

CASE REPORT

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A rare case of oral cavity lipoma in the inferior vestibule: a case report and literature review

O. Benhoummad , K. Cherrabi* , N. El Orfi and S. Berrichou

Abstract

Background: Lipomas are ubiquitous tumors. They are mostly present in the trunk as well as the proximal portion of the limbs. However, 15–20% are located in the head and neck regions, and only 4.4% are found in the oral cavity. They normally present as a bulge with signs of compression, without signs of infiltration.

Case presentation: This is the rare case of a 36-year-old man, who was admitted to ENT consultation for a bulge in the left inferior vestibule of the oral cavity. The bulging was not painful, with no inflammatory signs, no dental mobility, or abnormalities in the vestibular mucosa. No other masses were found in the general clinical exam. The patient underwent a dental panoramic X-ray, which showed a low opacity lesion next to the left inferior vestibular space, without signs of bone invasion. There was no hypoesthesia in the territory of the alveolar nerve was found. The patient underwent a craniofacial MRI which showed a mass which was hypointense on T1 and hyperintense on T2. Fine needle aspiration was not performed. The patient's pre-operative blood work showed no abnormalities. He underwent surgical excision under general anesthesia. The incision was carried out in the left inferior vestibular space, 1 cm away from the alveolar gingiva; careful dissection of the mucosa allowed the discovery of a large fatty mass, encapsulated, with a nourishing pedicle, without signs of invasion or ulceration in the adjacent structures. Careful dissection and hemostasis were performed, followed with the closure of the mucosa with Vicryl 4.0. The histopathological exam showed mature adipose tissue. The fat contains few small capillaries within thin fibrous strands. A thin fibrous capsule was seen. Clinical follow-up of 2 months showed no complications. The surgical incision healed perfectly, and no bulging was noticed upon palpation.

Conclusions: This is the rare case of a vestibular lipoma of the oral cavity; the clinical and radiological aspect allowed consideration of several differential diagnoses. The most important take away lesson is related to the very rich variety of differential diagnosis in oral cavity masses. Clinical and radiological aspects allow the clinician to have diagnostic theories. Only histological results allow to establish a definitive diagnosis.

Keywords: Oral cavity, Lipoma, Clinical, Radiology, Histology, Diagnosis, Treatment

Background

Lipomas are ubiquitous benign mesenchymal tumors composed of mature adipocytes [1–8]. They are mostly present in the trunk as well as the proximal portion of the limbs [3]. However, only 15–20% are located in the head

and neck regions [4, 5], and only 4.4% are found in the oral cavity [2–5, 8]. It appears as a bulge covered by normal oral mucosa [5, 8]. They are generally presented as a bulge without signs of extension to adjacent structures; in the oral cavity, they can be responsible for problems in swallowing and speech [2–5].

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Case presentation

This is a rare case of a vestibular lipoma in the oral cavity, in a 36-year-old man, who was admitted in ENT consultation for a bulge in the left inferior vestibule of the oral cavity. The bulge was not painful, no inflammatory signs, no dental mobility, or abnormalities in the vestibular mucosa. No hypoesthesia in the territory of the alveolar nerve was found. No other masses were found in the general clinical exam. The patient underwent a dental panoramic X-ray, which showed a low opacity lesion next to the left inferior vestibular space.

The patient underwent a craniofacial MRI which showed a mass which was hyperintense on T2 and hypointense on T1. Fine needle aspiration was not performed.

The patient's pre-operative blood work showed no abnormalities (Figs. 1, 2, and 3). He underwent surgical excision under general anesthesia.

The incision was carried out in the left inferior vestibular space, 1 cm away from the alveolar gingiva; careful dissection of the mucosa allowed the discovery of a large fatty mass, encapsulated, with a nourishing pedicle, without signs of invasion or ulceration in the adjacent structures. Careful dissection and hemostasis were performed, followed by closure of the mucosa with Vicryl 4.0.

The histopathological exam showed mature adipose tissue. The fat contains few small capillaries within thin fibrous strands. A thin fibrous capsule was seen.

A clinical follow-up of 2 months showed no complications. The surgical incision healed perfectly, and no bulging was noticed upon palpation.

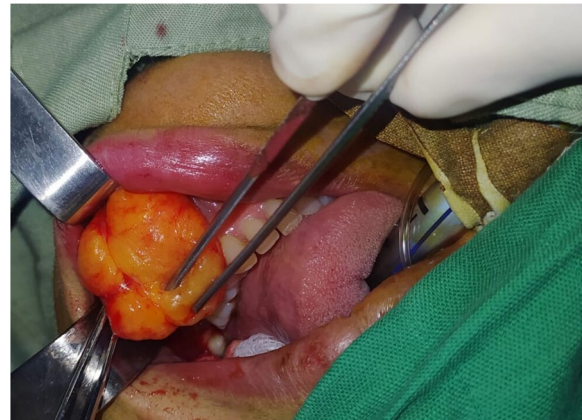


Fig. 2 Per-operative image showing the extirpation of a lipoma of the inferior oral vestibule

Conclusion

Oral cavity lipomas are very rare. Although their diagnosis is confronted with several differentials (Table 1), their management is very simple and the outcome is generally very good.

