

**CHRONIC ATYPICAL FACIAL PAIN CAUSED BY A NECROTIC BONE**NEKROTİK KEMİĞE BAĞLI GELİŞEN KRONİK ATİPİK YÜZ  
AĞRISI

Rinoloji

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**Leyla Kansu<sup>1</sup>**<sup>1</sup> Başkent Üniversitesi Tıp Fakültesi**Özet**

Yaklaşık 20-25 yıldır yüzünün sağ tarafında ağrı şikayeti olan 39 yaşında bayan hasta değerlendirildi. Ağrı yanak sağ tarafında, pulsatif, şiddetli ve ani atak şeklinde oluyordu. Ağrı atağı tipik olarak 3 saat ile 3 gün arasında sürüyor, ayda 1-3 kez görülüyor fakat hastayı uykudan uyandırmıyordu. Hastanın diş ve nörolojik muayenesi dahil fizik muayenesi normal idi. Paranasal sinus komputere tomografide sağ maksiller sinus içinde radyopak lezyon ve sağ maksiller sinus medyal duvarında kemik defekti saptandı. Nasal endoskopide sağ maksiller sinus içinde kitle görüldü. Eksize edilen kitlenin histolojik incelemesi nekrotik kemik dokusu olarak rapor edildi. Sonuç olarak, maksiller sinus içindeki nekrotik kemiğin kronik fasiyal ağrıya sebep olabileceği düşünüldü.

**Anahtar kelimeler:** maksiller sinus, nekrotik kemik başağrısı tanı endoskopi

**Abstract**

A patient of 39-year-old woman was evaluated for about 20- to 25-year history of pain on the right side of the face. The pain occurred as sudden attacks of episodic, severe, pulsating pain at the right cheek. The pain attacks typically persisted for 3 hours to 3 days and occurred from 1 to 3 times monthly but did not wake her up from sleep. Physical examination was normal, including dental and neurologic examination. Computed tomography scan of the paranasal sinuses showed a radiopaque lesion in right maxillary sinus and bone defect at the right medial maxillary wall. Nasal endoscopy showed a mass in the right maxillary sinus. Symptoms resolved after excision of the mass. Histologic examination of the excised mass was necrotic bone tissue. As conclusion, a necrotic bone inside the maxillary sinus may cause chronic facial pain was thought.

**Keywords:** maxillary sinus, necrotic bone headache diagnosis endoscopy

**Introduction**

Headache is a common complaint that may affect activities of daily living. Facial pain is a subtype of headache that may be very distressing for patients and may prompt medical evaluation [1]. Some patients may have vague and chronic facial pain of unknown cause. These patients may be diagnosed as having atypical or idiopathic facial pain because of limited knowledge about the cause [2,3].

Chronic facial pain may be a challenging diagnostic problem and frequently is misdiagnosed. A multidisciplinary approach, including a detailed history and physical examination, laboratory tests, and diagnostic imaging procedures, may be required to make a diagnosis [4]. We treated a patient who had undiagnosed chronic, atypical facial pain that mimicked migraine headache.

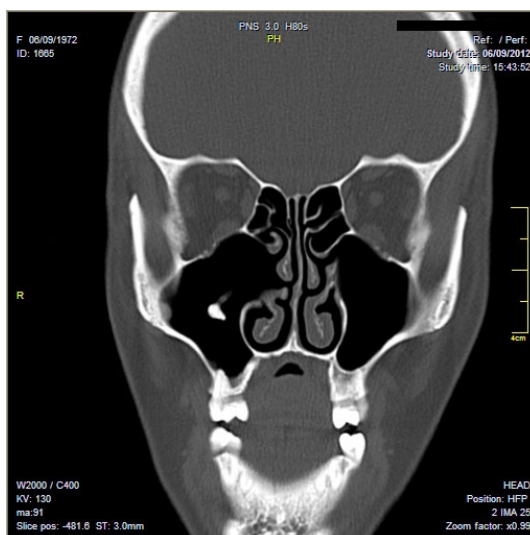
**Case Report**

Sorumlu Yazar: Leyla Kansu, Başkent Üniversitesi Tıp Fakültesi  
Saray mah Yunus Emre cad. No:1 Alanya  
leylakansu@hotmail.com

A 39-year-old woman was evaluated for a 20- to 25-year history of pain on the right side of the face. The pain occurred as sudden attacks of episodic, severe, pulsating pain at the right cheek, above the right upper teeth. The pain attacks typically persisted for 3 hours to 3 days and occurred from 1 to 3 times monthly but did not wake her up from sleep. The pain was associated with nausea but no vomiting or paresthesias. The pain was not aggravated or alleviated by eating, brushing the teeth, hunger, menstruation, or emotional stimulation. Analgesics including antimigraine drugs did not relieve the pain. The past medical history was normal, and there was no history of surgery or facial trauma. The patient did not smoke or use alcohol.

