Rare Fibrolipoma of Attached Gingiva: A Case Report and Review of the Literature

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Abstract

Fibrolipoma is a variant of lipoma that is relatively uncommon in the mouth tissues, especially the attached gingiva. It is diagnosed by histopathologic and immunohistochemistry evaluation and treated by total excision. A case of a patient with attached gingiva fibrolipoma, treated by surgical excision under local anesthesia is reported.

Introduction

Although lipoma (a benign soft tissue neoplasm) is common generally, it has a very low rate of incidence in the oral and pharyngeal region. Lipoma has numbers of variants: angio-lipoma, fibrolipoma, chondrolipoma, osteolipoma/chondrolipoma, adenolipoma, perineural lipoma, and myxoid lipoma [1,2]. Fibrolipoma (FL) is a histological subtype of lipoma that can be identified by a fibrous component that is mixed with adipose tissue lobules. Its consistency depends on the amount and distribution of fibrous tissue and the tumor depth. It may vary from soft to firm. The etiopathogeneses of lipoma and fibrolipoma remain unknown, but there are three possible reasons for the appearance of these lesions: it can be a congenital lesion due to the lack of endocrinal balance, which can arise with degeneration of a fibromatous tumor, or maturation of lipoblastomatosis. Moreover, mild trauma may cause adipose tissue proliferation; also, fibrolipoma can form beneath a complete denture. Magnetic resonance imaging (MRI) may be useful to diagnose the types of oral cavity lesions that are raised from adipose tissue. In general, lipoma displays high signal intensity and appears to be well-encapsulated masses on both T1- and T2-weighted images. Immunohistochemically can help to diagnose fibrolipoma by evaluating the expression rate of proliferating cell nuclear antigen (PCNA) and Ki-67. The expression of Ki-67 expression may indicate malignancy or recurrence. Fibrolipoma can show higher Ki-67 expression than classical lipoma and other variants of lipoma. Surgical excision must be operated to treat fibrolipoma. The prognosis of this type of lesion is generally favorable; if the surgery is performed well, it is not likely for this lesion to return. A follow-up must be considered. It can appear in all ages; although, it is mostly diagnosed in 40-60 years old patients. These lesions have a mean diameter of 2 centimeters (cm) in the oral cavity.

The lipoma of the mouth was described by Roux in 1848 as “yellow epulis” firstly. Among all benign oral lesions, oral lipoma has an incidence rate of about 1–4% and a prevalence rate of approximately 0.0002%. The review of English literature demonstrated a variable distribution of oral lipomas; however, about 50% of them were on the buccal mucosa. Other 50% of the oral lipomas were diagnosed in the tongue, floor of the mouth, lips, palate, and gingiva [1,2]. FL is a highly uncommon variation of lipoma and contains about 1.6% of all facial lipomas [3]. FL of the oral cavity has been infrequently reported. To the best of our knowledge, the review of the literature revealed a total of 33 cases of intraoral FL till now (Figure 1) [4].

As this lesion does not have any pain and grows slowly in the oral cavity, it is hard to clinically evaluate its

true incidence rate. Patients report the lesion to the dentist only when it turns symptomatic, for esthetics, or oral function.

Different studies were explaining their cases due to their rarity: Pereira reported a rare histologic variant of FL on the lingual marginal gingiva of the mandibular left third molar of a 35-year-old female patient in 2014 in India. Iaconetta also reported a rare FL of the tongue on the ventral surface of the tongue of a 71-year-old female patient in 2015 in Italy. Furthermore, Castellani reported a rare case of intraosseous fibrolipoma of the mandible in a 25-year-old female patient in 2015 in Italy. All these three cases were important to be reported because of the rareness of FL in the oral cavity and the site of FL in each of these presented cases.

As mentioned above, FL in the oral cavity is a rare case. In this paper, a case of gingival FL will be analyzed and its clinic and pathological features along with the patient management and follow-up will be discussed.

