

Case Report

Remitting Form of Hemicrania Continua With Seasonal Pattern

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Hemicrania continua is a primary headache syndrome characterized by a continuous, unilateral headache that is completely responsive to indomethacin. Hemicrania continua exists in continuous and remitting forms. Ten cases of the remitting form have been reported, none of which have had a seasonal pattern. We report a patient with remitting hemicrania continua with a clear seasonal predilection.

Key words: hemicrania continua, melatonin, indomethacin, seasonal variation

Abbreviation: HC hemicrania continua

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Hemicrania continua (HC) is a primary headache syndrome characterized by a continuous, unilateral headache that is completely responsive to indomethacin.

Hemicrania continua exists in continuous and remitting forms. In 1991, Bordini et al¹ proposed three subtypes of HC: the remitting form and the continuous form, which is subdivided into a form which is continuous from onset and a form which evolves from the remitting form (transformed).

In the remitting form of HC, distinct headache phases last weeks to months with prolonged pain-free remissions.² The first case of HC with a remitting course was reported by Sjaastad and Tjorstad³ in 1987, and was the third patient with HC reported in the literature. There was a gradual transformation from the remitting to the continuous stage, similar to the transformation of episodic into chronic migraine.⁴

The remitting type of HC is less common than the continuous form; the continuous: remitting ratio

is 8:1.⁵ Ten cases of remitting HC have been reported, none of which have had a seasonal pattern. We report a patient with remitting HC with a clear seasonal predilection.

CASE HISTORY

A 52-year-old white woman presented with a 19-year history of strictly unilateral left-sided headaches located in the ocular, frontal, and temporal regions. The pain was usually moderately severe, but the patient experienced intermittent severe exacerbations. The pain was continuous and accompanied by episodic symptoms of ipsilateral eye tearing, conjunctival injection, eyelid edema, and a feeling of "sand in the left eye." She also complained of marked photophobia, phonophobia, and nausea.

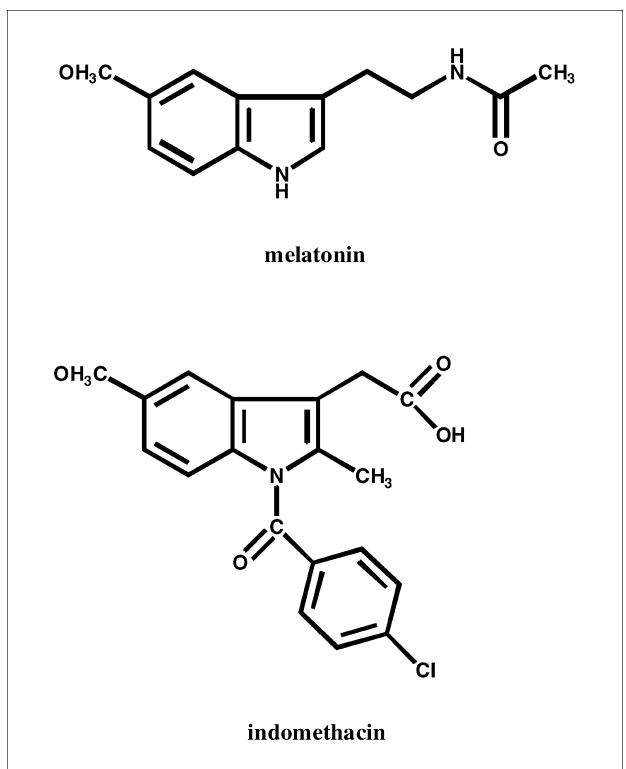
Since their onset, her headaches had presented in a seasonal pattern, starting in May and ending in July. She experienced the headaches at the same time every year, being completely headache-free from July to the next May. However, for the last 3 years, her headache periods were lasting longer and were now present until August.

Her past medical history disclosed a mild head trauma at the age of 9 and a recent diagnosis of hepatitis C. Neurologic examination was normal except for mild neck tenderness. Occipitonuccal trigger point

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Chemical structure of melatonin and indomethacin. Note the consistent methoxy indole nucleus.

palpation did not reproduce her pain. Magnetic resonance imaging of the head was normal. The patient's pain was completely responsive to indomethacin, 75 mg per day.

COMMENTS

Seasonal variability is reported in several neurological disorders including stroke,⁶ multiple sclerosis,⁷ Bell palsy,⁸ and seasonal affective disorder.⁹ In several headache disorders, a seasonal pattern is also present. It is a hallmark in cluster headaches¹⁰ and has been described in migraine.^{11,12}

A circannual (seasonal) variability has not previously been described in a patient with remitting HC. However, patients may not report this feature if they are not specifically questioned about it. All patients with remitting HC may have a seasonal predilection, which could represent a specific HC variant or a transition from HC to cluster headache.

The distinctive periodicity of cluster headache has strongly implicated a disturbance in the hypothalamus. The pathophysiology of HC is uncertain, but

our patient's remarkable circannual consistency supports hypothalamic involvement.

Melatonin is the most sensitive surrogate marker of circadian function in humans, and its rhythmic secretion is under the control of the hypothalamus.¹³ Melatonin secretion is markedly influenced by seasonal changes in the light-dark cycle and plays a role in the effects of season on animal physiology and behavior.¹⁴

Decreased melatonin levels have been reported in patients with cluster headache,^{15,16} and melatonin has been used for the prophylactic treatment of this type of headache.¹⁷ Melatonin has been shown to play a role in cluster headache circannual variation.¹⁸ Melatonin may have a role in the seasonal variation of remitting HC. Indomethacin and melatonin have a similar structural formula¹⁹ (Figure), so indomethacin responsiveness could be related to melatonin's biological activity. Further studies regarding the role of melatonin in HC are needed. Melatonin may have a therapeutic role in HC.

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