Journal of Oral and Maxillofacial Surgery, Medicine, and Pathology 31 (2019) 180–184

Contents lists available at ScienceDirect

Journal of Oral and Maxillofacial Surgery, Medicine, and Pathology

journal homepage: www.elsevier.com/locate/jomsmp

Case Report

A case of a large thrombosed lingual varix☆

Midori Eguchi[a], Hisao Shigematsu[a,*], Yuka Oku[a], Kentaro Kikuchi[b], Munehisa Okada[a,c], Hideaki Sakashita[a]

[a] Second Division of Oral and Maxillofacial Surgery, Department of Diagnostic & Therapeutic Sciences, Meikai University School of Dentistry, Japan
[b] Division of Pathology, Department of Diagnostic & Therapeutic Sciences, Meikai University School of Dentistry, Japan
[c] Department of Oral Surgery, Haga Red Cross Hospital, Japan

ARTICLE INFO

Keywords:
Thrombosed lingual varix
Venous thrombosis
Lingual varix
Tongue

ABSTRACT

Lingual varix is a condition characterized by purplish venous ectasia. It is usually found on the ventral surface of the tongue in elderly patients. On the other hand, thrombosed oral varices are small, localized, and probably not uncommon lesions. However, large thrombosed oral varices are very rare, and there have not been any reports about thrombosis in lingual varices. This report describes a rare case of a large thrombosed lingual varix involving the sublingual vein. A 75-year-old female presented with a mass on the ventral surface of her tongue. A lingual tumor was initially suspected based on echography and magnetic resonance imaging, and an excisional biopsy was performed under general anesthesia. A definitive histopathological diagnosis of venous thrombosis was made. We would like to emphasize that venous thrombosis should always be considered as a differential diagnosis in cases in which a dark blue or purple, painless tumor arises on the ventral surface of the tongue.

1. Introduction

Intravenous thrombus formation often occurs in patients with risk factors for the condition, and deep venous thrombosis can cause serious problems, such as venous thromboembolisms [1]. On the other hand, congenital venous deformities (so-called hemangioma) frequently arise in the tongue [2], and there have been many reports about cases of hemangioma involving venous stones [3]. Thrombus formation is also seen during the development of venous stones in hemangioma, and it is clinically important to distinguish between venous thrombosis occurring in a single blood vessel and hemangioma.

We consider that opportunities to encounter venous thrombosis in daily clinical practice are not uncommon [4,5]. There are some reports about venous thrombosis in the oral cavity. According to the literatures [4,6–8], thrombosed oral varices are small, localized lesions. Large thrombosed oral varices are very rare. In particular, as far as we know, there have been few reports about thrombus formation in a lingual varix involving the sublingual vein [9]. We experienced a case in which a large venous thrombosis developed on the ventral surface of the tongue. Here, we give a brief overview of this case and discuss associated considerations.

2. Case report

A 75-year-old female was referred to our hospital with a chief complaint of a tumor on the ventral surface of her tongue in December 2013. She had a history of hypertension, which was diagnosed at age 55, and hyperlipidemia, which was diagnosed at age 65. Therefore, she was taking an antihypertensive drug (5 mg/day amlodipine) for hypertension and pravastatin sodium (10 mg/day) for dyslipidemia. Although in 2011 it was determined that she had previously had a minor stroke, no specific treatment was administered. In addition, she had a penicillin allergy. It should be noted that the patient had never smoked or suffered a tongue trauma. Regarding her family history, her father had suffered from stomach cancer, and her mother had suffered a subarachnoid hemorrhage.

In July 2013, she noticed a thumb-sized tumor on the right side of her tongue. As it was painless, she had left it untreated, and it had temporarily shrunk. In August, she visited her dental clinic because her symptoms had not improved. The dentist diagnosed it as an ulcer caused by a denture or a collapsed mucous cyst. In December, she was referred to our department by her dentist because the tumor had increased in size again.