Case Description and Results

A 26-year-old woman, without any history of drug usage, was referred to Shiraz Oral and Maxillofacial Medicine department from the Periodontal department with a chief complaint of left lower attached gingiva swelling. Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy. The swelling had first been noticed two years earlier and had subsequently exhibited gradual, continuous enlargement. There was no pain or bleeding. The exophytic lesion was a dome-shaped base, smooth surface, non-homogenous color (pale pink-red and somewhere yellow), homogenous texture, soft in palpation but not fluctuant or mobile on the left lower gingiva next to the first and second mandibular molars (Figure 3). Its total measuring was $1.5 \times 1 \times 0.7$ cm. She had no medical problems and no familial history due to similar lesions. We asked our patient several clinical questions about the lesion's pattern of growth, general pain, bleeding, time of lesion existence, trauma, and fever; as already mentioned, it appeared two years ago and had a gradual enlargement, there was no evidence of pain, bleeding, trauma or fever. We operated some clinical and paraclinical examinations such as palpation, examination of other parts of her mouth, lymphadenopathy, aspiration, vitality test, probing, and periapical radiography. There was no other lesion in her mouth similar to our studied lesion, the aspiration was negative, teeth adjacent to the lesion were vital and did not have any periodontal problems. Regarding the differential diagnosis, the exophytic lesion could be reactive or tumoral; a reactive lesion was ruled out as there was no trauma or stimulating factor based on the patient's history; also, teeth adjacent to the lesion were vital. Therefore, the lesion could be tumoral: due to its continuous enlargement and lack of any stimulating factor. As its growth progress was slow, the tumoral lesion could be benign and as its consistency was soft, it could be a lipoma, neurofibroma, or pyoderma gangrenosum. For patient management, after signing the written consent form, we did an excisional biopsy and considered a follow-up. The tumor was excised under local anesthesia (by long buccal anesthesia or anesthetizing all around the lesion, the lesion was removed from its base with a blade); then, the specimen was placed in a formalin solution, and it was sent for a histopathological examination to the Pathology department. In the microscopic examination, sections showed a piece of oral mucosa covered by parakeratotic stratified squamous epithelium. The underlying connective tissue demonstrated abundant collagen fibers intermixed with lobules of fat cells (Figure 4). Therefore, a fibrolipoma was finally diagnosed. Patient was followed up to 3 months after excision, and no recurrency was reported.

Discussion

Lipomas are the most common benign tumors in almost all parts of the body that contain adipose tissue. They are relatively uncommon in the oral cavity (1-5% of all benign lesions in the mouth). Lipomas can be seen in different parts of the oral cavity. The literature review showed that half of the oral lipomas were related to the buccal mucosa, and the other half were found in the lips, tongue, floor of the mouth, palate, and gingiva. Oral lipomas were seen in all groups of age, but it has been most frequently reported after 40 years of age [5].

According to their histology, the WHO classifies lipomas in several groups: conventional lipomas, fibrolipomas, angiolipomas, pleomorphic lipomas/spindle cells, mixolipomas, condrolipomas, osteolipomas, miolipo-

mas, lipomatosis, lipomatosis of the nerve, lipoblastomas, and hybernomas. Other variants of lipomas than conventional lipoma is rare. As an example, FL, an uncommon variant of lipoma, is particularly rare in the oral cavity (a prevalence rate of only 1/5000 adults in the oral and oropharyngeal region). Its difference from conventional lipoma is in the way that the mature adipose tissue is interspersed by connective tissue bands [6]. FL has been reported to occur in the buccal mucosa, buccal vestibule, and tongue more frequently [7,8]. The reason for reporting our case is the rareness of the fibrolipoma in the mouth, especially in the gingival part of the oral cavity, and the importance of its differential diagnosis.

Lipomas are painless and freely mobile. Because of their thin overlying epithelium, they usually grow at a low rate and can be clinically seen in a semi-lucent yellow color; the presence and degree of the yellow hue depends on the degree and depth of fibrosis. Its consistency varies from soft to firm. This varies because of the depth of the tumor and the distribution and amount of fibrous tissue. Several cases have shown some grades of fluctuation as well. Lipoma and FL both usually have a thin capsule [9].

Regarding histology, FL consists of mature fat cells, which are divided into lobules by fibrous shoots. This lesion is generally oval-shaped [8].

Several cases of FL have been reported until this day (Figure 2). We reviewed 6 cases as described in Table 1. They aged from 25-75 years, and 50% of them, similar to our case, were females.

In common with Iaconetta, Kiehl, and Manjuantha, the lesion of our patient was yellow, capsulated, and movable. Its consistency, other than one case of Manjuantha, which was firm, was similar to our reviewed cases: soft.

In contrast to the other cases present in the literature described by Iaconetta, Kiehl, and Manjuantha, the FL of our patient did not show any mobility. Unlike our case, which was colored pink-red, the lesions described by Iaconetta and Kiehl were yellow.

The size of the lesions of the cases we reviewed was from 1 to 4 cm; similarly, the lesion of our patient measured 1.5 cm. Like Kiehl and Iaconetta's, our case did not show any pain.

For the management of our patient and all cases we reviewed (Castellani, Iconetta, Kiehl, Manjuantha), the lesions were removed under local anesthesia and sent to the Pathology department for further study about their microscopic characteristics [4,8,10,11].

