



A Case of Intramuscular Lipoma Arising in the Inferior Surface of the Tongue

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Abstract

Intramuscular lipoma is a benign soft tissue tumor that accounts for 1.8 to 2% of all lipomas, which often originates from the large muscles in the extremities, rarely in the oral region. This time, we experienced a case of intramuscular lipoma on the undersurface of the left side of the tongue and reported the overview with literal consideration. The patient was a 62-year-old male, and complained of indolent swelling on the undersurface of the left side of the tongue and sought medical attention. Relatively well-demarcated low-density area approximately 10 mm in greatest dimension was observed in the left side of the tongue tip on the CT images, while relatively well-demarcated area with hyperintensities in the inside and partially muscular tissues and partially linear portion showing the same signal with muscular tissues on T1- and T2-weighted MRI images. A diagnosis of lipoma was made based on clinical findings and imaging findings of the CT and MRI, and removal was performed using general anesthesia. Histopathologically, the fat tissues had no surrounding membranes and were lobulated and grew into the muscular layer, tumor cells were large, adipocyte-like cells with clear cytoplasm grew into the muscular layer, and a diagnosis of intramuscular lipoma was made. We searched for and collected case reports on intramuscular lipoma in the stomatognathic area in Japan and identified 14 cases including this case. Particularly, 3 cases of intramuscular lipoma on the undersurface of the tongue including this case were identified and the location was considered to be rare. Recurrence rate of intramuscular lipoma was high level of 3% to 62.5%, and it is important to examine much imaging information in detail from imaging diagnosis with CT and MRI examinations. This patient was well after surgery without signs of recurrence.

Keywords: Lipoma, Intramuscular lipoma, Inferior surface of tongue

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Conflict of interest and Ethical prospect

In this article, we do not have any conflict of interest (COI). As for ethical prospect, we have complied with the Declaration of Helsinki and have obtained informed consent from the patient for this case report. This article was disclosed partially at the 7th Clinico-pathological conference (CPC) at Matsumoto Dental University Hospital on February 19, 2015.

Introduction

Lipoma is the most commonly-observed nonepithelial benign tumor among soft tissue tumors which consist of mature adipose cells and develop subcutaneously. Lipoma is categorized to superficial lipoma which is observed in the subcutaneous tissues and to deep lipoma which is observed in the subfascial, intramuscular and intermuscular tissues. Mostly it is a soft single tumor which consists of mature adipose tissue, while it becomes multiple in a rare frequency. It likely develops in the subcutaneous tissue of back, shoulder, face, neck and thighs. Intramuscular lipoma is a benign soft tissue tumor which accounts for 1.8 to 2% of whole types of lipoma^[1]. It likely occurs in the muscles of extremities and rarely occurs in the oral area. At this time, we experienced a case of single intramuscular lipoma which occurred

in the left sublingual region; thus report the summary with the imaging and by adding some bibliographic consideration.

Clinical Case Report

A 62-year-old male patient found painless swelling in the sublingual region from the beginning of September 2014, which was disturbing; thus visited a nearby dentist then our hospital for a work-up. His general physical findings on the first visit were body height: 165 cm, body weight: 70.4 kg, body temperature: 35.1°C, and blood pressure: 145/85 mmHg. The local appearance was a pale-yellow, soft-elastic painless bump of 20 mm in longitudinal axis x 15 mm in horizontal axis in the left inferior surface of the tongue. No redness and haphalgesia was observed in the mucosa of that area, and nothing was palpable in the neck and the submandibular lymph nodes. His medical history was diabetes mellitus (HbA1c: 6.9, blood glucose level: 143) and hypertension since the age of 28-year-old, for which he has been

treated with pioglitazone hydrochloride, sitagliptin phosphate hydrate and voglibose and has been on regular visits for the treatment. There was no remarkable family history. Due to swelling observed in the left sublingual region at the consultation, CT and MR tests were performed for a work-up. In the CT imaging, a low density area with the greatest dimension of approximately 10 mm with relatively clear borders was found in the left proglossis part, and the internal CT value indicated -108 (Fig 1A, B). In the MR imaging, a round-like shaped tumor mass formation figure of around 1 cm dimension was observed in the slightly left area on the inferior/center of proglossis part, and a high signal area was found inside with relatively clear borders and a linear site which showed partially the equal signal to the muscular tissue was found in the T1-highlighted imaging (Fig 2 A, B) and T2-highlighted imaging (Fig 2 C, D). A low signal area was found in the fat-restraining T2-highlighted imaging (Fig 2 E, F).

