Symmetric Lipomatosis of the Tongue: Report of a Case

Yoshinori Jinbu¹, Kaoru Otawa¹, Yuri Obi¹, Taku Tsukahara¹, Mikio Kusama¹ and Hidemi Nakagawa²

¹Department of Dentistry, Oral and Maxillofacial Surgery, ²Department of Dermatology, Jichi Medical School, Tochigi, Japan

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A case of symmetric lipomatosis of the tongue is reported. The patient was a 72-year-old Japanese male who complained of a bilateral swelling of the tongue. The tongue of the patient was diffusely and markedly enlarged with somewhat nodular appearance in the lateral borders, presenting as a macroglossia.

Histopathologically, the biopsy specimen showed adipose tissues which consisted of mature lipocytes. Partial glossectomy was performed. Adipose tissues were interspersed with the muscles fibers and distinct capsule formation was not found. The patient was finally diagnosed as having symmetric lipomatosis of the tongue based on the clinical, radiological and histopathological findings.

Key words: symmetric lipomatosis of the tongue, benign symmetric lipomatosis, macroglossia

Correspondence: Yoshinori Jinbu, Department of Dentistry, Oral and Maxillofacial Surgery, Jichi Medical School, Yakushiji 3311-1, Minamikawachi-machi, Kawachi-gun, Tochigi, 329-0498, Japan
Phone: +81-286-58-7390, Fax: +81-285-44-8669

Introduction

Lipoma is one of the non-epithelial benign tumors occasionally seen in the oral cavity. However, symmetric lipomatosis, a subtype of lipomas, is an extremely rare disease in the oral cavity, and only 6 cases of symmetric lipomatosis of the tongue (SLT) have been reported in the literature (1-5).

This case report describes a patient who presented with a bilateral lingual swelling and who was finally diagnosed as having SLT.

Case Report

A 72-year-old Japanese male was referred to the Department of Dentistry, Oral and Maxillofacial Surgery, Jichi Medical School Hospital by his physician on December 27, 2002, with a complaint of tongue swelling. The patient first noticed the tongue swelling 2 years before. He was unaware of pain or difficulty in swallowing, but the swelling had gradually increased.

The patient was a mild diabetic, and his diabetes mellitus was well controlled by diet. He had no other remarkable medical history. Physical status was as follows: height 165cm; body weight 55.15kg; body temperature 36.6°C; pulse 66/min; blood pressure 132/74; and respiration 16/min. The tongue of the patient was diffusely and markedly enlarged with a somewhat nodular appearance

in the lateral borders, presenting as a macroglossia (Fig. 1). The surface of mucosa showed normal color with no erosions or ulcers. The lateral border of the tongue showed elastic soft to palpation and the lesion was unaccompanied by pain. There were no abnormal values in the general blood examination.

A biopsy was performed with a clinical diagnosis of amyloidosis of the tongue. After incision in the lateral border of the tongue, yellowish adipose tissues were seen just beneath the epithelium (Fig. 2). Histopathologically, the specimen showed adipose tissue consisting of mature lipocytes, and the histopathological diagnosis was lipoma (Fig. 3).

MR image showed diffuse and irregular high intensity areas in the lateral border of the tongue in the T1 weighted image (Fig. 4).

Although there was no interference with functions of the tongue and mouth, partial glossectomy was carried out under general anesthesia to reduce the volume of the tongue. Incision was made in the bilateral border and a proper amount of adipose tissue was removed. Adipose tissues were interspersed with the muscles fibers, and distinct capsule formation was not found (Figs. 5, 6). The wound healing process was normal.

The patient was finally diagnosed as having symmetric lipomatosis of the tongue based on the clinical,

Fig. 1: The tongue was diffusely and markedly enlarged.

Fig. 2: The yellowish adipose tissues were seen just beneath the epithelium in the lateral border of the tongue.

Fig. 3: The biopsy specimen showed adipose tissue consisting of mature lipocytes (H-E staining, ×100).

