Brief Report

Cephalalgia Reports

Cephalalgia Reports
Volume 1: 1–3

DOI: 10.1177/2515816318814226
journals.sagepub.com/home/rep
SAGE

Cluster-like headache secondary to maxillary sinusitis by Aspergillus

Laura Martinez Rodriguez, Angel Ignacio Perez Alvarez, and Jose Maria Sanchez Alvarez

Abstract

A symptomatic cluster headache (CH), caused by fungal maxillary sinus infection fulfilling the criteria of International Headache Society (ICDH-3) for CH is presented. A 35-year-old man without previous headaches reported episodes of pain in the right fronto-orbitary region, with severe eyelid oedema, photophobia, frontal sweating, lacrimation, conjunctival injection and rhinorrhoea, which lasted for 2–3 h. A brain magnetic resonance imaging (MRI) showed occupation of the right maxillary sinus, a mycetoma due to *Aspergillus fumigatus*. Although rare, secondary causes must be discarded before the diagnosis of a primary CH is made. Imaging study should always be performed, even though no atypical feature was present and ICDH-3 were fulfilled.

Keywords

cluster headache, differential diagnosis, infection, symptomatic, trigeminal nerve

Date received: 8 July 2018; Received revised October 9, 2018; accepted: 18 October 2018

Introduction

Cluster headache (CH) is the most frequent syndrome in the group of trigeminal autonomic cephalalgias. It is characterized by an excruciating and strictly unilateral headache, ipsilateral cranial autonomic features, circadian rhythmicity, lasting between 15 and 180 min.[1] CH is a primary headache, but secondary cases have been described, with lesions generally located in the sella turcica or the cavernous sinus.[2] Establishing a causal relationship between these lesions and the symptoms is not always clear. We present a case of symptomatic CH caused by fungal maxillary sinus infection, fulfilling the criteria of International Headache Society (ICDH-3) for CH.

Main text

A 35-year-old healthy man, without any concomitant disease or previous headache, reported episodes of right fronto-orbitary pain. They occurred every other day in two occasions separated by 1 week, without trigger factors recorded. After the fourth episode, verapamil was prescribed and pain did not recur. The pain was located in the right fronto-orbitary region associated with severe eyelid oedema, photophobia, frontal sweating, lacrimation, conjunctival injection and clear rhinorrhoea. All symptoms were ipsilateral to pain. It lasted for 2–3 h and occurred at any time of the day. Between the episodes of pain, he was asymptomatic.

He was evaluated by the hospital's emergency services. Basic laboratory investigation and neurological and systemic examinations were normal. He was diagnosed with CH and was given treatment with intravenous corticosteroids that improved the acute symptoms. Neither triptans nor oxygen was used. Prophylactic oral treatment with verapamil was started and pain did not recur.

Department of Neurology, Hospital Universitario Central de Asturias, Oviedo, Spain

Corresponding author:
Laura Martinez Rodriguez, Department of Neurology, Hospital Universitario Central de Asturias, Avenida de Roma s/n, Oviedo, Asturias 33011, Spain.
Email: lauramartinezrodriguez88@gmail.com

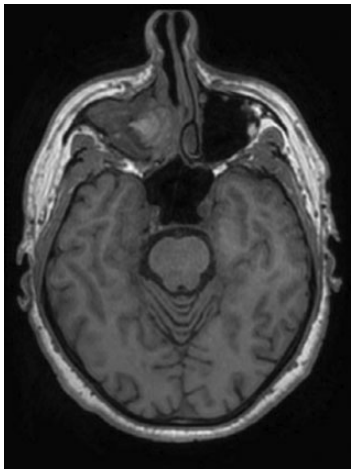

Figure 1. Occupation of the right maxillary sinus with distortion of its walls by mass protruding into the nasal fossa with complete obliteration of the middle infundibulum and reabsorption of the turbinate, all compatible with fungal sinusitis.

