



Case report

A case of large varix including partially organizing thrombosis on the oral floor

Riki Tomita^a, Naoya Kitamura^{a,*}, Yasumasa Yoshizawa^a, Eri Sasabe^a, Yasusei Kudo^b, Tetsuya Yamamoto^a^a Department of Oral and Maxillofacial Surgery, Kochi Medical School, Kochi University, Kochi 783-8505, Japan^b Department of Oral Molecular Pathology, Tokushima University Graduate School of Biomedical Sciences, Tokushima 770-8504, Japan

ARTICLE INFO

Keywords:

Large varix
Oral floor
Organizing thrombosis

ABSTRACT

Small thrombosed varices (diameter, < 10 mm) in the oral cavity are known to be prevalent in the tongue, lips, and buccal mucosa of elderly individuals. No past reports have described varices larger than 10 mm, except in the tongue. We report a very rare case in which a large varix (20 × 20 × 15 mm) in the midline of the oral floor and including partially organizing thrombosis occurred 3 days before the first visit in an 82-year-old man. This case is the first report of large oral varix in a region other than the tongue. Large oral varix may occur not only on the tongue, but also in any area of the oral cavity.

1. Introduction

Varix is a condition characterized by purplish venous ectasia. Small thrombosed varices (diameter, < 10 mm) in the oral cavity are known to be prevalent in the tongue, lips, and buccal mucosa of older individuals [1–3]. The prevalence of this pathology increases with age, with surveys indicating presence in up to 60 % of elderly patients, in both sexes [4–6]. Small varices are common, but large varix has been reported in the past only in several cases involving the tongue region. No past reports have described a varix larger than 10 mm in any sites other than the tongue. We report a very rare case of large varix in the midline of the oral floor that included partially organizing thrombosis in an elderly patient.

2. Case report

An 82-year-old man was referred to our hospital with a chief complaint of a dark-purplish mass on the midline of the oral floor in March 2014. His medical history included hyperlipidemia and prostatic hypertrophy. He was therefore taking pravastatin sodium (10 mg/day) and tamsulosin hydrochloride (0.2 mg/day). Three days before the first visit to our hospital, he noticed a thumb-sized purplish mass on the midline of the oral floor.

Clinical examination at the first visit revealed a well-circumscribed, elastic, slightly soft, movable, pendulous, dark-purplish mass,

measuring 20 × 20 × 15 mm in diameter, in the midline of the oral floor (Fig. 1). The upper and lower jaws had a single remaining incisor tooth and an edentulous jaw, respectively, with upper partial dentures and a lower complete denture used daily. No special abnormalities were observed in complete blood count or blood biochemistry.

Magnetic resonance imaging showed high signal intensity on T1-weighted imaging, high-low mixed signal intensity on T2-weighted imaging, and poor effective signal on both fat-saturation and gadolinium-enhanced images (Fig. 2).

Based on these findings, clinical differential diagnoses included cyst containing blood components, venous malformation (so-called hemangioma), or varix. Excisional biopsy was performed under local anesthesia on the day of the first visit. Using a bipolar electric scalpel, the mass was excised at the pedicle, and intraoperative blood loss was minimal.

Histopathologically, a markedly expanded vascular lumen (comprising both an internal elastic layer dyed with Elastica van Gieson staining and a vascular endothelial cell layer) was seen in the submucosal muscle layer (Fig. 3). The lumen was filled with partially organizing venous thrombosis containing platelets, fibrin, and cell components. The histopathological diagnosis was partially organizing venous thrombosis.

Based on these results, the final diagnosis was varix including partially organizing thrombosis on the oral floor. As of the time of writing, approximately 6 years postoperatively, no recurrence has been observed.

* Corresponding author at: Kohasu, Oko-cho, Nankoku-shi, Kochi 783-8505, Japan.

E-mail addresses: jm-rikitomita@kochi-u.ac.jp (R. Tomita), nkitamura@kochi-u.ac.jp (N. Kitamura), yoshizawa@kochi-u.ac.jp (Y. Yoshizawa), yoshieri@kochi-u.ac.jp (E. Sasabe), yasusei@tokushima-u.ac.jp (Y. Kudo), yamamott@kochi-u.ac.jp (T. Yamamoto).<https://doi.org/10.1016/j.ajoms.2020.03.003>

Received 4 December 2019; Received in revised form 3 March 2020; Accepted 13 March 2020

Available online 15 May 2020

2212-5558/ © 2020 Asian AOMS, ASOMP, JSOP, JSOMS, JSOM, and JAMI. Published by Elsevier Ltd This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).



Fig. 1. Intraoral photograph at first visit. Large (diameter, 20 × 20 × 15 mm), elastic, slightly soft, pedunculated, dark-purplish mass developed on the midline of the oral floor.