Evaluation included several dental examinations that were normal. General, neurologic and ear, nose, and throat examinations were normal and there were no trigger points about the face. Laboratory tests were normal. Magnetic resonance images of the brain with and without contrast, including thin sequences through the brainstem, were normal.

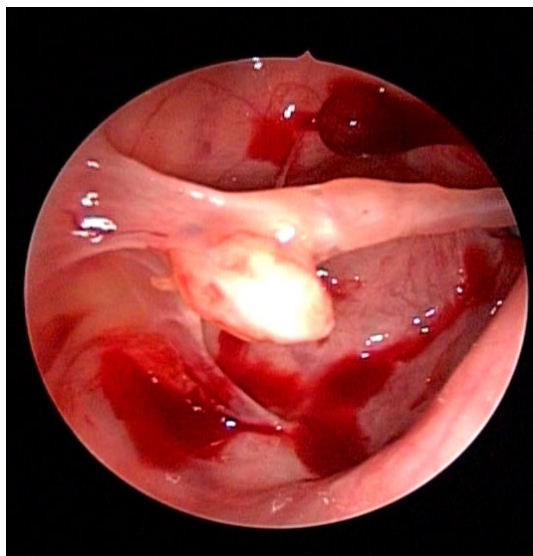
A computed tomography scan of the paranasal sinuses showed a radiopaque lesion and bone defect at the right medial maxillary wall (Figure 1).



**Figure 1**

A computed tomography scan of the paranasal sinuses showed a radiopaque lesion and bone defect at the right medial maxillary wall.

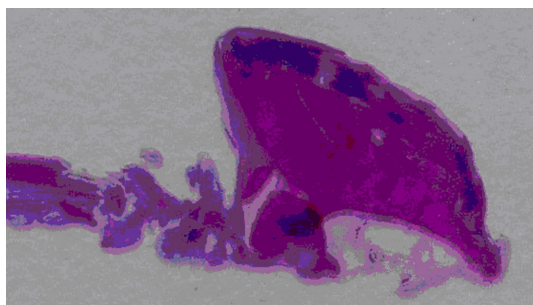
The radiopaque lesion had the appearance of a foreign body of the maxillary sinus, but the patient denied history of previous trauma or surgery. At surgery, nasal endoscopy showed a double middle turbinate and the maxillary sinus ostium was larger than normal. The right uncinate process was remote. Examination with a 70° endoscope showed a small, hard, cream-yellow mass in the right maxillary sinus that was attached by a mucosal bridge to the medial and lateral walls of the maxillary sinus (Figure 2).



**Figure 2**

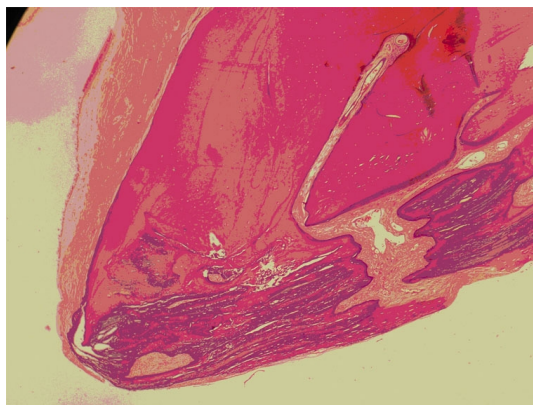
Endoscopic view of the right maxillary sinus showed a small, hard, cream-yellow mass that was attached by a mucosal bridge to the medial and lateral walls of the maxillary sinus.

