Head and Neck Pathology
https://doi.org/10.1007/s12105-022-01447-4

CASE REPORTS

Rapidly Growing Superficial Angiomyxoma in Mandibular Gingiva: A Case Report and Literature Review

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Received: 8 January 2022 / Accepted: 15 March 2022

Abstract

Superficial angiomyxoma (SA) is a benign tumor characterized by extensive myxoid stroma, numerous small blood vessels, sparse spindle-shaped fibroblastic cells, and inflammatory cell infiltrate. Oral cavity SA is extremely rare and typically presents as a painless, slow growth. We experienced SA in the mandibular gingiva that is rapidly growing. The patient was a 15-year-old female whose chief complaint was a painless mass in the lingual gingiva of the mandible that increased in size over 1 month. An excisional biopsy was performed under local anesthesia. According to histopathological examination, the mass was diagnosed as SA. The patient experienced recurrence twice because of positive margins. The second recurrent lesion, including periosteum, was resected, and no recurrence has been observed for 1 year. The cause of rapid growth was attributed to edematous changes due to tongue habit or traumatic stimuli. As this case exhibited repeated local recurrence, careful follow-up is required.

Keywords Angiomyxoma · Gingiva · Blood vessels · Recurrence · Alcian blue · Immunohistochemistry

Introduction

Superficial angiomyxoma (SA) is a benign tumor characterized by extensive myxoid stroma, numerous small blood vessels, sparse spindle-shaped fibroblastic cells, and inflammatory cell infiltrate [1, 2]. SA is most often found in the trunk, head, neck, and lower extremities but has rarely been reported in the oral cavity. SA typically presents as a painless mass with slow growth. We experienced a case of rapidly growing SA that occurred in the lingual gingiva of the mandible.

Case Report

This patient is a 15-year-old female with no extraordinary personal or family medical history. She became aware of a mass in the right mandibular gingiva, which rapidly grew for 1 month, after which its size remained stable. She was referred to our hospital 2 months after she first noticed the mass. Nothing extraordinary was noted about her systemic and extraoral findings. During the initial examination, a mass that was 30 mm in diameter and reddish, elastic, soft, lobulated, and painless was found in the lingual gingiva between the right lower lateral incisor and canine (Fig. 1A, B). No marginal periodontitis requiring treatment was observed in these teeth, and panoramic radiography revealed no alveolar bone resorption around the mass. Magnetic resonance imaging (MRI) demonstrated low signal on T1-weighted images and a well-defined, lobulated, relatively homogeneous lesion with high signal on T2-weighted images (Fig. 2A, B). Contrast-enhanced computed tomography (CT) showed heterogeneous enhancement within the lesion, which suggested abundant blood flow (Fig. 3). The clinical diagnosis

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Published online: 09 April 2022

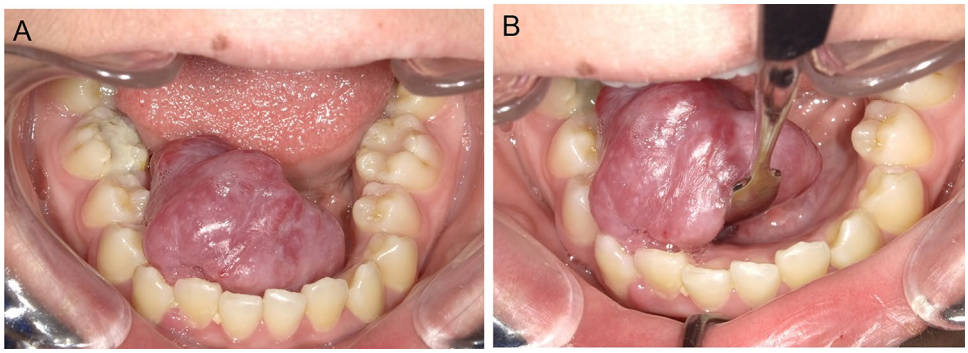

Fig. 1 **A** A reddish, elastic, soft, lobulated, painless mass 30 mm in diameter was found in the lingual gingiva of the mandible. **B** Its stalk was located between the right lower lateral incisor and canine

