Acute Rhinosinusitis as an Infrequent Cause of Symptomatic Cluster Headache: Report of Seven Cases

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Submitted February 7, 2018;
accepted December 25, 2018.

Cluster headache (CH) is a primary headache disorder characterized by unilateral headache attacks lasting 15 to 180 minutes, occurring between two and eight times a day, and accompanied by autonomic symptoms ipsilateral to the pain. However, cases of symptomatic CH that occur secondary to an underlying structural pathology have also been reported in the literature. In this report, seven patients are presented who were admitted with signs of CH and diagnosed with acute rhinosinusitis depending on extensive clinical and radiologic examinations. Symptomatic CH, though rarely reported in the literature compared to CH, should be kept in mind in patients presenting with the first attack of CH. Moreover, in such patients, whether the pain becomes worse when bending forward and becomes sensitive on palpation should be questioned, and a radiologic work-up should be performed to rule out secondary causes such as rhinosinusitis. In the present cases, the resolution of CH attacks with acute sinusitis therapy confirmed the diagnosis.
J Oral Facial Pain Headache 2019;33:408–412. doi: 10.11607/ofph.2175

Keywords: *cluster headache, inflammation, pain, rhinosinusitis*

Cluster headache (CH) is a primary headache disorder commonly occurring in the orbital, supraorbital, or temporal regions—or in any combination of these sites—characterized by unilateral headache attacks lasting 15 to 180 minutes and occurring between two and eight times a day accompanied by autonomic symptoms ipsilateral to the pain.[1] However, cases of symptomatic CH that occur secondary to an underlying structural pathology have also been reported in the literature, though rarely.[2] In patients presenting with CH, patient history and clinical examination are of vital importance, as they may reveal the secondary causes of CH. In some cases, although the clinical features of the patient may indicate primary headache, the diagnosis can be CH resulting from a secondary cause. This situation is valid not only for the tension-type headache that manifests with nonspecific symptoms, but also for the migraine and CH that manifest with more specific symptoms.[2]

Common causes of symptomatic CH include vascular, tumoral, infectious, posttraumatic, and dental pathologies.[2] Female patients describe CH attacks as more severe than labor pain; therefore, prompt diagnosis and treatment should be performed to eliminate the primary cause of the attack while keeping the secondary causes of CH in mind.[3] In this report, seven cases with CH who were admitted with signs of CH and diagnosed with acute rhinosinusitis depending on extensive clinical and radiologic examinations were presented.

Case Reports

Case 1

A 23-year-old male patient presented with a 2-week history of headache attacks with a severe stabbing pain around the left eye that occurred two times a day, mostly during lunchtime and before sleep, lasted for about 1 hour, and did not change when bending forward. The pain was accompanied by a stabbing sensation, lacrimation, and

