

# Well-Differentiated Liposarcoma/Atypical Lipomatous Tumor of the Oral Cavity: Report of Three Cases and Review of the Literature

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**Abstract** Atypical Lipomatous Tumor/Well Differentiated Liposarcoma (ALT/WDLS) is a soft tissue sarcoma of intermediate malignant behavior, most frequently affecting the retroperitoneum and lower extremities. Oral liposarcomas are very rare neoplasms, the most common histological subtype being ALT/WDLS. In this study, three additional cases of ALT/WDLS located on the tongue (2 cases) and the lower lip (1 case), respectively, are described. Analysis of the salient clinicopathologic features of 63 oral ALT/WDLS cases previously reported in the English language literature, as well as of the 3 cases presented in this study, indicates that the indolent biologic behavior of this tumor justifies its designation as a locally spreading malignant neoplasm, affording a rather conservative surgical approach.

**Keywords** Atypical lipoma · Liposarcoma · Oral neoplasms · Soft tissue sarcoma

## Introduction

Liposarcoma accounts for approximately 20% of all soft tissue malignancies, representing the most common type of sarcoma in adults [1, 2]. It usually involves the deep soft tissues of the trunk and lower extremities, in particular the retroperitoneum, thighs, and buttocks [2]. Liposarcomas are classified into several subtypes including: Atypical Lipomatous Tumor/Well-Differentiated Liposarcoma (ALT/WDLS), which is further subdivided morphologically into lipoma-like, sclerosing, inflammatory, and spindle cell variants; Dedifferentiated; Myxoid/Round-cell; Pleomorphic; and Mixed-type liposarcoma [3]. ALT/WDLS is the most common subtype of liposarcoma (40–45% of cases), arising usually in the retroperitoneum and thighs of middle-aged adults [3].

The concept of ALT was first introduced by Evans et al. [4] in 1979, who suggested that well differentiated liposarcomas of the subcutis and extremities show a more favorable clinical and biologic behavior compared to their retroperitoneal counterparts, and should be designated accordingly as *atypical lipomas* and *atypical intramuscular lipomas*. As the authors emphasized, although these tumors share a great tendency for local recurrence they do not metastasize unless dedifferentiation with development of a high-grade sarcomatous component occurs, thus radical surgical treatment should be avoided. The term *atypical lipomatous tumor* (ALT) was also advocated by Evans [5] in 1988, to describe both atypical lipomas and retroperitoneal tumors without lipoblasts. Subsequently, the term has been applied to lipomatous neoplasms with minimal nuclear atypia and few or no lipoblasts, regardless of location [1], while in the latest WHO classification (2002) the combined term ALT/WDLS is reserved for liposarcomas arising in surgically amenable locations, that despite

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their infiltrative growth pattern and locally aggressive behavior they do not tend to recur after complete surgical removal [3].

Clinical, morphological, and cytogenetic findings provide adequate evidence that ALT and WDLS are identical and both terms should be used synonymously [1, 6]. The theory of ALT/WDLS has gained acceptance, although issues like variations in diagnostic microscopic features and the relationship of the neoplasm to spindle-cell and pleomorphic lipoma have not been settled [1].

Liposarcomas of the head and neck are rare, representing 2–8% of all sarcomas in this region [7]. Oral liposarcomas show a much lower incidence, as relevant reviews have reported a limited number of documented cases [8–11]. In a review of head and neck liposarcomas between 1911 and 2005, approximately 46% of cases (36/78) were localized in the oral mucosa [11], while oral and salivary gland liposarcomas accounted for only 0.3% (18/5,435) of all liposarcomas diagnosed in the Armed Forces Institute of Pathology between 1970 and 2000 [8].

Although ALT/WDLS is the most common type of liposarcoma of the oral mucosa [1, 8–10, 12], pertinent data are limited because most relevant publications have been in the form of either single case reports or small case series. In this study, three additional cases of oral ALT/WDLS are described and a review of the reported oral ALT/WDLS cases in the English language literature is also presented.

## Case Reports

### Case 1

A 62-year-old female patient presented with an asymptomatic, slowly enlarging tongue lesion, first noticed approximately 1 year ago. Her medical history was non-contributory.