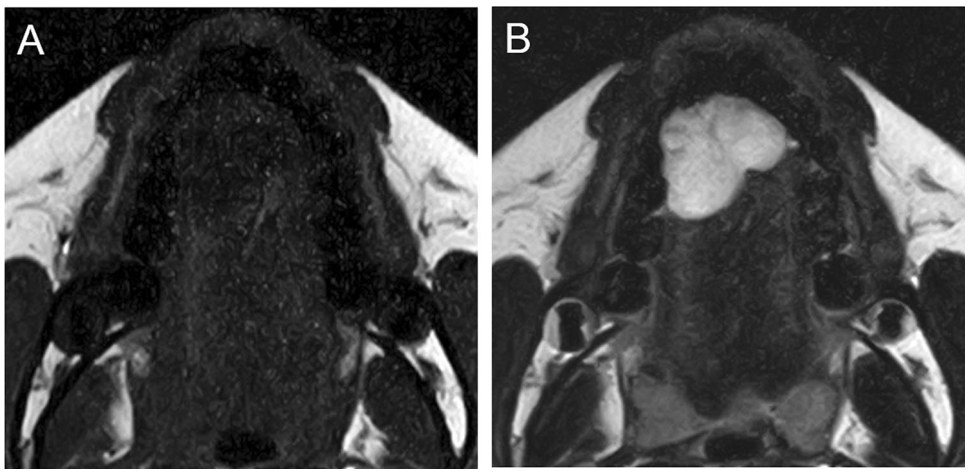

Fig. 2 **A** MRI demonstrated low signal on T1-weighted images. **B** A well-defined, lobulated, relatively homogeneous lesion with high signal on T2-weighted images

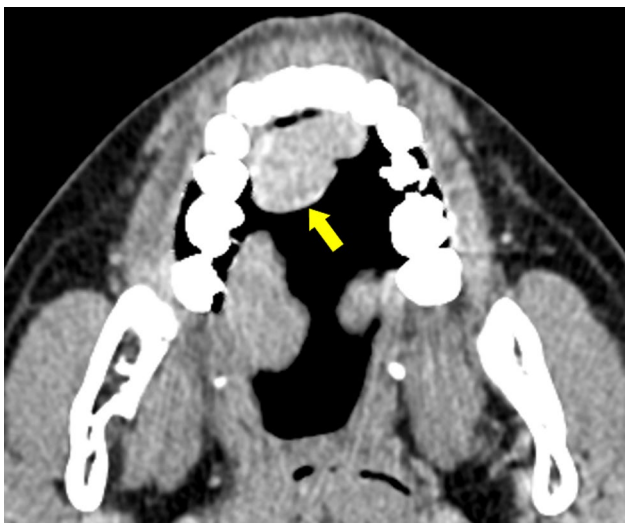

Fig. 3 Contrast-enhanced CT showed heterogeneous enhancement within the lesion, suggesting abundant blood flow

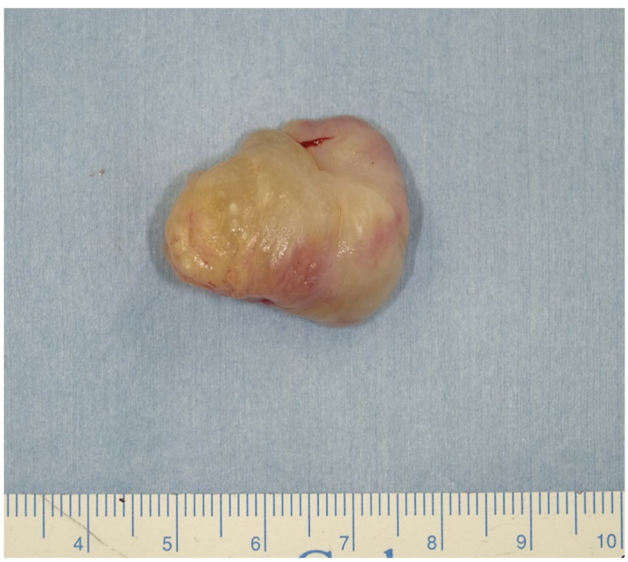

Fig. 4 The excised specimen was a slightly elastic, soft, nodular mass 27×18×23 mm in size with a smooth surface

was granulomatous epulis whose stalk was located in the lingual gingiva between the right lower lateral incisor and canine, and an excisional biopsy was performed under local anesthesia. The excision was performed on the marginal gingiva. The excised specimen was a slightly elastic, soft, nodular mass that was 27×18×23 mm in size with a smooth surface (Fig. 4). Hematoxylin and eosin (H&E) staining showed numerous slit-like blood vessels and fibrous tissue growth against a background of subepithelial myxoid stroma

