International Journal of Surgery Case Reports 84 (2021) 106030

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

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

Case report

A challenging diagnosis of a mucocele in the maxillary gingiva: Case report and literature review

Nagwan Elsayed [b], Tsuyoshi Shimo [a,*], Fumiya Harada [b], Daichi Hiraki [a], Masayasu Tashiro [c], Eiji Nakayama [c], Yoshihiro Abiko [d], Hiroki Nagayasu [b]

[a] Division of Reconstructive Surgery for Oral and Maxillofacial Region, Department of Human Biology and Pathophysiology, School of Dentistry, Health Sciences University of Hokkaido, Hokkaido 061-0293, Japan
[b] Division of Oral and Maxillofacial Surgery, Department of Human Biology and Pathophysiology, School of Dentistry, Health Sciences University of Hokkaido, Hokkaido 061-0293, Japan
[c] Division of Oral and Maxillofacial Radiology, Department of Human Biology and Pathophysiology, School of Dentistry, Health Sciences University of Hokkaido, Hokkaido 061-0293, Japan
[d] Division of Oral Medicine and Pathology, Department of Human Biology and Pathophysiology, School of Dentistry, Health Sciences University of Hokkaido, Hokkaido 061-0293, Japan

ARTICLE INFO

Keywords:
Mucocele
Maxillary gingiva
Differential diagnosis

ABSTRACT

Introduction: Mucoceles are mucous extravasation phenomena resulting from spontaneous ductal rupture or, less commonly, a traumatic cutting of a salivary excretory duct. Occasionally, the appearance of a mucocele closely resembles a neoplastic lesion, and it can be difficult to diagnose.
Presentation of case: A 74-year-old Japanese male patient was referred to our clinic with painless swelling related to the upper right canine-premolar area. Preoperative imaging of the lesion revealed that it was fluid-filled and the histopathological biopsy suggested a myxoid tumor. We excised the lesion with the patient under general anesthesia, using safety margins based on the histology of the biopsy specimen to reach the definitive diagnosis and treatment. The final pathological diagnosis was a mucocele in the vicinity with an aberrant small salivary gland. Follow-up visits showed complete healing of the epithelium, and no recurrence has been observed during the subsequent 30 months.
Discussion: In cases with a difficult definitive diagnosis based on medical history, diagnostic imaging and pathological examination, it is important to proceed carefully with the possibility of a tumor in mind.
Conclusion: This case emphasizes that some mucous cysts are challenging for clinicians in terms of diagnosis and treatment due to an uncommon presentation.

1. Introduction

Mucous cysts are classified into two types depending on the cause and the histopathological pattern. The first type is 'mucous extravasation phenomena,' or what is called a 'mucocele.' The other type is a mucous retention cyst. Mucoceles are pseudocysts that result from spontaneous ductal rupture or, less commonly, traumatic cutting of one or more salivary excretory ducts. For this reason, mucoceles appear commonly in the lower lip due to biting; the extravasated mucin elicits an inflammatory reaction and some fibrosis [1]. Mucoceles can occur at almost any site where minor salivary glands exist and are thus less likely to occur on the anterior hard palate and the gingiva, which do not typically have minor salivary glands [2]. Mucoceles can also develop in relation to the sublingual gland (called a 'ranula') and even the parotid gland (called a 'sialocele').

Mucoceles of a minor salivary gland origin present as painless or only mildly uncomfortable, soft, fluid-filled vesicles. The size usually ranges from a few millimeters to centimeters, but most are <1.5 cm in diameter [3]. They appear bluish in color because of the translucent overlying mucosa and the accumulated mucin [4]. Most mucoceles occur in children, teenagers, and young adults. They represent the 17th most common lesion of the oral mucosal diseases. The prevalence is 2.4 cases per 1000 people in the U.S. [5] A special type of mucocele known as a 'superficial mucocele' is formed after a duct rupture in the subepithelial layer; its preferred sites are the palate and retromolar areas [1]. Here, we describe the case of a mucocele in the maxillary gingiva for which

* Corresponding author.
E-mail address: shimotsu@hoku-iryo-u.ac.jp (T. Shimo).

https://doi.org/10.1016/j.ijscr.2021.106030
Received 22 April 2021; Received in revised form 23 May 2021; Accepted 23 May 2021
Available online 26 May 2021

preoperative imaging and a preoperative histopathological biopsy suggested myxoid tumor.