Antrostomy of the maxillary sinus (Caldwell-Luc procedure) was performed with a trocar above the right canine to the maxillary sinus. The maxillary sinus was visualized with a 0° endoscope, and the mass was noted hanging in the sinus with mucosal strings. The mass was removed totally with Stammberger forceps from the maxillary ostium without damaging nearby structures. The removed mass was 0.8 x 0.5 x 0.5 cm and was hard. It had a cord (length, 1.5 cm; diameter, 0.1 to 0.2 cm) covered with mucosa (Figure 3). Microscopic examination showed chronic inflammation and necrotic bone tissue (diameter, 0.7 x 0.5 cm) that was covered with respiratory epithelium (Figure 4).



**Figure 3**

Macroscopic appearance of the excised mass, including a cord that was covered with mucosa (hematoxylin-eosin, original magnification).



**Figure 4**

Microscopic appearance of the excised mass showing necrotic bone that was covered with respiratory epithelium (hematoxylin-eosin, original magnification x40).

The postoperative course was uneventful. After surgery the chronic facial pain had resolved completely. Evaluation at 20 months after surgery showed that her facial pain had not repeated again

## Discussion

The present patient had chronic facial pain that resolved after excision of a necrotic bone inside the maxillary sinus. There were no aggravating factors for the pain. The cranial nerve function, cervical spine, masticatory and neck muscles, temporomandibular joint, and complete intraoral and dental examination were normal. Computerized tomography of the paranasal sinuses facilitated the diagnosis. Literature review showed no previously published cases of maxillary sinus necrotic bone tissue causing chronic atypical facial pain.

Atypical facial pain of the cheek may be commonly caused by dental problems such as dental caries, infection, and inflammation about the root apex. Patients may have toothache associated with chewing, touch, and percussion. Periapical pathology may cause increased radiolucency on radiographs. Dental pain typically is unilateral, localized, and either intermittent or continuous [3]. The present patient did not have symptoms or signs of chronic dental pain, and dental examination was found normal.

Another common cause of facial pain of the cheek is trigeminal neuralgia. This condition typically is unilateral (bilateral in 4% patients) and causes pain that has the quality of a brief electric shock, limited to the distribution of  $\geq 1$  division of the trigeminal nerve [3]. The pain of trigeminal neuralgia may arise spontaneously or may be elicited by trivial stimuli including washing, shaving, smoking, talking, brushing the teeth, chewing, or talking. The pain may start abruptly and may remit for varied periods [5,6]. Trigeminal neuralgia typically begins between the sixth and seventh decades and is more common in women (female to male ratio, 3:2). The pain may be activated from a trigger zone, typically around the supraorbital or infraorbital foramen, the inner canthus of the eye, the region lateral to the ala, and the mental foramen. The second and third trigeminal nerve divisions are most commonly affected (first division, < 5% patients) [7]. There may be an ipsilateral reflex facial spasm (tic douloureux), and compression of the trigeminal and facial nerves may be implicated. The paroxysmal pain of trigeminal neuralgia may be confused with dental pain, and the patient may have multiple invasive dental procedures before the correct diagnosis is made [3-5]. Spontaneous remission may occur for months or years, and remission  $\geq 6$  months occurs in > 50% patients [7]. The present patient did not have pain or physical findings characteristic of trigeminal neuralgia.

Pretrigeminal neuralgia is a rare cause of facial pain. In this condition, there may be a dull continuous toothache in the upper or lower jaw, but evaluation may show normal neurologic and dental examinations and normal computerized tomography or magnetic resonance imaging scans [8].

Migraine is a highly prevalent, recurrent headache that involves the frontal, temporal, occipital, maxillary, and retro-orbital areas. It is most common in women aged 25 to 55 years. Migraine pain is unilateral, moderate to severe, pulsating, and aggravated by routine physical activity. The pain may be precipitated by stress, alcohol, tyramine-containing foods, menstruation, and bright lights. Migraine attacks persist for 4 to 72 hours and may be accompanied by sensitivity to light or sound, nausea, and vomiting. A positive family history of migraine may suggest a genetic basis for this disorder [9]. In the present patient, the facial pain was confused with migraine because of the patient's pain unilateral and it come with attacks , but there were no aggravating factors and the pain did not resolve with antimigraine therapy.

Episodic tension-type headache is a common primary headache that may affect the occipital, parietal, temporal, or frontal area. The patient may have  $\geq 1$  day but  $< 15$  days of headache per month. The pain typically is bilateral, tightening or pressing, and mild to moderate, and it may persist for several hours or days. In contrast with migraine, episodic tension-type headache is not associated with nausea, vomiting, or aggravation by routine physical activity. The pain may be precipitated by stress and associated with fatigue and poor sleep. Simple analgesics and nonsteroidal anti-inflammatory drugs may be effective in resolving the pain [4]. The present patient did not have episodic tension-type headache because her pain was unilateral, did not respond to analgesics, and had no aggravating factors.

