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Intraoral spindle-cell lipoma with chondroid differentiation: importance in the diagnosis of oral lesions presenting chondroid tissue

Lipoma de células fusiformes intraoral com diferenciação condroide: importância no diagnóstico de lesões orais contendo tecido cartilaginoso

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ABSTRACT

Lipomas are benign neoplasms of adipose tissue presenting several histologic variants, which can be rarely found in the oral cavity. We present a case of a 62-year-old woman with a submucous nodule located in the tongue. Histopathological examination revealed an encapsulated tumor composed of myxoid tissue, spindle cells and mature adipocytes in transition to cartilaginous tissue. The final diagnosis was spindle-cell lipoma with myxoid change and chondroid differentiation. No sign of recurrence was found after five years. The diagnosis of intraoral mesenchymal lesions with chondroid differentiation requires careful histologic examination, mainly to differentiate between benign and malignant lesions.

Key words: lipoma; diagnosis; microscopy; cartilage.

INTRODUCTION

Lipomas are benign neoplasms of mature adipose tissue, most commonly found in areas where adipose tissue is present, above all in the subcutaneous or submucous regions. They are somewhat rare in the oral region, representing about 1% to 4% of all cases⁽¹⁾. Lipomas often present as a slow-growing mass, almost always asymptomatic⁽²⁾. Their most common location in the oral region is the buccal mucosa, followed by floor of the mouth, tongue and lips^(1,2).

Microscopically, classic lipomas are composed of encapsulated, mature adipose tissue with variably sized adipocytes⁽¹⁾. According to microscopical features, several variants of oral lipomas have been described, including fibrolipoma, angiolipoma, myolipoma, spindle-cell/pleomorphic lipoma, salivary gland lipoma, osteolipoma and chondroid lipoma. Spindle-cell/pleomorphic lipomas are characterized by circumscribed lesions, composed of a variable admixture of adipocytes and spindle cells, hyperchromatic

rounded cells, and multinucleated giant cells associated with ropey collagen^(3,4).

Mesenchymal differentiation of lipomas into bone and cartilage is a rare event^(2,5). Chondroid lipoma is the main variant presenting chondroid tissue, with features of both embryonal fat and embryonal cartilage, and its recognition is important because it can be morphologically similar to sarcomas, particularly myxoid liposarcoma and myxoid chondrosarcoma⁽⁶⁾.

In order to avoid misclassification of different histological types of lipoma, spindle-cell lipoma with cartilaginous differentiation must be distinguished from chondroid lipoma. To the best of our knowledge, only two cases of spindle-cell lipoma with chondroid tissue have been published in the English language literature. Therefore, the purpose of this article is to report a case of spindle-cell lipoma with cartilaginous differentiation affecting the tongue, emphasizing the distinction between these lipomas and chondroid lipoma or other tumors with chondroid tissue.

CASE REPORT

A 62-year-old woman was referred for evaluation of a tongue nodule that had been present for about six months. Her medical history and systemic review were not significant. The patient had no history of trauma in the region. Physical intraoral examination revealed a painless well-delimited submucous nodule, firm on palpation, measuring about two centimeters in diameter noted on the left lateral border of the tongue (**Figure 1**). The main diagnostic consideration was a benign neoplasm, including salivary gland tumors or mesenchymal neoplasms such as lipoma, granular cell tumor, neurofibroma, schwannoma or ectomesenchymal chondromyxoid tumor. According to these hypotheses, an excisional biopsy was indicated. During surgery, a well-circumscribed yellowish mass was found. On gross examination the lesion did not float on formaldehyde. On microscopic examination, the lesion consisted of an encapsulated tumor with myxoid areas admixed with spindle and stellate cells and mature adipocytes in transition to cartilaginous tissue, which displayed well-differentiated areas preferentially located in the central portion (**Figure 2**). Immunohistochemical stains for S100 protein were positive in the adipocytic cells and cartilaginous tissue, and CD34 was found in spindle cells of the myxoid areas (**Figure 3**). Based on these histopathological and immunohistochemical features, a diagnosis of spindle-cell lipoma with prominent myxoid change and chondroid differentiation was rendered. No recurrence was identified during a five-year follow-up period.

DISCUSSION

Lipomas are the most common benign soft tissue mesenchymal tumors, and several microscopical variants have been reported,



FIGURE 1 – Submucous nodule on the left lateral border of the tongue

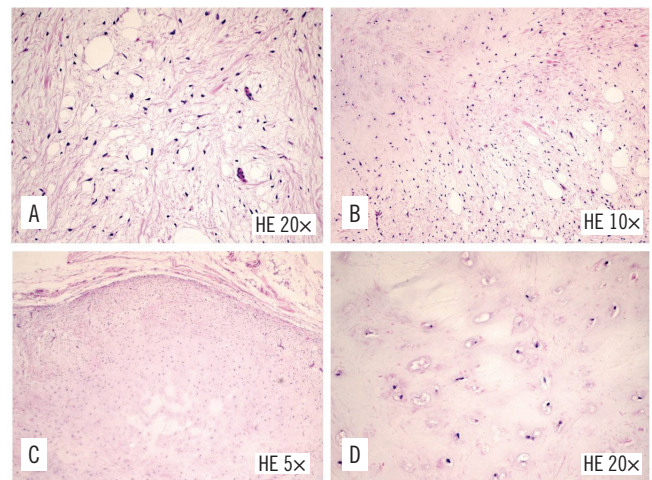


FIGURE 2 – Microscopical features of the spindle-cell lipoma with prominent myxoid change and cartilaginous differentiation