2. Case presentation

In October 2018, a 74-year-old Japanese male was referred to our department with the complaint of intraoral painless swelling of the right maxillary region at the junction between the free gingiva of the canine-premolar area and the buccal mucosa (Fig. 1). A visual inspection revealed that the overlying mucosa was intact and normal in color. On palpation, the lesion was movable and was soft to rubbery in consistency; no tenderness reported by the patient. No induration was found. The swelling's dimensions were nearly 18 × 8 mm. The result of the submandibular lymph nodes examination was negative. According to the data retrieved from the patient's local dentist, the patient required maintenance therapy. A periodontal examination showed that the overall pocket depth measurements were fair. The pocket depth of the related teeth (upper right canine and upper right first premolar) ranged between 3 and 4 mm, which we considered insignificant.

The patient's previous medical history revealed that he had undergone Endoscopic Submucosal Dissection for esophageal cancer in 2016 and gastric cancer in 2017. At our department, a periapical radiograph showed no osseous changes; only cervical bone resorption was evident and was related to the central incisors, and the canine and first premolar. Magnetic resonance imaging (MRI) demonstrated that the swelling appeared as a well-defined lesion that gave a low signal on T1-weighted images (T1WI) (Fig. 2A, arrowhead) and a high signal was on T2-weighted images (T2WI) indicating that the lesion was filled with fluid (Fig. 2B, arrowhead). In the short-TI inversion recovery (STIR) view, the lesion was bright and well-defined (Fig. 2C, arrowhead) that corresponded to the previous findings (Fig. 2A, B). Mild thickening of the mucosal lining of the right adjacent maxillary sinus was also identified.

Contrast-enhanced computed tomography (CECT) showed no bone resorption of the maxilla due to the lesion, but a mass with a low CT value and a clear non-contrast-enhanced boundary is observed in the area corresponding to the MRI findings. (Fig. 2D, arrowhead), which is why the possibility of a neoplastic lesion could not be denied. Light microscopy observation of the biopsy specimen showed that the lesion consisted of a mucoid matrix and fibrous connective tissue, and thus a myxoid tumor including odontogenic myxoma was suspected.

In November 2018, we excised the lesion with the patient under general anesthesia with safety margins based on the histology of the biopsy specimen, to reach the definitive diagnosis and identify the optimal treatment. After the lesion's removal, a feeding vessel that had been under the lesion was encountered, and its bleeding was stopped with electrocautery.

Fig. 3A is a photograph of the bony surfaces after enucleation; the arrow indicates the alveolar foramen where the feeding vessel came out and entered the lesion. The resected specimen (bone side) was bluish in color (Fig. 3B; the arrow indicates the entry of the resected feeding vessel). The defect was closed using a collagen membrane and antibiotic gauze to apply gentle pressure on the membrane and allow for secondary healing.

The pathological examination of the resected lesion revealed a mucocele and a thickened and well-defined cyst wall (Fig. 3C, D, asterisk) consisting of fibrous connective tissue in the vicinity with an aberrant small salivary gland and dilation of a salivary duct (Fig. 3C, D). Abundant blood vessels were found in the tissue around the cyst wall (Fig. 3D, arrowheads), suggesting that preoperative differential diagnosis from neoplastic lesion was difficult. As of 30 months since the surgery, the patient has reported no pain, good epithelialization developed from the surrounding tissue, and there has been no recurrence.