(Fig. 5A, B). Its tumor margins were exposed at the base. Inflammatory cells, such as lymphocytes and neutrophils, had infiltrated around the blood vessels, and spindle-shaped tumor cells were sparsely distributed in that area (Fig. 5C). No atypia was observed in the tumor cells. The myxoid matrix in the background was positive for Alcian blue staining (Fig. 6). Immunohistochemistry revealed that the spindle-shaped tumor cells were negative for S-100, α-SMA, desmin, CD34, pan-keratin, GFAP, and EMA (Fig. 7A–G). Moreover, the MIB-1 index was less than 5%. Based on these findings, we diagnosed the mass as SA. Recurrence occurred twice at 5 and 9 months after the first excision. At the second recurrence, the tumor, as well as the labial and lingual marginal gingiva, attached gingiva, and interdental papillae, was subperiosteally resected (Fig. 8). Considering the patient's wishes, her youth, and healthy dentition, we did not extract the right lower lateral incisor and canine and did not remove the alveolar bone. As of 1 year, no third recurrence has been observed.

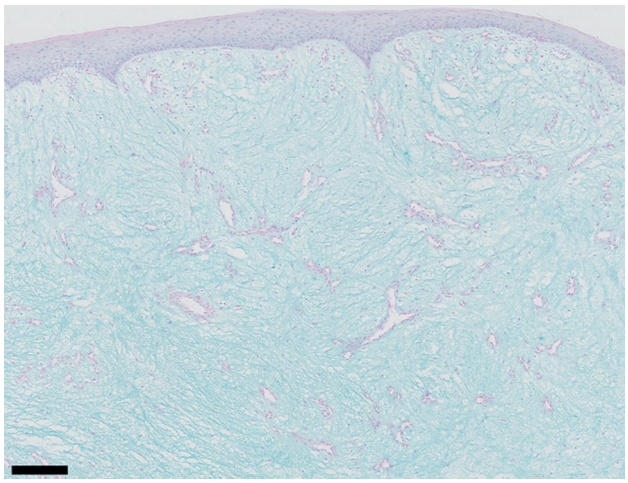

Fig. 6 Myxoid matrix in the background was positive for Alcian blue staining; scale bar 200 μm

Discussion

In this case, we experienced an atypical rapidly growing SA in the oral cavity. SA is a benign soft tissue tumor derived from fibroblasts, which was first proposed by Allen et al. in 1988 [1]. SA is most commonly found in middle-aged males, often on the skin of the trunk, but SA can also occur on the lower extremities, head, neck, and arms [1, 2]. The tumor size at initial examination ranges from 1 to 5 cm and is generally a small nodule or polypoid lesion with few subjective symptoms. SA typically presents with slow growth and often forms 3 months to 10 years before diagnosis [1, 2]. Eight cases of SA have been reported in the oral cavity [3–9] (Table). The reported occurrence sites are the buccal mucosa [3, 5], floor of the mouth [4, 6], upper posterior alveolar region [7], lower buccal vestibule [8], lower lip [9], and palate [9]. This is the first case of SA in the mandibular gingiva. As Table shows, previous