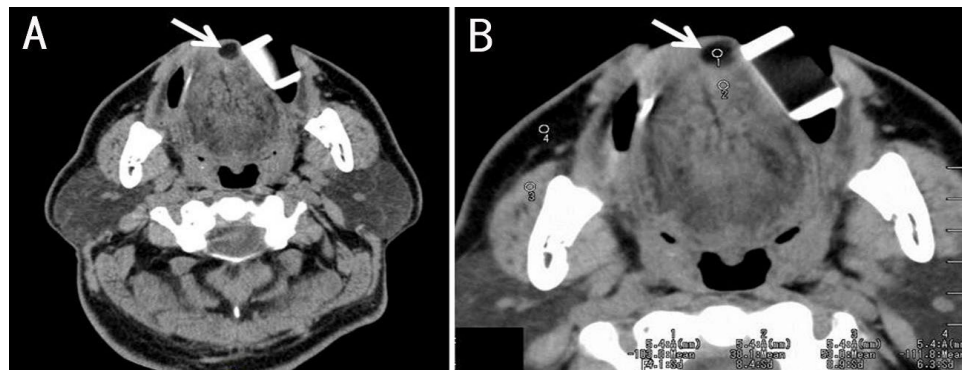


Figure 1: CT imaging (axial image)

(A) A low density area with the greatest dimension of approximately 10 mm with relatively clear borders was found in the left proglossis part (arrow).

(B) The internal CT value indicated -100, suggesting lipid (arrow).

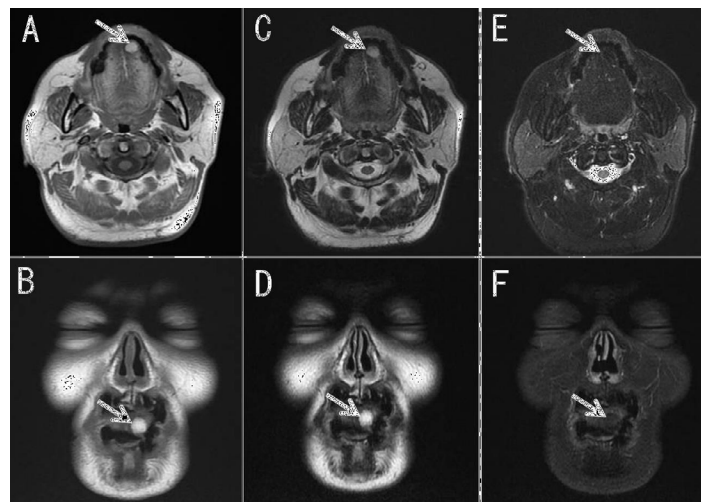


Figure 2: MR imaging (A, B: T1WI; C, D: T2WI; E, F: fat suppression T2WI)

In the MR imaging, a round-like shaped tumor mass formation figure of around 1 cm dimension was observed in the slightly left area on the inferior/center of proglossis part, and a moderate to high signal area was found inside with relatively clear borders and a linear site which showed partially the equal signal to the muscular tissue was found (arrow) in the T1-highlighted imaging (A, B) and T2-highlighted imaging (C, D). A low signal area was found in the fat-restraining T2-highlighted imaging (arrow) (E, F).

Suspected lipoma in the sublingual region was diagnosed according to the clinical diagnosis, CT and MR imaging diagnosis. Tumor mass enucleation was performed under local anesthesia in late October 2014. After peeling off the covering mucosa then confirming the adipose tissue, the tumor mass was enucleated as one mass by bluntly peeling off from the surrounding tissue. The macroscopical findings of the enucleated part from the left sublingual region were pale yellow, smooth on the surface and soft-elastic (Fig 3 A, B). In the histopathological diagnosis, the tumor mass consisted of adipose tissue without clear surrounding capsular structure. This adipose

tissue was separated lobularly with the fibrous tissue with observed proliferation into the muscular wall (Fig 3 C). The adipose tissue which composed the tumor was big and mature adipocyte, with big and clear cytoplasm, and with the nuclear compressed to the cellular rim (Fig 3 D). While these tumor cells were mildly different in size, it was not observed to be clearly atypical. In addition, no lipoblast-like cells were observed within the tumor tissue. The histopathological diagnosis was intramuscular lipoma. The healing process of the surgical wound was good, and the patient has been under regular outpatient observation by explaining that sufficient observation is required.