3. Discussion

When considering differential diagnoses of soft-tissue swelling in the buccal vestibule or particularly at the junction between the free gingiva and the buccal mucosa, the list should include a neoplasm of a minor salivary gland and a lesion of mesenchymal origin. Because acinic cell carcinoma clinically resembles a mucocele, it is easy to be mistaken for a mucocele. In our patient's case, the initial histopathological report mentioned that the specimen contained mucoid tissue, and a myxoid tumor was suspected. With the information available at that time point, the main differential clinical diagnosis of the lesion was a suspected peripheral odontogenic myxoma or acinic cell carcinoma. The

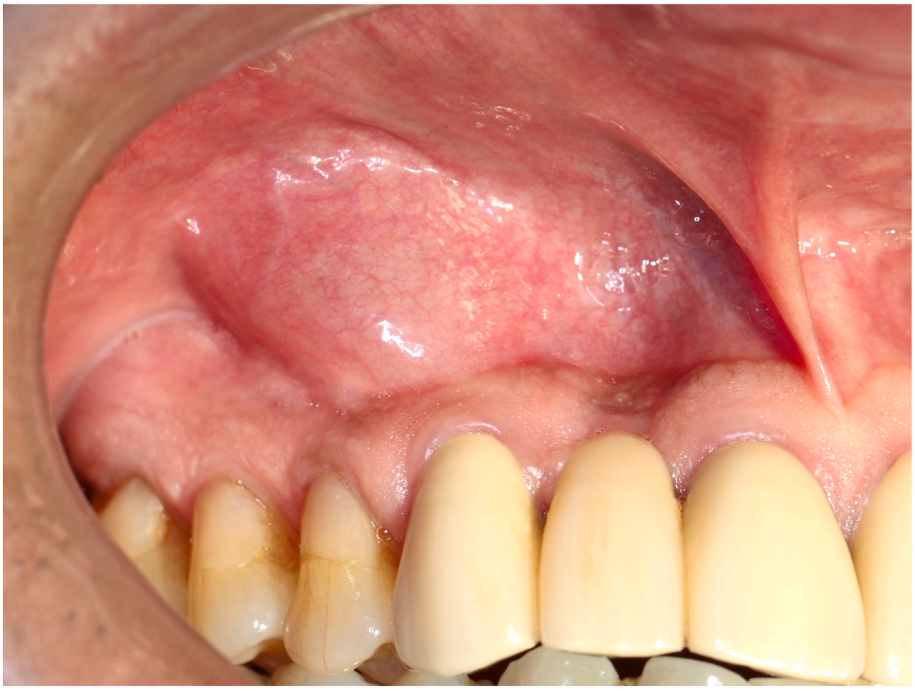

Fig. 1. Intraoral photograph at first visit. The mass was observed in the upper right canine-premolar area of the patient, a 74-year-old Japanese male.