Cluster headache is a rare primary headache that is characterized by unilateral pain attack and ipsilateral autonomic dysfunction. The pain may be localized to the orbital, supraorbital, and/or temporal region. Autonomic features include conjunctival injection, ipsilateral lacrimation, nasal congestion, rhinorrhea, and facial sweating. The pain attacks occur at night and awaken the patient [9].

Chronic paroxysmal hemicrania is a rare type of headache that is characterized by daily, multiple attacks of severe pain and associated autonomic symptoms. The pain is located most commonly in the ocular, temporal, maxillary, and frontal regions. Head flexion or rotation may precipitate the paroxysms in 10% patients [9].

Facial pain also may be caused by tumors. Osteoma is the most frequent benign tumor of the paranasal sinuses [10]. It is a slow growing tumor that becomes symptomatic after becoming large. The most common paranasal sinuses affected, in decreasing order of frequency, are the frontal, ethmoid, maxillary, and sphenoid sinuses. Maxillary osteomas near the trigeminal nerve branches may cause neuralgia [10]. In our patient, histologic examination of the excised mass had not found seem like osteoma that reported by pathologist. Nasopharyngeal carcinoma and cerebrovascular disease also may cause facial pain [1].

Acute sinusitis may cause periorbital pressure and pain at the affected sinuses. Maxillary sinusitis may cause referred pain to the maxillary teeth. The pain may be dull and constant, and the teeth may be sensitive to percussion and may feel extruded. Maxillary sinusitis is associated with malaise, fever, nasal obstruction, and purulent nasal discharge [9]. The present patient did not have physical and radiological findings characteristic of acute sinusitis. We thought that the necrotic bone tissue had fallen into maxillary sinus by pulling mucosal bridges after infection.

Facial pain typically does not cross the midline. Therefore, a complaint of bilateral pain may alert the clinician to the possibility of a psychosomatic cause [1]. Mental disorders have been associated with chronic facial pain including somatoform disorders, factitious disorders, and malingering. A diagnosis of psychogenic pain requires the exclusion of other organic disorders and the fulfillment of specific diagnostic criteria [9]. However, undiagnosed facial pain is caused most commonly by an organic problem that has not been identified.

## Conclusion

The different diagnosis of facial pain is long and includes conditions that are treatable or difficult to treat.

Therefore, when the source of facial pain is unknown from the history or physical examination, further detailed diagnostic evaluation is indicated. A complete examination includes careful evaluation of the cranium, brain, cervical spine, teeth, nose, eyes, throat, sinuses, ears, and chest. In the present patient, computed tomography and endoscopic surgery led to the diagnosis and resolution of symptoms.

## References

1. Quail G. Atypical facial pain- a diagnostic challenge. *Aust Fam Physician* 2005; 34:641-5.
2. Graff-Radford SB. Facial pain. *Curr Opin Neurol* 2000;13:291-6.
3. Graff-Radford SB. Facial pain. *Neurologist* 2009;15:71-7.
4. Sarlani E, Balciunas BA, Grace EG. Orofacial pain - Part I: Assessment and management of musculoskeletal and neuropathic causes. *AACN Clin Issues* 2005;16:333-46.
5. Evans RW, Graff-Radford SB, Bassiur JP. Pretrigeminal neuralgia. *Headache* 2005;45: 242-4.
6. Zakrzewska JM. Facial pain: neurological and non-neurological. *J Neurol Neurosurg Psychiatry* 2002;72(Suppl 2):ii27-ii32.
7. Hentschel K, Capobianco DJ, Dodick DW. Facial pain. *Neurologist* 2005;11:244-9.
8. Symonds C. Facial pain. *Ann R Coll Surg Engl* 1949;4: 206-12.
9. Sarlani E, Balciunas BA, Grace EG. Orofacial pain - Part II: Assessment and management of vascular, neurovascular, idiopathic, secondary, and psychogenic causes. *AACN Clin Issues* 2005;16:347-58.
10. Kayaci S ve ark. Primary osteoma of the orbit with atypical facial pain: case report and literature review. *Turk Neurosurg* 2012;22:389-92.