

A Slow Growing Ambiguous Soft Tissue Swelling of Buccal Mucosa; A Diagnostic Mêlée

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ABSTRACT

A 21-year-old patient reported with an asymptomatic painless swelling on left buccal mucosa present for past 4 years. Excision and subsequent histopathologic evaluation revealed a fibrolipoma of buccal mucosa. Microscopic examination revealed a parakeratinized stratified squamous epithelium in association with dense fibrous connective tissue. The epithelium appeared stretched with few areas exhibiting loss of rete ridges. Melanin pigment was also noted along the basal epithelial layer in one area. The connective tissue showed dense bundles of collagen fibers interspersed with lobules of adipocytes. Numerous blood capillaries, plump fibroblasts, and minimal inflammatory cell infiltrate were noted. The post-operative healing was uneventful, and no recurrence is reported till date.

Keywords: Adipocytes, Buccal mucosa, Benign tumor, Fibrolipoma, Lipoma

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INTRODUCTION

Oral lipoma, first described by Roux in 1848 as “yellow epulis,” is a mesenchymal benign neoplasm containing mature fat cells that may present as a developmental anomaly. Though lipomas can occur anywhere in the body, the most common site for oral lipomas is the buccal mucosa, followed by the tongue, the floor of mouth, buccal sulcus, palate, lips, and gingiva.^{1,2} Oral lipomas are relatively rare entities that constitute about 2.2% of all lipomas and 2.4% of benign tumors which occurs in the oral cavity.¹ Even though lipomas are usually present as soft nodular superficial swellings covered by normal mucosa, intramuscular, and infiltrative lesions have also been reported.² Fibrolipoma is an extremely rare subtype of lipoma, which accounts for 1.6% of all facial lipoma.³ The most common site of fibrolipoma is buccal mucosa followed by the tongue. Rare occurrences have been reported at the tip of the nose, and from the floor of the mouth.^{3,4} Fibrolipomas in the mouth for as long as 10 years, have been reported in literature.⁴ Many lipomas remain unrecorded as most cases are asymptomatic and grow slowly, and therefore, ignored by patients, so proper documentation is important. Fibrolipomas even though

benign in nature may cause difficulty in speech and mastication due to progressive enlargement. Therefore, early diagnosis and prompt treatment are essential. The diagnosis of oral lipoma is often made by clinical examination and for identification of the subtypes, a biopsy is still the gold standard. Advanced imaging modalities such as magnetic resonance imaging and computed tomography even though not used routinely, they may aid in the diagnosis and determination of the extent of the lesion, especially in cases of intramuscular and infiltrative lipomas. A histopathological differentiation is necessary to differentiate between the various types of lipomas. Fibrolipoma needs to be differentiated from spindle cell lipomas, which are composed of mature lipocytes and uniform spindle cells in a mucinous and fibrous background. Normal white fat consists of spherical cells containing one large lipid vacuole that displaces the thin oval nucleus to one side. On routine sections, the nucleus of most fat cells is barely perceptible. Surgical excision is the most common treatment of fibrolipomas. Most reports state that the recurrence was not found till a period of 5 years.⁴ A case of 21-year-old male patient with a swelling in the left buccal mucosa ultimately diagnosed as fibrolipoma is presented here.

CASE REPORT

A 21-year-old male patient reported to Department of Oral Medicine and Radiology, PMS College of Dental Science and Research, Vattapara, seeking treatment for decayed lower posterior teeth. The patient also mentioned about an asymptomatic painless swelling on the left buccal mucosa, which was present for the past 4 years with a history of cheek biting for 5 years. Even though it was slowly enlarging in size, the patient never had any difficulty in mastication and speech; moreover, there was no associated hemorrhage; ulceration or discharge (Figure 1).