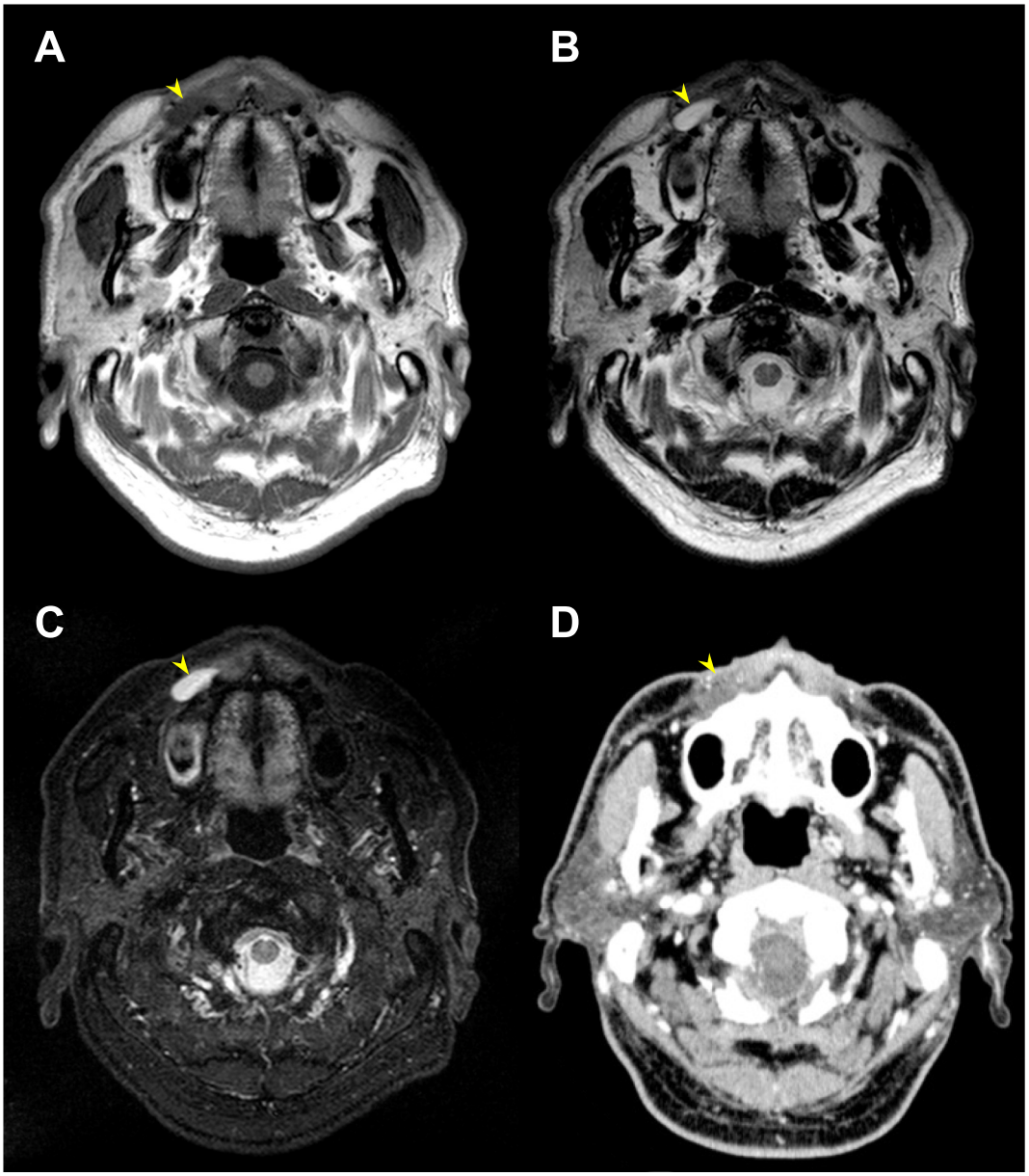

Fig. 2. Pre-operative MRI and CECT. A–C: Axial views on MRI. **A:** T1-weighted image (T1WI). **B:** T2-weighted image (T2WI) and short-TI inversion recovery (STIR). **D:** Axial view on CECT. *Arrowhead:* The mass lesion.

treatments and prognoses of these tumors differ, and it is therefore important to differentiate between them histologically. For this reason, we treated the present lesion with surgical excision with safety margins.

Acinic cell carcinoma is a rare tumor, and it usually involves the parotid gland and sometimes the minor salivary glands. Cho et al. reported a case of acinic cell carcinoma that presented as an asymptomatic mucocele-like mass on an unusual site, the lower lip; after a clinical and histopathological diagnosis of acinic cell carcinoma, they performed surgical excision with a safety margin [6]. Ishikawa et al. also reported a case of acinic cell carcinoma of the upper lip: the preoperative diagnosis was benign tumor or cyst, and the mass was thus surgically excised along the border with the normal tissues [7]. A case of mucoepidermoid carcinoma in which the clinical and radiographic investigations were suggestive of a mucocele (ranula) was described by Melo et al. [8] The treatment of choice was the complete excision of the mucocele and left submandibular salivary gland.

