A Case of Phlebolith in the Tip of Tongue

Hisashi KATO[*1, 2], Go OGURA[*3], Masahiro UCHIBORI[*1], Yasuhiro NAKANISHI[*1], Shohei SETA[*1, 2] and Akihiro KANEKO[*1]

[*1]*Department of Oral and Maxillofacial Surgery, Tokai University School of Medicine*
[*2]*Division of Oral and Maxillofacial Surgery, SUBARU Health Insurance Society Ota Memorial Hospital*
[*3]*Department of Pathology, Tokai University School of Medicine*

(Received March 30, 2020; Accepted September 20, 2023)

The differential diagnosis of a tongue mass containing calcified bodies includes a phlebolith associated with vascular lesions, such as hemangioma and vascular malformation, and diseases such as neoplasm, osseous choristoma and hypercalcemic states, including hyperparathyroidism. The appearance of the calcified bodies on plain radiographs may help to differentiate these entities. Computed tomography, magnetic resonance imaging, and ultrasonography are also useful for differentiating these soft tissue lesions. We report a 40-year-old man with a small mass containing a calcified body in the tip of tongue. The mass was surgically resected and histologically evaluated, confirming the diagnosis of phlebolith. Our case was a rare phlebolith that did not involve a vascular lesion.

Key words: phlebolith, tongue mass, calcified body, intravascular thrombus, vascular anomaly

INTRODUCTION

The pathological calcification of soft tissues occurs when calcium and other mineral salts are deposited in a tissue or passage. Dystrophic calcification occasionally occurs in degenerating and dead tissues, while metastatic calcification is the result of an excess of calcium salts in the circulating blood [1, 2]. The soft tissues rarely contain lesions that include calcifications. Phleboliths associated with vascular lesions are the most frequent [3, 4]. However, the differential diagnosis of phleboliths includes neoplasms and hypercalcemic states, including hyperparathyroidism [5–7]. Plain X-rays show the typical appearance of the calcified bodies and may help to differentiate these entities. Computed tomography (CT), magnetic resonance imaging (MRI), and ultrasonography are also useful for differentiating these lesions.

We report a patient with a small mass that contained a calcified body in the tip of tongue. The mass was resected and evaluated histologically, confirming the diagnosis of a phlebolith. We also discuss the differential diagnosis of tongue mass with calcification. Our case was a rare phlebolith that did not involve a vascular lesion.

CASE REPORT

A 40-year-old man presented to the Department of Oral and Maxillofacial Surgery, Tokai University School of Medicine, with a small, painless, movable, hard nodule in the tip of the tongue. It was first noticed by the patient when he rose on the same day. He complained of strangeness in his mouth because of the hard nodule. His past and family histories were unremarkable. On oral examination, there was no glossal asymmetry, restriction of mouth opening, swelling of the tongue, or associated lymph node enlargement. A soybean-sized, round nodule with firm consistency was palpable under the tip of tongue. The mucosa covering the lesion appeared normal. The mass was located far from Wharton's duct. The lost left first mandibular molar tooth was observed (Fig. 1A and B). However, there was no history of trauma, allergy, or other medical problems. Intraoral radiographs revealed a military radiopaque body with a radiolucent center in the tip of tongue (Fig. 2). CT showed a small radiopaque body in the left glossal submucosa (Fig. 3A and B). T1- and T2-weighted MRI revealed a round mass in the tip of tongue as a low-intensity signal. T2-weighted MRI showed a hyperintense signal at the margin of the mass (Fig. 4A and B). Imaging of the nodule by ultrasonography was performed with a curvilinear probe of 7 MHz, which showed a 7 × 5 × 7 mm mass with a smooth periphery in the tip of tongue. The lesion had low echogenicity and contained a calculus-like body (Fig. 5). Ultrasound and color Doppler studies suggested that it was not a vascular lesion. The rest of the clinical and laboratory examination was within normal limits. With the provisional diagnosis of a phlebolith, or tumor in the tip of tongue, the mass was resected under local anesthesia. A small round body was found in the left glossal submucosa and removed. There was little bleeding, and the wound was closed in layers. The resected lesion was a 5 × 5 × 4 mm in size, showing a globe-shaped, rubbery and bright reddish-brown mass with a smooth surface (Fig. 6A). In the cut surface, the mass was filled with bright brown and elastic soft material, which had an onion-like ap-

Hisashi KATO, Department of Oral and Maxillofacial Surgery, Tokai University School of Medicine, 143 Shimokasuya, Isehara, Kanagawa 259-1193, Japan
Tel: +81-463-93-1121 Fax: +81-463-90-5902 E-mail: hisashi@tokai-u.jp

