

Case Report

A case of phleboliths of the lower lip

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Abstract

We report a patient with phleboliths of the lower lip not associated with a hemangioma or vascular malformation. A 50-year-old male noticed a soft, elastic mass in the left lower lip, and repeatedly bit this area by mistake. A diagnosis of a mucocele was made, and cystectomy was performed under local anesthesia on an outpatient basis. Histopathological examination showed dystrophic calcification coated with fibrous connective tissue. His postoperative course has been favorable up to the present, without findings suggesting recurrence.

Keywords: Phleboliths; Hemangioma; Lower Lip; Mucocele; Calcification

Introduction

Phleboliths result from dystrophic calcification due to fibrin and thrombus formation caused by local abnormalities of the coagulation system, or impaired blood flow associated with hemangiomas or vascular malformations [1]. In the head and neck area, phleboliths often develop in the deep part of hemangiomas or vascular malformations; the complication rate was reported to be 5-20% [2]. In the oral area, phleboliths of the tongue and buccal mucosa have been reported [3-6], but those of the lip are rarely observed, and only a few patients have been reported [7-10]. Among them, there was no patient with phleboliths of the lower lip. In addition, our patient had no history of a hemangioma or vascular malformation, and phleboliths were superficially

present. We report a patient with phleboliths of the lower lip, showing no vascular lesion.

Case Report

A 50-year-old male visited our department due to a mass in the mucosa of the left lower lip. His past history contained nothing of note. Concerning the present illness, he noticed a mass in the mucosa of the left lower lip in July 2012, and repeatedly bit this area by mistake. Since the lesion size did not decrease, he visited our department for close examination. The mass (7 x 8 mm) was elastic and soft, and its surface was smooth, showing no induration (Figure 1). The diagnosis of a mucocele was made, and cystectomy was performed under local anesthesia on an outpatient basis. Since

there was no adhesion between the lesion and surrounding connective tissue, resection could be readily performed. No fluid content was observed. After resection, there was no swollen small salivary gland around the wound. As for the postoperative progress, he was good, and the recurrence was not accepted. Because the wound was good, operation one month later, I assumed follow-up the end.

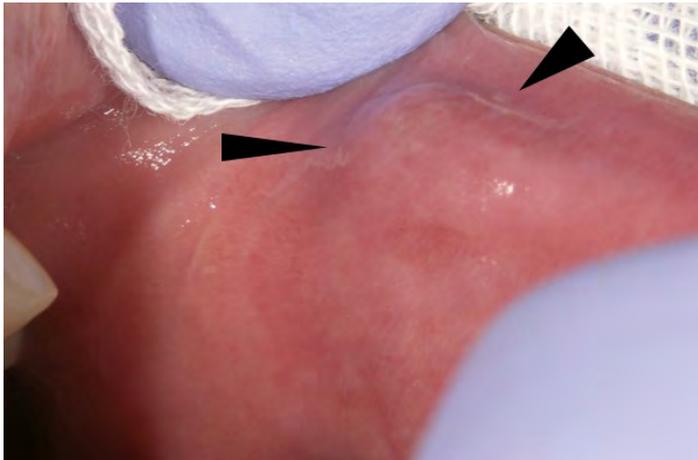


Figure 1. Oral findings at the first examination

A soft, elastic mass (7 x 8 mm) with a smooth surface was observed in the mucosa of the left lower lip (black arrowhead).

Histopathological examination showed hyalinized calcifications coated with fibrous connective tissue (Figure. 2a) and vein-like lumens around the calcifications (Figure. 2b).