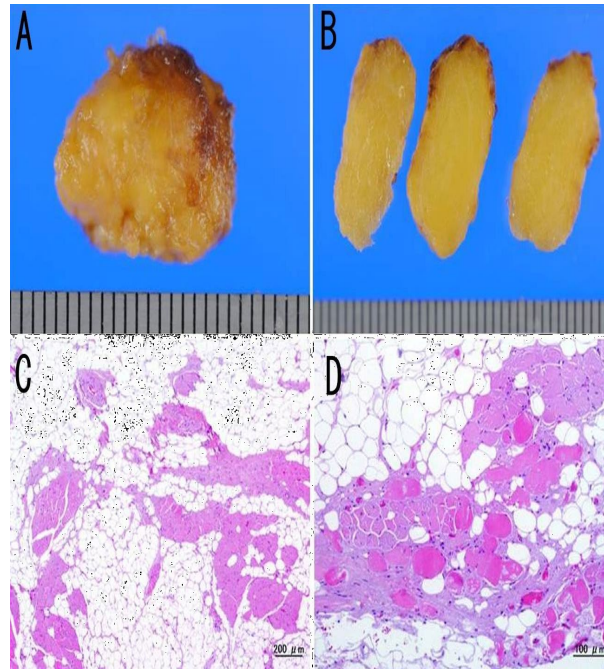


Figure 3: Imaging and pathologic findings of the enucleated part

The macroscopical finding of the enucleated part was pale yellow, smooth on the surface and soft-elastic (A, B). In the histopathological diagnosis, the adipose tissue was without surrounding membrane formation, was found lobular with the fibrous tissue and was with observed proliferation into the muscular wall (C). The tumor cells were mature adipose-like cells which possessed big and clear cytoplasm and the nuclear compressed to the cellular rim. The adipose-like cells were observed with proliferation into the muscular wall (D).

Discussion

Lipoma is a mesodermal, nonepithelial benign tumor which consists of mature adipose tissue, and has been reported to be one of the most frequent benign tumors^[1,2]. Lipoma grows relatively slowly to form painless swelling, which is palpable as a movable soft orbicular or lobular tumor mass with relatively clear borders.

Histopathological classifications include, according to the substrate and appearance of tumor cells, simple lipoma, fibrous lipoma, myxomatous lipoma, vascular lipoma, spindle cellular lipoma, polygonal lipoma, parosteal lipoma and muscular lipoma^[3-4]. With regard to histopathological incidence of lipoma in the mouth, Oku et al^[5] reported that simple lipoma is the most frequent with 62.2% (123/201 patients) followed by fibrous lipoma with 35.8% (72/201 patients), and these two types accounted for most of the cases. Muscular lipoma develops within the muscular tissue and grows due

to infiltrating mature adipose cells, which are tumor cells, deep into the muscle fibers. Histopathologically, it is characteristic that existing muscle fiber bundle remains within the tumor. Pélissier et al.^[6] reported about the incidence of intramuscular lipoma, that 4 out of 7 cases of intramuscular lipoma which developed in the mouth were on the tongue. In Japan, Kobayashi et al.^[7] reported 6 cases of intramuscular lipoma which developed on the tongue. In addition, Enjoji et al.^[3] reported that 27 cases of intramuscular lipoma (1.7%) were included in the 1615 cases of lipoma; thus, the incidence is considered “rare”. At this time we newly found 14 cases including self-experiments^[7-7] in the search and found by Pub Med, CiNii Article for case reports of intramuscular lipoma which developed on the tongue in the stomatognathic area (excluding reports at academic conferences) in Japan (Table 1). In addition, while some of the searched cases were

described as infiltrating or intramuscular lipoma, they were categorized into intramuscular lipoma according to the Shuman^[18] classification. The age was within 37 to 75 year-old and the mean age was 62 year-old. Since there are reports that lipoma in the oral area frequently occurs in the age range of 30 to 60 year-old and it is observed at the various age of 1 to 72 year-old^[19], the difference in age is not consistent among reports. However, lipoma mostly occurs in middle age to elderly people, which was not significantly different from the age for frequent occurrence of simple lipoma. With regard to the gender difference for lipoma, while Kobayashi et al.^[7] in Japan reported their determination with regard to glossal lipoma of the trend that it occurred more frequently in male (39 male subjects vs. 16 female subjects), no significant difference was observed for intramuscular lipoma which developed on the tongue (7 male subjects vs. 7 female subjects) in our determination. With regard to clinical symptoms, most

of them are painless tumor mass and swelling on the tongue, followed by less frequent articulatory disorder, lodging sensation of pharynx, and ear pain, which are observed to be normal clinical symptoms of lipoma and no specific clinical symptoms for intramuscular lipoma may exist. With regard to the onset site, onset in the glossal edge part was the most common with 10 cases. Those which developed in the inferior surface of the tongue were in 3 cases including self experiments; thus it is considered rare onset site. With regard to the size, while they were 50 to 7 mm in size and no specific size was observed, given that intramuscular lipoma is reported to be found mostly when they grow to a certain size compared to simple lipoma and intramuscular lipoma was reported to unlikely show manifestations due to its deeper location and slower growth than simple lipoma by Piattelli et al.^[20], no specific subjective symptoms were observed until the patient her/himself got aware in the self experiments as well.