Clinical examination revealed a submucosal swelling covered by normal mucosa at the left dorso-lateral border of the tongue. The lesion measured approximately 1 × 1 cm and on palpation was firm, circumscribed and painless. The rest of the oral mucosa was normal and no palpable neck lymph nodes were found. The differential diagnosis included benign tumors of mesenchymal origin, e.g. lipoma, neurofibroma, granular cell tumor, and surgical excision under local anesthesia was performed. The post-operative course of the patient was uneventful with no sign of recurrence 5 years after the excision.

### Case 2

A 36-year-old otherwise healthy female was referred for management of an asymptomatic nodule located on her

lower lip. The lesion had emerged 2 months previously after “a bite” in the area, as reported by the patient. Her medical history was non-contributory.

Clinically, a well-circumscribed submucosal mass measuring approximately 0.8 × 0.5 cm was present on her lower left lip. On palpation, the lesion was elastic and non-tender, while the overlying mucosa was normal. No cervical lymphadenopathy was found. With a provisional diagnosis of an “organized mucocele” the patient underwent complete surgical removal. The post surgical course was uneventful with no evidence of recurrence 2 years after the excision.

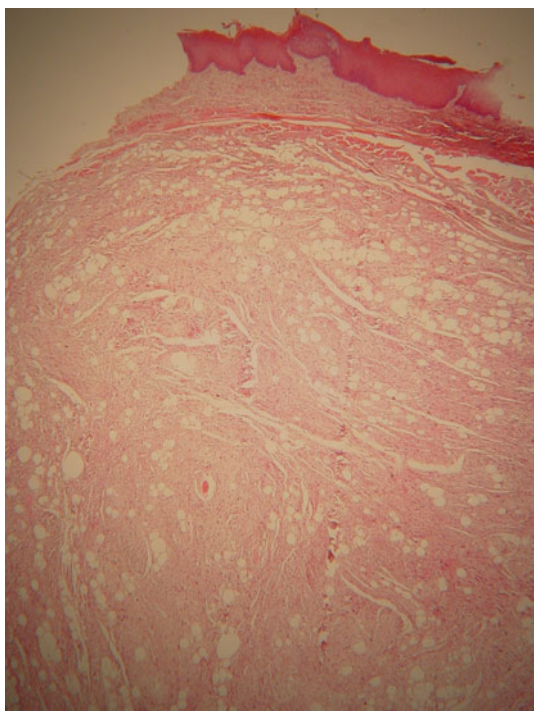
### Case 3

A 70-year-old female patient presented with a slowly growing tongue tumor of 4-years duration. The patient was under treatment for osteoporosis, hypothyroidism, and hypercholesterolemia.

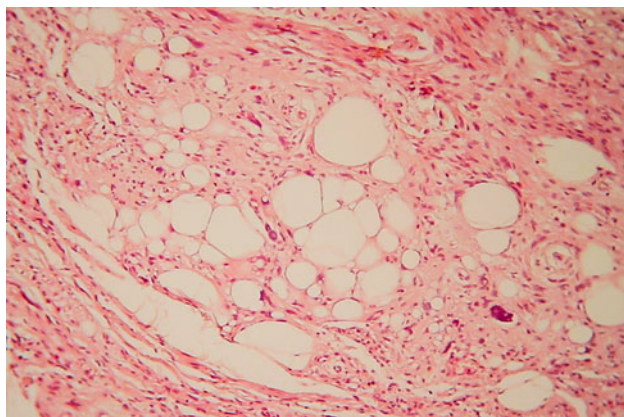
Clinically, a 1.5 × 1.5 cm, asymptomatic, firm, submucosal mass covered by normal mucosa was present on the right side of the anterior third of the tongue. The rest of the oral mucosa was normal and no regional lymphadenopathy was noted. The differential diagnosis included benign mesenchymal tumors and an excisional biopsy was performed. Upon histopathologic examination, infiltration of the surgical margins by neoplastic adipocytes was noted and a re-excision with a 0.5 cm wide clear resection margin was performed. Healing was uneventful and there has been no recurrence 18 months after surgery.