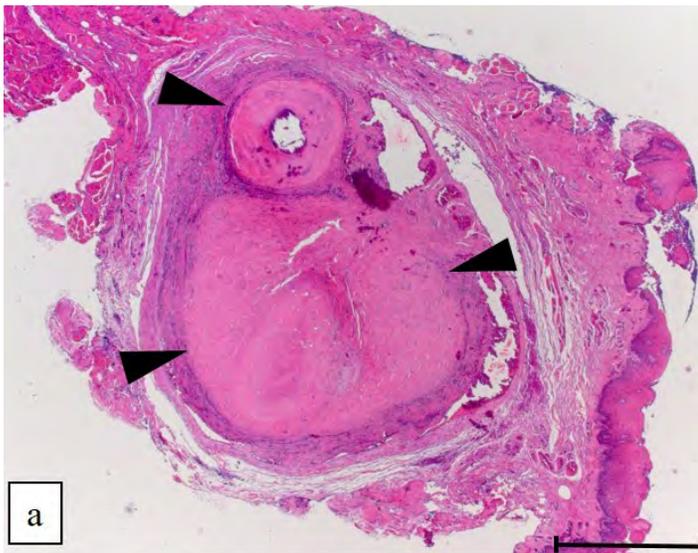
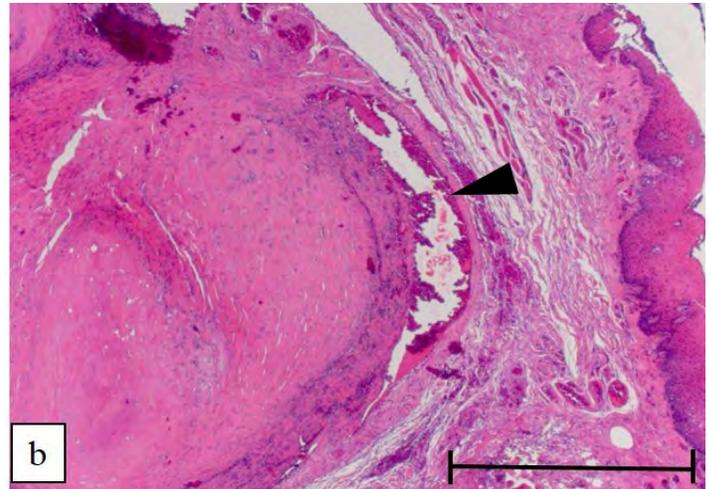


Figure 2. Histopathological image of a surgical specimen

a: Calcifications coated with fibrous connective tissue were observed (black arrowhead). bar = 1,000 μ m.



b: There was a vein-like vascular lumen around the calcifications (black arrowhead). bar =1,000 μ m.

Discussion

Phleboliths are calcifications resulting from concentric thrombus formation in the vascular wall due to abnormalities in the coagulation system or a decrease or stagnation of circulating blood flow in local areas. This disease is often detected in association with hemangiomas or vascular malformations [1, 11, 12]. In the head and neck area, phleboliths often develop in the deep part of hemangiomas or vascular malformations; the complication rate was reported to be 5-20% [2]. However, to our knowledge, there have been only 5 patients reported with phleboliths of the lip [7-10] including ours, and only our patient had phleboliths of the lower lip (Table 1). In the oral area, the incidence of hematomas of the lip is relatively high, being similar to that of hematomas of the buccal mucosa. However, the incidence of hematomas accompanied by phleboliths is very low. This was suggested to be because of marked changes in lip hemodynamics due to high-level movements and blood flow per unit area within hematomas [9], but no definite cause has been reported. In our patient, phleboliths alone developed in the absence of hemangiomas or vascular malformations, which is very rare. A medical interview with the patient could not clarify the cause of swelling of the lower lip mucosa and associated past history. However, since he repeatedly bit his lip by mistake, there is a possibility that organic injury led to hematoma formation. Since a patient in whom phleboliths developed due to a bite of the buccal mucosa was reported [11], there is a possibility that repeated biting by mistake also led to hematoma formation in our patient. We speculate that blood disappeared with time, and only the thrombus remained, leading to phleboliths.

Year	Author	Age	Gender	Site
1967	Masuyama	16	M	Upper lip
1980	Ivey	44	M	Upper lip
1988	Fujita	67	F	Upper lip
1994	Yamamoto	84	F	Upper lip
2014	Present case	51	M	Lower lip

Table 1. Reports of phleboliths of the lip.

In this patient, we could not suspect phleboliths until the examination of tissue specimens. In previously reported patients with phleboliths associated with previous hemangiomas or vascular malformations, indurations suggesting phleboliths were palpable. Therefore, the diagnosis of a hemangioma could be made in all patients, excluding the patient reported by Fujita et al. [9] who observed phleboliths using radiography, and confirmed their presence after resection [7, 8, 10]. However, clinical diagnosis may be difficult in patients such as ours with phleboliths not accompanied by a hemangioma. For our patient, we did not perform radiography because phleboliths of the lower lip are rare, no hemangioma or palpable indurations were observed, and the mass was elastic and soft. In this patient, en bloc resection of the mass containing that phleboliths was performed, and no postoperative recurrence was observed, suggesting that this treatment was appropriate. For the diagnosis of phleboliths, radiography is the most useful. In patients with a mass in the lower lip, diagnosis and treatment with consideration given to the possibility of phleboliths similar to those in our patient may be important.

Conclusion

We reported a patient with phleboliths of the lower lip without vascular lesions, along with a brief review of the literature. For the diagnosis of phleboliths, radiography is the most useful. In patients with a mass in the lower lip, diagnosis and treatment with consideration given to the possibility of phleboliths similar to those in our patient may be important.

Conflict of Interest

The authors declare no conflicts of interest.

Acknowledgements

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