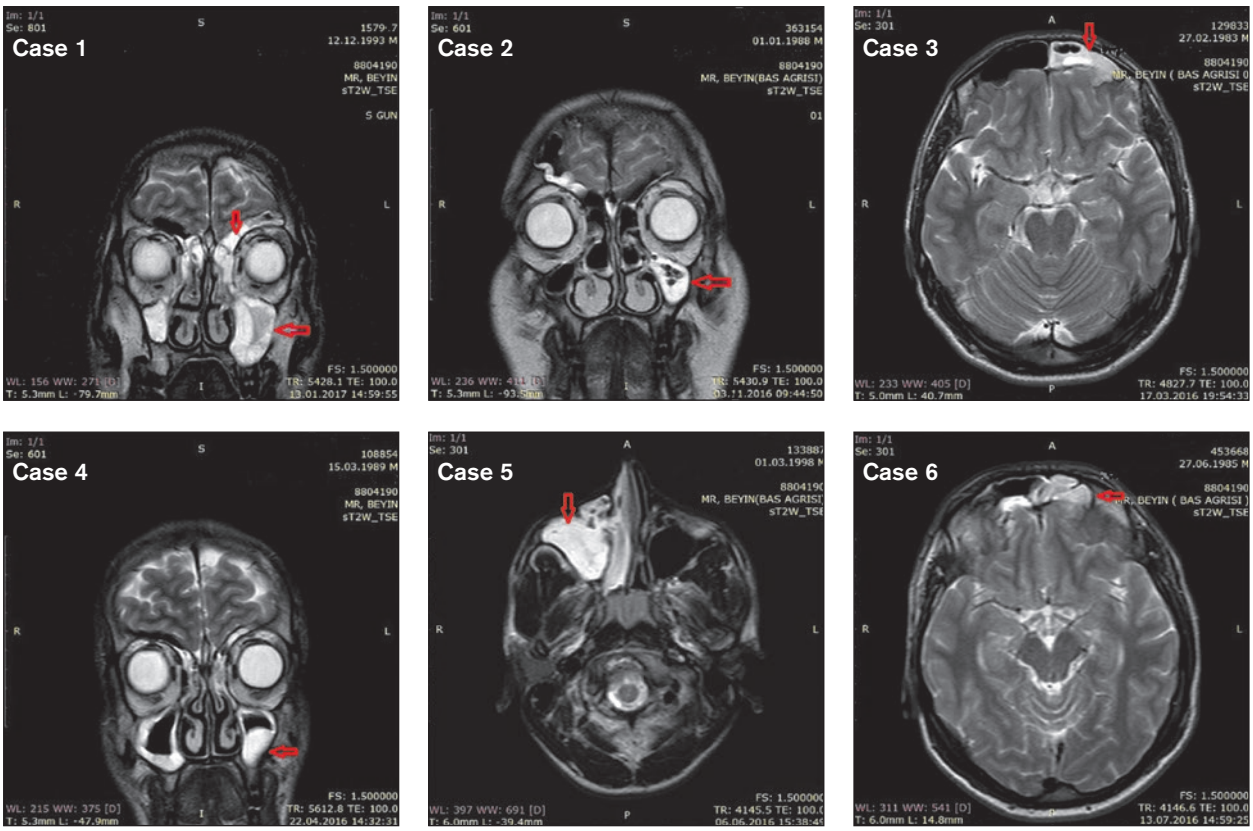
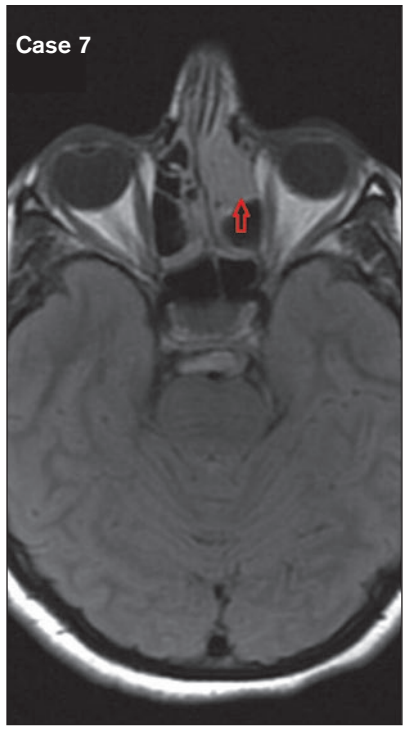


Fig 1 Magnetic resonance images of paranasal sinuses (cases 1 through 7).

redness in the left eye, with no nasal congestion or discharge ipsilateral to the pain. The patient also experienced extreme restlessness during the pain and described the pain as the most severe pain he had ever experienced. Neurologic examination and laboratory parameters (ie, sedimentation, C-reactive protein [CRP], whole blood count [WBC]) were normal. The patient had no significant history of disease and no history of similar attacks. Cranial magnetic resonance imaging (MRI) showed soft-tissue inflammation in the bilateral frontal, ethmoid, and maxillary sinuses, which was more pronounced on the left side (Fig 1). The patient was diagnosed as having symptomatic CH and was referred to the ear-nose-throat (ENT) department. Following antibiotic therapy, the complaints resolved completely, and no such headache attack has been observed throughout the 20-month follow-up period.