## Pathologic Description

Microscopic examination of formalin-fixed, paraffin-embedded tissue sections showed in all 3 cases an unencapsulated but circumscribed neoplasm with lobules separated by thin fibrous septae (Fig. 1). It was composed of mature adipocytes of variable size and shape (Fig. 2), intermixed with cytologically bland spindle cells (Fig. 3). Numerous multivacuolated lipoblasts with pleomorphic or hyperchromatic nuclei (Fig. 4a), some with a signet-ring appearance (Fig. 4b), were present. In Case 1, rare mitoses (<1 per 10 high power microscopic fields) were recognized (Fig. 4c). Focal infiltration of adjacent small vessels and nerves (Fig. 5), or muscle fibers (Fig. 6) was noted, but necrosis or hemorrhage were absent. Immunohistochemically, neoplastic adipocytes reacted positively with S-100 protein and vimentin. Histopathologic features were consistent with the diagnosis of lipoma-like ALT/WDLS in all cases. The surgical margins in Cases 1 and 2, and the re-excision in Case 3, were negative for tumor.



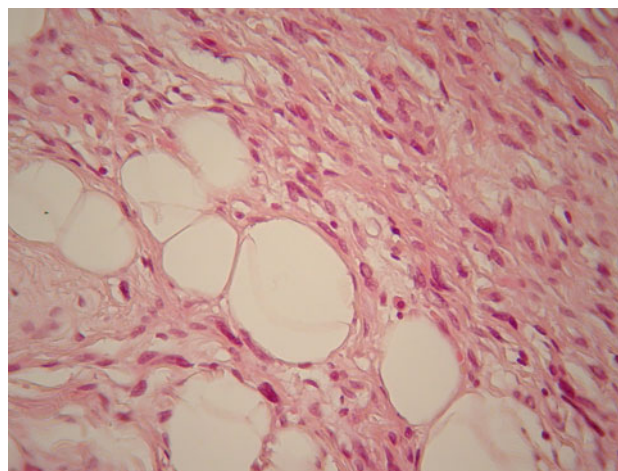
**Fig. 1** Case 1. An unencapsulated but circumscribed cellular mass (hematoxylin-eosin, original magnification  $\times 25$ )



**Fig. 2** Case 1. Mature adipocytes of variable size and shape in a fibrous stroma (hematoxylin-eosin, original magnification  $\times 100$ )

## Discussion

A Pubmed search of the English language literature up to July 2011 disclosed 63 cases of ALT/WDLS with a definite oral location. Age, gender, tumor size and location, histologic subtype, treatment modalities, and follow-up data of these 63 cases as well as of the 3 cases presented herein, comprising a total of 66 cases, are summarized in Table 1 [8–10, 12–39]. Five cases described previously as myxoid WDLS were excluded, since they most probably



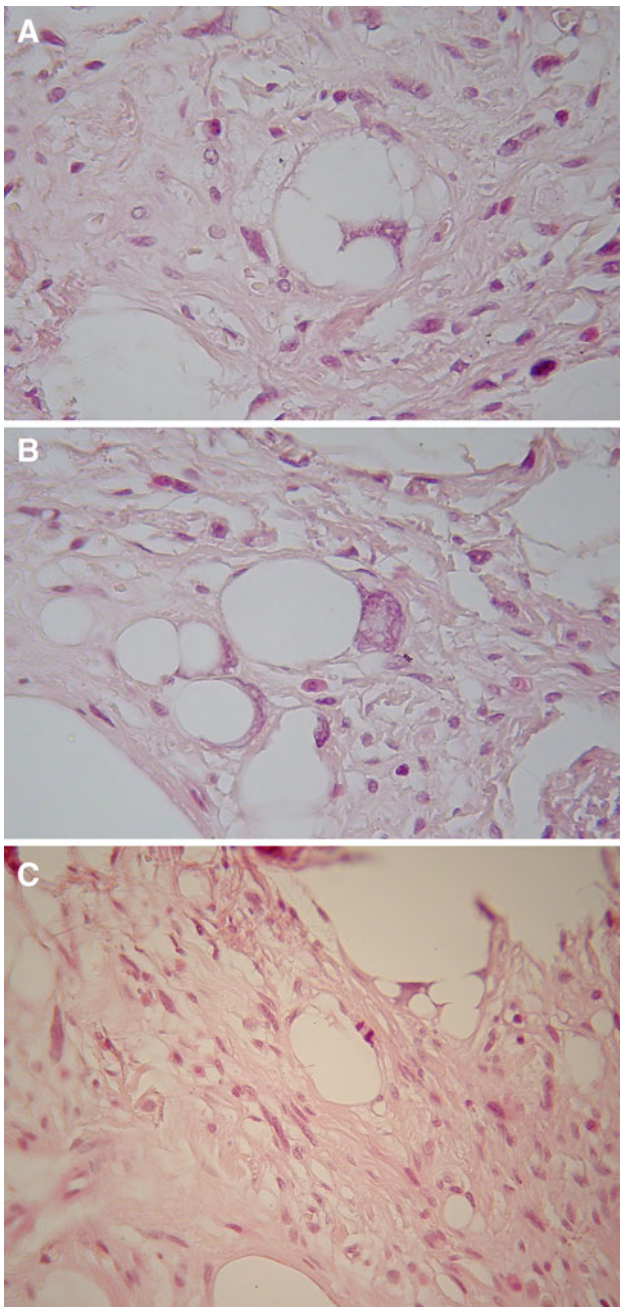
**Fig. 3** Case 1. Adipocytes intermixed with cytologically bland spindle cells (hematoxylin-eosin, original magnification  $\times 100$ )

