



Case Report

Cluster Headache and Insular Glial Tumor

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Summary

Cluster headaches are known to be induced by secondary reasons as a proportion of 3-5%. We present here a young patient applied with symptoms of left arm weakness and cluster headache, and he has lower grade glial tumour in the insular area in neuroimaging. With this exceptional case, we wanted to emphasize the importance of insula on pain and autonomic system, and to indicate the necessity to be careful about secondary reasons for the patients applied with cluster headache and additional neurological features.

Key words: Glial tumour, insular area, cluster headache, secondary headaches

Küme Baş Ağrısı ve İnsuler Glial Tümör

Özet

Küme baş ağrılarının %3-5 oranında ikincil nedenlere bağlı olarak ortaya çıktığı bilinmektedir. Sol kolda kuvvetsizlik ve küme baş ağrısı belirtileri ile başvuran ve nörogörüntülemeye insular bölgede düşük evrede glial tümör saptanan genç bir hastayı sunuyoruz. Bu nadir görülen olgu ile, insulanın ağrı ve otonomik sistem üzerine etkisini vurgulamak, küme baş ağrısı ve ek nörolojik bulgular ile başvuran hastalarda ikincil nedenler hakkında dikkatli olmanın gerekliliğine işaret etmek istedik.

Anahtar Kelimeler: Glial tümör, insuler bölge, küme baş ağrısı, ikincil baş ağrıları

INTRODUCTION

Cluster headache occurs in between average of 20 to 40 years of age. It is more common in men than in women. It is a headache characterized by unilateral orbital, supraorbital or sometimes temporal headache, accompanied autonomic features such as ipsilateral Horner, and ipsilateral conjunctival hyperaemia, eye tearing, continued for 15-180 minutes. It is severe attacks occurring between one times every other day to 8 times a day⁽⁹⁾.

Pathophysiology of cluster headache is not fully understood. Known can be

summarized as follows. Pain is triggered by the result of disruptions in the centre of circadian rhythm which is in the hypothalamus localized suprachiasmatic, with stimulation of trigeminal nerve nuclei, trigemino-vascular system is provided to activate and as a result of this activation, neurogenic inflammation occurs in the cavernous sinus and the veins around cavernous sinus. This initiates a typical cluster-type headache. Compression of the sympathetic nerve plexus occurs as a result of vasodilatation of blood vessels. This leads to show up of autonomic signs⁽⁹⁾. Although, the role of cerebral hemispheres on cardiac sympathovagal modulation, in

clinical and experimental studies, the insular cortex, amygdala and lateral hypothalamus were shown to be control centres for the autonomic function⁽¹²⁻¹⁶⁾.

The association of lower grade glioma or insular location and typical cluster headache has not been described. There are two broad reviews of symptomatic/secondary cases of cluster headache in the literature^(7,11). Briefly, in these reviews the low prevalence of glial tumours associated with cluster headaches is notable. Only one report has been published showing an association between cluster headache and a highly malignant glioma has been published⁽⁸⁾.

We presented the case applied for cluster headache that we thought occurred due to lower grade glial tumor placed in insular area.

CASE PRESENTATION

The male patient, age of 33 applied with complaints of pain localized around the right side of his head and right eye started 1 month ago which took place at 01.00 at night, 3 times a week. The patient said that; when his pain started he could not sleep, strolled around the house constantly, and could not release his pain. The pain was relieved in approximately 1.5 hours. The patient's headache was 8 according to visual analogue score. From patient history, it was learned that one year before he had headache similar to his present headache starting right side of his head at 01:00 o'clock in November. Pain always happened at the same time. Tearing and redness in the right eye accompanied to the headache. Then, he did not have pain again. The patient said that, he had feeling of heaviness at his left arm about one year that he could not notice when it exactly started.

There were no other features in the personal and family history of the patient. Physical and neurological examination was

normal. Cluster headache was considered for the patient according to the ICHD-2⁽¹⁰⁾. Cerebral MRI was performed due to complaint of weakness on the left arm of patient. In the cerebral MR at T2 FLAIR section hyper intense area existed in the right insular region without diffusion limitation at diffusion-sections (Figure 1).

Nevertheless, the aetiology of ischemic stroke was investigated. Complete blood count, antinuclear antibody, anti-DNA, Lupus anticoagulant and anticardiolipin antibodies, and other vasculitic markers were normal or negative. Electrocardiography revealed normal sinus rhythm. Carotid and vertebral artery Doppler ultrasound were normal. Transcranial Doppler ultrasound was performed for patent foramen ovale and was normal. The patient was undergone transthoracic echocardiography. Valve pathology and thrombus was not detected. The transition was not seen in the contrast echocardiography. Cerebral magnetic resonance angiography was performed and was normal. However, magnetic resonance spectroscopy revealed lower grade glial tumour because of the increased choline and lipid lactate concentration in the insular area (Figure 2). Therefore, secondary cluster headache was considered for the patient due to glial tumour. The decision of lower grade glial tumour was made by not only radiological features but also neurochirurgical consultation. Oral steroids and verapamil were started for cluster headache. No pain attacks were observed after the second day of steroid treatment. The steroid treatment was reduced after the fifth day and then stopped. The patient was followed clinically with verapamil 80 mg/day. After discharge, one month later the patient had no symptoms at his control, and his neurological examinations were normal. Verapamil was reduced and stopped. Clinical follow was recommended.

