

Oral Focal Mucinosi s Occurring in the Palate: A Case Report

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Abstract

Oral focal mucinosis (OFM) is a rare disease characterized by the localized accumulation of mucin within the oral connective tissue, which is thought to result from excessive production of hyaluronic acid by fibroblasts. Because its clinical features are non-specific, a preoperative diagnosis is often difficult. Herein, we report a case of OFM arising in the hard palate. A 38-year-old man was referred to our department with an asymptomatic, elastic, firm mass measuring 15 × 15 mm in the left hard palate. Computed tomography showed no evidence of bone resorption, whereas magnetic resonance imaging demonstrated a well-defined, contrast-enhancing lesion with low signal intensity on T1-weighted images and high signal intensity on T2-weighted images. A benign salivary gland tumor was clinically suspected, and an incisional biopsy was performed. Histopathological examination revealed a well-demarcated, non-encapsulated myxoid lesion composed of fibroblasts in an Alcian blue-positive myxoid matrix. Immunohistochemical staining was negative for S-100 protein, CD34, and α-SMA, confirming the diagnosis of OFM. The lesion was completely excised under general anesthesia, and no recurrence was observed during a 24-month follow-up period.

Categories: Pathology, Dentistry

Keywords: hyaluronic acid, immunohistochemistry, mucin, oral focal mucinosis, palate

Introduction

Oral focal mucinosis (OFM) is a rare disease characterized by a localized accumulation of mucin [1]. Although the etiology of OFM remains unclear, it is thought to result from excessive production of hyaluronic acid by fibroblasts [1]. Clinically, OFM usually presents as a painless, firm mass that is similar in color to the surrounding mucosa [2]. Because OFM is rare and lacks characteristic clinical features, it is often misdiagnosed as other mucosal lesions [2]. Histopathologically, OFM is characterized by well-demarcated myxoid tissue without a capsule, surrounded by dense, normal collagenous connective tissue [3]. OFM most commonly occurs in females in their fourth and fifth decades of life [3,4]. The gingiva is the most common site of occurrence, followed by the palate [3,4]. In Japan, only two cases of palatal OFM have previously been reported, excluding the present case [5,6]. To identify these cases, we searched Ichushi-Web, J-STAGE, and PubMed in August 2026 using the search term “oral focal mucinosis,” without restricting the publication start date.

Here, we report a case of OFM occurring in the hard palate of a 38-year-old man. The lesion showed contrast enhancement on magnetic resonance imaging (MRI) and was clinically suspected of being a benign salivary gland tumor. Histopathological examination following an incisional biopsy established the diagnosis of OFM. This case highlights the diagnostic challenge of palatal OFM, which may mimic a benign salivary gland tumor on clinical and imaging assessment, and emphasizes the importance of histopathological examination for definitive diagnosis.

Case Presentation

A 38-year-old man was referred to our department because of a mass in the left hard palate. He had no symptoms associated with the lesion and no significant medical history.

Clinical examination revealed a 15 × 15 mm mass in the left hard palate. The mass was elastic and firm, and the overlying mucosa appeared normal (Figure 1).

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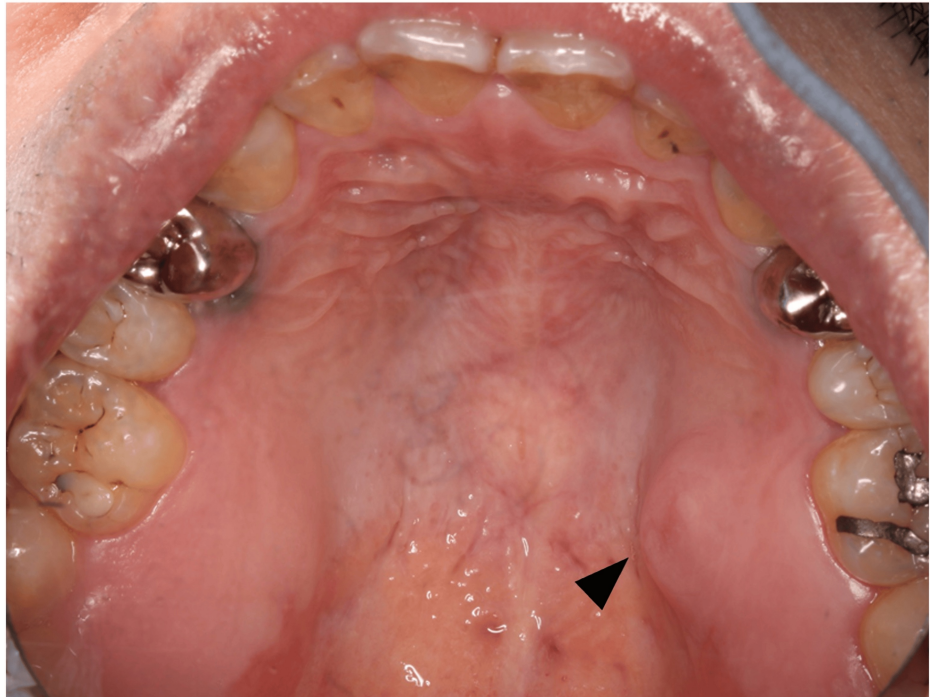