represented myxoid liposarcomas [40]. Case 4 of Nascimento et al. [12] and case 1 of Allon et al. [36] had been previously reported by other authors [10, 12] and as such they were only counted once. Additionally, case 1 of Minic [41] was excluded because it is more consistent with an extraoral location, as were the two cases (1 buccal and 1 lip) from the series of Davis et al. [7], since pertinent information was not given. Four cases described as ALT with dedifferentiation [12] were analyzed separately (Table 2), in order to compare their characteristics with those of the typical ALT.

According to available data in 64/66 cases, 37 patients (57.8%) were males and 27 females (male-to-female ratio approximately 1.37:1). As has been shown in previous studies, head and neck liposarcomas [7, 12] and intraoral liposarcomas [8–10] show a male predominance, in contrast to the reported female predilection for ALT/WDLS's of various locations [1, 6, 42]. The mean age of 64 patients with available information was  $54.05 \pm 17.84$  years (age range 6 months to 86 years; median 54 years), with 78.1% of patients (50/64) being  $\geq 40$  years-old. This finding is in accordance with the previously reported mean age at diagnosis for intraoral liposarcomas, regardless of histologic subtype [9, 38].

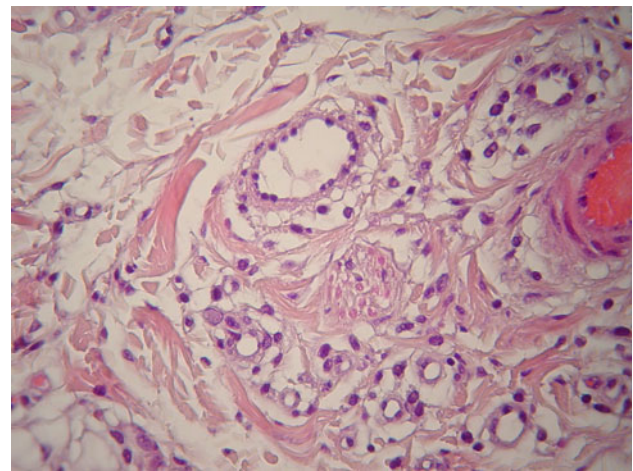
Oral liposarcomas have been reported to occur mainly on the buccal mucosa [8–10, 38, 43], but in the largest case series of ALT's, tongue was the favorite site of involvement [12]. In the present review, 36 out of 65 cases (55.38%) of oral ALT/WDLS were located on the tongue, with exactly 50% of lesions situated on the lateral border (18/36 cases), followed by the buccal mucosa (17 cases), the lips (4 cases), the gingiva/gingival sulcus (3 cases), the palate (3 cases) and the floor of the mouth (2 cases).

Oral ALT/WDLS usually appears as a circumscribed, slowly enlarging, painless, submucosal mass that may be