☆ AsianAOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

* Corresponding author at: Keyakidai 1-1, Sakado city, Saitama, 350-0283, Japan.
E-mail address: shigematsu@dent.meikai.ac.jp (H. Shigematsu).

https://doi.org/10.1016/j.ajoms.2018.11.003
Received 5 September 2018; Received in revised form 1 November 2018; Accepted 16 November 2018
Available online 22 November 2018

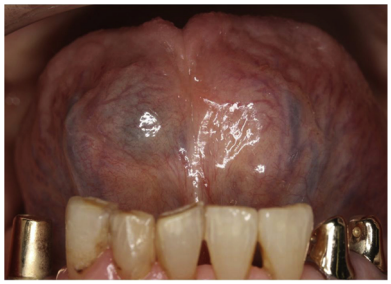

Fig. 1. Clinical findings at the first visit.
An elastic, firm, painless tumor was found on the right ventral surface of the tongue.

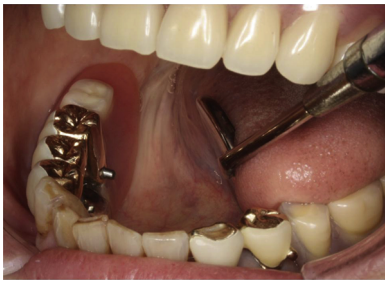

Fig. 2. Observations obtained while the patient was wearing a local floor denture.
The hook on the lingual side of the denture is shown.

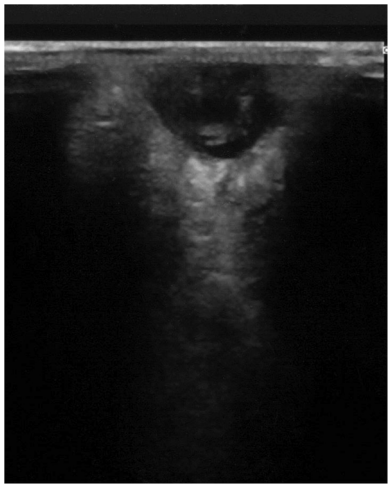

Fig. 3. Echographic findings of the tongue lesion.
The tumor exhibited internal heterogeneity, relatively clear boundaries, and echogenicity.

A physical examination revealed a height of 147.8 cm, a weight of 37.8 kg, and a body mass index of 17.49. She exhibited good blood pressure control. Her cholesterol levels were as follows: high-density lipoprotein cholesterol: 69 mg/dl, low-density lipoprotein cholesterol: 121 mg/dl. No other abnormalities were observed in blood tests or electrocardiographic examinations.

A clinical examination revealed a relatively well-circumscribed tumor, which measured approximately 2 cm in diameter, on the right side of the ventral surface of the tongue (Fig. 1). The lesion had a smooth surface and was dark purple. Palpation indicated that the lesion was round, elastic, firm, painless, and freely movable. The lesion was not compressible and did not fade when pressure was applied to it. To repair the mandibular molars on both sides, a Conus Krone local floor denture had been inserted, and the hook used to detach the denture was in contact with the lesion (Fig. 2).

Ultrasonography of the tongue showed a well-circumscribed solid lesion, measuring 2 × 1.5 × 1 cm, which exhibited internal heterogeneity and echogenicity (Fig. 3). Axial T1-weighted magnetic resonance imaging (MRI) showed a lesion that displayed weak signal intensity and little inherent contrast relative to the adjacent normal tissues. On T2-weighted MRI, the lesion produced strong non-homogenous signals and displayed well-circumscribed margins (Fig. 4a and b). A clinical examination did not detect any findings that were indicative of metastatic lymph nodes in the neck.

According to these findings, a clinical diagnosis of a tumor on the ventral surface of the tongue was made, and an excisional biopsy was performed under general anesthesia in February 2014. An epithelial incision was made around the tumor. The tumor was shelled out completely together with the overlying mucosa (Fig. 5). During the operation, the tumor was easily enucleated, and there was no abnormal bleeding. No blood vessels that required ligation were observed in the surrounding area. No drains were inserted into the wound site, and primary closure was achieved. The patient's postoperative course was uneventful. There was no evidence of recurrence after 4 postoperative years.

The gross surgical specimen contained a soft mass, measuring 20 × 10 × 11 mm in size (Fig. 6). The cross-section of the excised sample showed a cystic mass containing coagulated blood within the lumen of an enlarged blood vessel, and the central part of the lesion was slightly whitish in color.

