American Journal of Case Reports

ISSN 1941-5923
© Am J Case Rep, 2015; 16: 844-848
DOI: 10.12659/AJCR.895474

Received: 2015.07.27
Accepted: 2015.08.17
Published: 2015.11.30

Oral Spindle Cell Lipoma in a Rare Location: A Differential Diagnosis

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Conflict of interest: None declared
Source of support: This study was supported by grants from CAPES, CNPq (2973/2013), and FAPEMIG (CDS-PPM- 00221-13), Brazil

Patient: Male, 56
Final Diagnosis: Spindle cell lipoma
Symptoms: Asymptomatic
Medication: Not applicable
Clinical Procedure: Excisional biopsy
Specialty: Oral and Maxillofacial Surgery • Oral and Maxillofacial Pathology

Objective: Rare disease

Background: Spindle cell lipoma (SCL) is an uncommon and histologically distinct variant of lipoma. It usually occurs as a solitary, subcutaneous, and well-circumscribed lesion in the posterior neck, shoulders, and back of older men. SCL of the oral cavity is rare. We present the clinical-pathologic features of the third case of SCL located on the hard palate and discuss the histological differential diagnosis with other fusiform neoplasms.

Case Report: A 56-year-old man was evaluated for an asymptomatic swelling on the right side of the hard palate. The intraoral examination showed a 25×20 mm sessile and circumscribed tumor, underlying an apparently healthy mucosa of normal color. The lesion revealed a floating consistency during palpation. Excisional biopsy was carried out based on a clinical diagnosis of lipoma or a benign minor salivary gland tumor. The histopathology demonstrated a well-circumscribed but unencapsulated proliferation of bland spindle cells admixed with mature adipocytes in a collagenous/myxoid stroma. The spindle cells were uniform, exhibiting elongated nuclei and narrow cytoplasmic processes without atypia. They were positive to CD34 and negative to factor VIII, alpha-smooth muscle actin, S100, cytokeratin, and actin. Mitotic activity was low, as confirmed by Ki-67 immunostaining. No lipoblastic activity was found. The diagnosis of SCL was therefore established.

Conclusions: Oral spindle cell lipoma is a rare benign lipomatous tumor. The histologic picture shows a range of variations and the observation of morphological features is important to distinguish this lesion from other fusiform tumors. Immunohistochemistry should be helpful in this differentiation.

MeSH Keywords: Diagnosis, Differential • Immunohistochemistry • Lipoma

Full-text PDF: http://www.amjcaserep.com/abstract/index/idArt/895474

Background

Spindle cell lipoma (SCL) is an uncommon and histologically distinct variant of lipoma, initially described by Enzinger and Harvey [1]. It usually occurs as a solitary, subcutaneous, and well-circumscribed lesion in the posterior neck, shoulders, and back of older men [2]. The SCL is rarely found in the oral cavity. Histologically, SCL is characterized by replacement of mature fat cells by spindle cells that are closely associated with a mucoid matrix and collagen boundless [3,4]. Since the first report of an intraoral SCL, 43 new cases have been well documented and only 2 were located on the hard palate [5]. Here, we present the clinical-pathologic features of a third case of SCL located on the hard palate, and we discuss the histological differential diagnosis with other fusiform neoplasms.

Case Report

A 56-year-old black man with non-contributory medical history was examined for an asymptomatic swelling on the right side of the hard palate. The patient had noticed the lesion 3 months prior to the appointment and it further increased in size, preventing him from wearing the prosthesis. No abnormalities were noticed in the extraoral examination.

The intraoral examination showed a 25×20 mm sessile and circumscribed tumor, underlying an apparently healthy mucosa of normal color (Figure 1). The lesion revealed a floating consistency during palpation.

Excisional biopsy was carried out based on a clinical diagnosis of lipoma or a benign minor salivary gland tumor. The histopathology demonstrated a well-circumscribed but unencapsulated proliferation of bland spindle cells admixed with mature adipocytes in a collagenous/myxoid stroma (Figure 2A, 2B). Mast cells were common (Figure 2B). The spindle cells were uniform, exhibiting elongated nuclei and narrow cytoplasmic processes without atypia. They were positive to CD34 (Figure 2C) and negative to factor VIII, alpha-smooth muscle actin, S100, cytokeratin, and actin (Figure 2D). Mitotic activity was low, as confirmed by Ki-67 immunostaining. No lipoblastic activity was found. The diagnosis of SCL was therefore established. No recurrence was noticed up to 24-month follow-up.

