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LETTER TO THE EDITOR

Paroxysmal body pain without headache

Kubra Mehel Metin[1], Fatma Aysen Eren[2], Levent Ertugrul Inan[1]

[1]Ankara Training and Research Hospital, Department of Neurology, Ankara, Turkiye
[2]Kanuni Sultan Suleyman Training and Research Hospital, Department of Neurology, Istanbul, Turkiye

Dear Editor,

Recurrent pain in the body and extremities that improves with migraine-specific treatment in patients with migraine headaches may be associated with migraine (1-4). The prevalence of recurrent limb pain in children has been reported to be 2.6%, suggesting a close clinical and epidemiological relationship, as well as a shared pathogenesis, between recurrent extremity pain and childhood migraine (3). The literature also describes cases of extracranial (i.e., outside the head) pain occurring in association with migraine headaches (1, 5-7). The temporal relationship between headache and limb pain is defined as episodes of pain in one or more limbs occurring immediately before or after headache onset, during the headache, or shortly thereafter (5-7). We present the case of a woman with paroxysmal (sudden and transient) body pain without accompanying headache that responded positively to flunarizine treatment. The clinical features resemble migraine-associated extracranial pain described in the literature, suggesting that this presentation may represent a case of migrainous corpalgia.

A 41-year-old woman presented to the neurology outpatient clinic with complaints of body pain that had begun five years earlier and had increased in both intensity and frequency over the past three years. Prior to the onset of pain, she experienced symptoms including weakness, speech difficulty, numbness on the right posterior side of the head, and tingling in the right leg. These symptoms were followed by pain throughout the body, sparing the head. She described the pain as a pressure-like sensation enveloping her body, with greater intensity in the right arm and leg. Initially, the attacks occurred once every three to four months and lasted two to three days. By the time of presentation, however, they were occurring weekly and lasting up to five days. Pain intensity was rated as 8 on the Visual Analog Scale (VAS). The pain did not have throbbing, stabbing, burning, or dull characteristics, although it was occasionally accompanied by tingling sensations. After two to three days of continuous pain, severe exacerbations may recur several times during subsequent periods of weakness. During these episodes, the patient reports marked fatigue and an increased need for sleep. Although pain intensity gradually decreases over the course of each attack, she experiences significant difficulty with movement due to severe pain and requires bed rest. Following the attacks, she describes a generalized numbness sensation affecting the entire body, particularly the right arm and back, while sparing the head. The attacks are not accompanied by dizziness, headache, nausea, vomiting, or hypersensitivity to light, sound, or odors. There is no association with menstruation cycles, nor are there symptoms of morning weakness or stiffness. The attacks are stereotypical in nature, differing only in intensity. They may occur at various times throughout the day, but not during sleep. There is no evidence of allodynia (pain elicited by normally non-painful stimuli), and no triggering factors have been reported. The patient has no known chronic medical conditions and was not receiving any regular treatment.

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Correspondence: Kubra Mehel Metin, Ankara Training and Research Hospital, Department of Neurology, Ankara, Turkiye
E-mail: kubramehel@gmail.com

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The patient's neurological examination was normal, as were her laboratory tests and cranial magnetic resonance imaging (MRI) results. Cervical MRI showed minimal bulging at the C3-4 and C4-5 levels but was otherwise normal. Electroencephalography (EEG) and electromyography (EMG) results were normal, and bilateral median and tibial nerve SEP (somatosensory evoked potentials) studies were also considered normal. The patient was evaluated by the departments of rheumatology, physical therapy and rehabilitation (PTR), and psychiatry. The rheumatology department recommended steroid therapy for three months. Psychiatry prescribed duloxetine 60 mg, which resulted in approximately a 60% reduction in the frequency, duration, and intensity of the pain for eight months. However, the patient experienced no further benefit after this period. Subsequently, psychotherapy was recommended, but the patient did not find monthly or bimonthly sessions beneficial. A return to duloxetine treatment did not yield improvement. Therefore, flunarizine 5 mg daily was initiated, leading to an 80% reduction in pain intensity and a 50% reduction in duration. When the dose was increased to 10 mg daily, evaluation after seven months showed that the duration of attacks had decreased to 1-2 days, indicating a good response to treatment. The patient has been using flunarizine for five years. The dose of flunarizine was increased or decreased according to the frequency of attacks.