A brain magnetic resonance imaging (MRI) showed severe occupation of the right maxillary sinus with distortion of its walls, compatible with acute fungal-like maxillary sinusitis (Figure 1). He was treated with functional nasosinusal endoscopic surgery. The anatomopathological results and culture were compatible with a mycetoma (fungus ball) due to *Aspergillus fumigatus*.

Fifteen months after the surgery, the patient has not presented headache again and does not follow any prophylactic treatment.

Conclusion

Although cluster-like headache (CLH) is rare compared to primary CH, secondary causes must be discarded before the diagnosis of a primary CH was made.

The literature on CLH is mainly based on cases of lesions in the posterior fossa, and the aetiology varies widely from vascular, inflammatory, and neoplastic causes. Structural disease of sellar and parasellar structures, and carotid artery dissection have been reported too. Only a few cases of sinusitis mimicking CH have been collected in recent publications.[3]

It is assumed that in CH the origin of the headache is an initial activation of the hypothalamus that causes a secondary activation of the trigeminal autonomic reflex. In case of CLH, the physiopathology substrate is not well elucidated.

In CLH due to sinusitis, the pathogen growing may cause irritation of pain-sensitive structures and activation of trigeminal nerve endings.[4] Branches of the trigeminal nerve are widely distributed to the mucous surfaces of paranasal sinuses. If there is acute or chronic inflammation, trigeminal branches are being stimulated and are responsible for pain. The origin of autonomic symptoms is less defined, and it may be possible that nociceptive stimuli from inflamed sinus mucosa causes the activation of substance P neurons and parasympathetic neurons after the primary activation via the trigeminal afferent fibers of the Gasserian and sphenopalatine ganglions respectively. These types of neurons can elicit orthodromic and antidromic impulses along the nerve, inducing hyperactivity of neuronal network and could be responsible for the severe headache and autonomic symptoms.[5]

Mycotic infections of the paranasal sinuses are infrequent, but the most prevalent pathogen is *Aspergillus*.[6] This opportunistic fungal organism resides preferably in the nose and sinuses and is innocuous, but if conditions are favourable, it grows and develops, invading the nearby tissues.[7]

Headache due to sinusitis is characterized by different qualities and symptoms, such as protracted pain, purulent nasal discharge, local pain, and inflammation of the implicated sinus; and Horner's syndrome is not found.[5] In our case, the patient presented severe unilateral pain self-limited with clear nasal discharge, and none of the sinusitis headache characteristics was present.

Only three cases of CLH due to Aspergilloma sinuses infection[6–8] have been reported in the literature. The publications include three patients (two females and one male) between the ages of 62 and 68. One of the individuals had focal neurological symptom during episodes consisting of diplopia. Another patient presented episodes of pain of greater duration than expected (5 h), while the third one met with no exceptions, with all the criteria for the diagnosis of CH by the ICHD III. We have to emphasize that all of them were ages superior to those typically presented at the time of diagnosis of CH.

The case we presented has features suggestive of primary CH: sex, age of onset, duration, headache every other day, normal neurological examination, and the patient also responded to verapamil. However, the pain did not have a circadian rhythm, occurred both during day and at dawn, autonomic symptoms were maintained more than 12 h after the headache resolution, and the patient did not have restlessness. In addition, although the remission of the attacks was reached with steroid and verapamil, the response of the pain to CH medications neither excludes a symptomatic form. The present case evidences that imaging study should always be performed in patients with CH, even though no atypical feature was present and ICDH-3 were fulfilled, as in agreement with the European Headache Federation consensus.[9]

Clinical implications

- A secondary cause should always be considered in CH.
- MRI should always be performed.
- Response to usual treatment does not exclude a symptomatic form.

Author contributions

Laura Martinez Rodríguez contributed to acquisition of data and drafting content; Angel Ignacio Perez Alvarez helped in acquisition of data; Jose Maria Sanchez Alvarez proposed the original idea and was involved in study supervision.

Declaration of conflicting interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Funding

The author(s) received no financial support for the research, authorship, and/or publication of this article.

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