# Toothache Induced by an Angioleiomyoma of the Nasolabial Groove: A Case Report

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*Angioleiomyoma, a benign soft tissue tumor composed of smooth muscle cells and vascular endothelium, occurs most commonly in the extremities, the lower leg being a common site of occurrence. It rarely is found in the head and neck area, especially in the nasolabial groove. Surgical excision is the gold standard for diagnosis and treatment of angioleiomyoma; a preoperative diagnosis may be difficult. Here, a case of angioleiomyoma found in the nasolabial groove and associated with toothache is presented.* J OROFAC PAIN 2011;25:75–78

**Key words:** angioleiomyoma, nasolabial groove

The most common primary causes of toothaches include dental caries, pulpitis, apical periodontitis, and periapical abscess. Sinusitis from bacterial or fungal infection, postoperative cysts of the cheek, odontogenic cysts, and oroantral fistulas are also known to cause toothaches. However, in rare cases, a tumor may be the underlying cause.[1] An angioleiomyoma can develop in any smooth muscle cell; however, they rarely occur in the area of the head and neck.[2] Angioleiomyomas are painful subcutaneous nodules usually less than 2 cm in size that progress slowly; their size and progression depends on their blood supply.[3] Here, a case is presented of toothache associated with an angioleiomyoma of the nasolabial groove.

## Case Report

A 53-year-old male patient presented to the otorhinolaryngology outpatient clinic with a 3-year history of an intermittent left toothache worsening at night and a 6-month history of pain with mastication. He had previously been to a dental clinic and a neurology clinic but was referred to otorhinolaryngology because no abnormalities were previously detected. The initial pain was described as sharp with varying degrees of intensity; the pain started in the left maxillary premolar region and radiated to the left temporal areas. Specific triggers exacerbating the patient's symptoms included mastication with food and brushing of the teeth. The patient was prescribed a variety of nonsteroidal anti-inflammatory drugs (NSAIDs), but these medications failed to ease the patient's pain. The patient reported facial trauma after falling down the stairs 5 years previously but did not seek treatment because of the minor injuries. There was no other significant family or medical history. On physical examination, there was a solid

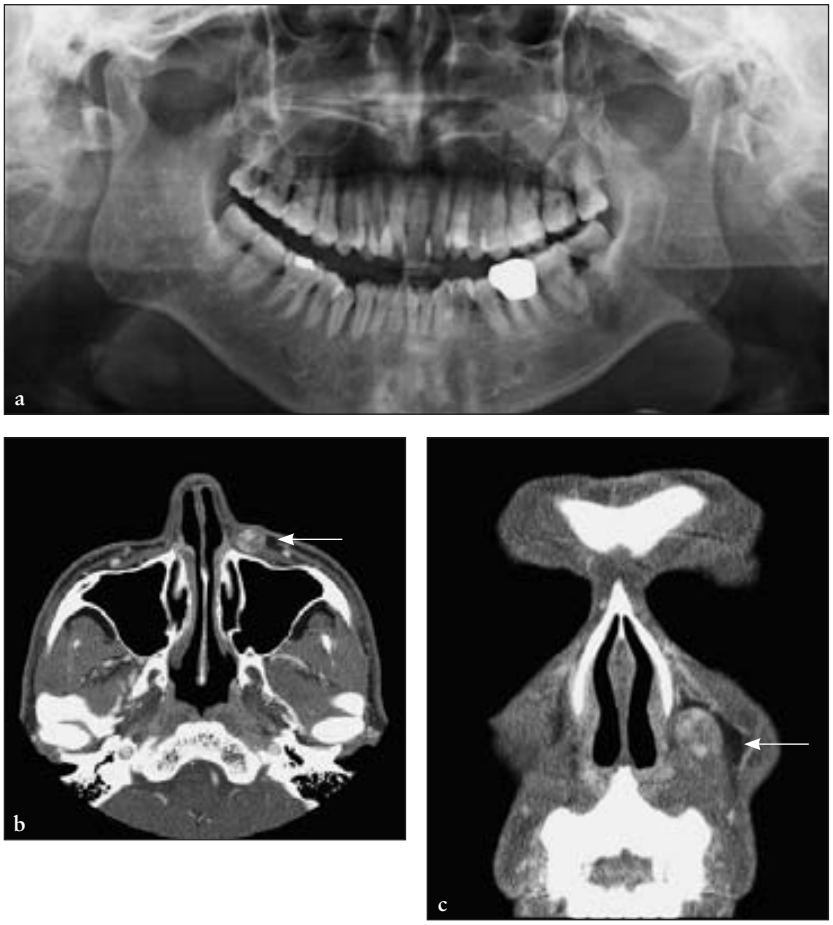


**Fig 1** Panoramic radiograph and computed tomography scan. *(a)* There were no abnormalities. *(b)* A 1.4 × 1.1 × 0.9 cm heterogeneously enhancing mass at the left nasolabial groove *(white arrow)*. *(c)* The base of the mass was adhered between the levator labii superioris and the levator labii superioris alaeque nasi muscles *(white arrow)*.

mass fixed to its nearby structures that measured 1 × 1 cm in the left nasolabial groove and that caused pain when palpated. A panoramic radiograph of the dental area showed no abnormality (Fig 1a), and consultation with a dentist resulted in no specific head and neck, temporomandibular joint, or dental findings. Only blood and inflammatory cells were found on the fine needle aspiration biopsy (FNAB) performed in the office. Computed tomography (CT) of the sinuses showed a round, heterogeneous, and irregularly enhancing mass in the left nasolabial groove that measured 1.4 × 1.1 × 0.9 cm and was fixed to the nearby muscles (Figs 1b and 1c).