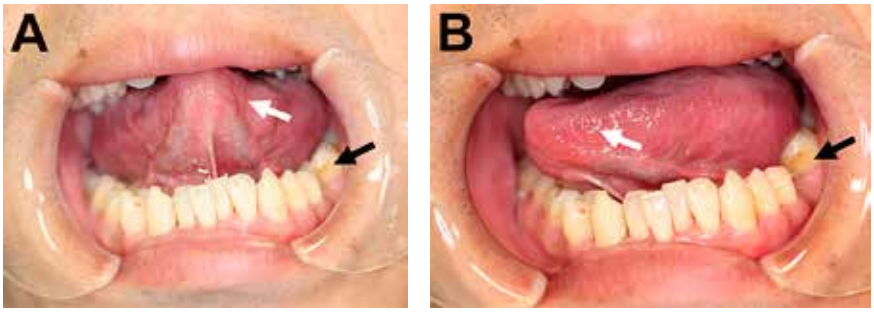

Fig. 1 A) and B) Intraoral picture showing no glossal asymmetry and swelling of the tongue. A soybean-sized, round nodule with firm consistency is palpable under the left tip of tongue (white arrow). The mucosa covering the lesion is normal. The left first mandibular molar has been lost (black arrow).

Fig. 2 Intraoral radiograph showing a spherical radiopaque body with a radiolucent center in the tip of tongue (white arrow).

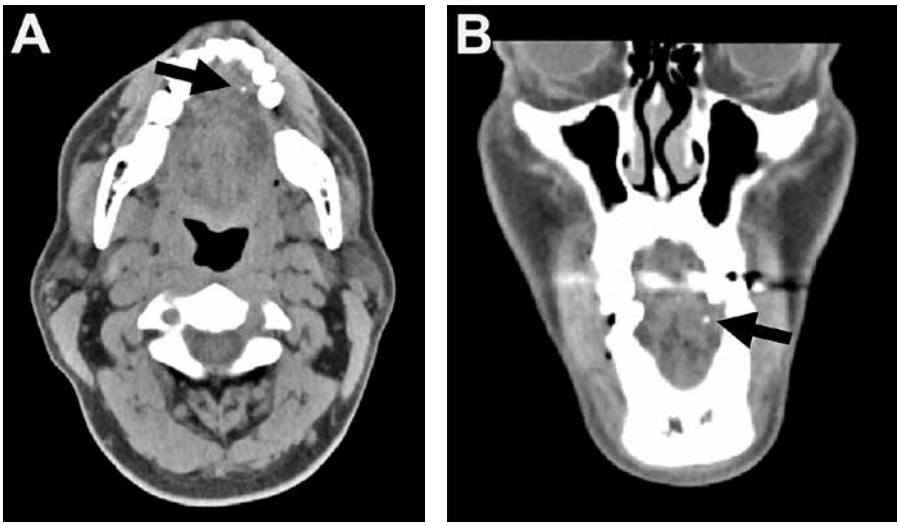

Fig. 3 A) and B) Axial CT image showing a small radiopaque body in the left glossal submucosa (white arrow).

pearance. A small calculus-like body was embedded in the center of the lesion (Fig. 6B). Histopathologically, the lesion seemed like a thrombus with lamellar fibrosis and calcification, focally surrounded by a papillary structure in hematoxylin and eosin (HE) stain (Fig. 7A). The papillary structure was composed of a fibrous core with endothelial lining (Fig. 7B). In Elastica-Masson (EM) stain, the thrombus-like lesion with papillary structure was surrounded by collagen fibers, and partially by elastic fibers (Fig. 7C), meaning that the

