



SYMMETRIC ATYPICAL LIPOMATOUS TUMOR OF THE TONGUE

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<p>Article Info <i>Received 16/11/2015</i> <i>Revised 29/12/2015</i> <i>Accepted 30/12/2015</i></p> <p>Key words: Atypical lipomatous tumor; Well differentiated liposarcoma, Tongue.</p>	<p>ABSTRACT Atypical lipomatous tumor/Well differentiated liposarcoma (ALT/WDLS) is a rare tumor in the tongue. We report a very rare case of bilateral involvement of ALT of the tongue. A 79-year-old Asian male presented at our department with slowly growing symmetric masses of the tongue. A Computed tomography (CT) scan demonstrated large inhomogenous low fatty density mass along the margin of the tongue. In histological examination, the tumor had proliferation of mature adipocytes, fibrous septa, entrapped striated muscle fibers in a cross section and admixture of lipoblasts and mature adipocytes. The histopathologic diagnosis was ALT/WDLS. Although recurrence of ALT/WDLS is thought to be unlikely after complete excision, close long-term follow-up is necessary due to its malignant potential when dedifferentiating.</p>
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INTRODUCTION

Liposarcomas are the most common soft tissue neoplasms of benign behavioral, approximately accounting for 20% of all soft tissue sarcomas [1]. Liposarcomas are classified into five subtypes: Atypical lipomatous tumor (ALT)/Well-differentiated liposarcoma (WDLS), which is further subdivided morphologically into lipoma-like, sclerosing, inflammatory, and spindle cell variants; Dedifferentiated; Myxoid/Round-cell; Pleomorphic; and mixed-type liposarcoma [2]. ALT/WDLS accounts for about 40-45% of all liposarcomas occurring as thighs or retroperitoneal tumor in late adult life, mainly between the fifth and the seventh decades [2]. Liposarcomas of the head and neck are rare, representing 2-8% of all sarcomas in this region [3]. Fanburg-Smith et al. reported oral and salivary gland liposarcomas accounted for only 0.3% of all liposarcomas [4].

CLINICAL REPORT

A 79-year-old Asian male presented at our department with slowly growing symmetric masses of the tongue. The patient had been aware of this slow enlargement for 4 years. He had occasional trauma from

accidental biting and difficulty in speech and swallowing. Clinical examination revealed symmetric bilateral tongue masses which were about 6×1.2 cm sized, painless, rubbery, yellowish in color and covered by normal mucosa [Fig. 1]. No other tumor-like masses could be identified on the trunk, head and neck or extremities. His medical history was non-contributory. Laboratory test results were within normal limits.

A Computed tomography (CT) scan demonstrated large inhomogenous low fatty density mass along the margin of the tongue [Fig. 2].

Under nasotracheal intubation, masses were entirely excised with surface mucosa. The tumor masses were not encapsulated, but easily separated from surrounding tissue without adhesion.

The excised masses were 6.5×2.8×1.8 cm (right side) and 6.3×2.6×1.5 cm (left side) sized, yellowish, non-encapsulated [Fig. 3]. In histological examination, the tumor had proliferation of mature adipocytes, fibrous septa, entrapped striated muscle fibers in a cross section [Fig. 4] and admixture of lipoblasts and mature adipocytes [Fig. 5]. The histopathologic diagnosis was ALT/WDLS.



Although we recommend clear marginal resection surgery for preventing recurrence, the patient refused further

operative treatment. The post-operative course was uneventful. The patient is currently on close follow-up.

Figure 1. The patient's tongue shows symmetric enlargement in both lateral borders.



Figure 2. Axial sectioned computed tomography showing inhomogenous fatty density lesion along the lateral border of the tongue.

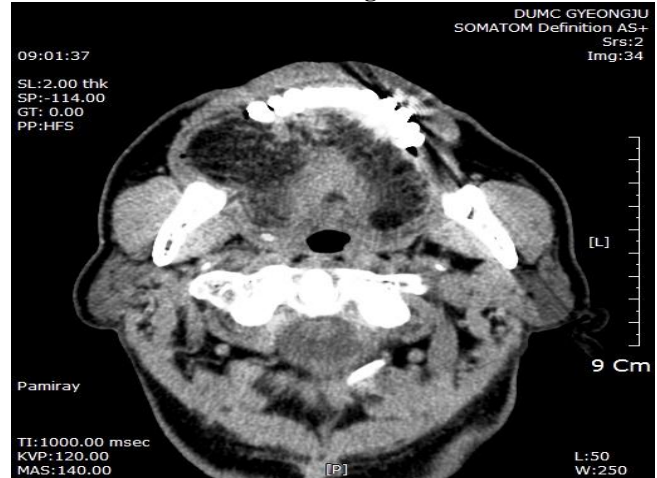


Figure 3. Specimen findings show 6.5×2.8×1.8 cm (right side) and 6.3×2.6×1.5 cm (left side) sized, yellowish, non-encapsulated, fatty tissues.



Figure 4. Mature appearing fat cells separated by fibrous septa. Entrapped striated muscle fibers in a cross section are evident (hematoxyline and eosin, original magnification 100×).