Discussion

SCL is an histological variant of lipoma, usually found in the trunk or neck. SCL of the oral cavity is a rare lesion and until now only 43 cases have been reported [5]. The tongue is the most commonly affected oral site, followed by the floor of the mouth and buccal mucosa [6–8].

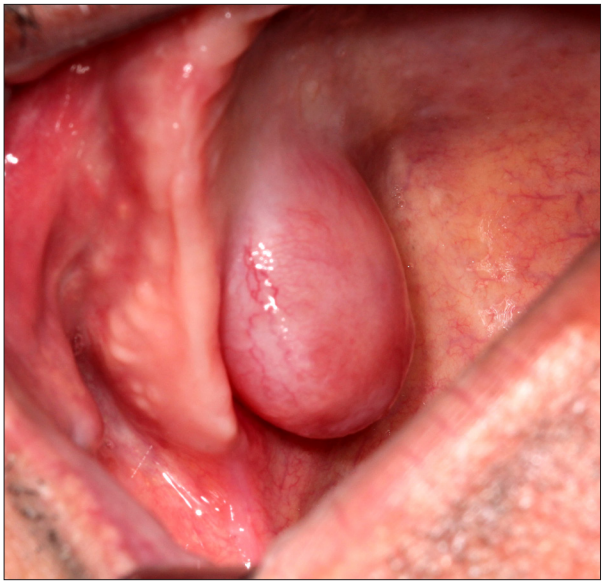

Figure 1. Clinical presentation: A 25×20 mm circumscribed swelling on the right side of the hard palate, covered by an apparently healthy mucosa with evident superficial vascularization.

There are only 2 published cases in which SCL was located on the hard palate [8,9]. Both cases were reported in male patients over the fifth and sixth decades of life, which is compatible with the present case. All tumors were circumscribed, covered by healthy mucosa, and ranged in size from 20 to 25 mm. Follow-up ranged from 6 to 24 months, and no recurrences are expected after local excision (Table 1). In previous reports, fibroma, salivary gland neoplasms, classic lipoma, and benign neural tumors were considered as clinical differential diagnosis of tumors located on the hard palate. All these lesions were included in our diagnostic hypotheses.

SCL is a neoplasm composed by fusiform cells, adipocytes, with a collagenous and occasionally mucinous matrix, presenting a spectrum of variation among these components [1,11]. When floret-like multinucleated cells are found, the SCL is classified as pleomorphic lipoma [5].

The fusiform component of SCL mimics other spindle cell lesions, demanding special attention in the histopathological differential diagnosis. The most important differentiation is with a well differentiated lipossarcoma (WDL), and subtle distinctions must be observed: SCL is located superficially in contrast with WDL. Spindle cells in SCL have an orderly appearance, with scarce mitotic figures and without pleomorphism [10,11]. Lipoblasts, represented by multivacuolated and pleomorphic adipocytes, are absent in SCL, but are a component of WDL [1,10,12]. The vascularization is also different between these lesions. While WDL exhibits a prominent plexiform capillary pattern, the vascular pattern in SCL is usually inconspicuous.