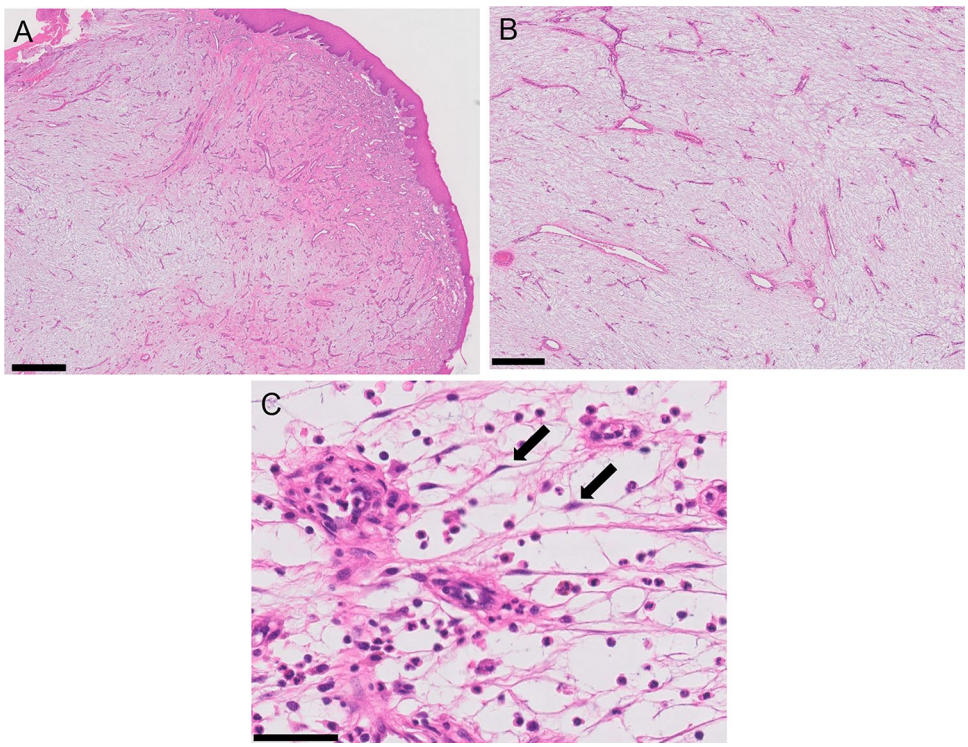

Fig. 5 H&E staining of the excised tissue. **A**, **B** Numerous slit-like blood vessels and fibrous tissue growth were observed against a background of subepithelial myxoid stroma. **A** Low magnification; scale bar: 1 mm. **B** High magnification; scale bar 500 μm. **C** Inflammatory cells, such as lymphocytes and neutrophils, had infiltrated the area around the blood vessels, and spindle-shaped tumor cells (arrow) were sparsely distributed; scale bar 50 μm