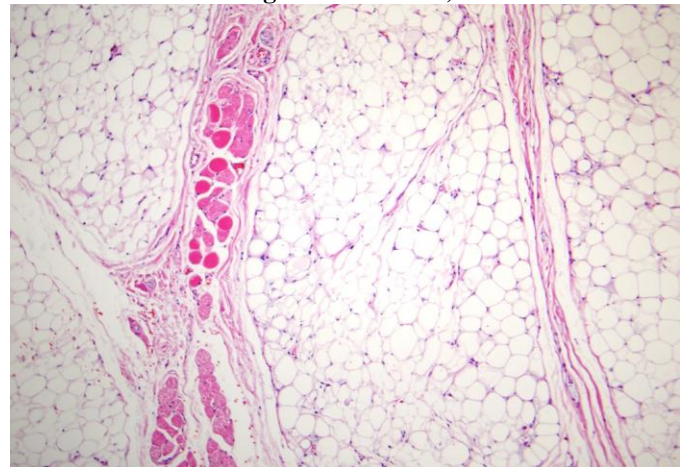
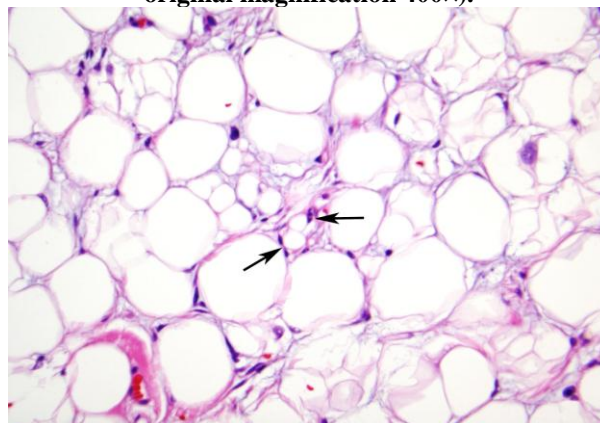


Figure 5. Admixture of multivacuolated lipoblasts (black arrows) and mature adipocytes (hematoxyline and eosin, original magnification 400×).



DISCUSSION

Liposarcoma is the most common mesenchymal malignancy of adulthood, arising in deep soft tissue of the extremities and in the retroperitoneum and in the abdominal cavity [2]. The occurrence in the head and neck region is very rare, accounting for approximately 4% of all liposarcoma cases [5]. DeWitt et al. [6] stated that in approximately 90% liposarcoma cases of the oral region, 38% involved the buccal mucosa, 33% involved the tongue, 7% involved the palate and 7% involved the floor of the mouth. ALTs/WDLs account for 40-45% of all liposarcoma cases [2].

Previous reports showed a male/female ratio of about 2:1 for liposarcoma in the head and neck region, indicating a male preponderance [7].

The liposarcomas within oral and maxillofacial region often appear as painless, slow-growing swelling or soft tissue masses. Its rarity and lack of characteristic symptoms and signs reduced the opportunity to early detect and management.

Clinical findings of the tongue indicate that lesions requiring differential diagnosis are slow-growing lesions, such as lipoma, lymphoepithelial cyst and neurilemmoma [8]. When liposarcoma is compared with lipoma, liposarcoma tends to be harder, to be more elastic and to adhere more to surrounding tissue. Therefore biopsy is necessary for differential diagnosis [9].

The presence of fat in CT and MRI may suggest a lipomatous tumor. Several characteristic findings including more than 75% fat, thick septa may contribute a lot to differential diagnosis between lipoma and ALT/WDLs [10]. However, it is very difficult to definitely diagnose such masses between liposarcomas and other soft tissue neoplasm based on radiography.

A proliferation of variable sized mature adipocytes with fibrous septa and presence of stromal cells with hyperchromatic nuclei and lipoblast cells with vacuolated nuclei are usually identified in a part of lipoma-type ALTs as a characteristic image. All these findings were considered to be consistent with ALT [8].

The lipogenic differentiation marker S-100, indicative of presence of lipoblasts, was stained positive in ALT/WDLs and myxoid liposarcoma, but negative in other subtypes [11]. Several tumor-associated genes including MDM2, HMGA2 and CDK4 were identified and proven beneficial for differential diagnosis by

immunohistochemical detection or fluorescent in situ hybridization [12].

Complete surgical excision with free margins is the primary treatment option for liposarcomas [7]. The values of adjuvant radiotherapy and chemotherapy for liposarcoma still remain controversial. Radiotherapy alone is occasionally considered as an alternative for selected cases especially for impossible total excision or recurrent tumors [11]. Golledge et al. [7] reported that patients treated with surgery alone had a 5-year survival rate of 83% as compared with 63% for those treated with combined surgery and radiotherapy. Local recurrence resulting from incomplete excision becomes a common challenge for successful management of liposarcoma. Although these tumors sometimes appear well encapsulated, wide excision should be emphasized to avoid microscopic residual disease.

The optimal treatment for ALT of the tongue is wide surgical excision with free margins. Among the reported cases of ALT of the tongue, 15% of tumors had recurred and 7.5% lesions demonstrated multiple recurrences [13]. No metastases from lingual ALT have been reported so far.

Careful follow-up after complete excision has been recommended for monitoring recurrence and distant metastasis [11].

Final comments

Although ATL is an extremely rare of the tongue and usually has benign behaviors, close long-term follow-up is necessary because of possibility of its malignant potentials.

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CONFLICT OF INTEREST:

The authors declare that they have no conflict of interest.

STATEMENT OF HUMAN AND ANIMAL RIGHTS

All procedures performed in human participants were in accordance with the ethical standards of the institutional research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

This article does not contain any studies with animals performed by any of the authors.

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