICHD-II) defines HC as a strictly unilateral persistent headache responsive to indomethacin (1). Early diagnosis is important because it is a disabling condition, allowing remission of symptoms (2,5,6). The diagnosis of HC includes three criteria: Presence of pain for at least 3 months, with exacerbations (1,6); Presence of two or more signs or symptoms ipsilateral to the pain (conjunctival hyperemia, tearing, nasal congestion, rhinorrhea, eyelid edema, facial and frontal sweating, facial flushing, sensation of ear fullness, miosis, ptosis, restlessness, agitation, or worsening of pain with movement) (1,2,6); Absolute response to therapeutic doses of indomethacin.

It is essential to consider HC as a chronic headache and to administer the INDOTEST (diagnostic test to detect indomethacin-responsive headaches) early (1) in cases of chronic unilateral headache, which may anticipate the identification of HC cases, along with a headache diary, using intramuscular indomethacin at a dose of 50 to 100 mg (6).

Although the patient had only 5 days of headache, she had other criteria that led to clinical suspicion of hemicrania. Therefore, early testing with Indomethacin was chosen to avoid waiting 3 months to establish the diagnosis (1,6), since it is a debilitating headache refractory to treatment with common analgesics.

Justified by the symptomatology and the varied expression of TACs, MRI was also requested to evaluate possible other disorders and to attempt to correlate the clinical presentation with possible findings (8). The absence of relevant findings reinforces the idea of the relationship between brainstem connections of the trigeminal system and the parasympathetic nervous system, justifying trigeminal headache with autonomic symptomatology (8,10).

Treatment, guided by retrospective case analysis, expert opinion, and clinical experience, ends up being empirical. Even with unknown mechanisms of response, indomethacin is the drug of choice for HC and, therefore, therapeutic testing is recommended in patients with chronic, continuous, unilateral headache (6). After establishing a safe and effective dose for the patient, dose reduction should be performed to determine the lowest dose capable of stopping the pain, along with the use of gastric protectors to prevent gastrointestinal side effects (6).

Conclusion

It is noted, therefore, that most patients do not meet the time criterion established for confirming the diagnosis of HC at the first consultation and, therefore, early testing with Indomethacin should be considered to assess response in patients presenting with daily, unilateral headache of moderate intensity, with exacerbations and the presence of at least two of the following signs: tearing, flushing, facial sweating, ptosis and/or miosis, and nasal congestion, considering the negative impact of continuous pain on the patient's life.

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