A) areas of lipoblast-like cells in a myxoid stroma; notice the atypical stromal cells; B) transition between the myxoid and cartilaginous areas; C) presence of a fibrous capsule; D) cartilaginous tissue showing mature-appearing cells in a homogeneous stroma. HE: hematoxylin and eosin.

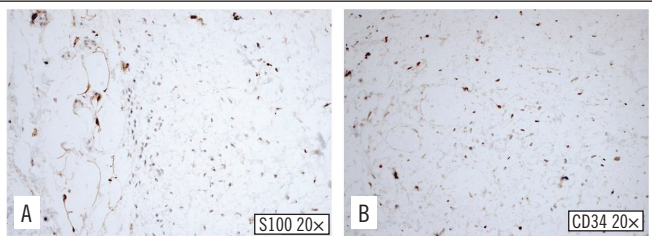


FIGURE 3 – Positive immunohistochemical staining for S100 protein (A) and CD34 (B)

including fibrolipoma, angiolipoma, myolipoma, sialolipoma, osteolipoma, spindle-cell/pleomorphic lipoma and chondroid lipoma. Spindle-cell lipoma is an uncommon variant, first reported by Enzinger and Harvey in 1975⁽⁷⁾. Microscopically, it is composed of mature fat cells, collagen-forming spindle cells with immunoreactivity for CD34 in a fibrocollagenous and myxoid background^(7,8). Some cases of spindle-cell lipoma show prominent myxoid changes⁽⁹⁾. Spindle-cell lipoma accounts for approximately 1.5% of all adipocytic neoplasms⁽¹⁰⁾, and typically occurs in elderly men as a solitary lesion in the posterior neck and back⁽¹¹⁾. It is less commonly found in the oral cavity^(12,13). Searching in the literature for reported cases of oral spindle-cell lipomas, we found 37 cases, which are summarized in the **Table**. From these 37 cases, 20 were situated in the tongue, followed by five cases on the floor of the mouth. Patients' mean age was 57.4 years, ranging from 23 to 88 years. Our case also affected the tongue of the 62-year-old woman, and the microscopic features were compatible with spindle-cell lipoma with prominent myxoid changes and chondroid differentiation.

TABLE – Summary of spindle-cell/pleomorphic lipoma of the oral region reported in the English-language literature

Authors	Year of publication	Location	Gender	Age
McDaniel <i>et al.</i> ⁽¹⁴⁾	1984	Floor of the mouth	F	33
		Tongue	M	52
Christopoulos <i>et al.</i> ⁽¹⁵⁾	1989	Hard palate	M	58
Levy and Goding ⁽¹⁶⁾	1989	Floor of the mouth	F	74
Lombardi and Odell ⁽¹⁷⁾	1994	Tongue	F	68
Khoo <i>et al.</i> ⁽¹⁸⁾	1995	Cheek	M	23
Tosius <i>et al.</i> ⁽⁹⁾	1995	Cheek	M	55
Dutt <i>et al.</i> ⁽¹⁹⁾	1999	Tongue	F	42
Piatelli <i>et al.</i> ⁽²⁰⁾	1999	Cheek	M	75
Piatelli <i>et al.</i> ⁽²¹⁾	2000	Cheek	M	63
Said-Al-Naief <i>et al.</i> ⁽¹²⁾	2001	Tongue	NI	NI
		Tongue	NI	NI
Agoff <i>et al.</i> ⁽²²⁾	2001	Gingivobuccal sulcus	F	61
Darling <i>et al.</i> ⁽²³⁾	2002	Alveolar mucosa	M	69
Piatelli <i>et al.</i> ⁽²⁴⁾	2005	Floor of the mouth	M	50
Billings <i>et al.</i> ⁽¹³⁾	2006	Tongue	M	45
		Tongue	M	67
		Tongue	F	31
		Tongue	F	75
		Lip	F	55
		Floor of the mouth	F	84
		Buccal mucosa	M	88
Coimbra <i>et al.</i> ⁽²⁵⁾	2006	Floor of the mouth	F	29
Imai <i>et al.</i> ⁽⁸⁾	2008	Tongue	M	72
Vecchio <i>et al.</i> ⁽²⁶⁾	2009	Buccal mucosa	M	52
Stokes <i>et al.</i> ⁽²⁷⁾	2011	Maxilla (intraosseous)	M	35
Junior <i>et al.</i> ⁽²⁸⁾	2013	Tongue	F	64
Al-Sheddi <i>et al.</i> ⁽²⁹⁾	2014	Mucogingival sulcus	M	68
Lau <i>et al.</i> ⁽³⁰⁾	2015	Tongue	M	62
		Tongue	M	62
		Tongue	F	61
		Tongue	M	80
		Tongue	F	65
		Tongue	M	35
		Tongue	F	47
		Tongue	M	47
Lin <i>et al.</i> ⁽³¹⁾	2015	Tongue	F	62

F: female; M: male; NI: no information.