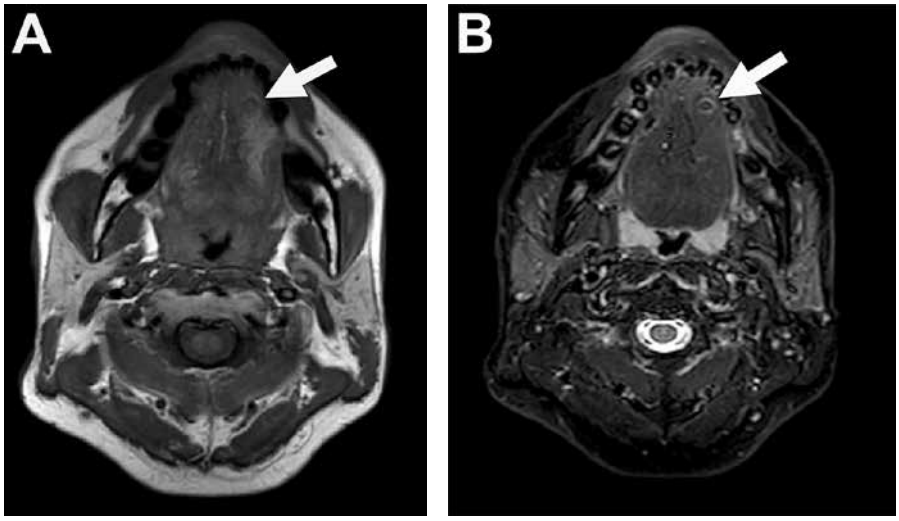

Fig. 4 MRI showing a round mass in the tip of tongue (white arrow). A) Axial T1-weighted MRI showing a mass as a low-intensity signal. The signal intensity is slightly lower than in the muscle. There is a further low-intensity area at the submarginal regions of the lesion. B) Axial T2-weighted MRI showing a well-circumscribed, low-intensity mass with a hyperintense signal at the margin.

Fig. 5 Grayscale sonograms showing a 7 × 5 × 7 mm mass with a smooth periphery in the tip of tongue (white arrow). The lesion has low echogenicity and contains a calculus-like body.

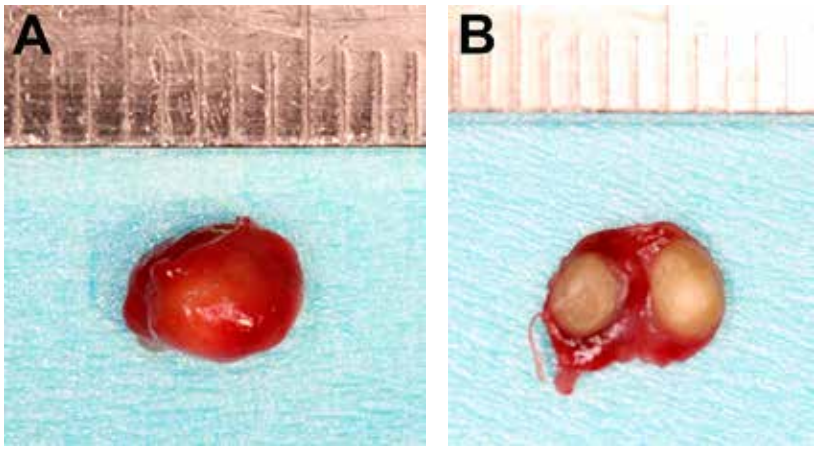

Fig. 6 Macroscopic appearance of resected specimen. A) The lesion measures 5 × 5 ×4 mm, showing a globe-shaped, rubbery and bright reddish-brown mass with a smooth surface. B) Cut surface of the lesion. The lesion is filled with bright brown and elastic soft material, and has an onion-like appearance with small calculus-like body embedded in the center of the lesion.