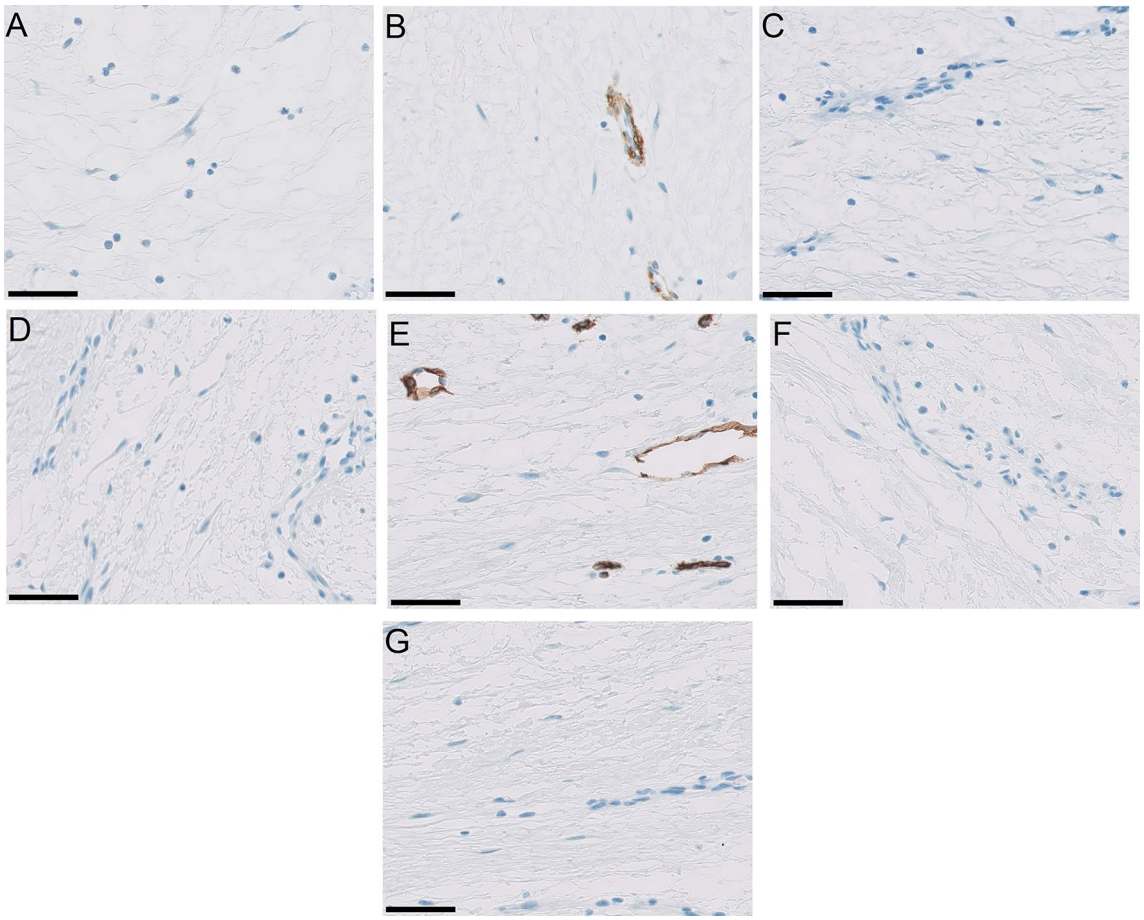

Fig. 7 Immunohistochemistry revealed that the spindle-shaped tumor cells were negative for **A** S-100, **B** α-SMA, **C** pan-keratin, **D** desmin, **E** CD34, **F** GFAP, and **G** EMA; scale bar 50 μm

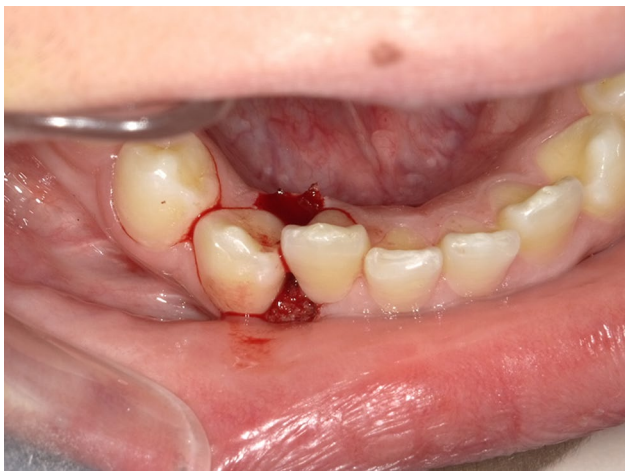

Fig. 8 At the second recurrence, the tumor, as well as the labial and lingual marginal gingiva, attached gingiva, and interdental papillae, was subperiosteally resected

reports of SA in the oral cavity also showed slow growth over several years, except for a case at 6 months of age [6]. In our case, the growth was rapid compared with that in previous cases, as the tumor increased in size within the first month. On gross examination, the tumor in our case was redder in color than those in previous reports, suggesting high blood flow, and a thin capsule. The pathology images demonstrated few cellular components, suggesting that tumor growth was associated with edematous changes rather than with cell proliferation. It has been reported that traumatic stimuli can be involved in the growth of soft tissue lesions [10]. Compared with SA on the skin, SA in the oral cavity, especially in the lingual gingiva of the mandible as in our case, is subject to external stimulation by tongue habit and brushing. This may have triggered inflammation and increased vascular permeability, which led to edema. In addition, the small size of the stalk relative to the tumor diameter could have also contributed to the edematous change.