As already mentioned, lipoma is one of the most common benign tumors in the body. Lipoma (specifically FL) is extremely rare in the oral cavity. This exophytic lesion can also be mistaken with reactive or other tumoral lesions: due to its adhesion to the surrounding tissues and pseudo-infiltrating characteristics of this lesion because of the abundance of collagen and connective tissue, it can cause doubts of differential diagnosis with malignant infiltrating lesions [11,12]. As a result of the lesion's adherence to the structures that surround it and its pseudo-infiltrating characteristics, a histological exam is necessary to clarify the nature of the neoformation and to resolve any doubt [13]. Therefore, it is mandatory to perform a biopsy and differential diagnosis and eventually diagnose FL carefully. Another importance of diagnosing FL is that this lesion is one kind of tumor; as a result, it has an increased growth potential. FL almost always grows slowly, but diagnosing it soon and performing the necessary management is essential for a better prognosis and patient's comfort. The treatment for this kind of lipoma in the oral cavity is a surgical incision under local anesthesia. Although commonly a good result can be observed after surgery, follow-up must be performed once in several months (depending on the lesion and patient's condition) due to its low recurrence rate.

Our study limitations included lack of genetic evaluation and short follow-up duration.

To diagnose accurately, clinical features and microscopic (histological) findings must be considered. FL is a rare benign tumor in the oral cavity with an increased growth potential compared to classical lipoma. It has a low chance of recurrence.

Lesions that look clinically similar to each other may demonstrate different and similar histopathological characteristics; they, therefore, can raise a diagnostic dilemma for a general dentist. Surgical excision may be an elective treatment for FL, but the examination of excised tissue along with consultation with an Oral Pathologist for an accurate diagnosis and careful follow-up is mandatory to provide a successful treatment and prevent any malignant transformation.

Our case adds to the few cases of gingival FL which have been reported in the English literature.

It is essential to document new cases of FL in the English literature so that better and more accurate treatments can be introduced to prevent any malignancy and further damage they may cause.

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Ethics approval

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Consent to participate

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Consent for publication

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Tables

Table Reviewed cases of FL in the English literature

Author	Age + / Sex ++	Site of the lesion	Size§	Characteristics	Symptoms	Consistency
Castellani et al. 2015 (14)	25/ F	Intraosseous of right mandibular ramus	-	radiolucency in the right mandibular ramus in the OPG¶ radiograph	None	-
Iaconetta et al. 2015 (8)	71/ F	Ventral surface of the tongue	40	curvy shaped-movable-covered by mucosa	dysfunction of phonation and swallowing, and a sensation of 'obstruction' of the oral cavity	Soft
Manjuanatha et al. 2010 (4)	75/ M	Right buccal mucosa	30	Pedunculated	None	Soft

Author	Age + / Sex ++	Site of the lesion	Size§	Characteristics	Symptoms	Consistency
Manjuanatha et al. 2010 (4)	55/ M	Right buccal mucosa	10	Sessile	None	Firm
Manjuanatha et al. 2010 (4)	70/ M	Soft palate	15	Sessile	None	Soft
Kiehl 1980 (11)	65/ F	Beneath a mandibular complete denture	15	Freely movable-yellow-encapsulated-covered by thin epithelium	None	Soft

+Age: in years- ++M: male- F: female- §size: in millimeters- ¶OPG: Orthopantomography

Figures

Author	***Age /Sex****	***Site***	***Duration***	***No. of Cases***	***Recurrence***
Saitoh et al 1995 [16]	3/F	Parotid	NA	1	NED 3 years
Dattilo et al 1996 [14]	45/M	Tongue	10 years	1	NA
Epivatianos et al 2000 [13]	NA	Tongue	NA	2	NA
Fregnani et al 2003 [1]	NA	Buccal Mucosa	NA	18	NED 26.5 months
Furlong MA et al 2004 [7]	NA	Parotid Buccal mucosa	NA	2	NA
Bandéca MC 2007 [23]	42/M	Lower lip	NA	1	NED 60 months
Capodiferro S et al 2008 [15]	43/M	Labial mucosa	8 months	1	NED 10 months
Freitas et al 2009 [9]	56/F	Buccal mucosa	NA	7	NA

*Age→in years, M→ Male, F→Female, NED--no evidence of disease; NA--not available

Figure 1 Summary of pervious reported cases of oral fibrolipoma (4)