Histopathologically, a markedly expanded vascular lumen was seen in the submucosal muscle layer. The lumen was full of erythrocytes and also contained a blood clot (Fig. 7a and b). The central part of the lesion was rich in fibrin. In other part of the lesion, granulation tissue and organizing thrombus were noted. The lesion only contained one expanded blood vessel. The histopathological diagnosis was venous thrombosis.

3. Discussion

Thrombosis is defined as a condition in which a thrombus forms in the heart/vascular lumen. A thrombus is a blood clot, containing platelets, fibrin, and cell components, that adheres to the vascular endothelium [4]. Venous thrombosis mainly occurs in the veins of the lower limbs (in ≥90% of cases), which is known as deep venous thrombosis [1]. On the other hand, in the mouth thrombosis is known to develop in the lips and buccal mucosa, but there have only been 6 reports about thrombosis in the tongue [4,8–12] (Table 1).

3.1. The causes of thrombus formation

Generally, there are three main causes of thrombus formation [1,5]:

(1) Abnormalities of the blood vessel wall (a failure of endothelial cells)
(2) Abnormalities of blood flow (venous blood flow stagnation)
(3) Abnormalities of blood composition (blood hypercoagulability)

Aging and long-term sitting are background factors that probably contribute to venous thrombosis. As for disease states, trauma, malignant tumors, varicose veins of the lower limbs, and historical venous thrombosis have been mentioned as risk factors for the condition [1,4]. Smoking is also a risk factor for thrombus formation as it damages blood vessel walls [13]. In addition, attention should be paid to antiphospholipid antibody syndrome, which is characterized by recurrent thrombosis [14]. It is an autoimmune disease caused by antiphospholipid antibodies, such as anti-cardiolipin antibodies, and it is considered to be one of the most important causes of thrombosis.

It is also known that many vascular malformations arise in the tongue. Misdirection of the tongue is one of the causes of vascular lesions that develop at the sides of the tongue, which usually occur after middle age [15]. In addition, lingual varicose veins (lingual varices) are

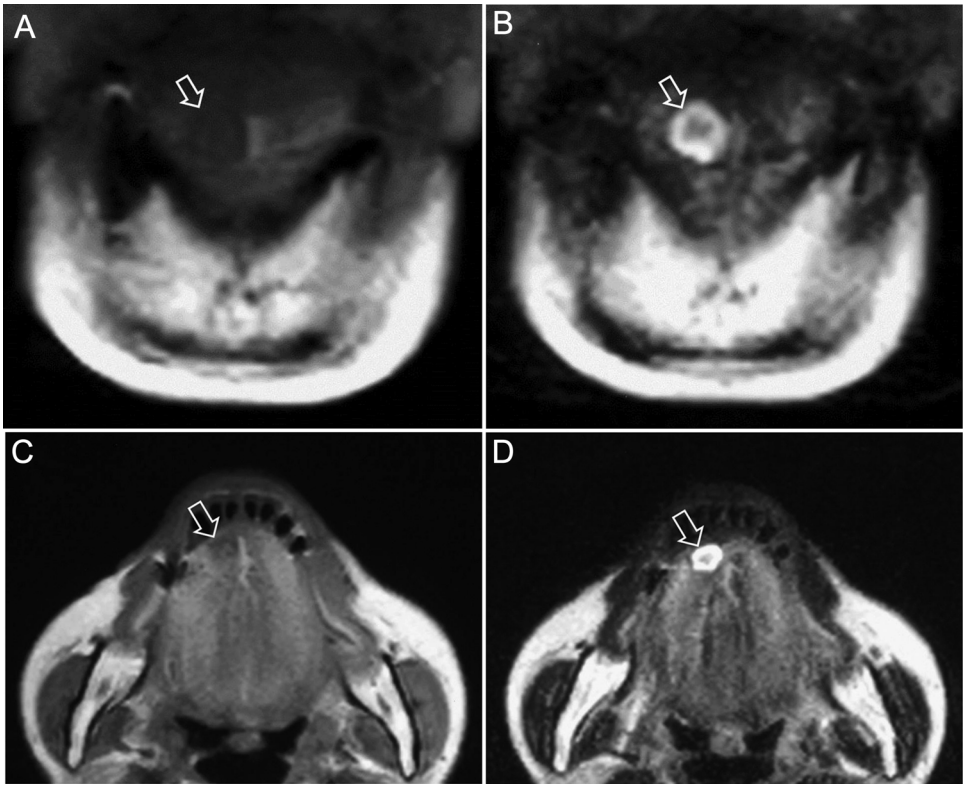

Fig. 4. Frontal cross-sectional and horizontal-section of MRI findings.
The lesion displayed low signal intensity on T1-weighted MRI (A, C) and high signal intensity on T2-weighted MRI (B, D).