Discussion

The first case of lipoma was described in 1848 by Roux [2, 8]; the age of onset is between 40 and 60 years old [1–3, 5, 7, 8]. They are very rare in children [4, 7]

Studies have suggested that there is no gender predilection. However, some series suggested that lipomas are generally predominant in males [1, 5, 7, 8]. Oral localizations are favorably found in female patients [4, 7].

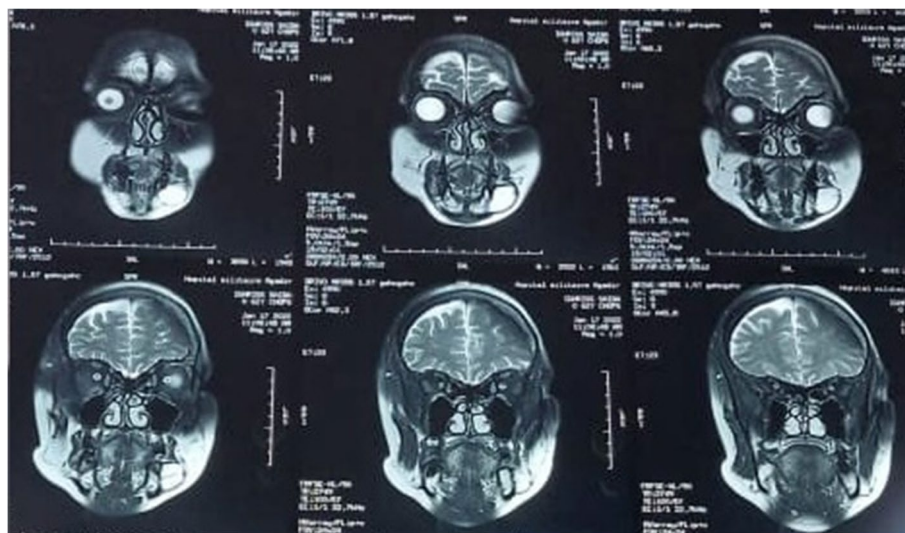


Fig. 1 MRI sections showing T2 hyper-intense lesions, in the left vestibular region

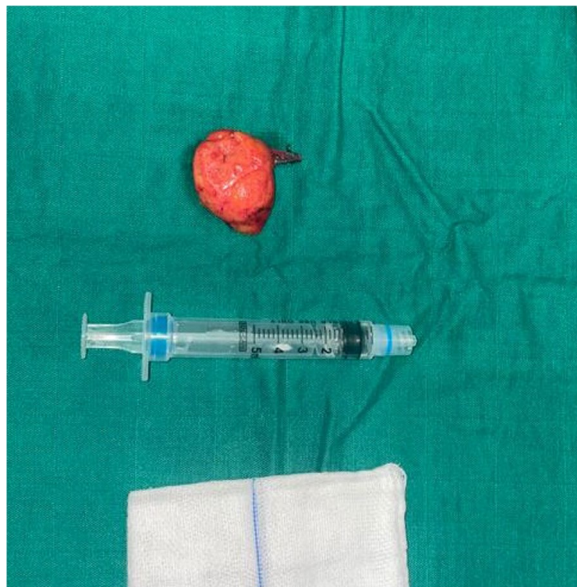


Fig. 3 Post-operative specimen

The most frequent mode of presentation is a slow-growing mass, covered by healthy mucosa, without any signs of invasion [1, 3, 5].

Lipomas of the oral cavity are most frequently localized in the buccal mucosa, especially in the cheek (50%) [1, 4] (Table 2). followed by other localizations such as the tongue, lip, floor of the mouth, buccal vestibule, palate, and retro-molar area [1, 2, 4, 5, 7].

The size usually fluctuates between 0.8 and 2.6 cm [3, 5, 7, 8]. Lipomas are rarely superior to 25 mm [1, 7]. Superficial presentations are generally yellowish well circumscribed, with a smooth nodular surface, that could be either peduncular or sessile. While profound forms are infiltrating, rarely typically yellowish and could present diagnostic challenge [4, 5].

Lipomas are generally clinically asymptomatic and slowly grows, until it manifests with signs of compression [1, 8]. Could present as solitary or multiple lesions [1, 8]. Multiple lesions could be associated with syndromes like Gardner's syndrome, Decrum's disease, multiple familial

Table 1 Differential diagnosis of lipoma of the vestibular region in the oral cavity [1–5, 8]