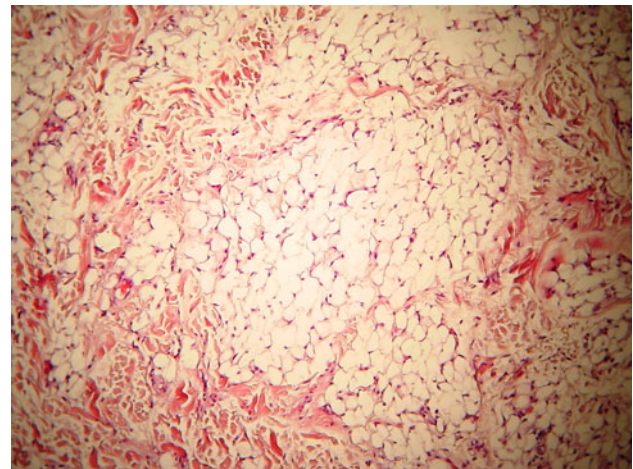


**Fig. 4** **a** Case 3. Lipoblasts with marked nuclear pleomorphism and vacuolated cytoplasm. **b** Case 3. Signet-ring cells. **c** Case 1. A rare mitotic figure (hematoxylin-eosin, original magnification  $\times 400$ )

present for several months or even years before being excised. Rare cases of bilateral or multinodular ALT/WDLS of the tongue have also been reported [22, 27, 36]. The tumor is generally asymptomatic and patients usually become aware of its presence, only when it reaches a size large enough to cause cosmetic or functional problems [10]. Superficial location may account for the small size at the time of diagnosis. In 60 lesions, the maximum diameter ranged from 0.5 to 12 cm, with a mean diameter of



**Fig. 5** Case 2. Small vessel and nerve infiltration by neoplastic adipocytes (hematoxylin-eosin, original magnification  $\times 100$ )



**Fig. 6** Case 3. Infiltration of adjacent muscle fibers by neoplastic adipocytes (hematoxylin-eosin, original magnification  $\times 100$ )

$3.31 \pm 2.64$  cm. The indolent growth and small size of the ALT may justify the usual provisional clinical diagnosis of an irritation fibroma or a benign mesenchymal neoplasm [2, 28, 41]. A provisional diagnosis of either a reactive or benign tumor or a cystic lesion was rendered in all three cases of our series.

Microscopically, lipoma-like was the most common histopathologic subtype, while 10 cases showed a spindle cell component, 6 displayed sclerosing areas, 2 focal myxoid changes, and 1 heterologous cartilage formation.

Four cases of ALT with dedifferentiation have been reported in the English literature (Table 2) [12, 44]. The mean age of patients was  $50.75 \pm 23.04$  (age range 29 months to 83 years; median 45.5 years), while 3/4 were females. The tumors were located one each on the tongue, floor of the mouth, buccal mucosa, and gingival sulcus, while their mean diameter was  $4.6 \pm 3.51$  cm (range

**Table 1** Clinicopathological features of 66 cases of oral ALT/WDLs

Author	No	Age/ gender	Size (greatest dimension in cm)	Location	Diagnosis	Follow-up (in months)
Henefer et al. [34]	1	39/M	7.5	Buccal mucosa	WDLS	18 NED
Larson et al. [35]	2	42/F	1.5	Tongue (ventral)	WDLS	N/A
Saunders et al. [36]	3	31/M	5	Soft palate	WDLS, sclerosing	18 NED
Yamada et al. [31]	4	51/F	12	Buccal mucosa	WDLS	12
Sadeghi and Sauk [18]	5	42/F	5.2	Buccal mucosa	WDLS	–
Suzuki et al. [37]	6	0.5/M	4.5	Buccal mucosa	WDLS	36 NED
Charnock et al. [38]	7	63/M	3	Buccal mucosa	WDLS	24 NED
Ruacan et al. [39]	8	32/M	3	Buccal mucosa	WDLS	10 NED
Nakahara et al. [40]	9	48/F	2.2	Floor of the mouth	WDLS with a myxoid variant	12 NED
Lucas et al. [41]	10	–/–	–	Buccal mucosa	WDLS	N/A
Kacker and Taskin [42]	11	78/M	6.0	Tongue (lateral)	Atypical lipoma	N/A
Saddik et al. [32]	12	76/M	2.5	Tongue (base/tonsillar fossa)	WDLS	4 recurrences in 23 years
Senyuva et al. [43]	13	71/M	7.9	Buccal mucosa	WDLS, sclerosing	12 NED
Kamikaidou et al. [23]	14	60/F	5	Buccal mucosa	WDLS	1 recurrence at 9 months as myxoid liposarcoma
Nelson et al. [25]	15	37/M	3.0	Tongue (anterior dorsolateral/ ventral)	WDLS or atypical lipoma	18 NED
Orita et al. [33]	16	70/M	3.5	Tongue (lateral)	WDLS, lipoma-like	8 NED
Gagari et al. [10]	17	73/M	2.0	Tongue (anterior dorsolateral)	WDLS, lipoma-like	LFU
Nikitakis et al. [11]	18	28/M	4	Buccal mucosa	WDLS	17 NED
Moore et al. [44]	19	43/M	0.8	Tongue (anterior dorsolateral)	Atypical lipoma	10 NED
Nunes et al. [45]	20	65/M	1.0	Tongue (anterior dorsal)	WDLS	18 NED
Nascimento et al. [12]	21	36/M	0.6	Tongue	WDLS, adipocytic (ALT)	108 NED
	22	48/F	3	n/a	WDLS, adipocytic (ALT)	N/A
	23	50/M	1.2	Tongue	WDLS, adipocytic (ALT)	48 NED
	24	47/F	2.2	Tongue	WDLS, adipocytic (ALT)	16 NED
	25	74/M	–	Tongue	WDLS, adipocytic (ALT)	4 recurrences in 17 months
	26	77/F	0.6	Tongue	WDLS, adipocytic (ALT)	20 NED
	27	43/M	1.0	Tongue	WDLS, adipocytic (ALT)	11 NED
	28	45/M	1.5	Gingiva	WDLS, adipocytic (ALT)	24 NED
	29	72/M	0.7	Tongue	WDLS, adipocytic with heterologous cartilage (ALT)	24 NED
	30	–/–	–	Buccal mucosa	WDLS, adipocytic with spindle cell features (ALT)	N/A