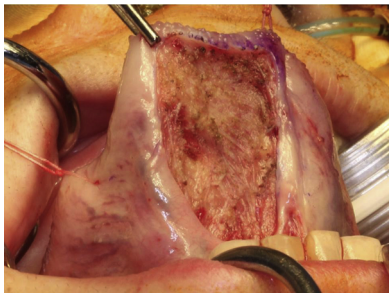

Fig. 5. Surgical findings.
There was no abnormal intraoperative bleeding, and the lesion was excised en bloc together with the overlying mucosa.

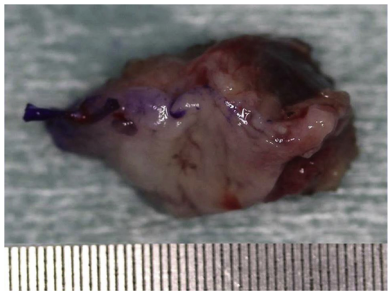

Fig. 6. Findings of the gross surgical specimen.
The excised specimen had a smooth surface and measured 20 × 10 × 11 mm.

relatively common in the tongue [16–18]. The mechanism responsible for the development of lingual varices is different from that responsible for the development of varicose veins in the lower limbs. Therefore, varicose veins are not directly related to oral venous thrombosis [4]. However, damage to the sublingual vein might be one of the local causes of blood flow stagnation in cases of lingual thrombus formation. If other local or systemic factors (vascular endothelial disorders, coagulation system elevation, etc.) are present, it is considered that thrombi can develop in the sublingual vein.

As for the systemic background factors for venous thrombus formation present in our case, the patient was elderly (age: 75 years), and she had been suffering from hypertension and hyperlipidemia for many years. Therefore, blood vessel wall abnormalities might have occurred.

In general, unconscious tongue trauma due to chewing and/or bad tongue habits is a local factor that can contribute to lingual venous thrombus formation. In our case, the patient was wearing a Conus Krone denture with a detachable hook. Since an ulcer was found at the site that came into contact with the hook before the patient visited our department, the hook was considered to have contributed to the onset of the lesion. There is also a possibility that the engorgement of the sublingual vein was caused by a blood flow disturbance or congestion. Thus, in the current case it was suggested that thrombus formation might have been induced by several overlapping factors.

Venous thrombosis in the oral cavity is known to be prevalent in the lips and buccal mucosa, and such lesions often measure < 10 mm in diameter [4,6,7,18]. To our knowledge, there is no report showing larger lesion than 10 mm except for lesions in the tongue (Table 1). However, in our case the thrombosis measured 20 × 15 × 10 mm; i.e., it was larger than most oral venous thromboses. The anatomical position of the ventral surface of the tongue might have contributed to this. The peripheral vein has a plexus, and an anastomosis between it and the deep artery of the tongue has been reported previously [15]. Thus, such anatomical features might have contributed to the enlargement of the lesion. Since lesions on the ventral surface of the tongue are not as conspicuous as those on the lips or buccal mucosa, there is also a possibility that the lesion became large simply because it was left untouched and untreated.