On inspection, the left buccal mucosa revealed a well-defined oval non tender pedunculated swelling of size 2 cm × 3 cm, pale pink in color with isolated erythematous and gray areas on the surface, opposite to the occlusal aspect of 37 and 38. On palpation, the swelling was well defined, non-tender, soft, pedunculated and mobile with a rough surface and occasional gray colored areas. Slip sign was positive. A soft tissue radiograph of the



Figure 1: A soft asymptomatic painless swelling on the left buccal mucosa



Figure 2: Excised specimen

swelling using intraoral periapical did not show any evidence of calcification. A provisional diagnosis of intraoral fibroma was made. Differential diagnosis of the fibro-epithelial polyp, lipoma, and fibrolipoma was considered. Routine blood examination results were found to be within normal limits. The lesion was then excised under local anesthesia, and the specimen was sent for histopathologic evaluation to the Department of Oral and Maxillofacial Pathology, PMS College of Dental Science and Research, Vattapara (Figure 2). The post-operative healing was uneventful, and no recurrence is reported till past 6 months.

Microscopic examination revealed a parakeratinized stratified squamous epithelium in association with dense fibrous connective tissue. The epithelium appeared stretched with few areas exhibiting loss of rete ridges. Melanin pigment was also noted along the basal epithelial layer in one area. The connective tissue showed dense bundles of collagen fibers interspersed with lobules of adipocytes. Numerous blood capillaries, plump fibroblasts, and minimal inflammatory cell infiltrate were noted (Figures 3 and 4). Correlating the clinical findings with histopathologic findings the mass was diagnosed as fibrolipoma.

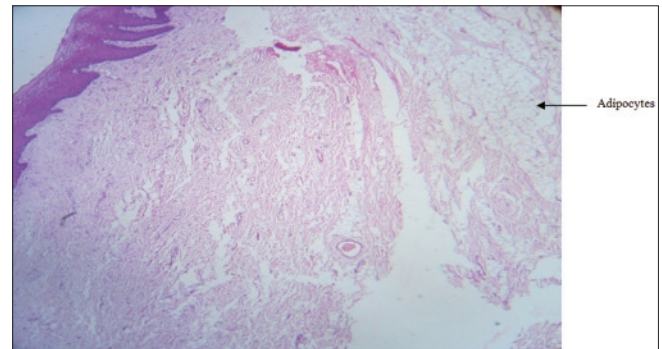


Figure 3: Surface epithelium with connective tissue showing lobules of adipocytes ($\times 4$ magnification)

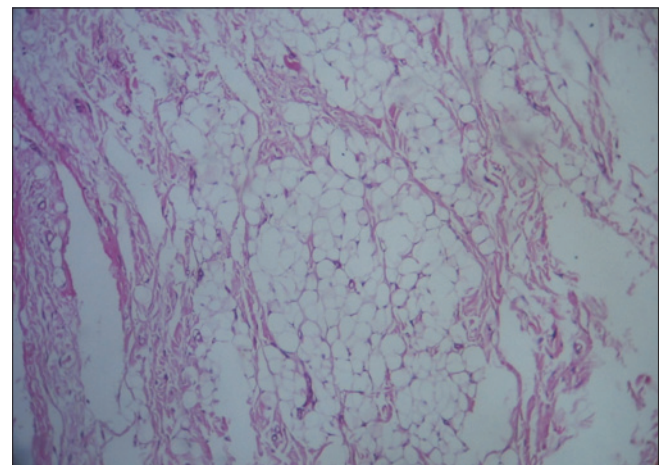


Figure 4: Adipocytes interspersed between collagen fibers ($\times 40$ magnification)