In our patient's case, mucocele was not our provisional diagnosis because of the clinical picture and the unusual presentation of the case. Mucoceles are known by their common occurrence on the lower lip (81.9%), followed by the floor of the mouth (5.8%) and the ventral surface of the tongue (5.0%) [9]. The most common size is 1 cm (40%) followed by 0.5 cm (34.3%) [5]. It was reported that 75% of the lesions are <1 cm [10]. Mucous extravasation cysts affect both genders in all age groups, with the peak incidence between the ages of 10 and 29 years [4]. We thus consider our patient's case unique for the above-mentioned reasons.

Mucoceles sometimes undergo spontaneous resolution and a subsequent accumulation of mucous. The lesions that do not undergo spontaneous resolution are those in which constant trauma persists. The spontaneous healing of a mucocele may be related to the degeneration of the adjacent acini because of the enzymes' action or the action of macrophages [11]. One of the interesting points about our patient's case is that it did not undergo spontaneous resolution, although the patient did not report any type of trauma.

We conducted a literature review concerning the uncommon presentation of mucoceles and identified a few reports (Table 1). In a case

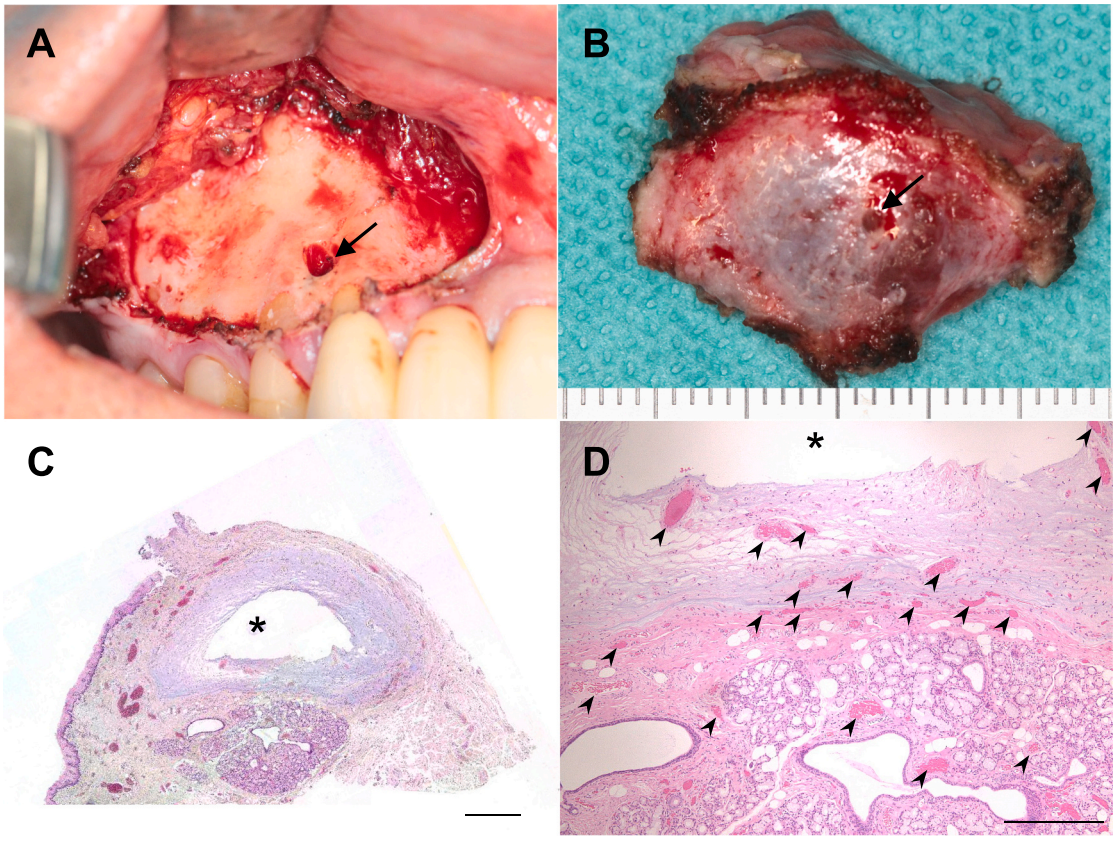

Fig. 3. Perioperative photo and histology of the lesion. **A:** After the resection, showing the bone surface. *Arrow:* Alveolar foramen of the feeding vessel that invaded the tumor. **B:** The resected specimen on the bone side. *Arrow:* Resected feeding vessel. **C,D:** Pathological findings of the resected tumor. *Cystic cavity. *Arrowheads:* blood vessel. Bars: 1 mm (C) and 500 μm (D).

Table 1
Characteristics of nine cases with a differential diagnosis between mucocele and tumor.

Author	Year	Age, yrs	Gender	Site	Size, mm	Provisional diagnosis	Final diagnosis
Cho et al. [6]	2005	64	F	Lower lip	10 × 10	Mucocele	Acinic cell carcinoma
Ishikawa et al. [7]	2016	31	M	Upper lip	12 × 10 × 10	Benign tumor or cyst	Acinic cell carcinoma
Melo et al. [8]	2018	49	F	Floor of the mouth	34 × 29 × 29	Mucocele	Mucoepidermoid carcinoma
Seo et al. [12]	2012	43	M	Buccal mucosa	35	Pleomorphic adenoma or lipoma	Mucocele
H. Lee [14]	2009	71	F	Upper lip	3 × 3	Mucocele	Adenocarcinoma
S. Gudi [15]	2013	21	F	Lower lip	10 × 10	Mucocele	Schwannoma
JL. Cunha [16]	2018	21	F	Lower lip	150	Mucocele	Neuroma
B. Menezes [17]	2020	7	M	Lower lip	80	Mucocele	Schwannoma