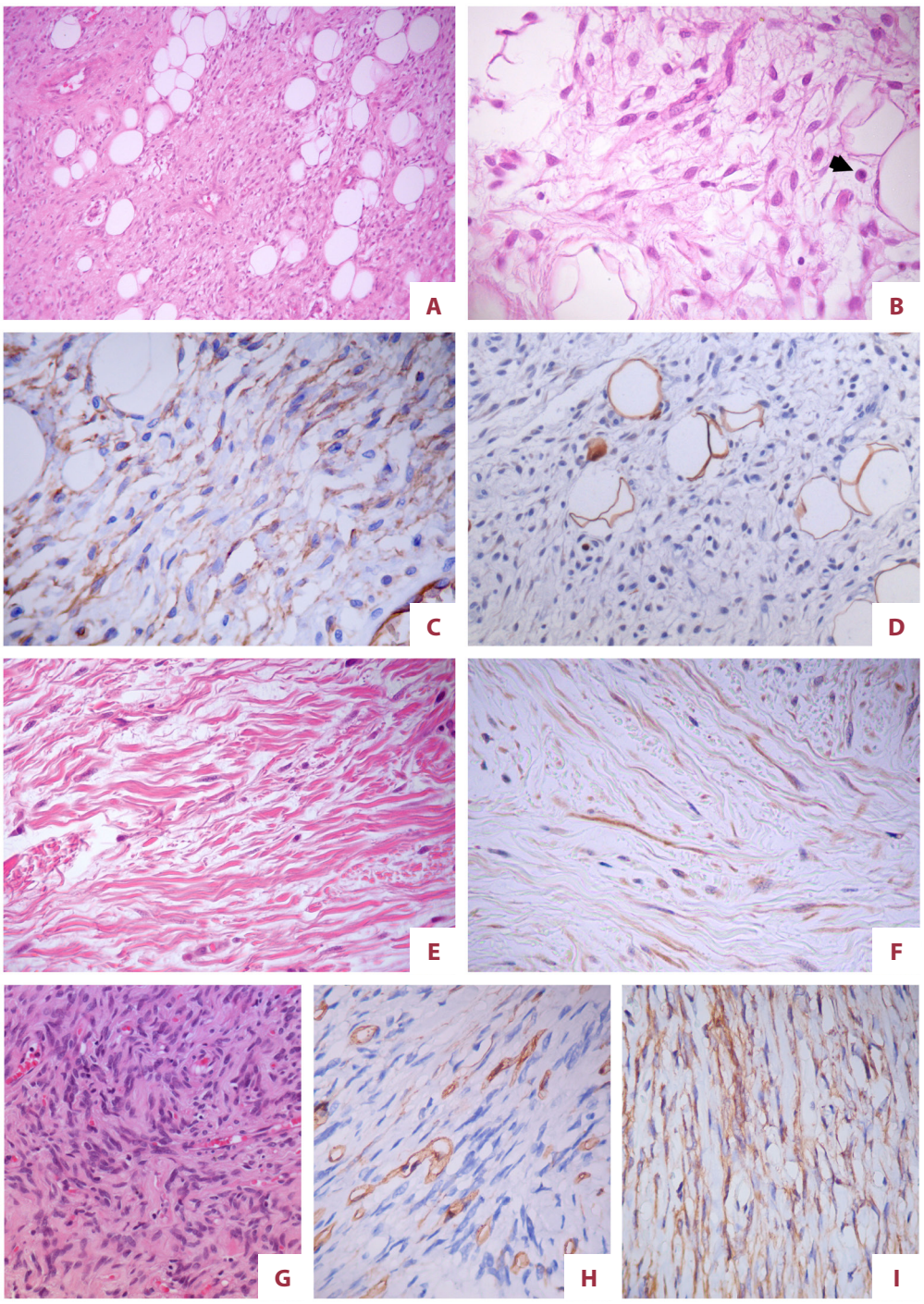

Figure 2. Microscopic findings and differential diagnosis: (**A–D**): Spindle cell lipoma; (**E, F**): Neurofibroma; (**G, I**): Solitary fibrous tumor. (**A**) Proliferation of bland spindle cells admixed with mature adipocytes in a collagenous stroma (HE, 100×). (**B**) Spindle cells exhibiting elongated nuclei and narrow cytoplasmic processes without atypia. A mast cell is evidenced (arrow) (HE, 400×). (**C**) Spindle cells immunopositive for CD34 (streptoavidin-biotin, 400×). (**D**) Adipocytes positive for S100. Spindle cells show negativity. (streptoavidin-biotin, 400×). (**E**) Fusiform component of neurofibroma exhibiting a wavy nuclei (HE, 400×). (**F**) Fusiform component of neurofibroma positive for S100 (streptoavidin-biotin, 400×). (**G**) Solitary fibrous tumor shows spindle cells in a variably vascular and collagenized stroma. The spindle cells often form whorls around small capillaries in a storiform pattern (HE, 400×). (**H**) Solitary fibrous tumor cells negative for CD34 (streptoavidin-biotin, 400×), and (**I**) Positive for CD99 (streptoavidin-biotin, 400×).

Table 1. Reported cases of oral spindle cell lipoma located in the hard palate (n=3).

