

Symmetric lipomatosis of the Tongue: Report of a Case

Goro Kawasaki^{1,*}, Saki Hayashida¹, Izumi Yoshitomi², Syuichi Fujita³ and Masahiro Umeda¹

¹Department of Clinical Oral Oncology, Unit of Translational Medicine, Nagasaki University Graduate School of Biomedical Sciences

²Department of Oral Surgery, Isahaya General Hospital, Nagasaki, Japan

³Department of Oral Pathology and Bone Metabolism, Unit of Basic Medical Sciences, Graduate School of Biomedical Sciences

*Corresponding author: Goro Kawasaki, DDS, PhD, Department of Clinical Oral Oncology, Unit of Translational Medicine, Nagasaki University Graduate School of Biomedical Sciences, 1-7-1 Sakamoto, Nagasaki, 852-8588, Japan, Tel: +81-95-819-7698; Fax: +81-95-819-7700; E-mail: gkawa@nagasaki-u.ac.jp

Received date: 09 Feb 2017; Accepted date: 23 Feb 2017; Published date: 28 Feb 2017.

Citation: Kawasaki G, Hayashida S, Yoshitomi I, Fujita S, Umeda M (2017) Symmetric lipomatosis of the Tongue: Report of a Case. J Clin Case Stu 2(1): doi <http://dx.doi.org/10.16966/2471-4925.138>

Copyright: © 2017 Kawasaki G, et al. This is an open-access article distributed under the terms of the Creative Commons Attribution License, which permits unrestricted use, distribution, and reproduction in any medium, provided the original author and source are credited.

Abstract

Symmetric lipomatosis of the tongue is a rare condition. We present a rare case of intramuscular lipomatosis of the tongue in a 70-year-old male. A clinical diagnosis of symmetric lipomas of the tongue was made. The tumors of the bilateral tongue were resected under general anesthesia. The pathological diagnosis was symmetrical lipomatosis of the tongue. There were no signs of recurrence 3 years after the operation.

Keywords: Lipomatosis; Tongue; Symmetric

Introduction

Lipomatosis is a condition characterized by benign tissue tumors on many parts of the body. It commonly affects the upper back and shoulder, but also occurs in the oral cavity at lower rates [1]. Multiple tongue lipomas, particularly of a symmetric tumor formation, are called symmetric lipomatosis of the tongue [1-3]. Lipomatosis of the tongue is extremely rare [4] (Figure 1).

Here, a case of symmetric lipomatosis of the tongue with intramuscular invasion is presented, and the current literature is reviewed.

Case Presentation

In November 2011, a 70-year-old man was referred to Nagasaki University Hospital complaining of bilateral swelling of the tongue. He had been aware of the swelling for many years, and he finally consulted his dentist.

On clinical examination, no tumor masses could be identified on the trunk, head, neck, or extremities. The tongue was diffusely enlarged and many small tumor-like lesions of yellowish color were observed bilaterally on the tongue. There was no dysphagia, ankyloglossia, or dyspnea. The patient had a history of hypertension, cerebral palsy after head injury, and moderate alcohol consumption for more than 50 years.

Magnetic resonance imaging (MRI) demonstrated multilocular lesions having high signal intensity on T1W1 bilaterally on the tongue, and the lesion was diminished by fat suppression (Figure 2).

Clinical diagnosis was symmetric lipomatosis of the tongue.

The tumors were resected under general anesthesia. Adipose tissue proliferation was unencapsulated (Figure 3).

Histopathological findings revealed adipose tissue consisting of mature adipocytes infiltrating between muscle fibers, confirming the diagnosis of symmetrical lipomatosis of the tongue (Figures 4 and 5).

Discussion

Lipoma of the tongue is rare and often presents as a single, superficial, pedunculated, and sessile lesion. Presentation as multiple tumors, infiltrating tumors, or macroglossia is infrequent [5]. Diagnosis of lipoma of the tongue is essentially clinical, but radiological investigations, especially magnetic resonance imaging (MRI), may be helpful in diagnosis [5,6].

Lipomas are classified into 5 categories: (1) lipoma, (2) variants of lipoma, (3) heterotopic lipoma, (4) infiltrating/diffuse, neoplastic/non-neoplastic proliferation of mature fat, and (5) hibernoma [7,8]. The fourth group includes benign symmetric lipomatosis, diffuse lipomatosis, and pelvic lipomatosis. Lipomatosis is characterized by multiple sites of involvement, invasiveness, and absence of encapsulation of the adipose tissue [4].

Our case had these characteristics, and thus we made a diagnosis of lipomatosis.



Figure 1: An intraoral view shows many yellowish tumors of the bilateral tongue.

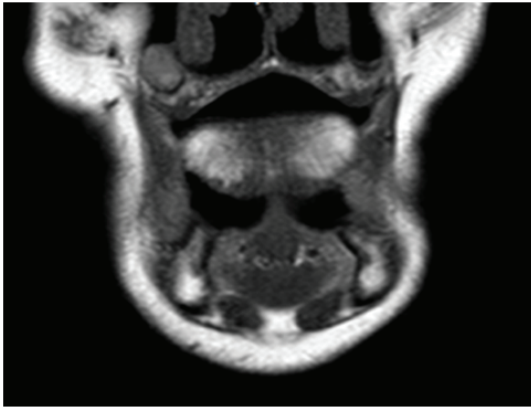


Figure 2: T1W2 MRI shows diffuse high intensity area on the tongue.