Migraine can be associated with limb pain in both adults and children. Extremity pain should be considered one of the periodic syndromes associated with migraine in childhood and part of the migraine spectrum in adulthood (8). The estimated prevalence of extremity pain among adults with migraine ranges between 2% and 4.4% (5, 6, 8). The literature has documented cases of body pain associated with migraine (1, 5-7). Cuadrado et al. (1) described three female patients aged 30 to 41 years with migraine with or without aura. In these cases, body pain occurred in various regions, including the face, back, and upper and lower extremities, and its onset could precede, accompany, or follow the migraine headache. The pain was described as heavy, pressing, burning, constricting, or throbbing in nature, and allodynia could accompany the pain in the affected area. The duration of body pain was irregular, often beginning in one area and spreading. Pain intensity was generally mild to moderate (rated 2–4 out of 10), although in one patient it reached 7 (1). Guiloff and Fruns reported 22 patients aged 19 to 79 years with diagnoses of migraine and/or cluster headache. In these patients, body pain occurred in the face, neck, shoulders, chest, abdomen, and upper or lower extremities. The pain could occur concurrently with the headache, follow it, or occasionally arise in isolation. The nature of the pain varied and was described as throbbing, dull, shooting, constricting, burning, tender, or prickling. The frequency and duration of attacks also varied (5).

Raudino identified two male and eight female patients aged 11 to 57 years with migraine with or without aura. In these cases, body pains occurred in the upper and lower extremities and could precede, accompany, or follow the headache (6). Prakash et al. (7) reported six cases of extremity pain associated with migraine headache. Attacks could occur before, during, after, or independently of the migraine headache. The duration of pain ranged from 1 to 72 hours, and limb pain was described as tearing, burning, or pressing in nature. Triggering factors included exposure to sunlight, weather changes, stress, fatigue, hunger, and sleep disturbances (7). A case of familial migraine with limb pain affecting eight family members, consistent with a dominant inheritance pattern, has also been reported in the literature (8). The characteristics of body pain in our case align with previously reported features of extracranial pain associated with migraine. The patient described the pain as a pressure-like sensation, sometimes accompanied by tingling. Interestingly, she did not report migraine headaches or other recurrent headache types commonly described in the literature.

In another study, levels of substance P (SP) and calcitonin gene-related peptide (CGRP) were measured in three children with extremity pain, both during the attacks and three days after resolution. SP levels increased during pain episodes, whereas CGRP levels were elevated both during and after the attacks. Thermographic assessments demonstrated a decrease in temperature in the affected area at pain onset, followed by an increase after pain resolution (4). The rise in plasma CGRP levels during extremity pain attacks suggests a pathophysiological process similar to that observed in migraine. These findings support the hypothesis that, analogous to the trigeminovascular theory of migraine, abnormal release of CGRP and SP within the vascular walls of the limbs could play a significant role in the pathophysiology of limb pain (4, 9, 10). The presence of cephalic and extracephalic allodynia, indicative of increased sensory sensitivity during migraine attacks, and reports of allodynia in areas of body pain support a potential relationship between spontaneous body pain and migraine. This suggests that spontaneous body pain associated with migraine may result from central sensitization. Migraine has been proposed to represent not merely a headache disorder

but a broader disorder of nociceptive processing (1). However, reports of extremity pain without accompanying allodynia indicate that central sensitization may not be the sole mechanism of underlying limb pain in individuals with migraine. The notion that extremity pain cannot be explained solely by activation of the trigeminovascular system suggests clinical heterogeneity. This observation raises the possibility that additional mechanisms, such as vasospasm, central sensitization, complex regional pain syndrome-like mechanisms, and ion channel dysfunction, may play a role in the etiology of extremity pain (7).

In the management of extracranial pain associated with migraine attacks, analgesics and acute migraine-specific treatments can be effective in reducing extremity pain, suggesting that migraine preventive therapies may also be recommended (1, 5, 7, 8).

The present case is noteworthy for demonstrating a significant reduction in the duration, frequency, and intensity of paroxysmal body pain following treatment with flunarizine, a medication used for migraine prophylaxis. This response supports the hypothesis that body pain may represent a component of the migraine spectrum. The literature indicates that paroxysmal body pain may occur before, during, after, or independently of migraine headache; in some isolated cases, patients have ultimately been diagnosed with migraine. However, no headache complaints were reported in the present case, reinforcing the possibility that extremity pain may occur without headache.

These findings support the view that migraine is not limited to headache but encompasses a wide spectrum of symptoms, including atypical presentations in certain patients. Therefore, individualized diagnostic and therapeutic approaches are essential, with treatment strategies tailored to the patient's clinical profile. Recognizing extracranial pain manifestations in migraine management underscores the importance of a holistic approach that may improve patients' quality of life.

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