Case 2

A 28-year-old male patient presented with a 10-day history of headache attacks with a stabbing pain around the right eye that occurred two times a day, mostly after waking up in the morning and during lunchtime, lasted for 45 minutes, and was mostly severe and intermittently mild. The pain was accompanied by redness and ptosis ipsilateral to the pain, with no burning or stabbing sensations or lacrimation. The pain was severe enough to awaken the patient and also caused restlessness. Neurologic examination and laboratory parameters (sedimentation, CRP, WBC) were normal. The patient had no significant history of disease and no history of similar attacks. Cranial MRI revealed right frontal and left maxillary sinusitis (Fig 1). The patient was referred to the ENT department. Endoscopic sinus surgery was performed since the patient was



unresponsive to antibiotic therapy. The complaints resolved after surgery, and no such headache attack has been observed throughout the 23-month follow-up period.

Case 3

A 33-year-old male patient presented with a 15-day history of headache attacks with a severe burning pain and redness around the left eye that lasted for 20 minutes and became worsened when bending forward but was alleviated with palpation of the frontal region. The pain started immediately after waking up in the morning and restarted after a 1-hour interval, with a total of six to seven consecutive attacks occurring during the day. The pain was accompanied by blocking of the left nasal passage and nasal discharge. Neurologic examination and laboratory parameters (sedimentation, CRP, WBC) were normal. The patient also experienced extreme restlessness during the pain and described the pain as the most severe pain he had ever experienced. As the pain was extremely severe, the patient considered suicide as the only way to escape the pain. The patient had no significant history of disease and no history of similar attacks. Cranial MRI showed a left frontoethmoidal mucocele (Fig 1). The patient was referred to the ENT department, and the mucocele was marsupialized with endoscopic sinus surgery. The complaints resolved immediately after the surgery, and no such headache attack has been observed throughout the 30-month follow-up period.

Case 4

A 27-year-old male patient presented with a 15-day history of headache attacks around the left eye that mostly occurred during lunchtime or before sleep, once a day, lasted for up to 2 hours, became worse when bending forward, and was sensitive on palpation. The pain was accompanied by a burning and stabbing sensation, lacrimation in the left eye, and blocking of the left nasal passage. The pain was also accompanied by forehead sweating, causing no sense of restlessness. The patient had no significant history of disease and no history of similar attacks. Neurologic examination and laboratory parameters (sedimentation, CRP, WBC) were normal, and cranial MRI showed bilateral frontal, ethmoid, and maxillary sinusitis, which was more pronounced on the left side (Fig 1). The patient was referred to the ENT department, and the complaints resolved completely after antibiotic treatment. No such headache attack has been observed throughout the 29-month follow-up period.

Case 5

A 16-year-old male patient presented with a 3-day history of headache attacks with a stabbing pain around the right eye that occurred two times a day at varying times, lasted for 1 hour, became worse when bending forward, and was sensitive on palpation. The pain was accompanied by lacrimation and ptosis in the ipsilateral eye, with no redness, burning, or stabbing sensations, nasal congestion, or discharge ipsilateral to the pain. The pain was extremely severe, causing a sense of restlessness. Neurologic examination and laboratory parameters (sedimentation, CRP, WBC) were normal. The patient had no significant history of disease and no history of similar attacks. Cranial MRI showed right maxillary and ethmoid sinusitis (Fig 1). The patient was referred to the ENT department. The complaints resolved completely after antibiotic therapy, and no such headache attack has been observed throughout the 27-month follow-up period.

Case 6

A 31-year-old male patient presented with a 1-week history of headache attacks with a stabbing pain around the left eye that occurred during evening hours once every other day, lasted for 2 hours, and did not change when bending forward but was sensitive on palpation. The pain was accompanied by a burning sensation, lacrimation, and redness ipsilateral to the pain, with no nasal congestion or discharge. The pain was extremely severe, causing a sense of restlessness. Neurologic examination and laboratory parameters (sedimentation, CRP, WBC) were normal. The patient had no significant history of disease and no history of similar attacks. Cranial MRI showed bilateral frontal sinusitis, which was more pronounced on the left side (Fig 1). The patient was referred to the ENT department, and the complaints resolved completely after antibiotic therapy. No such headache attack has been observed throughout the 26-month follow-up period.

