

Nighttime Facial Pain Induced by Myxoma of the Maxillary Sinus: A Case Report

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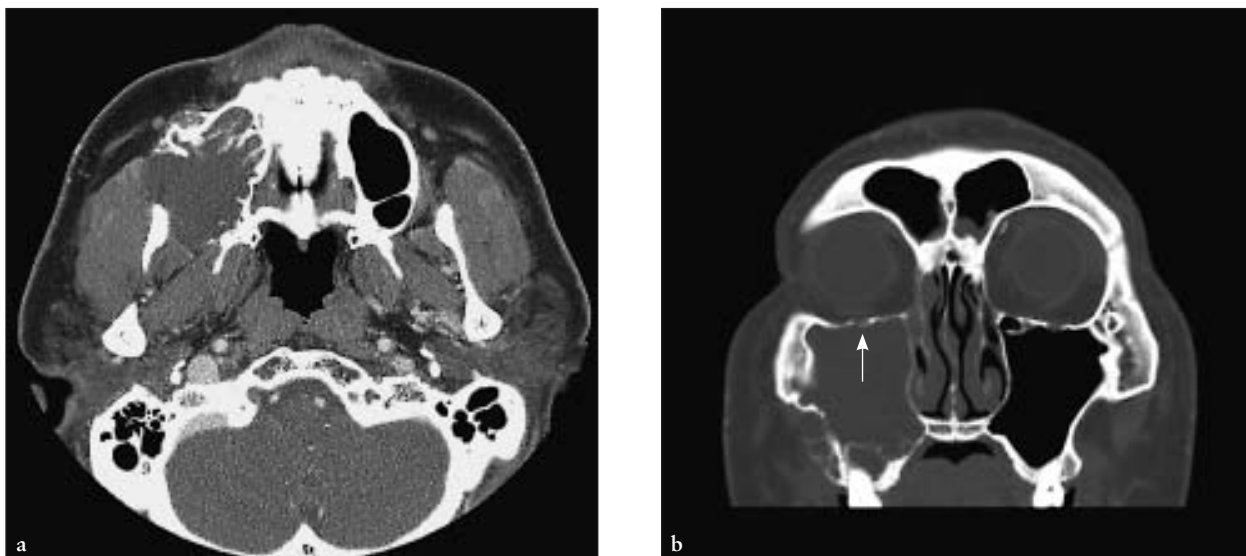
Myxomas of the maxillary sinus are uncommon benign tumors arising from the primitive mesenchyme. They may be related to dental malformations or missing teeth but may also occur without any abnormalities. They usually result in facial deformity manifested by slow, painless bony expansion. Although these tumors are well-circumscribed, they are more extensive than they appear due to their local aggressiveness and bone erosion. Therefore, it should be widely resected with maximal preservation of surrounding structures to prevent recurrence. This article reports a case of myxoma of the maxillary sinus and nighttime facial pain in a 33-year-old male who underwent curettage following debridement with a microdebrider via an intranasal endoscopic approach and surgery involving a modified Caldwell-Luc approach. J OROFAC PAIN 2011;25:170–173

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Myxoma of the maxillary sinus is a very rare mesenchymal tumor usually related to a sunken tooth arising from the mesenchymal tissue of the teeth.¹ In general, they are painless and grow slowly without specific clinical findings, so they are usually found to invade surrounding tissues and cause facial deformity by the time the patient visits the hospital.² They are histologically benign with clear margins, but they commonly invade the surrounding tissues, and wide resection is preferred due to risk of recurrence if they are not completely removed.³ The following describes an unusual case of a patient with nighttime facial pain in the right maxillary area with radiation to the right jaw induced by myxoma of the maxillary sinus and who underwent curettage via intranasal endoscopic approach and modified Caldwell-Luc approach.

Case Report

A 33-year-old male patient visited the otorhinolaryngology outpatient clinic. He complained of intermittent right facial pain worsening toward the night over the past 4 months and toothache arising after chewing food for the past 2 months. He had been to a neurology clinic and a dental clinic but was referred to the otorhinolaryngology clinic since he showed normal functions of the cranial nervous system and a panoramic radiograph showed no abnormality except for increased haziness in the right maxillary sinus. The patient described his initial pain as tender and sharp in the right maxilla premolar region that radiated to the right jaw. Eventually, he developed pain in his frontal and temporal areas. There were associated



Figs 1a and 1b Axial (*a*) and coronal (*b*) CT scans show a mass in the right antrum, without enhancement. Note destruction of infraorbital canal (*white arrow*).



Fig 2 MRI scans in axial view. On T1-weighted MRI (*a*), the mass shows lower signal compared to the adjacent tissue. On T2-weighted MRI (*b*), it shows higher lobulated signal. After gadolinium enhanced T1-weighted MRI (*c*), the mass is visualized with mild heterogeneous enhancement.

symptoms of right nasal obstruction and swelling of the isthmus 1 month prior to his visit, but he did not present any other nasal symptoms. There was no specific trigger exacerbating the patient's symptoms, but he suffered facial pain when he laid down to sleep. He was prescribed various nonsteroidal anti-inflammatory drugs (NSAIDs), but these medications provided relief only for a short period of time. The patient suffered facial trauma after falling down the stairs 6 months ago but had not received treatment since the injuries were minor. There was no family history or operation records. On physical examination, there was redness and swelling measuring 4×4 cm on the right maxilla with a solid mass caus-

ing pain on palpation. However, there were no specific findings in the nasal cavity or the nasopharynx. Computed tomography (CT) of the sinus showed a mass in the right maxillary sinus invading the surrounding tissues, and there was bone destruction of the posterolateral wall of the maxilla as well as of the infraorbital canal (Fig 1). T1-weighted magnetic resonance imaging (MRI) showed lower enhancement compared to surrounding structures and T2-weighted MRI showed a lobulated, highly-enhanced mass. In gadolinium enhanced T1-weighted MRI, there was partial enhancement of heterogeneous mass (Fig 2).

