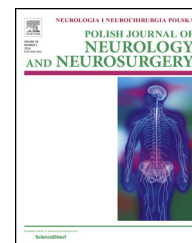


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Case report

A case report of an adolescent with cluster headaches following neck trauma: Coincidence or trigger?



Agnieszka Biedroń^{*}, Marek Kaciński, Małgorzata Steczkowska,
Anna Świerczyńska

Chair of Pediatric and Adolescent Neurology, Department of Pediatric Neurology, Jagiellonian University, Cracow, Poland

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ABSTRACT

Posttraumatic headaches usually have tension-type or migraine-like characteristics. A correlation between head trauma and cluster headaches (CH) has been previously reported. CH in children are rare and require thorough differential diagnosis. We present an original case of a 15-year-old boy with cluster headaches associated with allodynia probably evoked by a neck trauma. Severe headache attacks started one month after neck trauma. At the beginning clinical presentation of our patient's headaches was very misleading. Headaches were bilateral and associated with infection. Initial diagnosis of sinusitis was made. During further observation headaches have become unilateral with typical for CH associated symptoms and additionally with allodynia. Other causes of secondary CH like cervicogenic headaches, brain tumor and vascular malformation have been excluded. The boy has undergone prophylactic treatment based on flunarizine and gabapentin with good result. Possible pathogenesis of our patient's headaches has been proposed and diagnostic traps discussed.

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1. Introduction

Cluster headache (CH) is characterized by severe, unilateral pain localized in orbital, supraorbital or temporal region. It is associated with ipsilateral autonomic symptoms [1].

Sometimes CH can be associated with allodynia [2], however it is less common than in migraine [3].

Prevalence of CH in children is rare. In adults it is estimated for 0.12% and is more frequent in males [4].

Correlation of CH with head trauma has been previously described in the literature. Positive history of head trauma has

^{*} Corresponding author at: Chair of Pediatric and Adolescent Neurology, Department of Pediatric Neurology, Jagiellonian University, Wielicka Street 265, 30-663 Cracow, Poland. Tel.: +48 12 6581870; fax: +48 12 6581870.

E-mail addresses: aganow7@yahoo.com (A. Biedroń), neupedkr@cm-uj.krakow.pl (M. Kaciński), malgorzata.steczowska@uj.edu.pl (M. Steczkowska), annasmed@wp.pl (A. Świerczyńska).
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16.5% patients with CH [5], however its pathogenetic role is still not clear.

We present an unusual case of 15-year boy with cluster headaches associated with allodynia probably provoked by a neck trauma.

2. Case presentation

A 15-year-old boy with an 8-month history of headaches was admitted to the Department of Pediatric Neurology for further diagnostics. One month before headache attacks had started the boy was hit in the neck with a schoolbag by his classmate. The boy was admitted then to an emergency room of provincial hospital where cervical spine damage was excluded.

First headache attack occurred with association of upper respiratory tract infection and fever. The pain was localized in the fronto-orbital region bilaterally. The pain was severe, pulsating, associated with photophobia and lasted for one day with minor relief after paracetamol intake. The boy was diagnosed with acute sinusitis and treated with antibiotics (amoxicillin/clavulanic acid, azithromycin).

The next headache attack started 2 weeks later without signs of infection. Headache localization, character and intensity were similar however additionally bilateral eye reddening and eye lid edema appeared (more prominent on the left side). The boy was treated with paracetamol, antihistaminic drugs (desloratadine orally, azelastine eye drops) and intranasal steroid (fluticasone) with no result. 2 days later the boy was admitted to the pediatric department of provincial hospital. Focal neurological signs and meningeal signs were negative. Fundus of both eyes was normal. Blood pressure was normal. Neuroimaging studies (contrast enhanced head CT and MR) did not reveal any pathology. Lumbar puncture showed clear cerebrospinal fluid with negative bacteria culture. Leukocytosis and CRP were within normal limits. Otolaryngological examination revealed nasal polyps. Additionally sinuses CT was performed and revealed ethmoiditis and mild inflammatory changes in right maxillary and sphenoid sinuses. During hospitalization the headaches were present for 5 days. At the beginning of hospitalization oxygen therapy was tried with no result. Paracetamol brought him only short relief (2–5 h). Cessation of headaches was noticed after intravenous dexamethasone administration. Diagnosis of chronic sinusitis was made and the boy with recommendation of multidisciplinary (laryngology, allergology and neurology) outpatient care was discharged home.

