

Rare Variant of Spindle Cell Lipoma of the Lip: Utilizing Immunohistochemistry as an Essential Diagnostic Tool

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ABSTRACT

Introduction: The histopathological variant of lipoma identified as spindle cell lipoma invariably begins to develop deep within the skin in the back of the neck, upper back, or shoulder region. It develops seldomly in the oral cavity yet extremely frequently on the lip. Predominantly, these lesions manifest mostly as painless or slowly gaining masses.

Case report: A 75-year-old woman had single swelling in the left lower lip which had existed there for four months. On a microscopic level, these tumors are well structured and represented in varied amounts of mature adipocytes, spindle cells, and irregular bundles of thick collagen fibres. The spindle cells are S-100 protein, desmin, and smooth muscle actin-negative whilst immunopositive to CD34. Treatment primarily involves local excision, and no reports of recurrence have been made.

Conclusion: With a study of the available literature on diverse histological variants, differential diagnosis, and the adoption of IHC for diagnostic purposes, we present a rare case of spindle cell lipoma of the lip.

Keywords: Spindle cell, Lipoma, Lip, Immunohistochemistry, CD-34, IHC

INTRODUCTION

The Greek word for "Liparein," which implies "to persevere" or "to persist," is the root of the English term "lipoma," which also means "fat." Moreover, the word "Leip" has Proto-Indo-European roots and means "to stick" or "to cling". A benign tumor made up entirely of fatty tissue is termed a lipoma.¹ Merely 14% of occurrences of lipomas, regardless of gender, occur in the oral cavity. In the oral cavity, they make up between 0.5% and 5% of all benign neoplasms. Oral spindle cell lipomas (SCL), which comprise only about 7% of all cases and range from 2.17% to 9.8% of all oral lipomas, are relatively rare among these.²

SCLs are typically diagnosed in older men between the fifth and seventh decades of life, as they possess a propensity to develop slowly. Mature adipocytes, much less often encased in a thin fibrous capsule, contribute to the majority of these benign mesenchymal neoplasms. The most common sites for oral SCL lesions to surface are the buccal mucosa, mouth floor, or tongue. These lesions have often been painless, well-defined, and circumscribed.³

Spindle cell lipomas can have many different characteristics, resulting in them being potentially harder to discern within other spindle cell neoplasms, both lipomatous and non-lipomatous. This contrasts with traditional lipomas, which are simple to comprehend. We, therefore, emphasize a lip-related spindle cell lipoma, which the literature asserts seems to be infrequent.⁴

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How to cite the article: Doddawad VG, Patil K, Sanjay CJ, Shivananda S. Rare variant of Spindle cell lipoma of the lip: Utilizing Immunohistochemistry as an essential diagnostic tool. Oral Maxillofac Pathol J 2024; 15(2):274-279.

Source of Support: Nil

Conflict of Interest: None

CASE PRESENTATION

The chief concern of a 75-year-old woman who visited the Department of Oral Medicine and Radiology was swelling in the left lower lip which had existed there for four months. She gives a history of swelling just on the left lip's side for 4 months, which was not preceded by a history of trauma. The swelling was asymptomatic. It was a single,

soft-diffuse, dome-shaped, sessile swelling of insidious onset and measuring approximately 2 x 3 cm. The surface over the swelling and surrounding structures appears normal and the swelling is non-tender with no evidence of bleeding/discharge on digital palpation. (as shown in Figure-1) Ultrasonography showed well-defined hyperechoic area with multiple intense internal fine echogenic lines. (as shown in Figure-2)

A differential diagnosis was considered based on the clinical examination and included mucocele, liposarcoma, and myxoma. Under local anesthetic, an excisional biopsy was carried out with the patient's prior consent, and the specimen

was further forwarded for histopathological analysis. A benign encapsulated tumor with homogeneous, mature adipose tissue lobules mixed with spindle cells within a fibrous myxoid stroma was reported, according to the histopathological evaluation. No mitotic activity or necrosis was observed (as shown in Figure 3). Spindle cells tested positive for CD34 however significantly negative for S-100 protein by immunohistochemical (IHC) staining (as shown in Figure-4). The lesion's identification as a spindle cell lipoma was validated by these results. Three months following the surgical procedure, the patient was reevaluated, and there were no discernible signs of any