The presence of cartilaginous tissue within a lipoma is a relatively rare finding⁽⁵⁾. Chondroid lipoma and chondrolipoma are the main variants that exhibit chondroid tissue formation. Chondroid lipoma is a rare variant, mainly in the head and neck region, and was included in the World Health Organization (WHO) classification of soft tissue tumors in 2002⁽³⁾. Microscopic examination of chondroid lipomas shows mature nests of vacuolated lipoblasts, in a prominent myxoid to hyalinized chondroid matrix, and a variable amount of mature adipose tissue. The tumor is usually circumscribed by a fibrous capsule and there is no cell atypia or mitotic figures⁽³²⁻³⁴⁾. Chondroid lipomas may show positivity for vimentin, S100 protein (in lipoblasts), and CD68

in vacuolated cells. Chondroid lipoma should be distinguished from chondrolipoma, which is a lipoma with cartilaginous metaplasia. In chondrolipomas there is absence of lipoblasts and myxoid matrix, and a clear separation between the cartilaginous tissue and the fatty component⁽⁵⁾. Cases of chondrolipomas have also been reported in the intraoral region⁽³⁵⁻³⁹⁾. Our case does not represent a chondroid lipoma, due to lack of lipoblast-like cells. The presence of chondroid tissue in spindle cell lipoma is very uncommon. Lau *et al.* (2015)⁽³⁰⁾ reported eight cases of spindle-cell lipoma of the tongue, and they found two cases containing a tissue imparting chondroid appearance.

The presence of chondroid differentiation in an oral lesion requires special attention. A variety of benign and malignant tumors, including pleomorphic adenoma, ectomesenchymal chondromyxoid tumor, myxoid liposarcoma, well-differentiated liposarcoma, pleomorphic liposarcoma and extraskeletal myxoid chondrosarcoma, may present chondroid differentiation^(1, 32, 33). Therefore, some histopathological details may be important to differentiate them. Pleomorphic adenoma can resemble, clinically and microscopically, lipoma with chondroid differentiation, mainly because of the myxoid and chondroid stroma. However, this salivary gland tumor shows ductal/epithelial elements which are not found in lipomas⁽⁴⁰⁾. Ectomesenchymal chondromyxoid tumor is a rare benign intraoral mesenchymal neoplasm almost exclusively seen on the dorsum of the tongue⁽⁴¹⁾. Histopathologically, this tumor is usually unencapsulated but well-demarcated, with lesional cells proliferating in a lobular pattern and arranged in cords, strands, and sheets in a myxoid to chondromyxoid background. The cells are either round, oval, polygonal or spindled in morphology. They may have multilobulated nuclei and may occasionally show atypia⁽⁴²⁾. However, mature fat cells are not seen in ectomesenchymal chondromyxoid tumors. The presence of ropey collagen, seen in spindle-cell/pleomorphic lipomas, is useful for differential diagnosis since it is not seen in well-differentiated, myxoid or pleomorphic liposarcoma; moreover, spindle-cell lipomas lack lipoblasts, which can be seen in liposarcomas⁽⁴³⁾. Myxoid liposarcoma is a malignant tumor composed of uniform round- to oval-shaped primitive mesenchymal cells and a variable number of small signet-ring lipoblasts in a prominent myxoid stroma^(6, 32). Well-differentiated liposarcoma, also known as atypical lipomatous tumor, is a locally aggressive malignant mesenchymal neoplasm composed of mature adipocytic proliferation showing significant malignant cytological features; it can sometimes show areas of chondroid metaplasia but without extensive myxochondroid matrix⁽⁴⁴⁾. Extraskeletal myxoid chondrosarcoma is a malignant soft-tissue tumor characterized

by a multinodular architecture, abundant myxoid matrix, and malignant chondroblast-like cells⁽⁴⁵⁾. Lipomas with chondroid differentiation may be confused with this malignant neoplasm

due to the presence of scattered nests of lipoblast-like cells and myxochondroid matrix. However, lipomas are well-circumscribed tumors and always contain a mature fatty component⁽¹⁾.

RESUMO

Lipomas são neoplasias benignas de tecido adiposo que podem apresentar diversas variantes e raramente são encontradas na cavidade oral. Apresentamos o caso de uma mulher de 62 anos de idade com queixa de um nódulo na língua. A análise histopatológica da lesão revelou tumor encapsulado composto de tecido mixoide, células fusiformes e adipócitos maduros em transição para tecido cartilaginoso. Nenhum sinal de recorrência foi observado após acompanhamento de cinco anos. O diagnóstico de lesões mesenquimais intraorais contendo diferenciação condroide requer atenção especial, principalmente para a diferenciação de lesões benignas e malignas.

Unitermos: lipoma; diagnóstico; microscopia; cartilagem.

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