Headache attacks occurred 9 times for the next 7 months (every 3–4 weeks). The attacks lasted from 1 to 10 days with temporary relief (2–3 h of pain then 6–8 h pain free period) after analgesic drugs (paracetamol, ibuprofen). The headaches were unilateral, localized in temporal or supraorbital and temporal region with headache side shifting. They were associated with ipsilateral reddening of the eye and/or ipsilateral allodynia. Usually they were severe and pulsating and associated with photophobia. Only three attacks were evaluated as moderate.

During this period the boy underwent allergological diagnostics and treatment. Skin prick test to food and inhalant

allergens were positive to mites, grass, corn, artemisia, hezel and cat's fur. Antihistaminic drug (rupatadine) was introduced and intranasal corticosteroid (mometasone) with no influence on headache frequency and intensity. The boy underwent allergen immunotherapy against mites. Finally, inhalant steroid was introduced (ciclesonide).

Follow-up laryngologist examination was normal, X-ray of sinuses did not reveal inflammatory process. Follow-up ophthalmologic and neurologic examination was normal and suspicion of cluster headaches was made.

Additionally, two months before admission to our Department the boy started rehabilitation therapy of the cervical spine. During the therapy shift of headache side was observed. The headaches started to be less intense and headache periods were shorter.

On admission to our Department the boy was in good general condition, neurological examination did not reveal pathological signs. Transcranial doppler examination of intracranial arteries and MR angiography did not show any pathology.

During hospitalization the boy experienced one episode of headache attack localized in the left temporal area associated with ipsilateral allodynia. The intensity of headache was mild to moderate. The pain lasted for 2 days with night time break. Oxygen therapy was ineffective. The boy responded well to paracetamol.

One month later the boy was admitted again to our Department for follow up. In the mean time he continued rehabilitation of cervical spine. There were no episodes of headache attacks since last hospitalization. The boy underwent MR of cervical spinal cord which did not reveal pathology of spinal cord, only abolition of physiological lordosis with slight kyphosis of C3–C4 segment associated with reduction of dural sac fluid space at this level. At the level of C5/6 and C6/7 slight posterior left-sided bulging of intravertebral discs was noticed however with no signs of compression of spinal cord or nerve roots. Moreover, video-EEG examination was performed and revealed no characteristic changes. During one-week hospitalization there were no episodes of headache. The boy was discharged home in good general condition with recommendation of keeping headache diary and abortive treatment of headache attacks based on paracetamol and ibuprofen.

Further history of patient's headache attacks we collected on the basis of a telephone conversation with his mother.

The boy remained headache free for nine months. In the meantime he underwent complex cervical spine rehabilitation (massages, swimming) in the local rehabilitation sanatorium.

After that period headache attacks occurred again. Initially headaches were moderate, however in few months they developed into severe cluster headaches. During 18-month observation the boy experienced 6 cluster periods. Prophylactic treatment with flunarizine has been introduced (5 mg/day and 10 mg/day during cluster period). Sometimes laser therapy was used during cluster periods with good result (shortening of cluster period, diminishing headache intensity). The headaches were localized in the orbital and temporal region (headache side shifting was observed) associated with reddening of the eye, when severe additionally associated with lacrimation, eyelid edema, rhinorrhoea. Sometimes

allodynia was reported. Abortive treatment with acetaminophen plus caffeine plus codeine was used (headache relief within 0.5–1 h after taking the drug, painless period of 3–8 h). The most severe cluster attack lasted 16 days. Gabapentin was used to control the headaches with good result. The boy was on antiepileptic treatment for four months (maximal dose of gabapentin 1200 mg/day for 3 days, then gradual reduction). After withdrawal of gabapentin only one mild cluster headache attack occurred (observation period 3 months). 3 months later flunarizine was withdrawn as well, and no cluster attacks have been observed since then (observation period 2 months).

3. Discussion

According to The International Classification of Headache Disorders, 3rd edition (beta version) (ICHD-3) headaches can occur secondary to head and/or neck trauma [1]. The time interval for onset of this type of headache after an accident is strictly defined to maximum 7 days. Acute headaches subside within 3 months whereas persistent headaches last longer. Characteristics of posttraumatic headaches are not defined however usually they have tension-type or migraine-like clinical presentation [6]. There have been reports of posttraumatic CH, short-lasting unilateral neuralgiform headache attacks with conjunctival injection and tearing (SUNCT) and paroxysmal hemicrania [7–9]. Positive history of head trauma in patients with diagnosed CH, with longer interval between headache onset and the accident may be considered as risk factor for CH manifestation in prone patients [10].