Previous reports of SA in the oral cavity

References	Age	Gender	Site	Size (mm)	Growth period	Clinical diagnosis	Treatment	Recurrence
Chen et al. [3]	19 years	Male	Buccal mucosa	50×35×30	2 years	Benign mesenchymal soft tissue tumor	Excised	No
Gardner [4]	69 years	Female	Floor of the mouth	10×12×12	3 years	Lipoma	Excised	No
Meer and Beavon [5]	37 years	Female	Buccal mucosa	45×32×20	2 years	Lipoma/other myxoid benign soft tissue neoplasm	Excised	No
Mokhtar et al. [6]	6 months	Male	Floor of the mouth	50×36×26	1 month	Soft tissue tumor/cyst	Excised	No
Ravindra et al. [7]	30 years	Male	Upper posterior alveolar region	30×30	2 years	Soft tissue tumor	Incisional biopsy	–
Anehosur et al. [8]	32 years	Male	Lower buccal vestibule	60×40	3 years	Mucous retention cyst	Excised	–
Singhota et al. [9]	47 years	Female	Lower lip	10×10	14 months	Mucocele	Excised	No
	58 years	Male	Palate	40	-	Cyst	Excised and alveolectomy	No
Present case	15 years	Female	Mandibular gingiva	27×18×23	2 months	Epulis	Excised	Twice

This table is based on Anehosur et al. [8] with some changes and additions

This case was diagnosed as granulomatous epulis before excision. Differentiating epulis from SA in a gingival mass in the absence of alveolar bone resorption is clinically difficult. Histopathological analysis, including immunohistochemistry, is essential to confirm the diagnosis. Histopathologically, SA is characterized by a focally lobular or multinodular growth pattern; extensive myxoid changes; numerous small blood vessels; inflammatory cell infiltration, such as lymphocytes and neutrophils; and spindle-shaped cells without nuclear atypia [2]. Stromal mucin is positive with Alcian blue staining. A diagnosis of SA is often difficult because it exhibits no specific immunohistochemical staining pattern. Herein, the tumor cells were determined to be negative for S-100, α-SMA, pan-keratin [2], desmin, CD34 [11], GFAP, and EMA [12]. However, S-100, α-SMA, CD34 were also found to be positive [13]. These results provide clues for differentiation of SA from soft tissue myxoma, angiomyolipoma, myxoid nerve sheath tumor (neurothekeoma), myxoid neurofibroma, myxofibroma, odontogenic myxoma, oral focal mucinosis, and aggressive angiomyxoma, AA [7]. One study reported oral focal mucinosis with a clinical diagnosis of epulis in a lesion similar to the one in our case [14]. AA is a locally invasive tumor with a high recurrence rate and most commonly found in the pelvis and perineum of women [15]. Histopathologically, the sparse growth of spindle-shaped or stellate cells and the myxoid matrix in the stroma are similar to those of SA, but large, thick-walled blood vessels are observed in AA [15]. Immunohistochemistry determined that tumor cells of AA are positive for desmin, unlike SA [2]. Only one case of AA in the oral cavity has been reported thus far [16].

In our case, local recurrence occurred twice. It has been reported that 20–40% of SA recurs after incomplete resection [2, 17]. The periosteum was not resected during the first and second resections, and the tumor was exposed at the base, suggesting that the recurrence was caused by incomplete resection. During the third resection, the periosteum was included in the resection, and the tumor has not recurred a third time.

Although SA in the oral cavity is rare, it may rapidly grow and recur, as in our case. Therefore, careful follow-up is necessary even after excision. If recurrence occurs in the future, tooth extraction or alveolar bone removal should be considered.

Conclusion

We experienced a SA in the lingual gingiva of the mandible. Because of its rapid growth and recurrence, careful follow-up is required.

Acknowledgements We thank Enago (www.enago.jp) for editing a draft of this manuscript. This study received no funding, financial relationships from any sources.

Author Contributions SM performed excisional biopsy. MM, MT, TS, and SM performed follow-up observations. TK performed pathologic diagnosis. MM and SM drafted the manuscript. All authors approved the final version of the manuscript.

Funding None.

Data Availability Not applicable.

Code Availability Not applicable.

Declarations

Conflict of interest None.

Ethical Approval Not applicable.

Consent to Publication The patient and their family have consented to the submission of the case report to the journal.

Informed Consent Written informed consent was obtained from the patient and their family.

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