Case 7

A 39-year-old female patient presented with a 2-day history of headache attacks with a severe stabbing pain localized in the left orbital and supraorbital regions that occurred in the morning hours and at lunchtime, two to three times a day, lasted for 2 hours, and did not change when bending forward but was sensitive in the left supraorbital region. The pain was accompanied by lacrimation and redness in the ipsilateral eye, with no ptosis, burning or stabbing sensation, nasal congestion, or discharge ipsilateral to the pain. The patient described the pain as the most severe pain she had ever experienced. The pain also caused a sense of restlessness. Neurologic examination and laboratory parameters (sedimentation, CRP, WBC) were normal. The patient had no significant history of disease and no history of similar attacks. Cranial MRI showed mucosal thickening in the left sphenoid and ethmoid sinuses (Fig 1). The patient was referred to the ENT department. The complaints resolved completely after antibiotic therapy, and no such headache attack has been observed throughout the 20-month follow-up period.

Table 1 Clinical and demographic characteristics

Case no.	Age (y)	Gender	NE	TR	OR	Restlessness	TA	TP	Worsening with bending forward or sensitive on palpation	Medical history
1	23	M	N	NA	NA	Yes	NA	First attack	Yes	No features
2	28	M	N	NA	NA	Yes	NA	First attack	None	No features
3	33	M	N	NA	NA	Yes	NA	First attack	Yes	No features
4	27	M	N	NA	NA	None	NA	First attack	Yes	No features
5	16	M	N	NA	NA	Yes	NA	First attack	Yes	No features
6	31	M	N	NA	NA	Yes	NA	First attack	Yes	No features
7	39	F	N	NA	NA	Yes	NA	First attack	Yes	No features

NE = neurologic examination (including trigeminal nerve exits); TR = triptan response; OR = oxygen response; TA = triggering with alcohol; TP = temporal pattern; N = normal; NA = not available/unknown.

Table 1 presents the clinical and demographic characteristics of the patients.

Discussion

Secondary headache should be considered if the headache arises de novo as a result of another clinical condition, even when the headache is phenomenologically similar to migraine, tension-type headache, or CH.[1] Common causes of secondary headache include head and neck injuries, intracranial vascular (eg, stenosis, hemorrhage) or nonvascular disorders, headaches secondary to drug abuse or withdrawal, intracranial and systemic infections, hemostatic disorders, and psychiatric, craniofacial, or cervical disorders. On the other hand, common nasal-sinus pathologies causing secondary headache include acute, chronic, and recurrent rhinosinusitis.[1] In a recent study, Lee et al evaluated 1,235 patients who presented to the emergency service and were diagnosed with primary headache by neurologists and reported that rhinosinusitis was the underlying cause of headache in 172 (13.9%) patients.[4] Another study that was conducted in 2002 reported that in 90% of the patients diagnosed with sinus headache, the clinical signs and symptoms were consistent with migraine according to the International Headache Society (IHS) criteria.[5]

Mainardi et al identified a total of 156 CH cases published between 1975 and 2008. Based on the order of frequency, the secondary causes associated with CH included:

- Vascular diseases, including intracranial aneurysm, dural fistula, ischemic injuries, cerebral venous thrombosis, arteriovenous malformation, and carotid artery dissection
- Tumoral disorders, including pituitary adenomas, meningiomas, and carcinomas of the paranasal structures
- Infectious pathologies, including sphenoidal aspergillosis, ophthalmic herpes zoster, and maxillary sinusitis
- Posttraumatic pathologies, including orbital enucleation, penetration of a foreign body in the maxillary sinus, and concussive and nonconcussive cranial trauma
- Other neurologic pathologies, including multiple sclerosis, syringomyelia, and idiopathic intracranial hypertension
- Iatrogenic or drug-related conditions, including cocaine use, chemotherapy, warfarin therapy, and intraocular lens implant
- Tooth extraction