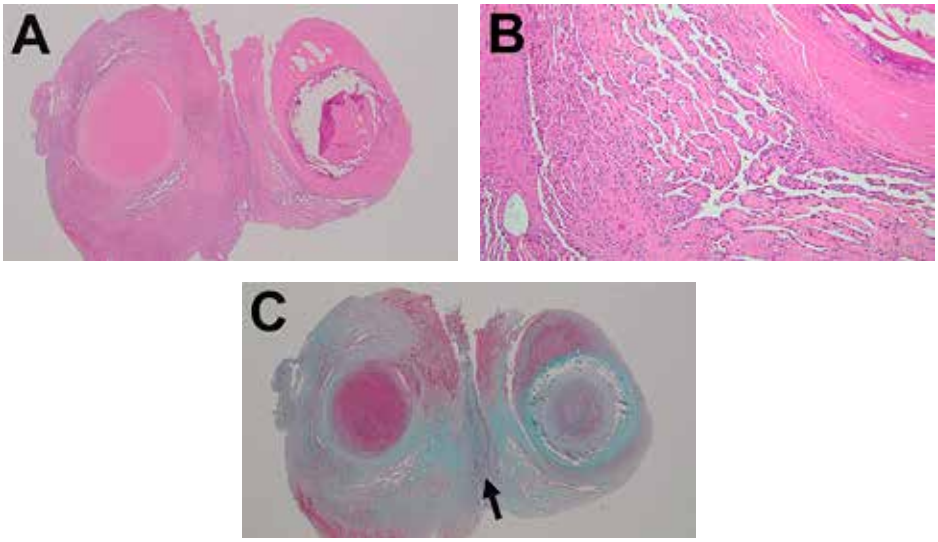

Fig. 7 Histopathology of the lesion. A) The lesion shows a thrombus-like appearance with fibrosis and calcification. Papillary structure is noted around thrombus-like material (hematoxylin-eosin stain, original magnification, × 2). B) Papillary structure is composed of a fibrous core with endothelial lining (hematoxylin-eosin stain, original magnification, × 20). C) Around the thrombus-like material, elastic fiber is partially observed (black arrow). This means that the lesion is intravenous (Elastica-Masson stain, original magnification, × 2).

lesion was intravenous. Additionally, lamellar fibrosis was noted in the thrombus-like lesion as it was in the HE stain, so this lamellar fibrosis seemed to represent the macroscopic onion-like appearance of this lesion. The lesion was then considered a venous thrombus with fibrosis and calcification, and the papillary structure was considered the organizing process. Finally, the diagnosis of phlebolith was made. There has been no recurrence in the 12 months since surgery.

DISCUSSION

When evaluating a tongue mass with a calculus-like body, phleboliths associated with vascular lesions are the most frequent. However, the mass must be differentiated from neoplasms, osseous choristoma, and metastatic calcifications, which occur in case of hypercalcemic states, including hyperparathyroidism [5–7]. Osseous choristoma could be easily ruled out based on clinical findings because it exists in the dorsal surface near the circumvallate papillate of the tongue [8, 9]. Metastatic calcifications also could be easily ruled out based on medical history. Metastatic soft-tissue calcifications are almost exclusively found in uremic patients, such as patients with renal failure with a long history of elevated calcium-phosphate product and secondary hyperparathyroidism. However, improvements in the management of end-stage renal disease have significantly reduced the incidence of these complications [6].

The appearance of calcified bodies on a plain radiograph may help to differentiate other entities. A phlebolith is usually laminated, with a radiopaque center, although the center is sometimes radiolucent; a small phlebolith is uniformly radiopaque. Moreover, multiple phleboliths are often present when associated with vascular anomalies [3]. In our case, the plain radiograph showed a round radiopaque body with a radiolucent center. The mass was resected with the provisional diagnosis of a phlebolith, or tumor, and the diagnosis of phlebolith was confirmed histologically.

The pathogenesis of phleboliths is thought to involve an organized thrombus produced when the peripheral blood flow slows [3, 4]. The thrombus calcifies, forming the core of the phlebolith. Then, the fibrinous component undergoes secondary calcification and becomes attached. Repetition of this process causes enlargement of the phlebolith [10, 11]. Therefore, most phleboliths are found along with vascular anomalies.

In the head and neck area, vascular anomalies and phleboliths are relatively common. In fact, intramuscular hemangiomas most frequently occur in the masseter muscle, and phleboliths are frequently found at the masseter and/or buccinators muscles [12–16]. In addition, occasionally there is masticatory trauma to the oral mucosa, leading to thrombus formation in this area [14]. Our case was not associated with a vascular anomaly. The occurrence of this solitary phlebolith without vascular lesions in the tongue is rare. To our knowledge, besides this case, there are two previously reported cases of solitary phlebolith in the tongue. [12, 17–19]. A venous thrombus caused by masticatory trauma to the tongue and consequent stasis of the blood flow could result in phlebolith formation [18, 19]. Although there was no history of trauma, there might have been unidentified masticatory trauma to the tongue because of the lost left first mandibular molar tooth. As another possible cause, a childhood hemangioma with phlebolith could have gradually become smaller and eventually the phlebolith may be the only residual sign in the adult [12].

Diagnostic imaging modalities, such as X-rays, CT, MRI, and ultrasonography, are useful for differentiating these soft tissue lesions with calcifications. The most cost-effective modality of them to diagnose a phlebolith is plain X-ray because of the typical appearance of the calcified bodies, while CT, MRI, and ultrasonography have significantly increased the accuracy of the differential diagnosis [10]. Vascular lesions are characteristically much more hyperintensity on T2-

weighted images than T1-weighted images due to the increased free water present within stagnant blood in the vessels. Ultrasound and color Doppler studies can also suggest a vascular lesion [15]. In our case, T1- and T2-weighted MRI revealed a round mass in the tip of tongue as a low-intensity signal, and T2-weighted MRI showed a hyperintense signal only at the margin of the mass. Ultrasonography showed a small round mass containing a calculus-like body. However, ultrasound and color Doppler studies suggested that it was not a vascular lesion. In the preoperative assessment, CT, MRI, and ultrasonography accurately showed that the mass did not involve vascular anomalies and contained a calculus-like body.