FIGURE 1: Intraoral photograph at the initial presentation.

A well-defined mass is visible in the left palate (arrowhead).

Although computed tomography (CT) revealed no definite soft tissue lesion or bone resorption (Figures 2A, 2B), MRI demonstrated a well-defined lesion with low signal intensity on T1-weighted images (Figure 3A), high signal intensity on T2-weighted images (Figure 3B), and contrast enhancement (Figure 3C).

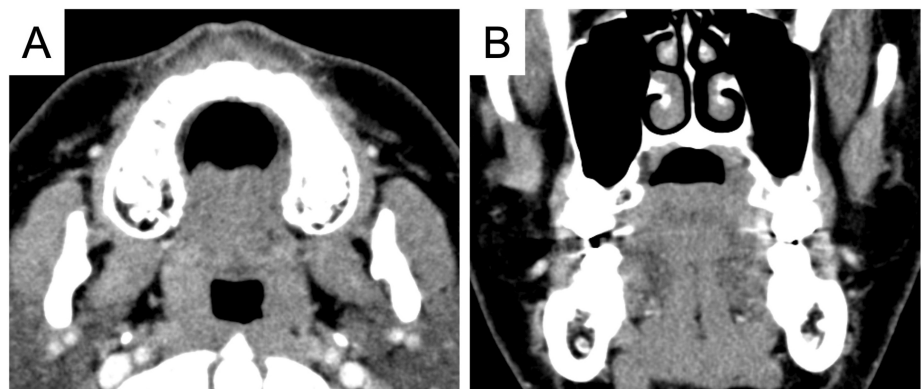


FIGURE 2: CT images of the left palate (A, axial image; B, coronal image).

No definite soft tissue lesion or bone resorption is observed.

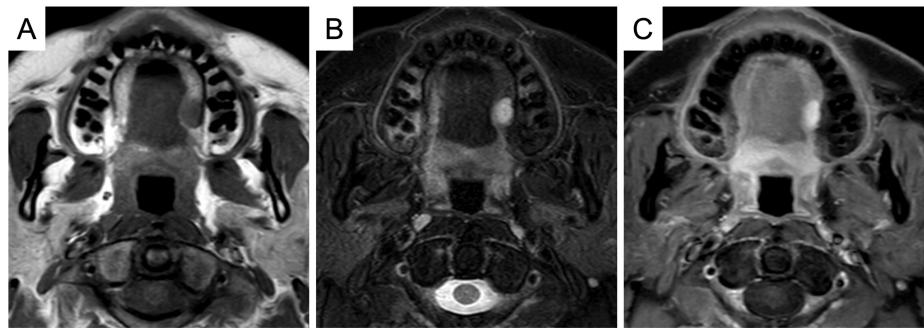


FIGURE 3: Magnetic resonance (MR) images of the left palate.

A well-defined lesion is observed, showing (A) low signal intensity on T1-weighted images, (B) high signal intensity on T2-weighted images, and (C) contrast enhancement.