3. Discussion

Varix is a condition characterized by purplish venous ectasia. Small thrombosed varices (diameter, < 10 mm) in the oral cavity are known to be prevalent in the tongue, lips, and buccal mucosa of elderly individuals [1–3]. The prevalence of this pathology increases with age, with surveys indicating presence in up to 60 % of elderly patients, in both sexes [4–6]. We encountered a case of large varix in the midline of the oral floor with partially organizing thrombosis that occurred 3 days before the first visit in an 82-year-old man. Eguchi et al. suggested that venous thrombosis can occur in any part of the oral cavity, but large thrombosed oral varix is very rare [7]. No previous reports appear to have described varix larger than 10 mm except in the tongue region, and they considered a total of 7 cases of thrombosed lingual varix (including their own case) [7–12].

Chronic mechanical stimulation is thought to be one cause of oral varix. Eguchi et al. considered chronic mechanical stress on the sublingual vein by the hook of a Conus Krone denture as a cause of varix [7]. In our case, the lingual midline edge of the lower complete denture was always in contact with the lingual frenulum, which might have caused chronic irritation of the peripheral branch of the facial vein at

the oral floor. Alternatively, the single remaining maxillary central incisor tooth (Fig. 1) may have damaged the mucosa of the oral floor when the patient was not wearing dentures.

Past reports of organized thrombosed large lingual varix have displayed signal hypointensity on T1-weighted imaging and signal hyperintensity on T2-weighted imaging. However, our case displayed signal hyperintensity on T1-weighted imaging, mixed high-low signal intensity on T2-weighted imaging, and poor effective signal on both fat-saturation and gadolinium-enhanced imaging. This was attributed to our oral floor varix representing organizing thrombosis rather than organized thrombosis.

Histopathologically, as with past reports of varix, a markedly expanded vascular lumen comprising both an internal elastic layer dyed with Elastica van Gieson staining and a vascular endothelial cell layer was seen in the submucosal muscle layer. In our case, the varix lumen had not yet been completely organized and showed partially organizing venous thrombosis, because only 3 days had passed since varix onset.

4. Conclusion

We reported a very rare case in which large varix including partially organizing thrombosis was identified in the midline of the oral floor. This report offers the first description of large oral varix arising in a region other than the tongue. Large oral varix may occur not only on the tongue, but also on any area of the oral cavity.

Ethical approval

The patient has been informed that there is no risk to their anonymity in association with publication of this report [but were they asked for permission and did they give written informed consent for publication?].

Funding

The authors declare that no funding was provided for the performance of this study.

Transparency document

The [Transparency document](#) associated with this article can be found in the online version.

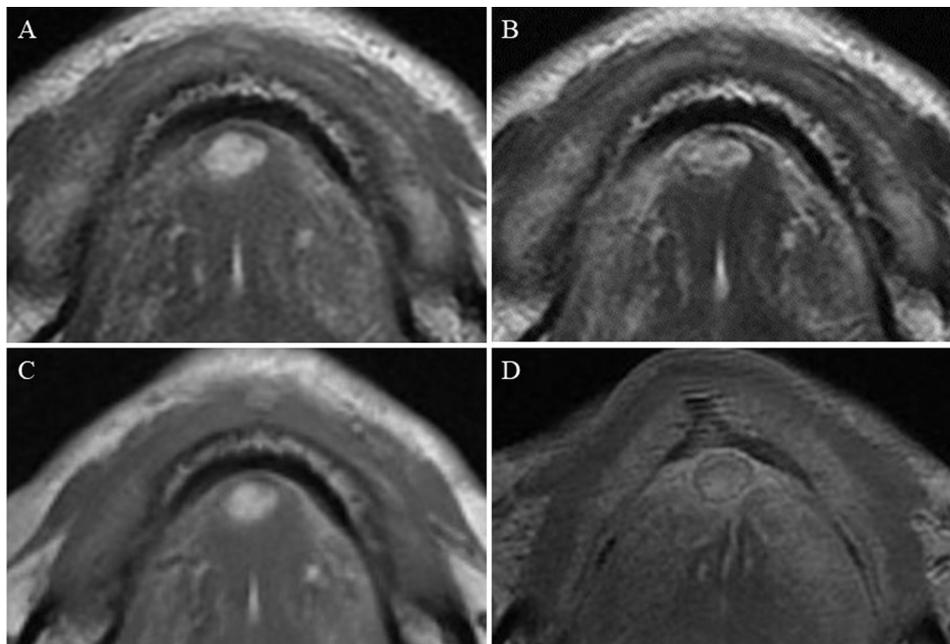


Fig. 2. Magnetic resonance imaging. Signal hyperintensity on T1-weighted imaging, high-low mixed signal intensity on T2-weighted imaging, and poor effective signal on both fat saturation and gadolinium-enhanced imaging. A) T1-weighted image; B) T2-weighted image; C) fat-saturation image (T1 base); D) gadolinium-enhanced image (T1 base).