Authors	Year	Age (years)	Gender	Size (mm)	Clinical differential diagnosis	Follow-up (months)
Christopoulus et al. [9]	1989	58	Male	20	Lipoma Fibroma Salivarygland tumor	24 NR
Caldeira et al. [2]	2011	60	Male	23	Fibroma Salivary gland tumor Benign neural tumors	6 NR
Present case	2015	56	Male	25	Lipoma Salivarygland tumor	24 NR

NR – no recurrence.

Table 2. Immunohistochemical profile of the fusiform component in spindle cell lipomas and other spindle cell lesions.

Lesion	Main histopatologic feature	CD34	Bcl-2	F VIII	CD99	S100
SCL	Mature adipocytes and spindle cells	+	+	–	–	–
SFT	Fusiform cells forming whorls around capillaries	+	+	–	+/–	–
NFB	Fusiform component with wavy nuclei	–	–	–	–	+
DFS	Entrapping feature of fusiform component	+	–	–	–	–
WDL	Lipoblasts, pleomorphic adipocytes, plexiform capillary pattern	–	–	–	–	–

SCL – spindle cell lipoma; STF – solitary fibrous tumor; NFB – neurofibroma; DFS – dermatofibrossarcoma protuberans; WDL – well differentiated liposarcoma; +, positive; –, negative; +/–, variable.

Other differential diagnoses to be considered are neurofibroma, dermatofibrossarcoma protuberans (DFS), and solitary fibrous tumor (SFT). Neurofibroma is a relatively common lesion in palate, but the fusiform component exhibits a wavy nuclei which is not observed in spindle cells of SCL (Figure 2E). Mast cells are common in both tumors.

There is also an overlap between histologic features of DFS and SCL, however, DFS is a cutaneous tumor exceptionally seen in oral tissues [13]. The fusiform component of DFS diffusely infiltrates skeletal muscle bundles, peripheral nerves, and salivary glands, mimicking its cutaneous counterpart. Indeed, this entrapping feature is the distinguishing characteristic between DFS and other fusiform lesions [14].

SFT is an uncommon neoplasm within the oral tissues, and it is mostly located in the buccal mucosa [15]. SFT usually shows circumscription and is encapsulated. It is characterized by a proliferation of spindle cells in a variably vascular and collagenized stroma. The spindle cells often form whorls around small capillaries in a storiform pattern in hypercellular areas (Figure 2G). In contrast, vascularization is not a prominent feature in SCL and mature adipocytes are always present. However, entrapped fat cells can also be seen in SFT and mast cells are shown in both neoplasms [14,15]. Cellularity is variable between SCL and SFT; nevertheless, overall cellularity is consistently lower in SCL. Collagen characteristics also vary between the neoplasms, but the “ropey” collagen bundles are seen exclusively in SCL [14].

The origin of the fusiform component in SCL is uncertain; however, ultrastructural studies have shown that spindle cells are modified fibroblasts that originated from mesenchymal cells located around vessels, which have lost their ability to differenciate into lipocytes but are capable of collagen synthesis [1,12]. These spindle cells show immunoreactivity to CD34, bcl-2 [16] and are negative to, alpha-smooth muscle actin, Factor VIII, cytokeratin, and S-100 [5,9]. The positivity to CD34 can be helpful in the confirmation of diagnosis [17], but the immunohistochemical profile alone is not enough to distinguish SCL from other fusiform lesions (Table 2).

WDL rarely shows positivity to CD34; however, other fusiform neoplasms are immunopositive to CD34, such as SFT (Figure 2H)

and DFS. Fusiform cells in SFT may also show positivity for bcl-2 and CD99 (Figure 2I). In these situations, the morphological differences should be observed to establish the final diagnosis, although immunohistochemistry can contribute to specific differential diagnosis. For example, bcl-2 is expressed in SCL but is not observed in dermatofibroma, nodular fasciite, or in malignant and benign smooth muscle proliferations [5]. Although adipocytes in SCL are positive to S-100 (Figure 2D), the fusiform cells are negative to this antigen, differentiating SCL from neurofibroma (Figure 2F).

Conclusions

In summary, oral spindle cell lipoma is a rare benign lipomatous tumor. Although it presents a predilection for the tongue and cheek, occasional SCL can occur in the palate. The histologic picture shows a range of variations and the observation of morphological features is important in distinguishing SCL from other fusiform tumors. Immunohistochemistry should be helpful in this differentiation.

Conflict of interest

None declared.

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