Because a benign tumor, such as a pleomorphic adenoma, was suspected, a biopsy was performed. Histopathological examination revealed a well-demarcated but non-encapsulated lesion characterized by myxoid connective tissue containing fibroblasts (Figures 4A, 4B). No epithelial or myoepithelial components or ductal structures were identified. The myxoid matrix was positive for Alcian blue staining (Figure 4C). Immunohistochemical staining was negative for S-100 protein (Figure 4D), CD34 (Figure 4E), and α -SMA (Figure 4F).

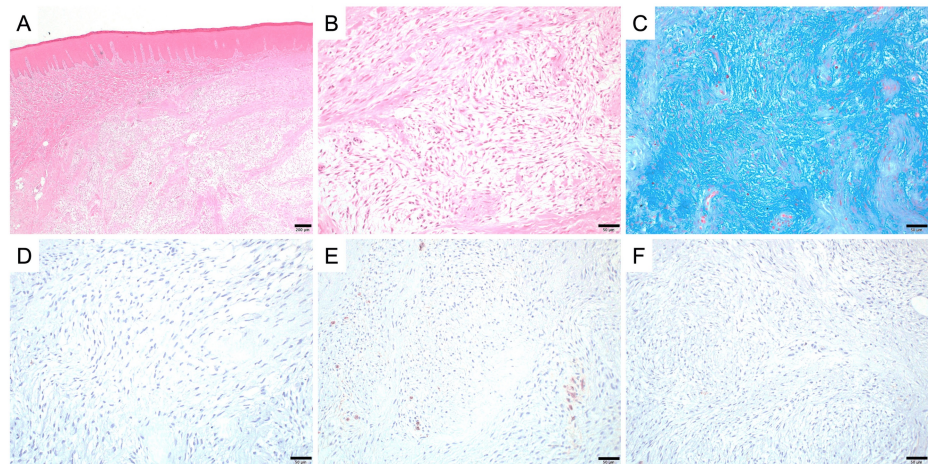


FIGURE 4: Histopathological findings.

(A) Myxoid stroma is observed in the subepithelial region (H-E staining; scale bar = 200 μ m); (B) proliferation of fibroblasts within the stroma (H-E staining; scale bar = 50 μ m). No capsule is identified.

(C) The myxoid stroma is positive for Alcian blue staining (scale bar = 50 μ m).

Immunohistochemical analysis shows that the fibroblasts are negative for (D) S-100 protein, (E) CD34, and (F) α -SMA (scale bar = 50 μ m).

Based on these findings, a diagnosis of OFM was made, and the lesion was excised under general anesthesia (Figure 5A). Complete removal of the lesion was confirmed clinically, and histopathological examination of the excision specimen confirmed the diagnosis of OFM and clear surgical margins. The cut surface of the resected specimen was homogeneously white and solid (Figure 5B). The wound was covered with a collagen-based artificial dermis graft. Complete epithelialization was achieved approximately three weeks postoperatively, and no recurrence was observed during the 24-month follow-up period.