Table 1: Characteristic features of various types of lipomas^{5,12}

Types of lipoma	Characteristics
Infiltrating lipoma/ intramuscular lipoma	<ul style="list-style-type: none"> • Invades the muscles or grows between them • Involves the skeletal muscle fibers.¹¹ Rarely occurs in head and neck • Located within striated muscle³ • No malignant transformation reported.²
Fibrolipoma	<ul style="list-style-type: none"> • Asymptomatic, painless swelling, slow growing • Lobules of mature fat cells surrounded by thin fibrous connective tissue septa.¹
Spindle cell lipoma	<ul style="list-style-type: none"> • Not encapsulated • Rarely infiltrates adjacent muscles, • Multiple lesions, asymptomatic¹² • Accounts for approximately 1.5% of all adipocyte tumors, • 1/60th of incidence of ordinary lipoma.²
Angiolipoma	<ul style="list-style-type: none"> • Multiple subcutaneous nodules, pain can be present • Characteristic feature of prominent vascular components² • Subcutaneous nodule consisting of mature fat cells, intermingled with small, and thin-walled vessels; a number of which contain fibrin thrombi.¹¹
Pleomorphic lipoma	<ul style="list-style-type: none"> • Spindle-shaped appear dysplastic or mixed with pleomorphic giant cells with or without hyperchromatic and enlarged nuclei⁹ • Asymptomatic, mobile dermal or subcutaneous mass¹²
Myxoid lipoma	<ul style="list-style-type: none"> • Myxoid background stroma⁹
Osteolipoma	<ul style="list-style-type: none"> • Occurs rarely with chondroid or osseous metaplasia in lipoma⁹ • Painless, affects scapula, neck, skull, suprasellar region, tuber cinereum.¹⁵
Myelolipoma	<ul style="list-style-type: none"> • Usually adrenal glands, pelvic soft tissue¹¹ • Lipoma presents with bone marrow components.⁹
Adenolipoma	<ul style="list-style-type: none"> • Ductal or tubular adnexal structures when scattered in fat cells⁹
Chondroid lipoma	<ul style="list-style-type: none"> • Painless mass of variable duration, recent h/o enlargement • Usually occurs in proximal extremities and limb girdles¹¹ • Benign adipose tissue tumor containing lipoblasts, mature fat, and chondroid matrix.¹²
Myolipoma of soft tissue	<ul style="list-style-type: none"> • Also known as extrauterine lipoleiomyoma • Involve abdominal cavity, retroperitoneum, inguinal areas • Occur in adults. Females are most affected • Exhibit features of mature smooth muscle and mature adipose tissue.¹²
Hibernoma	<ul style="list-style-type: none"> • Brown fat neoplasm⁹ • Composed of brown fat cells with granular, multivacuolated cytoplasm.¹ Brown fat component is admixed in variable proportion with white adipose tissue.¹²
Perineural lipoma	<ul style="list-style-type: none"> • Also known as lipomatosis of nerve • Associated with motor or sensory deficits • Imaging studies show fusiform enlargement of nerve with fatty infiltration • Infiltration of epineurium by adipose and fibrous tissue. The tissue grows between and around nerve bundles, causing enlargement of affected nerve.¹²
Sialolipoma ¹³	<ul style="list-style-type: none"> • Usually asymptomatic, slow-growing • Involving the major and minor salivary glands • Characterized by well-demarcated proliferation of mature adipocytes with secondary involvement of salivary gland parenchyma.¹⁴
Atypical lipoma ^{6,7}	<ul style="list-style-type: none"> • Also called well-differentiated liposarcoma is an intermediate malignant mesenchymal neoplasm composed either entirely or in part of mature adipocytic proliferation showing significant variation in cell size and at least focal nuclear atypia in both adipocytes and stromal cells.¹²
Lipomatosis ¹²	<ul style="list-style-type: none"> • Present with massive accumulation of fat in the affected areas that may mimic a neoplasm, neuropathy, and CNS involvement • Affects different anatomic regions • Diffuse overgrowth of mature adipose tissue.¹²
Benign lipoblastoma ¹¹	<ul style="list-style-type: none"> • Occurs in infants¹¹ • Also known as fetal lipoma, embryonic lipoma, infantile lipoma¹² • Slow growing, well circumscribed, may compress adjacent structures.¹² • A lobulated, localized or diffuse tumor, resembling fetal adipose tissue.¹²

DISCUSSION

The location of the lesion in buccal mucosa and probabilities of trauma required intraoral fibroma to

be in the diagnosis list. Intraoral fibromas are round to oval, asymptomatic, smooth surfaced, firm sessile or pedunculated swelling and can occur at any age due to trauma.⁴ Since slip sign was positive, in addition, to

other findings lipoma was considered next. A lipoma is a benign tumor composed of mature fat cells, and often appears as a yellowish mass with a thin epithelium occurring on the buccal mucosa and tongue, with a predilection to occur above 40 years. The hemangioma was not considered as the swelling was non-compressible and non-reducible; moreover, the appearance was non-contributory. In this case, the variants of lipomas were taken into consideration. A definitive diagnosis could be made only based on the histopathologic examination.