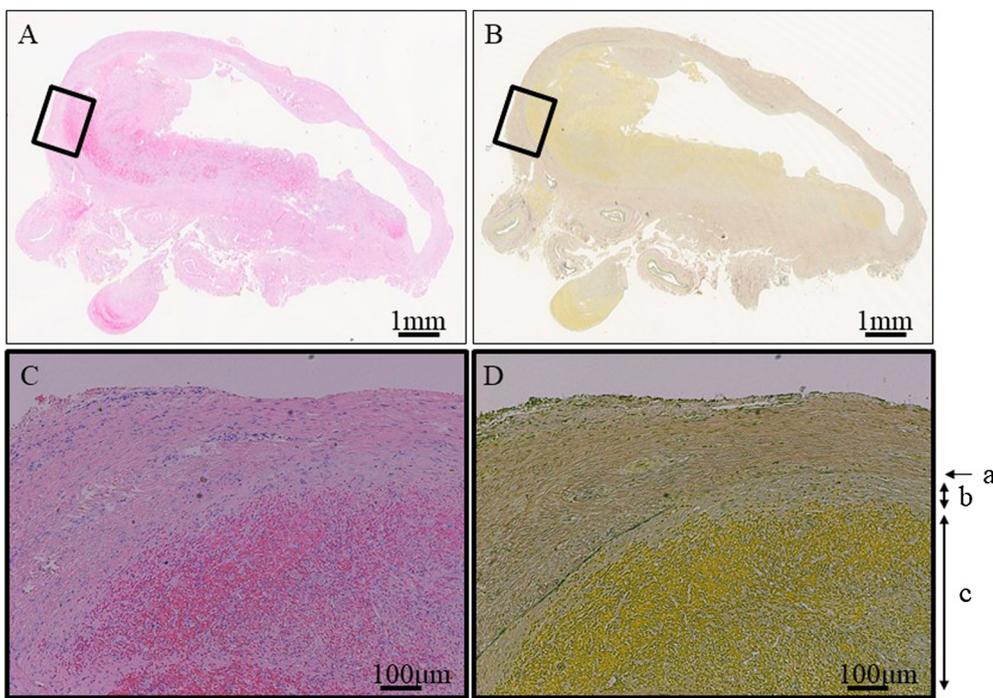


Fig. 3. Histopathological findings. A markedly expanded vascular lumen, comprising both an internal elastic layer dyed with Elastica van Gieson staining and a vascular endothelial cell layer, is seen in the submucosal muscle layer. The lumen is filled with partially organizing venous thrombosis containing platelets, fibrin, and cell components. A) Hematoxylin and eosin (HE) staining, low magnification (bar: 1 mm); B) Elastica van Gieson (EVG) staining, low magnification (bar: 1 mm); C) HE staining, high magnification (bar: 100 µm); D) EVG staining, high magnification (bar: 100 µm). a, Internal elastic layer; b, vascular endothelial cell layer; c, organizing thrombosis.

Declaration of Competing Interest

The authors declare there were no conflicts of interest during the performance of this study.

Acknowledgments

The authors express their sincere thanks to H. Toi, MD, PhD, from the Laboratory of Diagnostic Pathology at Kochi Medical School Hospital for advice on pathological findings in the present this article.

References

[1] Nasir A, Gupta D, McBride G. Minerva picture case. *BMJ* 2006;333:104. <https://doi.org/10.1136/bmj.333.7558.104>.
 [2] Michael NP. Sublingual varices are not unusual. *BMJ* 2006;333:202. <https://doi.org/10.1136/bmj.333.7560.202>.

[3] Nasir A, Latif A, McBride G. Lingual varices. *J Coll Physicians Surg Pak* 2008;18:197–8. PMID: 18460259.
 [4] Kleinman HZ. Lingual varicosities. *Oral Surg Oral Med Oral Pathol* 1967;23:546–8.
 [5] Kovac-Kavcic M, Skaleric U. The prevalence of oral mucosal lesions in a population in Ljubljana, Slovenia. *J Oral Pathol Med* 2000;29:331–5.
 [6] Ettinger RL, Manderson RI. A clinical study of sublingual varices. *Oral Surg* 1974;38:540–5.
 [7] Eguchi M, Shigematsu H, Oku Y, Kikuchi K, Okada M, Sakashita H. A case of a large thrombosed lingual varix. *J Oral Maxillofac Surg Med Pathol* 2019;31:180–4.
 [8] Weathers DR, Fine RM. Thrombosed varix of oral cavity. *Arch Derm* 1971;104:427–30.
 [9] Kc Tjioe, Oliveira DT, Santos PS. Tongue phlebothrombosis: pathogenesis and potential risks. *Quintessence Int* 2015;46:545–8. PMID: 25918751.
 [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 Vol. 1. 2014. p. 67–70 <http://crp.sciedupress.com>.