Fig. 4: MR image showed diffuse and irregular high intensity areas in the tongue (T1 weighted image).

radiological and histopathological findings. After the operation, re-examination of the whole body was carried out by dermatologist and physician. No fatty deposits were noted in other parts of the body, but a small hemangioma and cyst were detected in the liver.

Discussion

Benign symmetric lipomatosis (BSL) was first reported by Brodie in 1846 (6). Since then, approximately 200 cases of BSL have been reported in the literature (7). Some were reported as Madelung's disease (8, 9). BSL is characterized by diffuse symmetric deposits of adipose tissues in the neck. The neck masses achieve great size and become cosmetically deforming over several years, and the upper aerodigestive tract may be compressed in advanced disease, causing dyspnea and dysphagia. The fatty deposits are uncapsulated with multiple projections infiltrating and displacing the surrounding structures without destruction. Association with alcoholism has been reported in 60% to 90% of the patients (10). Hyperruricemia, gout, liver disease, polyneuropathy, diabetes, glucose intolerance, peripheral insulin resistance, renal tubular acidosis, hypertension, hyperthyroidism, and

hyperlipidemia have been identified in patients with BSL (10).

On the other hand, symmetric lipomatosis of the tongue (SLT) is an extremely rare disease. Recently, Calvo-Garcia *et al.* (5) reported a case of SLT and reviewed the literature (Table 1). According to Calvo-Garcia *et al.*, in 6 cases of SLT, all patients were male and the mean age was 67 years (61-71 years). Four of the patients were Japanese and none of the 6 cases had the typical features of BSL: deposition of fat in the neck, shoulder and upper trunk. Only one Japanese patient was an alcoholic (3). The present case showed clinical features similar to those patients previously reported.

Histopathological findings of BSL and SLT were essentially the same: multiple site of involvement, invasiveness and absence of encapsulation of the adipose tissues. None of the patients with SLT showed the typical features of BSL, and lingual lipomatosis has not been reported in patients with BSL (5). Therefore, there has been some confusion on the etiology. Ogawa *et al.* (2) suggested that BSL and SLT are the same entity, but Katou *et al.* (3) and Calvo-Garcia *et al.* (5) suggested that they are different. Regarding the etiology of BSL, Pollock *et al.*

Fig. 5: The adipose tissues infiltrated into the lingual muscles, and capsule formation was not found.

Fig. 6: The adipose tissues were interspersed with the muscle fibers (H-E staining, ×100).

Table 1: Summary of reported cases of SLT (modified from Calvo-Garcia *et al.* 1999)

Author	Year	Gender	Age	Alcoholism	Dysarthria	Dysphagia	Systemic disease	Treatment
Desmond	1944	Male	67	—	+	+	· Diabetes · Mental disorder	Glossectomy
Ogawa <i>et al.</i>	1988	Male	67	+	Not clear	Not clear	· Stroke	Glossectomy
Yoshimura <i>et al.</i>	1988	Male	71	—	+	+	· Angina · Raeder syndrome	Glossectomy
Katou <i>et al.</i> (Case 1)	1993	Male	61	—	+	+	· Gastric cancer	Glossectomy
Katou <i>et al.</i> (Case 2)	1993	Male	71	Mild drinking	—	—	· Hyper-tension	Glossectomy
Calvo-Garcia <i>et al.</i>	1999	Male	67	—	±	—	—	Glossectomy
Jinbu <i>et al.</i>	2003	Male	72	—	—	—	· Diabetes	Glossectomy

(11) suggested an abnormality in the amount and/or function of the Gs-protein, or in the catalytic unit of adenylate cyclase found in adipocyte membranes. Recently, Berkovic *et al.* (12) concluded that BSL is a manifestation of brown fat mitochondrial dysfunction. Although further analysis of more cases of SLT and BSL are needed, SLT may not be a neoplastic lesion but a fatty metabolic dysfunction. On the treatment of SLT, all cases have been managed by glossectomy (Table 1). As the adipose tissues

infiltrate deep into the lingual muscles, complete resection is impossible. As the possibility of regrowth of the adipose tissues is undeniable and malignant transformation of SLT to myxoid liposarcoma occurred in one case, (13), long term follow-up is important.

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