Fig. 1: Swelling in the lower left labial mucosa

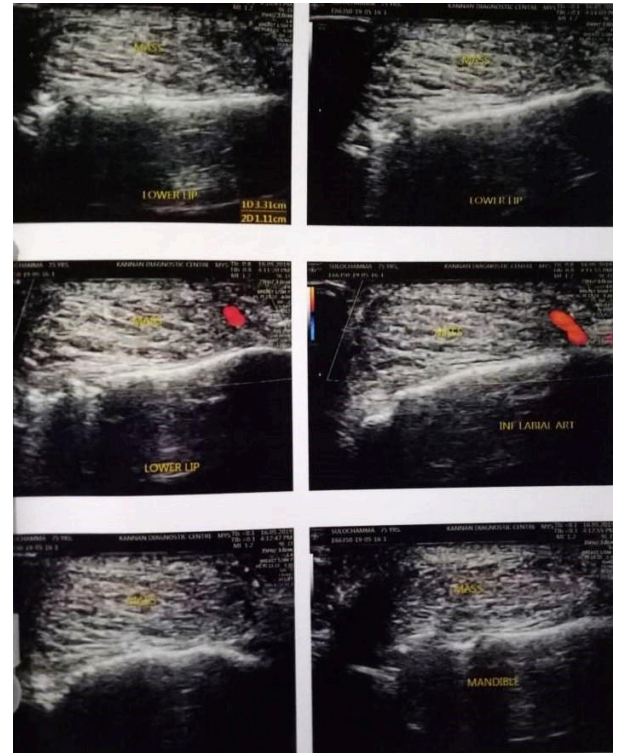


Fig. 2: Hyperechoic area with multiple intense internal fine echogenic lines

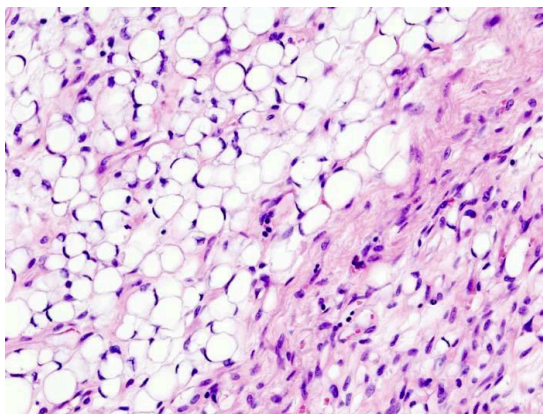


Fig. 3: Mature adipose tissue intermingled with spindle cells within a fibrous myxoid stroma.

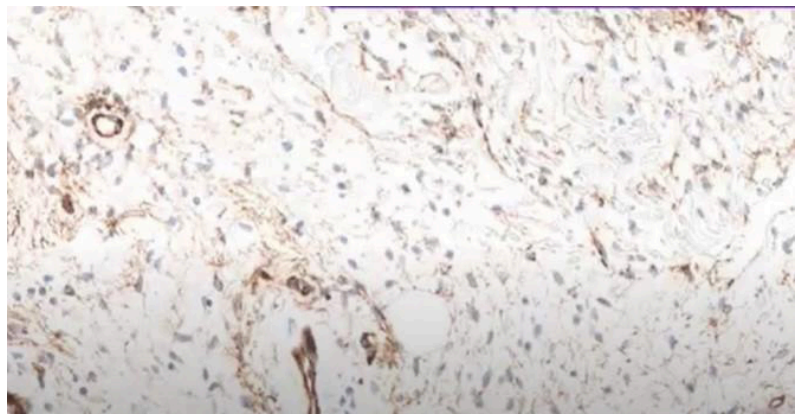


Fig. 4: Positive staining for CD34 in the spindle cells

postoperative sequelae. A four-year follow-up was carried out to assess for any indications of adverse outcomes or recurrence.

DISCUSSION

SCL represents a unique entity characterized by its specific clinicopathological and immunohistochemical features. It originates from prelipoblastic mesenchymal cells and was first documented by Enzinger and Harvey in 1975. However, McDaniel et al. in 1984 documented the first occurrence of SCL in the oral cavity.^{5,6}

Lipomas are observed in a multitude of pathological forms, such as fibro-lipoma, sialolipoma, myelolipoma, osteolipoma, chondroid lipoma, angioliipoma, angiomyolipoma, myelolipoma, intramuscular lipoma, pleomorphic lipoma, and spindle cell lipoma (SCL).⁷ The World Health Organization (WHO) eventually categorized benign lipomatous pathologies into several lesions, such as lipoma, lipoblastoma, lipomatosis, lipomatosis of a nerve, myolipoma of soft tissue, angioliipoma,