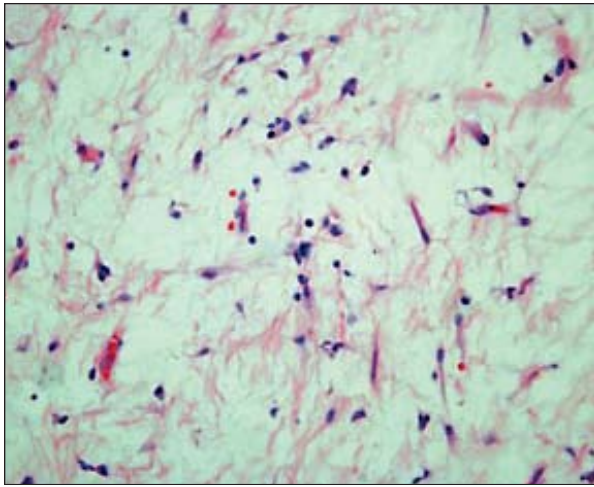


Fig 3 Histologic section of the tumor showing abundant mucoid ground substance between stellate and spindle-shaped cells (hematoxylin & eosin stain, original magnification $\times 400$).

Incisional biopsy with transseptal approach under general anesthesia was performed, and after confirming that the mass was a benign tumor suggestive of myxoma, the mass was removed by curettage via an intranasal endoscopic approach and surgery involving a modified Caldwell-Luc approach. The mass was removed with maximal preservation of the surrounding tissues by using a microdebrider on the invaded soft tissues and bone tissues. Incisional biopsy of the mucosa of inferior, posterior periosteum and muscle tissues during surgery confirmed no remnant mass.

Histologic findings confirmed that the mass was a myxoma composed of slightly basophilic, unevenly lined spindle cells in a low-density mucous substance (Fig 3). The patient was discharged 3 days after the operation without any complications. The patient had resolution of his symptoms within a few days, which confirmed his diagnosis as myxoma. For the 24 months of follow-up since the operation, there has been no use of medication, and the patient has been free of pain in his right maxilla and has normal mastication skills. Also, observing the maxillary sinus by middle meatal antrostomy showed no signs of recurrence, showing complete healing of the jaws.

Discussion

Myxoma is a mesenchymal tumor arising from the tooth and is composed of spindle cells in the mucous stroma.¹ Its etiology remains unclear and its occurrence in the head and neck region is very uncommon.² It usually arises in the 20- to 30-year-old age group

with no difference in sex.^{1,2} Myxomas are thought to originate from the mesoderm since they usually occur from dental defects.³ Also, they occur mostly in teeth, and the presence of enamel, dentin, cementum, pulp, and periodontal ligament in the mass supports the view of their dental origin.^{3,4}

Since a myxoma of the maxillary sinus grows slowly without specific clinical findings, the mass may be big enough to cause facial bone deformity by the time a diagnosis is made.² It may cause nasal obstruction, swelling of the palate, and pain may not always be present.^{2,4} The patient in the present case complained of intermittent right facial pain worsening toward the night and toothache arising as a result of chewing food. The pain was eventually relieved with NSAIDs. The pain is thought to originate from the stimulation of nerves after destruction of the infraorbital canal, and pain occurring at night may be due to the pressure effects of a dependent position, ie, the tilting of the head to one side of the body when the patient lies down to sleep.

Invasiveness and destruction of the cortical bone may be shown through CT findings, and low enhancement in T1-weighted MRI scans with high enhancement in T2-weighted MRI scans showing a homogeneous mass help to differentiate margins from surrounding normal tissues.⁵ Clinically and histologically, myxomas should be differentiated from odontogenic fibromas, which are mesenchymal odontogenic tumors. Odontogenic fibromas are characterized as usually arising in the young and adolescent age groups, originating from the mandible, and being constituted of fibrous collagenous tissues.⁶ Diagnosis is confirmed by the histological documentation of spindle cells and astrocytes in mucous tissues composed of collagenous fibrils that react with vimentin and actin in immunohistochemical staining.⁴

Although there is no standard treatment, radical excision of the tumor is preferred due to local invasion and risk of recurrence if it is not removed completely.³ However, careful consideration should be taken since radical excision not only causes cosmetic problems, but also deterioration of speech and masticatory skills as well.⁴ In the present case, the mass was removed by curettage via an intranasal endoscopic approach and a modified Caldwell-Luc surgical approach that made a prompt recovery possible and that, by direct access to the mass, spared normal tissues and produced satisfactory cosmetic results. Since myxomas are benign tumors occurring at young ages, radiotherapy does not seem warranted because it may be completely treated by surgical removal.⁷ Myxomas are known to recur in less than 2 years, so it is imperative to carefully monitor the patient for evidence of recurrence.⁴

In conclusion, myxomas arising in the maxillary sinus are very uncommon, and their delayed clinical manifestations make diagnosis difficult. However, they should be differentiated from other diseases that cause progressive intermittent facial pain and toothache arising as a result of mastication.

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