Of the 156 CH cases, only 4 were diagnosed with sinusitis.[2] Of these, 2 cases were reported by Takeshima et al in 1988, of which one case presented with frontal, ethmoidal, and maxillary sinusitis and the other presented with frontal and maxillary sinusitis.[6] The remaining 2 cases were later reported by Molins et al, who evaluated a large series of 100 CH patients and revealed that 4 of them were present with symptomatic CH, including 2 patients with maxillary sinusitis.[7] Nevertheless, the prevalence of symptomatic CH remains unknown, since there are no prospective population-based studies in neurology including neuroradiology.[8]

In the present study, a total of 84 patients were diagnosed with CH between June 2014 and January 2018 depending on the IHS criteria. Of these, 69 patients underwent cranial MRI after admission following the first attack of CH, whereas no radiologic examination was performed in the remaining 15 patients since they had previously undergone cranial MRI and were present with episodic or chronic CH. Of the 84 patients, 77 (91.7%) had primary CH, and only 7 (8.3%) were diagnosed with symptomatic CH secondary to acute rhinosinusitis. The resolution of CH attacks with the acute sinusitis therapy confirmed the diagnosis. These 7 patients comprised 6 men

and 1 woman, which implicates that symptomatic CH is more common in men, as in primary CH. No other secondary causes associated with symptomatic CH were detected in any patient. Accordingly, symptomatic CH secondary to acute rhinosinusitis, which is reported to have a low incidence, has a higher incidence than reported in the literature.[2]

In the present study, patients diagnosed with acute rhinosinusitis had normal CRP and sedimentation levels. An increased CRP level is significant in acute rhinosinusitis; however, it may not increase in all cases. In a recent study, Gowri and Muthiah reported normal CRP levels found in 66% of the patients diagnosed with sinusitis.[9]

The exact pathophysiology of CH remains unclear.[8] Moreover, whether the CH pain is of a central or peripheral origin remains controversial.[10] Nevertheless, "the pain-processing areas of the central nerve system" and the hypothalamus, as well as peripheral structures—including the trigeminal nerve, parasympathetic nerves, and the cranial-vascular structures—have been implicated in the pathophysiology of CH. The trigeminal-parasympathetic reflex projects to the trigeminocervical complex after the nociceptive stimulation induced by the sensory fibers arising from the trigeminal nerve traverse the trigeminal sensory ganglion. The fibers arising from the trigeminocervical complex project to higher brain regions through the ascending route and simultaneously project to the neurons of the superior salivatory nucleus. The parasympathetic system, activated by the stimulation of the superior salivatory nucleus, produces autonomous symptoms such as rhinorrhea, lacrimation, and nasal congestion through the mediation of the sphenopalatine ganglion.[10] As a result, unilateral headache attacks accompanied by severe rhinorrhea and lacrimation occur.[11] Moreover, in the presence of headache secondary to acute rhinosinusitis, CH may occur by the initiation of a parasympathetic reflex through trigeminal nerve activation. On the other hand, the response of the headache to sumatriptan and other typical CH medications does not rule out symptomatic CH.[12]

Cluster headache is typically diagnosed based on the information obtained from patient history regarding the localization and timing of the pain and the accompanying conditions; therefore, the diagnosis is often made based on patient history, and further questioning is often overlooked. Nevertheless, questioning the presence of local tenderness and whether the pain becomes worse when bending forward and performing a radiologic examination are highly important for avoiding a missed diagnosis of CH secondary to acute rhinosinusitis, which has a higher incidence than reported in the literature. Moreover, as steroids are commonly used in the treatment of primary CH, misdiagnosis of CH secondary to acute rhinosinusitis may aggravate the infection. For these reasons, questioning whether the pain becomes worse by bending forward and becomes sensitive on palpation is of paramount importance in patients presenting with the signs of CH.

Conclusions

Symptomatic CH, though rarely reported in the literature compared to CH, should be kept in mind in patients presenting with the first attack of CH. Moreover, in such patients, whether the pain becomes worse by bending forward and becomes sensitive on palpation should be questioned, and radiologic work-up should be performed to rule out secondary causes of CH such as rhinosinusitis.

Acknowledgments

The authors declare no conflicts of interest.

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