spindle cell/pleomorphic lipoma, chondroid lipoma, and hibernoma.⁸

Table 3: Histological variants of Lipoma.¹⁷

S. no.	Type	Features
	Fibrolipoma (most common)	Fibrous component + Adipose tissue.
	Angioliipoma	Vascular component + Adipose tissue.
	Myoliipoma	Smooth muscles + Adipose tissue
	Spindle cell lipoma	Variable amounts of uniform appearing spindle cells + adipose tissue Subtypes: Classic Spindle cell, rope-like collagen, adipose tissue. Fibrous Massive collagen fibers. Myxoid stroma. Low-fat Little or almost no adipose tissue. Fat-rich A large amount of adipose tissue. Pseudo angiomatous Highly vascular with staghorn type bloodvessels
	Pleomorphic lipoma	Pleomorphic spindle cells.
	Intramuscular/ Infiltrating lipoma	Muscle gets replaced by mature adipose tissue. More deeply situated and having an infiltrative growth pattern.
	Chondroid lipoma	Chondrocytes + Adipose tissue.
	Perineural Fibro lipoma	Abundant adipose tissue surrounding the nerves displaying concentric perineural fibrosis
	Osteoliipoma	Osseous component + Adipose tissue.
	Myxoliipoma	Myxoid stroma + Adipose tissue
	Myeloliipoma	Bone marrow component + Adipose tissue.
	Adenoliipoma	Ductal component + Adipose tissue.

Table 1: Clinical features of Spindle cell lipoma in the lip reported in the literature

S. no.	Authors (Year)	Age/sex	Clinical Symptoms
1	Girgis S et al.,[3]	38-year/ Female	Pedunculated, slow-growing mass on her lower lip with intermittent change in size (6 × 6 mm in diameter) and bleeding.
2	Val-Bernal JF et al.,[15]	49-year/ Male	smooth nodule of the mucosa of the lower lip that was gradually increasing. nondescript smoothsurfaced, non-tender nodule measuring 1.5 cm
3	Rocha AFL et al.,[16]	29-year/ Male	asymptomatic nodule, with gradual increases in size, and was asymptomatic, lesion presented as a sessile nodule covered by normal-appearing oral mucosa and measuring approximately 1.0 cm in diameter.

Table 2: MRI findings for differentiating various lesions from spindle cell lipoma.

	Internal uniformity	Fat content	Tumor boundary	Contrast of tumor boundary	Peripheral infiltration	Septum like apperaence	Nodular non-fat tissue
Lipoma	Homogenous	High	Regular	No	No	No	No
SCL	Homogenous	varies	Regular	No/Yes	No/Yes	Yes	No
Liposarcoma	Homogenous	varies	Regular	Yes	Yes	Yes	Yes

The exact causes and underlying mechanisms of lipoma formation remain unclear. Lipomas are thought to arise from nests of lipoblastic embryonic cells. However, several factors have been suggested as potential contributors to their development. These factors include persistent trauma, chromosomal abnormalities, hereditary predisposition, chronic irritation, hormonal imbalances, metabolic disorders, and the development of adipose tissue. Some researchers have suggested that common genetic backgrounds that predispose individuals to spindle cell lipoma (SCL) may be distinguished by hereditary chromosomal aberrations, such as translocations involving chromosome 12q 14.3 or absence of chromosomes 13q (particularly 13q12 and 13q14-q22) and/or 16q (16q13-qter).⁹

According to sources, men are more likely than women to have tongue spindle cell lipomas, which are typically observed in elderly individuals with an average age of 58.2 years. (male-to-female ratio 1.9 to 1). This gender bias may be partially attributed to the reactivity of androgen receptors (AR), although the exact mechanism remains unknown. But syed et al. explained that the growth of SCL may be stimulated by the action of endogenous sex steroids (or even exogenous androgenic-anabolic steroids) via ARs. Androgen receptor proteins exist in both the nuclear and cytosolic fractions of human preadipocytes and adipocytes.¹⁰

The oral location most commonly affected by spindle cell lipoma (SCL) is the tongue with 37% of cases, Q3 followed by

the buccal mucosa with 31% and the floor of the mouth with 15%. SCL has been similarly reported in other locales, such as the hard palate and alveolar ridge.³ To the extent feasible, there have been a mere two occurrences of SCL which involved the lip. Billings et al. described an elderly woman of 55 with SCL of the lower lip, while Manor et al. reported a patient who was a 23-year-old man with SCL of the upper lip. Clinically, intraoral lipomas (OLs) typically manifest as painless, solitary, soft-textured nodules and tend to be either sessile or pedunculated. These lesions exhibit a gradual growth pattern over time and superficial OLs often present as yellowish nodules and enlisted the list of articles that are available in the scientific literature as shown in Table 1.^{4, 11, 12} The case described here of a 72-year-old female patient with SCL on the lip represents a particularly rare occurrence at this specific location. Depending on just the anatomical site, the following conditions will indeed be considered part of the clinical differential diagnosis of intraoral SCL: tumors of the salivary glands, benign mesenchymal neoplasms, mucous extravasation anomalies, and mucous cysts.² The surgical management undertaken was an excisional biopsy seeing as the lesion appeared pedunculated, even though the clinical diagnosis in the current report was a mucous extravasation cyst and the lesion was 5.0 cm in size.