The patient had the nasolabial groove mass removed by the gingivobuccal approach. During surgery, there was significant bleeding since the base of the mass adhered between the levator labii superioris muscle and the levator labii superioris alaeque nasi muscles. The bleeding was controlled by cauterization, and the rest of the lesion was removed without complication. The removed mass was oval in shape and almost completely encapsulated. The base of the mass had an irregular margin due to the adherence to muscles. Light microscopy examination revealed thickened blood vessel walls and irregular benign proliferation of leiomyofibers, confirming the diagnosis of an angioleiomyoma (Figs 2a and 2b).

The patient was discharged 3 days after surgery with resolution of the toothache by 2 weeks after the procedure. The postoperative course was uneventful and the patient had no recurrence of the mass or complications during 1 year of follow-up.

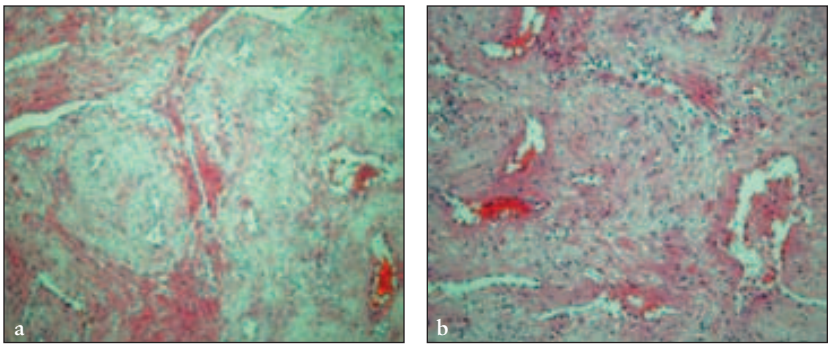


**Fig 2** Microscopic findings. *(a)* The mass was composed of smooth muscle bundles that surrounded the vascular channels (H-E; ×100). *(b)* The mass was composed of thick-walled vessels with partially patent lumens. The wall of the vessels showed marked proliferation of cigar-shaped spindle cells (H-E; ×200).

## Discussion

According to the World Health Organization classification of soft tissue lesions, leiomyomas are classified as simple leiomyomas, angioleiomyomas, and leiomyoblastomas.[4] Angioleiomyomas account for 5% of all benign neoplasms of the soft tissue and can occur wherever smooth muscle cells are present.[2] They are more common in women during the fourth to sixth decades of life and frequently are diagnosed in the lower extremities.[2] They are relatively rare in the head and neck area, with only 8.5 to 10% of reported cases occurring in the head and neck region.[3]

The etiology of angioleiomyomas continues to be debated; however, it is widely thought that they originate from the smooth muscle cells in the walls of vascular channels.[5] Some have suggested smooth muscle proliferation from an angioma, and others suggest that these lesions result from infiltration of chronic inflammatory cells associated with trauma, venous stasis, and hormonal changes, especially related to progesterone.[6] In this case, the trauma that occurred 5 years previously may have played a major role. However, the pathogenesis remains unclear.

Pain is the most striking clinical feature of an angioleiomyoma.[5] The pain is often paroxysmal and provoked by exposure to cold air.[3] The pain is thought to be due to the active contraction of smooth muscle that results in local ischemia.[6] The patient presented here had toothache that was worse at night and after mastication. The patient was incorrectly diagnosed by the local neurology clinic as having trigeminal neuralgia because the symptoms were relieved after taking carbamazepine.

The diagnosis of an angioleiomyoma is made using FNAB, ultrasound, CT, or magnetic resonance imaging (MRI). However, in cases that develop in the nasolabial groove, the diagnosis may be difficult. First, angioleiomyomas are very rare compared to other diseases that cause toothaches. Secondly, they show irregular signal density in the CT or MRI. The histological findings show a well-defined border surrounded by a fibrous capsule.[7] They are composed of muscle bundles that are surrounded by vascular channels.[8] Spindle-shaped cells are set in bundles and coils surrounded by numerous vessels with cigar-shaped nuclei.[9] Angioleiomyoma found in deep connective tissue are histologically variable, with changes including fibrosis, giant cell reactions, and accumulation of mucous lesions.[7] Hematoxylin and eosin (H-E) staining may be used to confirm the diagnosis; however, it may not differentiate vascular muscle cells from myopericytes.[9] Immunohistochemical staining using antibodies such as SMA (α-smooth muscle stain), HHF35 (muscle-specific actin), CALP (calponin), and h-CD (h-caldesmon) help confirm the diagnosis.[9] In the present case, the diagnosis was confirmed by H-E staining, which showed proliferation of vascular smooth muscle cells, a unique feature of angioleiomyomas.

The treatment of angioleiomyoma is complete surgical excision of the mass.[2,5] The nasolabial groove mass was successfully removed in the present case by the gingivobuccal approach. The mass was completely removed without any difficulty, and the patient's symptoms resolved. The prognosis for these lesions is favorable, and recurrence is rare after early detection and complete excision.[2] The recurrence rate in the head and neck area is reported to be less than 5%,[9] and as in this case, after 1 year, there is usually no recurrence.

Angioleiomyomas arising in the nasolabial groove are rare, and the nonspecific clinical manifestations make the diagnosis difficult. However, they should be included in the differential diagnosis of toothaches.

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The editors of the *Journal of Orofacial Pain* gratefully acknowledge the efforts put forth by the reviewers in 2010.

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