

**Case Report**
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# Episodic Unilateral Limb Pain and Migraine—A Case Report, Hitherto an Unnamed Entity!

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**ABSTRACT**

**Background:** Migraine limb pain is a very under-recognized manifestation of migraine in which episodic pain happens in the peripheral limbs. Despite its description for many years, only limited clinical details are known. Only case reports and case series with limb pain as a symptom of migraine are reported in the literature. In this case report, important features of episodic unilateral limb pain and its association with migraine has been discussed.

**Case presentation:** Here the author (a neurologist) describes episodic limb pain from his own experience, which started in childhood and used to be episodic, unilateral and with changing sides. In view of various features mimicking migraine, classical manifestation of migraine in later life and a strong family history, episodic unilateral limb pain in childhood is likely a form of peripheral migraine.

**Conclusion:** Unilateral episodic limb pain with various features mimicking migraine is a very important clinical entity. This case should help in understanding the clinical details of peripheral limb pain - a relatively under-diagnosed symptom of migraine.

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**Introduction**

Migraine limb pain is a very under-recognized manifestation of migraine in which episodic pain happens in the peripheral limbs. Though it has been described as early as 1857 [1]. Only case reports and case series with limb pain as a symptom of migraine are reported in the literature [2]. It has mostly been described in association with the headache. Here, the author (a neurologist) describes his own experience of episodic, unilateral limb pain without any associated headache in the childhood and episodic headache in adulthood as a manifestation of migraine. This should help in understanding the clinical details of peripheral limb pain - a relatively under-diagnosed symptom of migraine.

**Case Presentation**
**Demographic Details**

The case details are of the author himself. The author is currently 42 years age, was born of non-consanguineous parentage. There was no significant history to suggest any childhood insult. The milestones were normal.

**Medical History**
**Symptoms and Signs**

It all started in the early teenage with episodic unilateral pain the lower extremities on the either side, mainly affecting the calf region. At the onset, the pain used to be mild but will slowly become severe enough to disturb the day to day activity. The pain

used to be dull boring in character affecting the posterior and sometimes the anterior aspect of the calf region but at times will also ascend proximally to involve the anterior aspect of the thigh (mainly quadriceps region). But it never involved the hip or pelvic region. However, it has rarely involved the ipsilateral upper limb as well. In the upper limb, the commonest area affected was between the elbow to the wrist. It used to last for 4-5 hours but occasionally continued even for the whole day or so. If pain persists for more than one day then it may disappear in the affected limb and start in the other lower limb. However, it was not associated with any weakness and there was no aggravation by prolonged walking, running, or climbing stairs.

**The commonest precipitating factor** for this peripheral pain was direct exposure of limbs to cold air especially while sleeping. The other precipitating factors were stress, fasting, and sleep deprivation. The relieving factors were compressing the affected leg by either tying a tight cloth around it or by sitting in a way to put own body weight on the affected leg. However, the symptoms may reappear soon after relieving the compression. The best relief used to happen with low dose non-steroidal anti-inflammatory drugs (NSAIDs) like paracetamol or acetylsalicylic acid. The pain frequency was once in 2-3 months.

Subsequently, at around the age of 25 years, episodic headache started with typical symptoms like severe headache on either side, with photophobia, phonophobia, and nausea lasting for 4-5 hours to 48 hours at a frequency of 2-3 per month fulfilling the ICH criteria of classical migraine [3]. The headache is still there at

the same frequency. The peripheral limb pain is occurring rarely, almost 1-2 times in a year that too after exposure to cold air.

### Family History

The author is having five brothers and three sisters. All of them are having episodic unilateral headache without any aura qualifying for the criteria of common migraine. All brothers and one sister used to have leg pain at the age of 10 to 20 years but 2 elder brothers are still getting leg pain rarely in their forties and fifties. The next generation is also getting leg pain in childhood and headache at a later age.

### Differential Diagnosis

Various differentials for episodic limb pain were excluded by appropriate investigation. There was no vascular etiology, no vitamin deficiency or nocturnal leg cramps noted. There was no spondylosis or root compression noted by appropriate imaging. There were no features to suggest fibromyalgia or metabolic etiology.

As described above various characteristic clinical features can be noted for this peripheral limb pain (peripheral migraine).

**Migrating Unilateral Pain-** episodic, unilateral pain with occasional migration to the other side.

**The Character of the Pain-** Starting with dull boring pain in the leg and then increasing in intensity and may involve ipsilateral thigh followed by upper limb.

**Duration-** Typical duration of a few hours to one to two days

**Precipitating Factors-** exposure to cold, sleep deprivation, and stress.

**Relieving Factors-** Relief with NSAIDs or local compression. Applying a band over the head is known to produce relief in headache [4].

**Anatomical Localization-** Ipsilateral involvement of upper and lower limb will localize the pathology clinically above the cervical region, indicating thalamic dysfunction

**Family History -** A strong family history of headache and episodic unilateral leg pain, especially in childhood will again favor some relation of this pain to the migraine.

### Discussion and Conclusion

This case represent clinical features of unilateral episodic limb pain in detail and highlights similarity with unilateral headache seen in migraine. It's very important to replicate these findings in a large series. A genetic study may help to find a unique genetic variation.

There is very limited data on limb pain in migraine. Prakash et al (2009) described recurrent limb pain and migraine which is quite similar to these details [5]. However they have described pain mainly in the upper limb and some of the subjects had migraine in the beginning and limb pain later in the life. While in this case, it was unilateral lower limb pain in childhood and headache started quite late. Recently familial limb pain has been described in four generations (Heather Angu-Leppan) [6]. It mentions that children are more likely to have leg pain while adults suffer mainly from headache as a manifestation of migraine.

As migraine can have various other peripheral manifestations like episodic abdominal pain, cyclic vomiting, and episodic vertigo, this peripheral pain can be explained with the same theory. The author would like to use the term 'Peripheral Limb Migraine' as this can explain the episodic unilateral limb's symptoms in a better way. In migraine pathophysiology, trigemino-thalamic connections are involved [7]. Similarly spinothalamic connections might be

involved in peripheral limb pain.

**Conclusion-** Episodic unilateral limb pain is similar to unilateral headache in migraine. It is more common in childhood and may precede by many years before onset of classical headache.

### Clinical Implications

- Episodic unilateral limb is a unique symptoms seen in patients with migraine.
- It may be the only symptom in childhood and can be considered as migraine variant.
- A proper diagnosis will help in prognostic the patients at an early stage.

### Declarations

- Ethics approval and consent to participate- The author is describing his own history so not applicable
- Consent to publish-Yes , consent is there
- Availability of data and materials-reference added
- Competing interests
- Funding-NIL
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