The treatment for phlebolith is not necessary to be resected after fairly accurate preoperative diagnosis as phlebolith if the patient has no problem with phlebolith. However, the patient complained of strangeness in his mouth because of the phlebolith, and surgical removal of the lesion was performed.

When evaluating a tongue mass with a calculus-like body, the lesion should initially be considered a phlebolith. In addition, a detailed interview of the history of the illness, such as injury and masticatory trauma to the tongue, may help to differentiate several entities. Plain X-rays may also help with the diagnosis because of the typical appearance of the calcified bodies, while CT, MRI, and ultrasonography are more useful for making an accurate diagnosis.

REFERENCES

1) Scarpelli DG, Chiga M. Cell injury and errors of metabolism. In: Anderson WAD, Kissane JM, editors. Pathology. Saint Louis: C. V. Mosby; 1977. p. 90–147.
2) Sencimen M, Gulses A, Ogretir O, Gunhan O, Ozkaynak O, Okcu KM. Dystrophic calcifications arising in the masseter muscle: a case report. Quintessence Int 2010; 41: 295–7.
3) O'Riordan B. Phleboliths and salivary calculi. Br J Oral Surg 1974; 12: 119–31.
4) Raymond AK, Batsakis JG. Angiolithiasis and sialolithiasis in the head and neck. Ann Otol Rhinol Laryngol 1992; 101: 455–7.
5) Stewart JG, MacMahon H, Vyborny CJ, Pollak ER. Dystrophic calcification in carcinoma of the lung: demonstration by CT. AJR Am J Roentgenol 1987; 148: 29–30.
6) Walker RP, Kooiker PD, Chow JM, Sizemore GW, Solans E, Paloyan E, *et al*. Metastatic soft tissue calcification presenting as a tongue mass. Otolaryngol Head Neck Surg 1993; 109: 540–2.
7) Keberle M, Robinson S. Physiologic and pathologic calcifications and ossifications in the face and neck. Eur Radiol 2007; 17: 2103–11.
8) Andressakis DD, Pavlakis AG, Chrysomali E, Rapidis AD. Infected lingual osseous choristoma. Report of a case and review of the literature. Med Oral Patol Oral Cir Bucal 2008; 13: e627–32.
9) Gorini E, Mullace M, Migliorini L, Mevio E. Osseous choristoma of the tongue: a review of etiopathogenesis. Case Rep Otolaryngol 2014; 2014: 373104.
10) Kanaya H, Saito Y, Gama N, Konno W, Hirabayashi H, Haruna S. Intramuscular hemangioma of masseter muscle with prominent formation of phleboliths: a case report. Auris Nasus Larynx 2008; 35: 587–91.
11) Mulliken JB, Fishman SJ, Burrows PE. Vascular anomalies. Curr Probl Surg 2000; 37: 517–84.
12) Zachariades N, Rallis G, Papademetriou J, Konsolaki E, Markaki S, Mezitis M. Phleboliths. A report of three unusual cases. Br J Oral Maxillofac Surg 1991; 29: 117–9.
13) Rossiter JL, Hendrix RA, Tom LW, Potsic WP. Intramuscular hemangioma of the head and neck. Otolaryngol Head Neck Surg 1993; 108: 18–26.
14) Scolozzi P, Laurent F, Lombardi T, Richter M. Intraoral venous malformation presenting with multiple phleboliths. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2003; 96: 197–200.
15) Rai P, Setia S, Kalra N, Upreti L. Intramuscular vascular malformation of the masseter muscle presenting with turkey wattle sign. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2006; 102: 618–24.
16) 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.
17) Gouvêa Lima Gde M, Moraes RM, Cavalcante AS, Carvalho YR, Anbinder AL. An Isolated Phlebolith on the Lip: An Unusual Case and Review of the Literature. Case Rep Pathol 2015; 2015: 507840.
18) Kurita H, Chino M, Kurashina K, Kotani A. Phlebothrombosis with phlebolith of the tongue. Oral Surg Oral Med Oral Pathol. 1994; 77: 552.
19) Tjioe KC, Oliveira DT, Santos PS. Tongue phlebothrombosis: Pathogenesis and potential risks. Quintessence Int. 2015; 46: 545–8.