MRI scans of spindle cell lipomas (SCL) and pleomorphic lipomas (PLs) can reveal internal heterogeneity within the tumor. In T1-weighted images, non-adipose components among these lipomas tend to exhibit muscle-like signal

Table 4: Histopathological differential diagnosis of spindle cell lipoma.¹⁷

Lesion	Lipocytes	Spindle cells	Collagen bundle	vascularity	Infiltration
Spindle cell lipoma	+	+	+	-	-
Classic lipoma	Localised	-	-	-	-
Solitary fibrous tumor	+	-	-	High	-
Neurofibroma	-	Wavy nuclei	-	-	-
Leiomyoma	-	+	-	-	-
Fibrosarcoma		Arrange in herringbone pattern	-	-	+
Liposarcoma	-	Cigar shaped nuclei	-	High	+
Vascular tumor	-	-	-	High	+
Dermotofibrosarcoma protuberance	+	Fascicular pattern	-	-	+

Table 5: Immunohistochemical markers for differentiating various lesions from spindle cell lipoma.¹⁸

Lesion	CD34	Bcl2	FVIII	S-100	CD-99	Alpha SMA	Ki67
Spindle cell lipoma	+	+	-	-	-	-	-
Solitary fibroma	+	+	-	-	-/+	-	-
Neurofibroma	-	-	+	-	-	-	-
Leiomyoma	-	-	-	-	-	+	-
Liposarcoma	-	-	-	-	-	-	+
Dermotofibrosarcoma protuberance	+	-	-	-	-	-	-



density, and they can additionally exemplify powerful signals in T2-weighted images. This variation in signal density is attributed to the existence of myxoid and vascular components. Antagonistically, standard lipomas typically have peculiar borders and signal densities that are equatable with those found in subcutaneous fat, presenting a homogeneous appearance on MRI.¹³ Due to the different proportions of adipose and nonadipose tissue in SCL and PLs, the differential diagnosis is based on MRI findings, as shown in Table 2.

Spindle cell lipomas (SCL) are well-defined neoplasms that are capable of being expressed from other tumors by the coexistence of both mature adipocytes and spindle cells that are cytologically viable. These two components may be found in differing levels in a particular tumor, with one component seldom prevailing over another. The spindle cell component of the tumor is portrayed by bundles of vivid eosinophilic collagen that are sinewy or rope-like in appearance and are predominantly encountered surrounding the spindle cells in the SCL.⁴ These features were also observed in the present case.

The spindle cells in SCL, according to Beham et al. may originate from fibroblastic cells or resemble non-lipoblastic mesenchymal stellate cells, which are found in primitive fatty lobules and hold the ability to yield collagen. In their analysis using electron microscopy, Domanski et al. indicated that whilst certain spindle cells reflect attributes of fibroblasts, others exhibit non-membrane-bound lipid droplets, signifying a prelipoblastic provenance. However, the origin of stromal cells in SCL remains a matter of debate due to variants of SCL as shown in Table 3. Table 4 details the pathological differential diagnosis of spindle cell lipoma.

Spindle cells in spindle cell lipomas diligently have a significant impact on CD34 and vimentin while evaluated immunohistochemically. However, most other mesenchymal markers, including actins and desmin, are usually negative. On the contrary end, the tumor's fat cells are S100 protein positive. It is noteworthy to emphasize that, regardless of its predictive relevance, CD34 expression plays a tiny portion in the detection of spindle cell lipomas.^{3,4} The immunohistochemistry screening is performed as stated in Table 5 to contribute to the differential diagnosis of soft tissue spindle cell tumors which are both lipomatous and non-lipomatous.

The most preferred treatment for intraoral SCL is conservative surgical excision, which means that once the lesion is dealt with effectively, there ought to be no recurrences.² It seldom tends to affect the oral cavity, and SCL hasn't yet been reported to transform malignant.³ The prognosis of these cases is generally favorable, with rare relapses.

According to studies by Fregnani et al., intraoral spindle cell lipomas (SCL) have been observed to have a faster cell proliferation rate compared to traditional intraoral lipomas. Therefore, we must continuously pursue further assessment of SCL. Saponification, myxomatous degeneration, calcification, infection, persistent trauma-induced ulceration, and malignant transformation are some of the modifications (liposarcoma).¹⁴

CONCLUSION

Rarely does such histologically markedly different lipoma characterized as spindle cell lipoma (SCL) originate in the oral cavity. It is a benign neoplasm that often has no symptoms and is characterized by gradual growth. The underlying causes or etiological factors contributing to its development remain uncertain.

A conclusive diagnosis of spindle cell lipoma relies on a combination of clinical presentation and microscopic examination of tissue samples. This approach ensures a clear identification of the condition.

Spindle cell lipoma is usually benign and poses minimal health risks. Therefore, the primary treatment approach is conservative surgical removal. Furthermore, individuals with this ailment can be at firmly convinced that this surgical management does indeed have a low rate of recurrence.

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