Table 1: Article for case reports of intramuscular lipoma which developed on the tongue in the stomatognathic area in Japan

Case No	Authers	Age	Gender	Chief Complaint	Site	Magnitude
1	Nakayama et al, 1986	65	Male	Painless swelling	Bilateral tongue edge	Right:12 mm: Left:7mm
2	Saka et al, 1988	56	Female	Painless swelling	Left tongue lower surface	18×10×10 mm
3	Kanekawa, 1989	67	Male	Painless swelling	Left tongue edge forward, tongue lower surface	Right: 30×20×10 mm Left: 20×20×10 mm
4	Shirasuna et al,1989	56	Female	Painless swelling	left ventral side of the tongue	17×20×0.6 mm
5	Takeda, 1989	37	Male	Painless swelling	Right tongue edge	10×13×14 mm
6	Hiramatsu et al, 1996	70	Male	Swelling of the tongue	Bilateral tongue edge	red bean
7	Kobayashi et al, 2002	66	Female	Swelling of the tongue	Left tongue edge	27×23 mm
8	Nakano et al, 2002	65	Female	Swelling of the tongue Dysarthria	Left tongue edge	25mm
9	Tanabe et al, 2006	72	Female	Feeling of pharynx holding, Right earache	Right side tongue half side	52×21 mm
10	Naruse et al, 2012	58	Female	Painless swelling	anterior left region of the tougue border	35×30×20 mm
11	Kamatani et al, 2014	60	Female	Swelling of the lower tongue surface	Under tongue	25×18×10 mm
12	Kinukawa et al, 2014	63	Male	Discomfort of the tongue edge	Bilateral tongue edge	rice-grain ~red bean
13	Kohinata et al (present), 2018	62	Male	Swelling of the lower tongue surface	Left tongue lower surface	20×15 mm

With regard to the mechanism of development of lipoma, while there are hypotheses such as ectopic development theory^[21], and hyperplasia theory of normal tissue^[22], the causes such as inherited predisposition, congenital intrinsic factor, endocrine deconditioning, impaired metabolism of lipid, trauma, and chronic stimulation^[23] have been reported; however, the causality has not been currently clarified. In the self experiments, the medical history included diabetes mellitus and hypertension, the causes could not be confirmed.

Lipoma is categorized to superficial lipoma which is observed in the subcutaneous tissues and to deep lipoma which is observed in the subfascial, intramuscular and intermuscular tissues^[24]. While superficial lipoma is relatively easy to be diagnosed due to its likely clearer swelling compared to deep lipoma, deep lipoma may be difficult to be diagnosed depending on its onset site; thus, CT and MR tests are very useful imaging tests for the diagnosis of lipoma. It is reported specific for intramuscular lipoma to have a CT imaging finding of a high density linear figure which is assumed to be residue muscular

tissue in the tumor showing a uniform lipid concentration^[25,26]. In addition, it has been reported that some irregular parts in the low-density tumor mass with clear borders, which indicated equal density as the muscular layer, were observed in contrast CT imaging^[7]. While we did not perform a contrast CT test for this patient, a high density linear figure which is assumed to be residue muscular tissue in the tumor was shown in the simple CT imaging finding. With regard to the MR imaging findings, it has been reported that equal signal parts to the muscular tissue were mixed with unclear borders in the same high signal area as adipose tissue in the both T1 and T2-highlighted imaging, and that linear figures with equal signal to muscles were partially observed in the T2-highlighted imaging coronal section^[27]. In this case, MR findings showed the tumor mass with clear borders in both T1-highlighted imaging and T2-highlighted imaging, with internal area mostly shown as a high signal, and parts of equal signal to the muscular tissue mixed with unclear borders and linear sites with equal signal to the muscular tissue were observed inside. While it is considered that our case at this time showed imaging findings which are characteristic for intramuscular lipoma, it is essential for a final diagnosis to determine by a histopathological search.

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