

## Case Report

### Secunder paroxysmal hemicrania caused by mucocele

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**Abstract.** Paroxysmal hemicrania (PH) is classified as a subgroup of trigeminal autonomic headaches among primary headaches. The pain is unilateral, located frequently at the orbital, supraorbital and/or temporal regions. It is sharp, severe, and these headaches tend to occur at least 5 times a day, which last for 2 – 30 minutes, accompanied by autonomic findings such as conjunctival injection, lacrimation, nasal congestion, nasal discharge, ptosis and orbital edema. Secondary PH is associated with tumor or vascular pathologies. Most frequently hypophyseal tumors were detected. In our case, there was a mucocele originating from the left ethmoidal sinus and compressing on the medial rectus muscle in the cranial MR examination of a patient admitted with clinical features of PH. In this case, it was seen that neuroimaging is mandatory in patients with TOB, in order to exclude secondary causes. Also, nociceptive impulses originating from surrounding tissues with the compression effect of the mucocele induce trigeminal autonomic reflex, supporting the hypothesis that autonomic symptoms and pain occur with hypothalamic and trigeminal connections.

**Keywords:** Trigeminal autonomic cephalalgia, paroxysmal hemicrania, mucocele

#### Introduction

Paroxysmal hemicrania is classified among trigeminal autonomic cephalgias subgroup among primary headaches [1]. TOS is a kind of headache characterized by autonomic symptoms at trigeminal nerve somatic distribution [2]. The pain is frequently seen at the ophthalmic region of the trigeminal nerve. IHSS-3 have remarked that PH pain may be seen as orbital, supraorbital, temporal or any combination of the trigeminal nerve [1]. PH is characterized by very severe pain accompanied by sharp, short unilateral autonomic symptoms [3]. The pain is typically described as severe, throbbing, and squeezing type of sensation [3, 4, 5, 6]. Pain is intermittent and starts suddenly. It frequently lasts for 10-30 minutes, with a range duration between 2-45 minutes. More than 60% of patients experience restlessness and pain between episodes [7]. IHSS-3 beta criteria requires accompaniment of at least one autonomic symptom such as ipsilateral conjunctival injection, lacrimation, nasal congestion, rinorrhea, sweating of forehead and face, myosis, ptosis and/or eye edema [1].

The prevalence of PH is not as low as it may be thought. Either the cases were not recorded or the diagnosis was not considered. Case series have described both typical and atypical cases, and 22% of cases show an atypical presentation [8].

Secondary PH is typically associated with tumors and vascular pathologies. Most frequent causes of PH and other

TOS include pituitary gland pathologies, and for this reason cranial MR imaging is mandatory [8].

In our case, there was a mucocele which originated from the left ethmoidal sinus and compressed the medial rectus muscle at cranial MR imaging in a patient admitted with clinical features of PH.

#### Case report

A 52-year-old female patient was admitted at our outpatient clinics with complaints of headache which had started 3 months ago, and showed an increase in frequency and severity in the last 15 days. The pain was experienced around the left eye, lasted for 5-10 minutes, and had occurred for at least 10-20 episodes per day. The patient described the pain as very severe in intensity and stinging/stabbing. There was injection and ptosis at the ipsilateral eye during an episode. Treatment was initiated with indomethacin 75 mg/day and complete remission was obtained on the second day. A lesion originating from the left anterior ethmoid sinus and compressing the left medial rectus muscle was detected in cranial MRI (Figure 1).

The patient had a surgical intervention at the ENT department, and histopathological examination revealed a mucocele. The patient did not experience pain during one-year of follow-up.

#### Discussion

PH is a rarely encountered type of headache, which

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Figure 1. Cranial MR a) T1, b) T2, c) T2 Flair imaging: Mucocele originating from the left anterior ethmoid sinus and compressing the left medial rectus muscle

## Discussion

PH is a rarely encountered type of headache, which may be due to missing this diagnosis or not recording it. There may be a group of patients, which may not exactly fulfill the IHSS-3 diagnostic criteria [8].

There is overlap in clinical findings and treatment of TACs, which may be caused by a common physiopathology of TACs [9, 10]. In a PET study in patients with PH, activation was observed in the contralateral posterior hypothalamus, ventral midbrain, red nucleus and substantia nigra [11]. Trigeminal autonomic reflex (TAR) was held responsible for the symptoms of trigeminal autonomic pain syndromes. TAR activation is responsible for the acute attacks of TACs [12].

This reflex is probably triggered due to activation of hypothalamus and structures closely related with it [12]. PH and other autonomic findings of TACs probably develop with central dysinhibition of TAR by direct hypothalamic – trigeminal connections of the hypothalamus [13].

Pituitary gland lesions constitute the most frequent cause of secondary PH and TAC. For this reason, cranial MRI should be part of our imaging studies in PH and TACs [8].

In conclusion, a mucocele originating from the ethmoidal sinus and compressing the rectus muscle induces TAR with nociceptive impulses that originate from the surrounding tissues. Autonomic symptoms and pain occur as a result of hypothalamic – trigeminal connections. Neuroimaging should be done in order to exclude secondary causes in all patients with a diagnosis of PH.

## Conflict of Interest

The authors declare no conflicts of interest.

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