In the presented patient the time interval between neck trauma and headache onset was one month which is longer than defined time in the criteria for posttraumatic headache.

Association between headache onset and neck injury made us also consider cervicogenic headache as possible diagnosis. However results of cervical spine X-ray and MR did not indicate pathology which could explain appearance of this type of headache. Additionally the patient did not experience neck pain. Positive response to rehabilitation of cervical spine may be coincidental.

Unilateral headache with ipsilateral autonomic symptoms has been evoked in patients after stimulation of great occipital nerve (GON) with sterile water injection in the territory of the nerve [11]. The pathomechanism of this phenomenon is explained by connections between the nerve and trigeminal nerve via trigeminocervical complex and its connections with other brain structures which take part in nociception. Rozen [12] reported a patient who developed CH after bilateral GON blockade. The author hypothesized that GON was stimulated during the procedure (needle insertion) or when the blockade was over and generated CH attack. Cluster period was probably due to secondary bilateral activation of hypothalamus, which could explain its atypical course (irregular cluster pattern, side shifting).

Based on these observations we find it possible that neck trauma which our patient experienced could trigger CH attacks by GON stimulation and secondary hypothalamus activation.

Our patients' headache attacks fulfilled diagnostic criteria for CH except for two first headache attacks which were

bilateral and of longer duration (when medication was not used headaches lasted more than 3 h). The patient did not manifest restlessness or agitation during the attacks. Additionally no effect after oxygen introduction could be observed.

Also very confusing was co-occurrence of acute sinusitis during the first headache attack. The following headache attacks become cluster-like with unilateral presentation and autonomic features. Cluster-like headaches have been described in the course of sinusitis or other pathological processes localized in the sinuses (sphenoid aspergilloma, meningioma) [13,14] or presence of foreign body [15].

The head CT and MR was performed in our patient two weeks after acute sinusitis, which may explain still present changes in the sinuses. Persistence of headache attacks and lack of clinical and radiological signs of sinusitis during follow-up visits made us reject sinusitis as a possible cause of the headaches.

Differential diagnosis of CH includes brain tumors and vascular malformations.

Secondary CH have been reported in the course of pituitary tumor [16], meningioma [17] or metastatic tumor [18]. Arteriovenous malformations, artery dissections and aneurysms have been described in patients with cluster-like headaches as well [19–21]. Brain MR of our patient and MR angiography were normal.

Management of cluster headaches comprises acute and prophylactic treatment. Oxygen therapy has been proved to be effective in cessation of cluster attacks in about 70% of patients [22]. Our patient did not respond to the oxygen therapy however good response to steroids was observed. Short term early prophylaxis with steroids has good results in cluster headaches however does not maintain the remission period [23]. For long term treatment calcium channel blockers (verapamil) have proven efficacy [24]. In CH treatment antiepileptic drugs may be used when standard treatment is ineffective. Our patient was initially treated only with flunarizine however introduction of gabapentin resulted in cessation of the most severe cluster period and full remission for 4 months was achieved. After discontinuation of gabapentin treatment only one mild attack has occurred (3-month observation period). Positive response to gabapentin in cluster headaches has been previously reported in the literature [25].

Laser therapy has been used in acute and chronic pain disorders like headaches, myofascial pain, neck and back pain. Positive effect of laser therapy in chronic migraine has been reported [26] however we have not found reports concerning application of laser therapy in patients with cluster headaches [PubMed]. Response to laser therapy was good in our patient.

4. Conclusions

Cluster headache belongs to autonomic cephalalgias and its prevalence in children and adolescents is rare. Thorough differential diagnosis must be conducted to exclude pathologies mimicking the primary headache. A correlation between head trauma and cluster headache has been previously observed. We present an unusual case of an adolescent with

cluster headaches associated with allodynia following neck trauma.

Conflict of interest

None declared.

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None declared.

Ethics

The work described in this article has been carried out in accordance with The Code of Ethics of the World Medical Association (Declaration of Helsinki) for experiments involving humans; Uniform Requirements for manuscripts submitted to Biomedical journals.

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