Headache Medicine

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Cluster headache with side-changing attacks: a case report and diagnostic challenges

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Introduction
Cluster headache is a primary headache disorder characterized by severe unilateral pain associated with autonomic symptoms and/or motor restlessness. Its estimated prevalence ranges from 41 to 381 cases per 100,000 inhabitants. However, it remains an underdiagnosed condition, with patients experiencing an average diagnostic delay of 5.7±6.7 years. In this context, atypical presentations may make diagnosis even more challenging, highlighting the importance of reporting such cases.

Case presentation
A 63-year-old male presented with a 10-year history of headache characterized by unilateral, excruciating, and disabling pain associated with conjunctival injection, lacrimation, and nasal congestion. During attacks, the patient exhibited agitation and photophobia. Episodes lasted 15–20 minutes and occurred approximately three times daily. He experienced cluster periods lasting about two months, separated by pain-free intervals of approximately ten months. During these active cluster periods, the patient reported alternating pain sides within the same cluster period. Neurological examination revealed no significant abnormalities. MRI showed signs of global cerebral atrophy (GCA 1) and microangiopathy (Fazekas 1). The patient fulfilled diagnostic criteria for cluster headache. Significant improvement was observed after treatment with verapamil and dexamethasone during cluster periods.

Discussion
Cluster headache classically presents with unilateral pain. Most patients (84–91%) do not experience changes in pain laterality, while the remaining patients present side-shifting attacks between different cluster periods. Alternating pain laterality within the same active cluster period is considered a rare presentation in the literature. The pathophysiology supports its unilateral presentation and involves a complex interaction between the trigeminovascular system, parasympathetic pathways, and the hypothalamus, with PET and functional MRI studies demonstrating unilateral hypothalamic activation during attacks, thereby reinforcing the lateralized nature of the disorder. However, the pathophysiology underlying side-shifting attacks remains unknown. Previous reports suggest a higher association between alternating laterality and chronic forms of the disease or prior occipital nerve corticosteroid injections, characteristics that were absent in the present case. It is likely that, in clinical practice, identification of side-shifting attacks is underestimated; in a study by Youn et al., 13 out of 124 patients presented alternating laterality within the same cluster period. Patients with these atypical phenotypes are at increased risk of remaining unrecognized.

Conclusion
This case illustrates an atypical presentation of cluster headache characterized by alternating laterality within the same cluster period. Recognition of the phenotypic variability of cluster headache is essential to avoid underdiagnosis and ensure appropriate diagnosis and treatment.

Keywords:
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