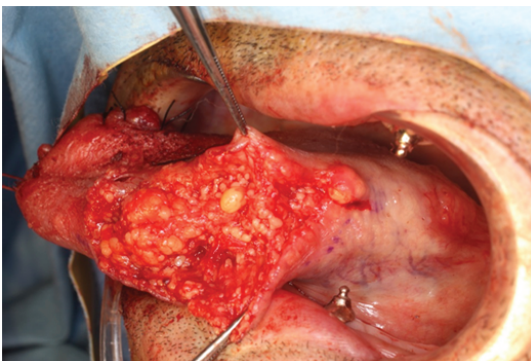


Figure 3: Yellowish adipose tissues were seen just beneath the epithelium on the lateral border of the tongue. The adipose tissues infiltrated the lingual muscles.

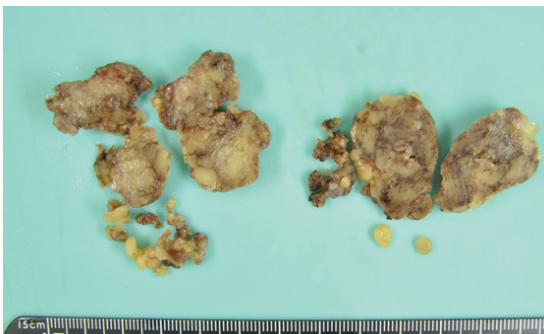


Figure 4: The excised tumors showed no capsule formation.

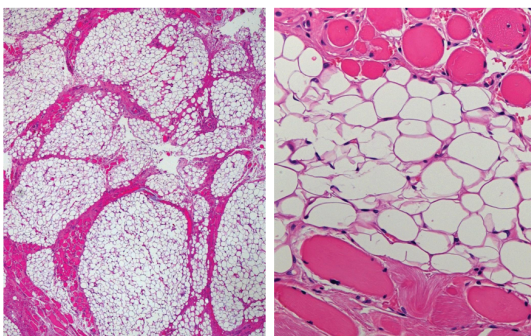


Figure 5: Pathological findings: Adipose tissues were interspersed with the muscle fibers (H-E staining, × 100).

Symmetric lipomatosis of the tongue is reported to be an extremely rare disease [9]. Calvo-Garcia et al. [4] described only 6 cases of symmetric lipomatosis, including one case reported in the English literature. On the other hand, tongue symmetric lipomatosis has been reported more in Asia [10]. We reviewed 28 cases of symmetric lipomatosis of the tongue in the Japanese literature [11-29]. All of the patients were male, and 17 of 28 patients suffered from alcoholic hepatitis and/or alcoholism.

Acetyl CoA, an organic compound, is a metabolite of alcohol in the liver, and it produces neutral lipids [30]. It is generally known that an increase of acetyl CoA is associated with fatty deposition [30]. In our case series, there were many cases of alcoholic hepatitis and alcoholism; hence, we suggest that symmetric lipomatosis of the tongue is associated with alcohol metabolic deposition.

With regard to treatment, the gold standard is complete surgical resection [1]. In our case series, there was surgical resection of the tumor in 18 of 28 cases. The reasons for non-surgical treatment in our case series were poor general condition, unwillingness of the patients to undergo surgical treatment, and lack of subjective symptoms. If a patient has a functional disorder, such as dysarthria and/or dysphagia, surgical treatment is recommended. In the present case, the patient desired surgery because of difficulty in food intake due to tongue enlargement. Since surgery, there have been no signs of recurrence.

Conflict of Interest

There are no conflicts of interest to report.

References

1. Imai T, Michizawa M, Shimizu H, Imai T, Yamamoto N, et al. (2008) Bilateral multiple spindle cell lipomas of tongue. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 106: 264-269.
2. Ogawa A, Nakamura H, Takahashi H (1988) Benign symmetrical lipomatosis of the tongue: report of a case. *J Oral Maxillofac Surg* 46: 502-504.
3. Katou F, Shirai N, Motegi K, Satoh R, Satoh S (1993) Symmetrical lipomatosis of the tongue presenting as macroglossia. Report of two caases. *J Craniomaxillofac Surg* 21: 298-301.
4. Calvo-Garcia N, Prieto-Prado M, Alonoso-Orcajo N, Junquera-Gutierrez LM (1999) Symmetric lipomatosis of the tongue. Report of a case and review of the literature. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 87: 610-612.
5. Thomas S, Varghese BT, Sebastian P, Koshy CM, Mathew A, et al. (2002) Intramuscular lipomatosis of tongue. *Postgrad Med J* 78: 295-297.
6. Norris DA, Doku C (1993) Orofacial lipomas diagnosed by CT and MRI. *J Am Dent Assoc* 124: 110-115.
7. Enzinger FM, Weiss SW (1995) Lipomas. In: Gay SM, Gery L (eds) *Soft Tissue Tumors*. 3rd edition, St. Louis: Mosby 381-431.
8. Vasileiadis I, Mastorakis G, Ieromonachou P, Logothesis I (2013) Symmetrical lipomatosis of the tongue- a rare cause of macroglossia: diagnosis, surgical treatment, and literature review. *Laryngoscope* 123: 422-425.
9. Jinbu Y, Otawa K, Obi Y, Tsukahara T, Kusama M, et al. (2004) Symmetric lipomatosis of the tongue: report of a case. *Oral Med Pathol* 9: 123-126.
10. Tohya T, Kawahara K, Hotta F, Shirozu T, Sano D (2001) Bilateral lipomas of the tongue: report of a case. *Aichi Gakuin J Dent Sci* 39: 519-523.
11. Aoki N, Endo M, Endo Y, Nagamori T, Fujita K (1985) A case of lipoma in bilateral tongue margin. *Jpn J Oral Maxillofac Surg* 31: 858-862.