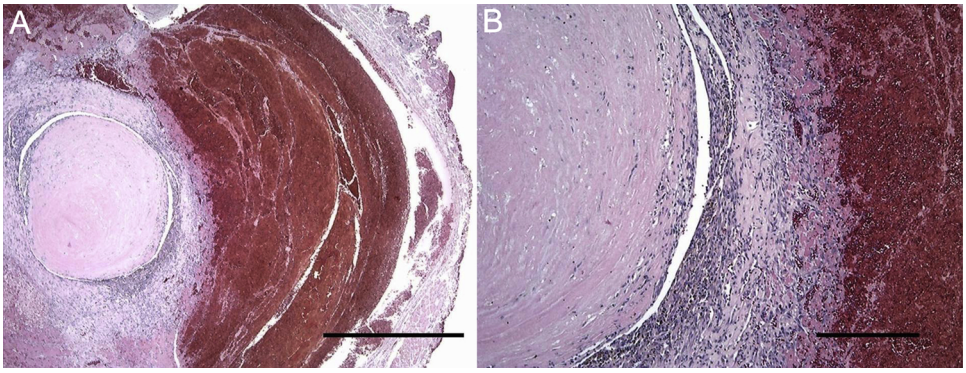

Fig. 7. Histopathological findings.
The interior of the lesion was filled with erythrocytes, and organization was seen in the center of the lesion. (A: low magnification, bar: 2 mm; B: high magnification, bar: 200 μm).

Table 1
Lingual thrombosis.

	Authors		Age	Sex	Site	Clinical description	Initial diagnosis	Remarks
1	Lasho JW et al. [9]	1964	63	F	left ventral surface	2 cm in diameter, purphish blue, firm nodule and slightly tender	no description	hypertension
2	Weathers DR, et al. [4]	1971	65	F	lateral tongue	blue, asymptomatic	no description	
3	Kurita H, et al. [10]	1994	69	F	left side of tongue	painless, domed-shaped swelling	no description	
4	Lekovic JP, et al. [11]	2013	28	F	right lateral tongue	**purple** discoloration and very painful swelling	lingual thrombosis	antiphospholipid syndrome, pregnant
5	Arndal E, et al. [12]	2014	57	M	left side of tongue	painfull swelling and discolored tongue with a necrotic lesion	tongue carcinom	alcohol abuse and heavy smoking
6	Tjioe KC, et al. [8]	2015	61	M	right dorsum of the tongue	1.5 cm in diameter, firm purple, slight painful nodule	vascular malformation	hypertension, atherosclerosis, habit of chewing his tongue
7	present case	2018	75	F	right ventral surface of the tongue	2 cm in diameter, dark purple, elastic firm, painless nodule	tumor of the tongue	hypertension, hyperlipidemia

3.2. Differential diagnosis and treatment policy

Venous thromboses in the oral cavity are usually dark bluepurple, elastic, hard, and painless nodules, and they exhibit mobility without adhering to the mucosa. They are also characterized by an older age at onset compared with congenital vascular malformations and hemangiomas [4,5,15]. The clinical differential diagnoses for lesions in the oral cavity include congenital vascular malformations and hemangiomas, mucous cysts, and tumors. In particular, it is important to differentiate venous thromboses in the oral cavity from minor salivary gland tumors, including salivary gland malignancies, such as mucoepidermoid carcinoma [4]. The sizes and hardness of venous thromboses vary, and their imaging findings differ depending on the degree of thrombus organization, so the differential diagnosis of such lesions is not always easy [4,5,16]. A case of thrombosis of the lingual artery that had to be differentiated from oral cancer due to necrosis has been reported [12]; therefore, it is necessary to take sufficient care when diagnosing venous or arterial lingual thrombosis.

In our case, there was a relatively large bluish lesion on the ventral surface of the tongue, which was found to be elastic and firm by palpation and had not adhered to the mucosa. On the other hand, the lesion was not compressible and did not fade when pressure was applied to it, which are seen in hemangioma. On diagnostic imaging, internal heterogeneity and a solid lesion were suspected. Accordingly, a malignant salivary gland tumor could not be completely ruled out. Therefore, a clinical diagnosis of a sublingual tumor was made, and an excisional biopsy was performed.

The clinical findings of venous thrombosis in the oral cavity are similar to those of hemangioma and congenital vascular malformations [2]. However, venous thrombosis is based on thrombus formation, and its etiology differs from those of the above-mentioned conditions [15]. In addition, while hemangioma and congenital vascular malformations occur more often in relatively young people, venous thrombosis in the oral cavity is known to occur in older patients. Therefore, pathological examinations are indispensable in cases involving oral lesions, especially when venous thrombosis is suspected in middle-aged or elderly patients.

Generally, oral venous thrombosis does not cause large lesions, and it can be definitively diagnosed based on a pathological examination after excision or resection [4,6–9]. In our case, we performed an excisional biopsy, and the resultant defect healed without causing postoperative dysfunction or esthetic issues. Treatment with a YAG or CO_2 laser is also worth considering in such cases. However, when laser treatment is performed without a biopsy, a definitive diagnosis cannot be obtained.