**Table 1** continued

Author	No	Age/ gender	Size (greatest dimension in cm)	Location	Diagnosis	Follow-up (in months)
Fanburg-Smith et al. [9]	31	80/M	1.0	Tongue	WDLs, adipocytic with spindle cell features (ALT)	2 NED, then LFU
	32	35/F	4	Floor of the mouth	WDLs, adipocytic with spindle cell features (ALT)	25 NED
	33	49/F	3	Buccal mucosa	WDLs, adipocytic with spindle cell features (ALT)	24 NED
	34	51/F	0.9	Lip	WDLs, adipocytic with spindle cell features (ALT)	3 NED, then LFU
	35	53/M	0.8	Tongue	WDLs, adipocytic with spindle cell features (ALT)	2 NED, then LFU
	36	47/M	6	Buccal mucosa	WDLs, adipocytic with spindle cell features (ALT)	Multiple recurrences in 7 years
	37	28/F	2	Lip	WDLs, adipocytic with spindle cell features (ALT)	9 NED, then LFU
	38	55/F	1	Soft palate	WDLs, adipocytic with spindle cell features (ALT)	N/A
	39	36/F	6	Palate	WDLs, sclerosing	LFU
	40	41/M	–	Buccal mucosa	WDLs	276 NED
	41	43/F	2.0	Tongue (lateral)	WDLs	24 NED
	42	52/M	5.5	Mandibular soft tissues	WDLs	192 NED
	43	64/F	1.5	Tongue	WDLs	192 NED
	44	67/F	6	Buccal mucosa	WDLs	180 NED, 2 recurrences at 4 and 5 years
	45	69/M	–	Buccal mucosa	WDLs, focal myxoid features	LFU
	46	70/M	2.1	Buccal mucosa	Atypical lipoma, sclerosing	144 NED
	47	81/M	1.2	Lower lip	ALT/WDLs	LFU
	Hattori [46]	48	58/F	2.5	Tongue (lateral)	WDLs
49		86/M	0.5	Tongue (anterior/middle lateral)	ALT	N/A
Allon et al. [18]	50	68/F	1	Tongue	ALT	12 NED
	51	72/M	0.7	Tongue	ALT	12 NED
	52	57/M	2	Tongue	ALT	12 NED
Chan et al. [49]	53	58/M	1	Anterior mandibular alveolus	WDLs, lipoma like	12 NED
	54	58/F	0.6	Tongue (lateral)	WDLs/ALT	18 NED
DeWitt et al. [21]	55	62/F	0.7	Tongue (lateral)	WDLs/ALT	12 NED
	56	36/F	–	Tongue	WDLs/ALT	N/A
Paragis Sanchez et al. [50]	57	78/F	1.1	Tongue (lateral)	ALT, lipoma like	63 NED
	58	27/F	0.7	Tongue (lateral)	ALT, lipoma like	58 NED