12. Shinbo T, Kikuchi S, Nakajima Y, Shinoki K, Fujita Y (1985) Multiple lipomas of the tongue: a case report. *Jpn J Oral Maxillofac Surg* 31: 1421-1424.
13. Nakayama Y, Nishijima K, Nabeyama H, Kayano T, Ikegami N, et al. (1986) A case of bilateral infiltrating lipoma of the tongue. *Jpn J Oral Maxillofac Surg* 32: 1068-1073.
14. Ogawa A, Nakamura H, Sano K, Inokuchi T, Takahashi H (1986) Benign symmetric lipomatosis of tongue: Report of a case. *Jpn J Oral Maxillofac Surg* 32: 1030-1033.
15. Yoshimura Y, Miyakawa A, Odani M, Odajima T, Yamamoto E, et al. (1988) A case of macroglossia caused by benign symmetrical lipomatosis. *Jpn J Oral Maxillofac Surg* 34: 1061-1065.
16. Kanekawa A (1989) A case of benign symmetric lipomatosis of the tongue. *Jpn J Oral Maxillofac Surg* 35: 1535-1537.
17. Tateishi A, Mitsuse K, Nodai T, Futamura H, Kikuta T, et al. (1993) Clinicopathologic observations of oral lipomas 20 cases in our department over 15 years. *Jpn J Oral Maxillofac Surg* 39: 276-280.
18. Sugisawa Y, Matsuyama T, Hayashi N (1997) A case of lipomatosis of the tongue. *Jpn J Clin Dermatol* 51: 662-664.
19. Honda Y, Ozeki S, Oobu K, Goto K, Yoshimura K, et al. (1998) Two cases of benign symmetric lipomatosis of the tongue. *Jpn J Oral Maxillofac Surg* 44: 342-344.
20. Inoda H, Yamamoto G, Yoshitake K (2000) A case of benign symmetric lipomatosis of tongue. *Jpn J Oral Diag/Oral Med* 2000; 13: 206-209.
21. Kawai T, Yamashita C, Yosue T (2000) A case of multiple lipomas of the tongue. *Jpn J Oral Diag/Oral Med* 13: 531-534.
22. Kurokawa H, Kimura K, Miura K, Takeda S (2000) Lipomas of the tongue presenting as a macroglossia: report of a case. *Jpn J Oral Diag/Oral Med* 13: 451-454.
23. Irisawa Y, Horiuchi C, Taguchi T, Kawai S, Satou M (2003) Lipomatosis of the bilateral sides of the tongue: a case report. *Oto-Rhino-Laryngol* 46: 363-367.
24. Sato Y, Takahashi E, Takahashi Y, Sakamoto I, Omura K, et al. (2004) A case of symmetric lipomatosis of the tongue. *Jpn J Oral Maxillofac Surg* 50: 297-300.
25. Kurabayashi H, Yamazaki H, Otsuru M, Sasaki M, Kaneko A, et al. (2005) A case of nodular type lipomatosis arising from both borders of the tongue. *Jpn J Oral Maxillofac Surg* 51: 348-351.
26. Ishikawa Y, Katada Y, Enomoto M, Kawasa K, Okamoto Y, et al. (2006) A case of symmetrical lipomatosis of the tongue presenting as macroglossia. *Oral Sci Int* 3: 90-93.
27. Miyake S, Takeda Y, Fujimoto Y, Ito K, Tobe M, et al. (2008) Bilateral lipomatosis of the tongue: a case report. *J Jpn Oral Muco Membr* 14: 9-13.
28. Igarashi Y, Iida A, Kobayashi T, Saku T, Hayashi T, et al. (2011) Symmetric lipomatosis of the tongue arising in a patient with alcoholic liver cirrhosis. *Jpn J Oral Maxillofac Surg* 57: 119-123.
29. Kato R, Jinbu Y, Ioka Y, Miyagi T, Kusama M (2011) Symmetric lipomatosis of the tongue: report of a case. *J Jpn Oral Muco Membr* 17: 17-21.
30. Lieber CS (2004) Alcoholic fatty liver: its pathogenesis and mechanism of progression to inflammation and fibrosis. *Alchol* 34: 9-19.