Present Case	2021	74	M	Maxillary gingiva	8 × 18	Myxoid tumor	Mucocele

report by Mustapha et al., a mucocele in the upper lip was observed [2]. Our patient's mucocele appeared at the junction between the buccal mucosa and the free gingiva, which is rare because of the absence of minor salivary glands in this region. Seo et al. reported a rare case of a 35-mm-dia. mucocele on the buccal mucosa of a 43-year-old male [12]. A rare case of bilateral mucoceles in a 40-day-old infant was described by Abdelsalam et al. [13] The present patient was a 74-year-old male. Two different cases that were clinically diagnosed as mucoceles were reported by Lee et al., but the histological examination revealed oral cysticercosis and minor salivary gland adenocarcinoma [14].

.udi et al. explained a case of schwannoma in the lower lip and titled their report "Swelling on lower lip…not always a mucocele." [15] On the other hand, a neuroma on the lower lip that clinically looked like a mucocele was reported by Cunha et al. [16] In 2020, Menezes et al. observed a schwannoma that simulated a mucocele in a 7-year-old child [17]. The characteristics the above-cited reports of different diagnoses at both preoperative and postoperative stages are summarized in Table 1.

The present case has been reported in line with the SCARE 2018 criteria [18].

4. Conclusion

It is crucial to emphasize that some non-neoplastic salivary gland diseases pose a challenge to clinicians regarding both diagnosis and treatment due to the uncommon presentation. If a diagnosis is difficult despite a thorough detailed medical history, diagnostic imaging and clinicopathological correlation, it is important to proceed carefully while keeping in mind the possibility of a tumor.

Authors' contributions

All authors contributed equally to the data collection, data analysis or interpretation, and writing of this paper.

Source of funding

None to declare.

Ethical approval

Ethical approval was exempted by our institution.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request. Identifiable patient information has been removed.

Registration of research studies

NA.

Guarantor

Tsuyoshi Shimo.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Declaration of competing interest

The authors report no conflicts of interest regarding this case report.

Acknowledgements

We thank all of the clinicians and staff of our institute for the care of the patient.

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