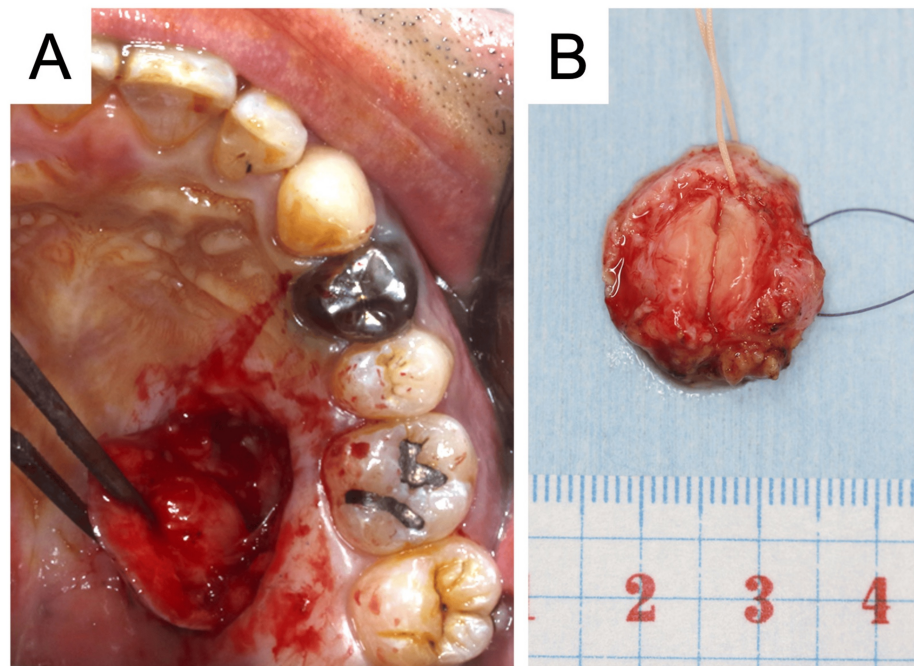


FIGURE 5: Intraoperative photograph and photograph of the resected specimen.

(A) The lesion is well demarcated from the surrounding tissue.

(B) The cut surface is homogeneously white and solid.

Discussion

OFM is considered to be the oral counterpart of cutaneous focal mucinosis and is characterized by the localized accumulation of mucin within the connective tissue [1,7]. It was first reported by Tomich et al. in 1974 [7]. Although cases have subsequently been reported from various countries, a recent systematic review identified only 113 reported cases [3], indicating that OFM remains relatively rare. According to this review, the gingiva is the most common site of occurrence (52%), followed by the palate (17%) and alveolar ridge (10%). In that review, five cases were reported from Japan [3]. To further examine cases reported in Japan, we conducted an independent literature search of Ichushi-Web, J-STAGE, and PubMed in August 2026 using the search term “oral focal mucinosis” without restricting the publication start date. Based on this process, we identified 15 cases, including the present case, to the best of our knowledge (Table 1) [5,6,8-15]. The five cases reported in the systematic review and the 15 cases identified through our literature search represent counts obtained using different review approaches and should therefore be interpreted in the context of their respective search methods. Of the 15 cases, eight occurred in the gingiva, whereas only three occurred on the palate. The male-to-female ratio was 3:2, indicating a slight male predominance, and the mean age was 53 years. These findings differed slightly from those of the systematic review [3]. Although there may be differences among populations, the number of reported cases remains small, and further accumulation of cases is needed to clarify the clinicopathological characteristics of OFM. Because there are no specific clinical findings for OFM, some lesions have been completely excised without prior biopsy because they were clinically diagnosed as fibromas or other benign tumors. In the present case, a benign salivary gland tumor, such as pleomorphic adenoma, was clinically suspected; therefore, an incisional biopsy was performed, which led to the diagnosis of OFM.

Case	Author	Year	Age	Sex	Site	Size (mm)	Clinical diagnosis	Treatment	Recurrence
1	Saito et al. [8]	1985	35	M	Gingiva	Unknown	Fibroma	Excision	None
2	Saito et al. [8]	1985	50	F	Gingiva	Unknown	Fibroma	Excision	None
3	Suzuki et al. [9]	1993	70	M	Buccal mucosa	15 × 10	Fibroma	Excision	None
4	Ohta et al. [10]	2000	52	M	Gingiva	15 × 10	Epulis	Excision	Unknown
5	Ohta et al. [10]	2000	49	M	Gingiva	15 × 10, 5 × 5	Epulis	Excision	Unknown
6	Okamoto et al. [5]	2007	77	F	Palate	20	Benign tumor	Excision	None
7	Ibaragi et al. [11]	2012	1	F	Gingiva	12 × 12 × 6	Tumor	Excision	None
8	Higuchi et al. [12]	2016	67	M	Tongue	3	Fibroma	Excision	None
9	Suzuki et al. [13]	2017	75	M	Gingiva	30 × 25	Epulis	Excision	None
10	Higuchi et al. [14]	2019	26	M	Retromolar	17 × 12	Tumor	Excision	None
11	Higuchi et al. [14]	2019	60	F	Gingiva	7 × 6	Epulis	Excision	None
12	Higuchi et al. [14]	2019	47	F	Gingiva	10 × 10	Epulis	Excision	None
13	Akimori et al. [6]	2022	73	M	Palate	13 × 9	Benign tumor	Excision	None
14	Kobayashi et al. [15]	2024	71	F	Tongue	2 × 2	Fibroma	Excision	None
15	Present case	2026	38	M	Palate	15 × 15	Benign tumor	Excision	None

TABLE 1: Reported cases of oral focal mucinosis (OFM) in Japan.