As mentioned above, a history of thrombosis needs to be recognized as a risk factor for future venous thrombosis in other parts of the body [1,5]. In addition, regarding lesions that develop on the ventral surface of the tongue, there have been some reports of malignant tumors that caused similar symptoms to those seen in our patient. Thus, pathological examinations are also useful for ruling out other neoplastic diseases.

4. Conclusion

We reported a rare case in which a large venous thrombosis occurred on the ventral surface of the tongue. It is considered that venous thromboses can occur in any part of the oral cavity and should be included among the differential diagnoses for mass lesions arising in the tongue.

References

[1] Nicolaides AN, Breddin HK, Fareed J, Goldhaber S, Haas S, Hull R, et al. Prevention of venous thromboembolism. International consensus statement Guidelines complied in accordance with the scientific evidence. Int Angiol 2001;20:1–37.

[2] Enjoras O, Wassef M, Chapot R. Introduction: ISSVA classification. Color atlas of vascular tumors and vascular malformations. New York: Cambridge University Press; 2007. p. 1–18.

[3] Kamatani T, Saito T, Hamada Y, Kondo S, Shirota T, Shintani S. Intramuscular hemangioma with phleboliths of the tongue. Indian J Dent 2014;5:100–1.

[4] Weathers DR, Fine RM. Thrombosed varix of oral cavity. Arch Derm 1971;104:427–30.

[5] Tobouti PL, Pigatti FM, Marthins-Mussi MC, Machao de Sousa SCO. Oral Thrombus: report of 122 cases with clinically descriptive data. Med Oral Patol Oral Cir Bucal 2017;22:e366–70. https://doi.org/10.4317/medoral.21627.

[6] Greer RO. Thrombosed oral varix. J Oral Surg 1974;32:688–9.

[7] Goodman RS, Kahn E, Valente P. Thrombosis of an oral varix. J Otolaryngol 1982;11:283–5.

[8] Tjioe KC, Oliveira DT, Santos PS. Tongue phlebothrombosis: pathogenesis and potential risks. Quintessence Int 2015;46:545–8.

[9] Lasho JW, Mariner J. Thrombosis in the tongue. New York State Dent J 1964;30:71–2.

[10] Kurita H, Chino M, Kurashina K, Kotani A. Phlebothrombosis with phlebolith of the tongue. Oral Surg Oral Med Oral Pathol 1994;77:552.

[11] Lekovic JP, Friedman CM, Desancho MT. Lingual thrombosis in a woman with antiphospholipid syndrome. Am J Obstet Gynecol 2013;208:e3–4.

[12] Arndal E, Barfoed C, Rasmussen ER. Lingual thrombosis presenting as tongue carcinoma. Case reports in clinical pathology 1. 2014. p. 67–70Http://crcp.sciedupress.com.

[13] Pavilidis P, Gouveris C, Kekes G, Maurer J. Changes in electrogustometry thresholds, tongue tip vascularization, density and form of the fungiform papillae in smokers. Eur Arch Oto-Rhino-Laringol 2014;271:2325–31.

[14] Cervera ER, Font J, Go´mez-Puerta JA, Espinosa G, Cucho M, Bucciarelli S, et al. Validation of the preliminary criteria for the classification of catastrophic antiphospholipid syndrome. Ann Rheum Dis 2005;64:1205–9.

[15] Wiegand S, Tiburtius J, Zimmermann AP, Güldner C, Eivazi B, Werner JA. Localization and treatment of lingual venous and arteriovenous malformations. Vasc Med 2014;19:49–53.

[16] Hedström L, Albrektsson M, Bergh H. Is there a connection between sublingual varices and hypertension? BMC Oral Health 2015;15:78–83.

[17] Shafer GW, Hine MK, Levy BM, Tomich CE. Lingual varices. A textbook of oral pathology. Philadelphia: W.B. Saunders; 1983. P. 30.

[18] Regezi JA, Sciubba JJ, Jordan RCK. Varix and other acquired vascular malformations. Oral pathology St. Louis. Elsevier; 2012. P. 118.