	Age	Gender	Most frequent location	Clinical feature	Histological aspect
Fibroma	> 20 years old	Same incidence	- Mucosa - Tongue	Pink, firm, could be ulcerated	Stratified squamous lining covering dense connective tissue
Lymph-angioma	Congenital Mostly children	Same incidence	- Anterior 2/3 of the tongue - Center of the tongue - Buccal mucosa	Hypo-chromatic sometimes purple mass Could cause difficulty swallowing and interfere in phonation	Dilation of lymphatic vessels
Rhabdo-myosarcoma	Fourth and fifth decades	More in females	Lip, tongue, buccal mucosa	Slow growing, asymptomatic, firm	Numerous thin-walled vessels Muscle bundles
Neuroma	Second to fifth decades	Same incidence	Tongue, palate buccal mucosa	Semi-elastic consistency, firm, mobile, painful on palpation	Remanence of nerve endings Double cell distribution histological pattern: Antoni A, Antoni B
Dermoid cyst	Congenital	Same incidence	Tongue, lip, palate	Mobile slow-growing, painless Problems in speech and swallowing	Keratinized stratified squamous epithelioma Follicles, sweat, and sebaceous glands
Benign tumors of minor glands	Fourth decade	Female more than male	Palate	Mobile, slow-growing, and painless Isolated lesion	Histological variable Epithelial component Large number of patterns
Mucocele	First and second decade	Same incidence	Lip, buccal mucosa, floor of the mouth	Sessile, fluctuating, painless, rapidly growing mass	Cyst wall without epithelial lining
Hemangioma	Congenital (children)	Same incidence	Lip Tongue Buccal mucosa	Reddish or bluish lesion, the compression of the lesion produces whitish aspect which could lead to ischemia	Capillary hemangioma. (capillary of endothelial origin): superficial Cavernous hemangioma, poorly defined Mixed hemangioma

lipomatosis, encephalo-cranio-cutaneous lipomatosis, Pai syndrome, and neurofibromatosis [5].

Etiopathogenesis

While a definitive etiology of lipoma formation remains unclear [2, 5, 6, 8], two theories have been associated with the formation of lipomas [2, 5, 6, 8].

The hypertrophy theory stipulates that lipomas are advertent growths of adipose tissue; it explains the formation of lipomas by the onset of obesity and the adipose tissue associated with it [2, 5, 6, 8].

The metaplasia theory stipulates that lipomas are due to aberrant differentiation of mesenchymal cells into lipoblasts [2, 5, 6, 8].

Besides, a congenital origin has been suspected where embryogenic cells differentiate into lipomas under hormonal influence [6].

Several factors have been associated with the onset of oral cavity lipomas: obesity, hereditary, injuries, infection, muscle cell metaplasia, residual embryonic fat cells, chronic irritation, fatty degeneration, and hormonal abnormalities [2–4, 6, 8].

Several differentials could be lymphoepithelial cysts, epidermoid cysts, dermoid cysts, and granular cell tumors, neurofibroma, traumatic fibroma, and salivary gland lesions such as mucocele and mixed tumors [1–5, 8].

Clinical aspect of lipomas generally allows diagnosis. However, association with X-ray and sonography is necessary for unusual localizations, sometimes even CT scan and MRI are necessary to rule out more frequent lesions [4]. Histological aspect remains the gold standard to establish a definitive diagnosis [4, 5].

Several histological patterns depend on the histological subtype [1–5] Table 2.

Macroscopically, lipomas are generally made of benign mesenchymal neoplasms composed of fatty cells, generally surrounded by a thin fibrous capsule. The following are the different histological sub-types: simple lipoma, spindle cell lipoma, fibro-lipoma, angiolipoma,

polymorphic myxoid lipoma, and atypical lipomas [1, 2, 4, 6, 8]. Sialo-lipoma and chondro-lipomas are very rarely reported in the literature [4–6, 8].

Liposarcomas are rare in the oral cavity [1].

Proliferating cell nuclear antigen (PCNA) and ki67 are found in lipomas [5]. PCNA could suggest faster-growing lipomas [5]. Microscopical aspect is a circumscribed but not encapsulated aggregate of mature adipocytes with large and clear cytoplasm, without vascularity [1].

Spindle cell lipomas are related to 13q or 16q chromosome abnormalities [6, 8].

Treatment

Surgical excision is the treatment of choice, as a complete excision allows to bring recurrence rate to very low values. Infiltrating lipomas are very hard to extirpate [1, 8]. Recurrence after complete excision is rare; infiltrative lipomas have more recurrence rates as they are not encapsulated [8].

Secondary atrophy can be spotted and should not be confused with malignant degeneration [5]. No incidence of malignant transformation has been cited [5, 8].

Medical treatment based on steroid injection could be recommended for spindle cell lipomas inferior to 2.5 cm [8].

Conclusions

Differential diagnosis of oral cavity benign masses is diverse; lipomas are very rare in this localization. However, their management is very simple, and the outcome is generally very good.

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Authors' contributions

O.B performed the surgery. K.C contributed to the clinical management and investigation and analyzed and interpreted the patient data. NO contributed to the clinical and surgical management of the patient. SB contributed to the clinical management of the patient. All authors have read and agreed to its content. The authors read and approved the final manuscript.

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Declarations

Ethics and consent to participate

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Consent for publication

The subject provided written consent for participation and publication of the case report.

Competing interests

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Table 2 Histological variations of lipomas of the oral cavity [1–5]

Histological sub-types of lipomas of the oral cavity

- Simple lipoma
- Fibro-lipoma
- Spindle cell lipoma
- Intra-muscular lipoma
- Chondro-lipoma
- Pleomorphic lipoma
- Myxoid lipoma
- Angiolipoma
- Sialo-lipoma

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