Although the etiology of OFM remains unclear, mechanical stimulation has been suggested to induce fibroblasts to overproduce hyaluronic acid [1,2]. However, mechanical stimulation was unlikely to have contributed to the development of the lesion in the present case, and its etiology therefore remained unknown.

Histopathological examination is essential for the definitive diagnosis of OFM. OFM is characterized by well-demarcated, non-encapsulated submucosal accumulation of mucin and is thought to result from excessive production of hyaluronic acid by fibroblasts. The mucin in OFM stains positively with Alcian blue [2,3]. In the present case, the lesion showed these characteristic features, and no epithelial or myoepithelial components or ductal structures were identified in the examined tissue. Pleomorphic adenoma was considered in the clinical differential diagnosis because of the palatal location and imaging findings. Histologically, pleomorphic adenoma typically exhibits epithelial and myoepithelial components, often with ductal structures and variable myxoid or chondromyxoid stroma. The absence of these cellular components and structures in the present specimen, together with the characteristic morphology of OFM, supported the diagnosis of OFM rather than pleomorphic adenoma.

It is important to differentiate OFM from other myxoid lesions, including odontogenic myxoma, peripheral nerve sheath tumors, such as nerve sheath myxoma, and solitary fibrous tumor (SFT) [6,15]. Because these lesions are also typically positive for Alcian blue staining, additional special stains and immunohistochemical analyses are important for the differential diagnosis. Odontogenic myxoma, an intraosseous tumor with locally aggressive behavior, can usually be distinguished from OFM based on its morphological features and clinical or radiographic findings [7,14]. Reticulin staining is also useful, as odontogenic myxoma is reticulin-positive, whereas OFM is reticulin-negative [6,14]. S-100 protein is positive in peripheral nerve sheath tumors and can be used to distinguish these lesions from OFM, which is S-100-negative [6,14]. SFT, which is classified by the WHO as a tumor of intermediate malignant potential, is characterized by CD34-positive spindle cells and can therefore be distinguished from CD34-negative OFM [6,16]. However, these immunohistochemical markers are not entirely specific, and negative staining alone cannot exclude all lesions within these groups of diseases. Therefore, the diagnosis of OFM should be based on the combined assessment of its characteristic morphological features and immunohistochemical findings, rather than on any single stain.

To date, surgical excision has been the treatment of choice for all reported cases of OFM [3]. Only one recurrence, which was attributed to incomplete excision, has been reported, whereas all other cases have had favorable outcomes [3]. Although no recurrence has been observed in the present case, long-term follow-up is warranted.

Conclusions

Although OFM generally has a favorable prognosis, it is important to distinguish it from other lesions, particularly salivary gland tumors such as pleomorphic adenoma, that may present with similar clinical and radiological features. Careful histopathological evaluation is essential to establish an accurate diagnosis and guide appropriate management.

Additional Information

Author Contributions

All authors have reviewed the final version to be published and agreed to be accountable for all aspects of the work.

Concept and design: Sho Akashi, Katsuhiko Amano

Drafting of the manuscript: Sho Akashi

Acquisition, analysis, or interpretation of data: Kazuki Nakatsuji, Aiko Hyodo, Shuichi Ishida

Critical review of the manuscript for important intellectual content: Kazuki Nakatsuji, Aiko Hyodo, Katsuhiko Amano, Shuichi Ishida

Disclosures

Human subjects: Informed consent for treatment and open access publication was obtained or waived by all participants in this study. **Conflicts of interest:** In compliance with the ICMJE uniform disclosure form, all authors declare the following: **Payment/services info:** All authors have declared that no financial support was received from any organization for the submitted work. **Financial relationships:** All authors have declared that they have no financial relationships at present or within the previous three years with any organizations that might have an interest in the submitted work. **Other relationships:** All authors have declared that there are no other relationships or activities that could appear to have influenced the submitted work.

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Sho Akashi and Kazuki Nakatsuji contributed equally to the work and should be considered co-first authors.

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