Benign lipomas are the most common mesenchymal tumor affecting the soft tissues. Lipomas consist of mature adipocytes, surrounded by a thin fibrous capsule.⁵

In a literature review of fibrolipoma by Manjunatha *et al.*, intraorally buccal mucosa is the most common site and tongue is the second most common site.⁶ They usually occur on the floor of mouth, gingiva, mucobuccal/labial fold, major salivary gland, lips, oropharynx, palate, and vestibule.⁷ Hard palate is rarely affected due to the presence of very little fatty tissue.¹ In the oral cavity and oropharyngeal region it is relatively uncommon, the prevalence rate being 1 in 5000 adults.⁸ According to Fregnani *et al.* 45.7% cases were lipomas and 39.1% were fibrolipomas.⁹

Hatziotis reported that 80% of patients were over 40 years of age, 64% were over 50 years and 40% over 60 years, with a general age range of 2-87 years for the various variants of lipoma.⁶ Even though oral lipomas occur at all ages, it frequents at 40 years. Mean age in case of fibrolipoma is 34 years with a range from 3 to 56 years.⁶ Some reports also suggest that it occurs in fourth and fifth decade.¹⁰ Ozturk *et al.* reported that fibrolipomas rarely occur below the age of 20 years.^[10,11]

In general, lipomas do not have any gender predilection while Manjunatha *et al.* has reported a male predilection, other studies showed a female predilection.^{6,7,10}

Lipomas mostly present as asymptomatic, slow-growing mass in the oral cavity and in the subcutaneous tissue. The size of fibrolipoma depends on location, but it is rarely >25 mm in diameter.⁶ Complications are rare and few. Long-standing cases may turn into liposarcomas.^{3,6}

The prognosis of the neoplasm is considered good as it is slow growing and rarely recurs after surgical excision.⁹

Etiology of lipoma is varied. Origin from lipoblastic embryonic cell nests, metaplasia of muscle cells, and fatty degeneration are some of the causative factors for simple lipoma. Others include trauma, infarction, infection, chronic irritation, and hormonal imbalance.

In most cases, they represent a developmental anomaly but they can arise as a result of trauma and rearrangement of chromosomes number 12 q, 13 q, and 6 p. It may also arise by preadipocyte differentiation and proliferation mediated by cytokines following soft tissue damage when blunt trauma and hematoma formation occurs.^{1,2,10}

The different theories for the development of fibrolipoma are - it can be congenital, caused by endocrinal imbalance, a product of degenerated fibrous tumor or it can arise from maturation of lipoblastomatosis. Lipoblastomatosis is an infiltrative type of benign neoplasm with lobules of immature fat cells separated by connective tissue septa, and areas of loose myxoid matrix.²

Variants of lipomas can be divided based on morphological and histological differences. Morphologically, intraoral lipomas are classified as diffused form, which affects deeper tissues, and superficial and encapsulated form.⁵ Depending on the site, it can be as superficial, deep, and periosteal.¹

After a thorough review of the literature, we have attempted to summarize the characteristic features of various histological variants of lipoma (Table 1).

CONCLUSION

A case of 21-year-old male patient reporting with an asymptomatic, well defined, soft pedunculated swelling on the left buccal mucosa is presented. Fibrolipoma is an extremely rare subtype of oral lipoma with no pathognomic clinical characteristics. The diagnosis is ultimately by histopathological examination. The management is by excision. The present case of fibrolipoma was also managed by excision of the lesion and subsequent histopathological examination which confirmed the diagnosis. Recurrence is very rare. The patient is still under follow-up and no recurrence has been reported till date.

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