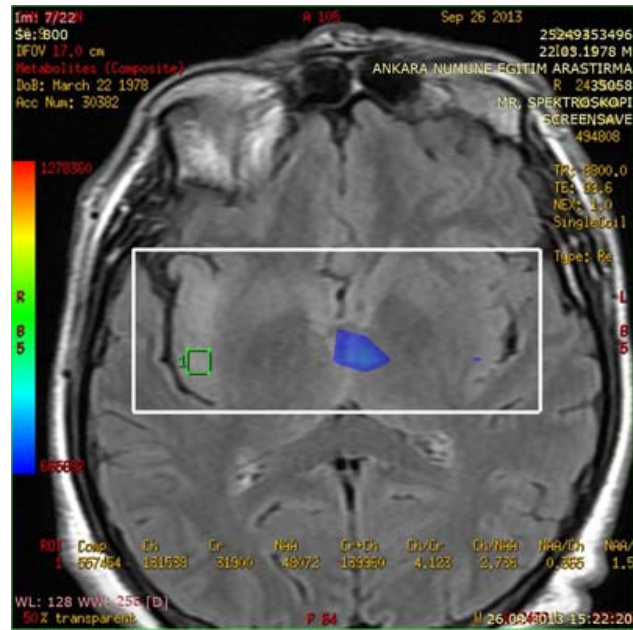


Figure 1: The right insular region is seen hyper intense on T2 FLAIR section.

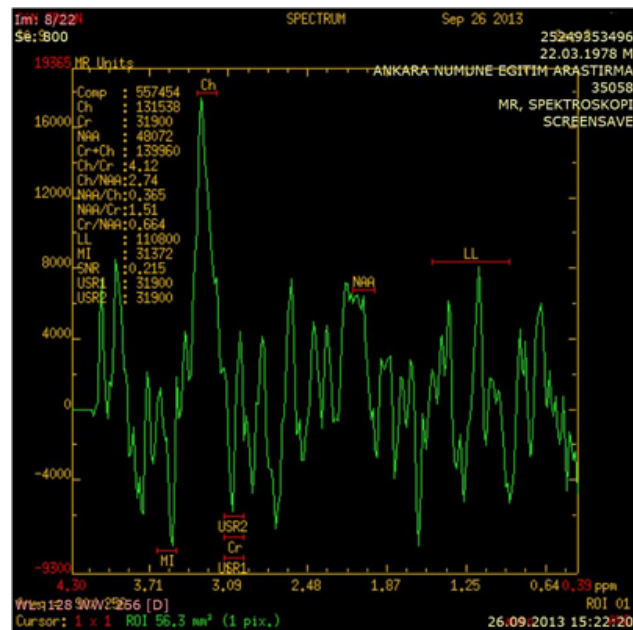


Figure 2: The MR Spectroscopic measurements of the lesion.

DISCUSSION

In experimental and clinical studies, it was shown that, insular cortex was the most important cortical area for the sympathetic and parasympathetic cardiovascular regulation⁽¹⁴⁾. It was shown that, tonus of sympathetic cardiovascular increased with the stimulation of right insular cortex, whereas the parasympathetic activity occurred with the left insular cortex stimulation. Insular cortex is often activated bilaterally during nociceptive somatosensory stimulations and is thought to play an important role in pain process⁽²⁾. The afferent nociceptive inputs may spread as rostral to posterior and anterior insula from second somatosensory cortex⁽¹²⁾. Activation of insular cortex was correlated with the severity of nociceptive stimulation and this structure may play a role for pain intensity^(6,15). Experiences of pain in patients with insular lesions with painful stimuli show more complex differences⁽⁵⁾. It is asserted that extensive connections of the insula may play a versatile and complex role in pain. For example, it is asserted that the insula may be important in both, pro-nociceptive processes and as well as the anti-nociceptive processes; in placebo, opioid analgesics, pain expectancy and the duration of hypnosis⁽³⁾. Until now, in few studies the changes of brain activation related pain after insula damage and the integrity of the pain perception was investigated. Functional neuroimaging studies suggested that the insular cortex is important for pain processing^(1,4,17). When we considered complex relation explained above between insular cortex and both pain and the autonomic system, in our case, would not be wrong to think cluster headache which is probably due to glial tumour in the insular area.

Headache is a common manifestation of brain tumours, however, tension-type headache is the most common phenotype. To our knowledge, this is the first report

showing a lower grade glioma resulting cluster headache, and also the first description of insular location causing cluster headache. With this case, we wanted to emphasize the importance of insula on pain and autonomic system and to indicate the necessity to be careful about secondary reasons for the patients applied with cluster headache and to perform detailed history and neurologic examination as well as proper neuroimaging investigations for them.

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