Author	Number of cases
Horton et al. 1968 [5]	1
Dattilo et al. 1996 [3]	1
Epivatianos et al. 2000 in Manjunatha et al. [2]	2
Said-Al-Naief et al. 2001 [6]	3
Fregnani et al. 2003 [7]	1
Juliasse et al. 2010 [9]	1
Manor et al. 2011 [10]	3
Shi et al. 2014 [4]	1
Camacho et al. 2014 [8]	1

Figure 2 Summary of previous reported cases of tongue fibrolipoma (8)

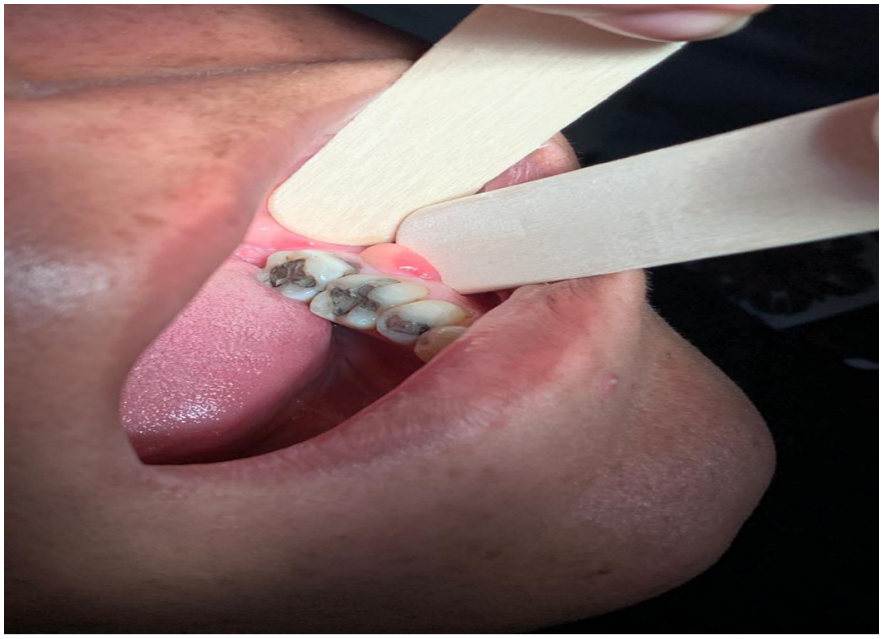

Figure 3 A photograph of our case showing an exophytic lesion with a dome-shaped base, smooth surface, non-homogenous color (pale pink-red and somewhere yellow), and homogenous on the left lower gingiva next to the first and second mandibular molars

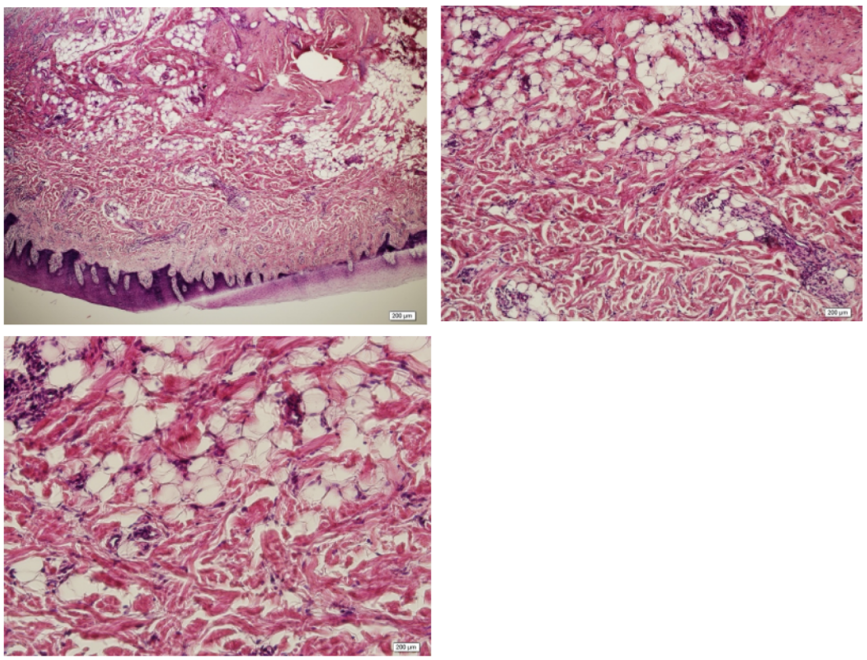

Figure 4 Microphotograph showing piece of oral mucosa covered by parakeratotic stratified squamous epithelium. The underlying connective tissue demonstrates abundant collagen fibers intermixed with lobules of fat cells. (H & E stain, 40X (upper- left figure), 100X (upper- right figure), and 200X (lower- left figure) magnification)

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