Table 1 continued

Author	No	Age/ gender	Size (greatest dimension in cm)	Location	Diagnosis	Follow-up (in months)
	59	65/M	2	Tongue (multiple nodules, bilaterally)	ALT, lipoma like	Recurrence at 59 months
	60	11/M	0.8	Tongue (lateral)	ALT, sclerosing	N/A
	61	75/M	1	Tongue (lateral)	ALT, sclerosing	159 NED
	62	50/F	1.2	Tongue (lateral)	ALT, lipoma like	4 NED
Moritani [51]	63	68/M	1.3	Tongue (lateral)	ALT	36 NED
Present report	64	62/F	1.0	Tongue (anterior lateral)	ALT-lipoma like	60 NED
	65	36/F	0.8	Lower lip	ALT-lipoma like	24 NED
	66	70/F	1.5	Tongue (anterior lateral)	ALT-lipoma like	18 NED

F female, M male, N/A not available, NED no evidence of disease, LFU lost to follow-up

1–8 cm). In all 4 cases, dedifferentiation was observed during initial histopathologic diagnosis and in 2/4 cases was described as “incipient”, characterized by small areas of increased cellularity with focal pleomorphism [45]. This finding is possibly consistent with the reported potential of gradual dedifferentiation in some cases of ALT/WDLS’s, although the prognostic significance of small foci of dedifferentiation remains undetermined [46]. The concept of low-grade dedifferentiation, consisting of fibroblastic areas with mild nuclear atypia, as a precursor lesion of high-grade dedifferentiation has also gained acceptance, although no differences regarding the clinical outcome seem to exist between the two grades [46–48]. It is noteworthy that these cases of dedifferentiation reveal a younger median age, female preponderance, and a greater diameter compared to ALT/WDLS’s, although the number of cases is too small for conclusions. Prognostically, for an average of 27.6 months of follow-up, none of these cases was reported to recur or metastasize.

The histopathologic differential diagnosis of ALT/WDLS includes benign adipose tissue lesions, such as: the ordinary lipoma; spindle-cell, pleomorphic or chondroid lipoma; lipogranuloma; lipoblastoma; angiolipoma; fibrolipoma; hibernoma; and intramuscular lipoma [1, 2, 12, 22]. ALT/WDLS is usually well-circumscribed but unencapsulated, similar to an ordinary lipoma, while the presence of muscle infiltration may mimic the features of an intramuscular lipoma. Additionally, mitoses are rare and necrosis or hemorrhage are absent, while sclerosing ALT/WDLS may show extensive fibrosis simulating degenerative changes noted in lipomas. On the other hand, lipoblasts, varying from primitive mesenchymal cells with few, inconspicuous lipid droplets to signet ring cells with laterally displaced or indented nuclei, are only occasionally seen in ALT’s [2]. Interestingly, Fanburg-Smith et al. [8] have noticed that tongue ALT/WDLS contain more lipoblasts, compared to liposarcomas of other oral and extraoral sites. However, lipoblasts are not diagnostic for ALT/WDLS as they may be found in benign adipocytic lesions, such as lipoblastoma, pleomorphic lipoma, and chondroid lipoma [1, 8], while cells mimicking lipoblasts may be seen in spindle cell lipoma (“pseudo-lipoblasts”) [49] and various reactive conditions (“lipoblast-like cells”), such as fat necrosis and granulomatous foreign-body reactions (e.g. silicone granuloma) [50]. Careful inspection and extensive sampling allow the recognition of atypical cells with large, hyperchromatic and bizarre nuclei, as well as a great variation in cell size and shape, which are diagnostic for ALT/WDLS [12]. These findings and the presence of dense rather than wiry collagen fibers separating lobules of neoplastic cells [49] proved useful in Case 1 of the present study, where an abundance of spindle cells was strongly reminiscent of a spindle cell lipoma.

**Table 2** Clinicopathological features of 4 cases of oral ALT/WDLS with dedifferentiation reported in the English literature

Author	No	Age/ gender	Size (greatest dimension in cm)	Location	Diagnosis	Follow-up (in months)
Da Cuhna et al. [26]	1	42/F	1	Gingival sulcus	WDLS with angiosarcomatous dedifferentiation	12 NED
Nascimento et al. [12]	2	29/M	5	Buccal mucosa	WDLS, adipocytic (ALT), “incipient” dedifferentiation	5 NED, then LFU
Nascimento et al. [12]	3	83/F	2.5	Tongue	WDLS, adipocytic with dedifferentiation (non-lipogenic, round cells)	N/A
Nascimento et al. [12]	4	49/F	8	Floor of the mouth	WDLS, adipocytic with spindle cell features (ALT), “incipient” dedifferentiation	66 NED

*F* female, *M* male, *N/A* not available, *NED* no evidence of disease, *LFU* lost to follow-up

Immunohistochemistry is rather of minimal value in the diagnosis of ALT/WDLS [12]. Lipoblasts may be identified by S-100 protein positivity, while HMB-45 negativity may be of help in the differential diagnosis from angiolipoma [3]. CD34 immunostaining is also of limited use in cases of ALT resembling spindle cell lipoma, since CD34 positive ALT/WDLS's have been reported [12, 49].

The histologic classification, as well as differential diagnosis, of liposarcomas may be facilitated by the recognition of distinct cytogenetic phenotypes [1, 51]. ALT/WDLS features ring or giant marker chromosomes, composed of DNA sequences derived from the 12q14–15 region. Overexpression of MDM2 (murine double minutes) nuclear phosphoprotein, CDK4 (cyclin-dependent kinase 4), and HMGI-C glycoprotein (High-mobility group protein isoform I-C), suggesting loss of control at the G1-S checkpoint, are frequently found in ALT/WDLS, and their prognostic value remains to be decided [42]. Immunohistochemical expression and amplification by FISH analysis of MDM2 and CDK4 have also been described in oral ALT/WDLS [22]. Myxoid and round cell liposarcomas show a characteristic t(12;16) translocation or, more rarely, a t(12;22) translocation, leading to the fusion of the transcription factor gene CHOP on 12q13 with the transcription factors TLS/FUS on chromosome 16p11 or EWS on chromosome 22, respectively. Complex rearrangements have also been identified in pleomorphic liposarcomas.

Wide surgical excision with free margins is usually curative for ALT/WDLS [4] and inadequate excision is considered the main reason for local recurrence, estimated at approximately 30% [1, 10]. Post-operative selective radiation may be of some benefit in certain cases, while the role of adjuvant chemotherapy has not been adequately evaluated [7, 41, 43].

Follow-up, ranging from 2 months to 23 years, was available in 52 out of 66 oral ALT/WDLS. Six tumors recurred (11.53%) within a mean time of 40 months, most of them many times during the follow-up period, but none was reported to metastasize. One tumor recurred as a

myxoid liposarcoma [25, 52] while in another case of recurrent tumor situated in the base of the tongue, the diagnosis of ALT/WDLS was established after the last recurrence [25, 52]. As recurrence may develop many years after initial treatment, a continuous and close follow-up is necessary.

A better prognosis for liposarcomas of the head and neck has been associated with the superficial location, small size, and predominance of the ALT/WDLS variant [29, 41]. In general, superficially located liposarcomas behave in a rather benign manner, in contrast to deep-seated lesions, such as retroperitoneal or mesenteric liposarcomas. Most intraoral liposarcomas are usually superficial and well circumscribed, although they lack a true capsule and satellite nodules may be seen infiltrating adjacent tissues [9, 10]. A  $\leq 5.0$  cm maximum diameter is considered a favorable prognostic marker for oral liposarcomas [8]. ALT/WDLS and myxoid liposarcomas have a much better prognosis than the other histologic subtypes and they rarely metastasize [7, 10, 12, 43]. In contrast, round-cell, pleomorphic, and dedifferentiated liposarcomas show higher rates of recurrence and metastasis, particularly to the lungs, brain, spine, liver, pleura, and kidney [10, 29]. It should be noted though, that even in the three cases of oral ALT/WDLS with dedifferentiation and available follow-up data, no evidence of local recurrence or metastasis was reported.

In conclusion, most of the hitherto reported oral ALT/WDLS have been small-sized non-metastasizing tumors that have exhibited an excellent outcome after complete excision and a low recurrence rate. The clinicopathologic features, as well as the indolent biological behavior of oral WDLS's, justify their designation as ALT; thus, conservative surgical excision with close follow-up rather than a radical surgical approach should be preferred and oral surgeons should be informed accordingly by the oral pathologist.

**Conflict of interest** The authors declare that they have no conflict of interest.



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