

Solitary myofibroma of the oral cavity; a rare case report



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Abstract

Myofibroma is an uncommon benign neoplasm of myofibroblastic origin that primarily affects children and adolescents. While it most frequently arises in the skin and subcutaneous tissues, its occurrence in the oral cavity, particularly in the buccal soft tissue is exceptionally rare. Owing to its clinical similarity to other benign spindle-cell lesions, such as irritation fibroma and neural tumors, establishing an accurate diagnosis can be challenging. This report describes a rare case of solitary oral myofibroma presenting as a buccal soft tissue swelling of the left mandible. The clinical, radiographic, histopathologic, and immunohistochemical features are presented, together with a discussion on differential diagnosis, management, and prognosis.

Introduction

Myofibroma is a rare, benign mesenchymal neoplasm of myofibroblastic origin that most commonly develops during infancy or early childhood, typically before the age of two years (1). The term myofibroma was first introduced by Smith et al in 1989 to describe a solitary lesion, whereas myofibromatosis refers to multiple lesions; both are currently recognized by the World Health Organization (2). These tumors predominantly affect the head and neck region (approximately 36%) and the trunk, while intraoral involvement is relatively uncommon (3). Within the oral cavity, solitary myofibromas are especially rare in the buccal soft tissue. Clinically, they usually present as firm, asymptomatic swellings that can closely mimic other benign spindle-cell lesions, such as fibroma, neurofibroma, leiomyoma, or lipoma (1).

Given their nonspecific clinical presentation, accurate diagnosis can be difficult. Advanced imaging modalities, particularly cone-beam computed tomography (CBCT), are valuable in assessing the extent of the lesion and its relationship to adjacent anatomical structures (4). Nonetheless, histopathologic evaluation combined with immunohistochemistry remains essential for confirming myofibroblastic differentiation

(1). Awareness of this rare entity is crucial to ensure timely diagnosis and appropriate surgical management, as complete excision typically results in excellent functional and esthetic outcomes with minimal risk of recurrence.

Case Report

A 15-year-old male presented to the department of oral and maxillofacial surgery with a firm, asymptomatic swelling in the left buccal region of the mandible, which had been present for approximately two months. Clinical examination performed by the attending oral and maxillofacial surgeon revealed a well-circumscribed, non-tender, firm mass in the buccal vestibule adjacent to the left premolar–molar region. The overlying mucosa appeared intact, with normal color and texture, and there were no signs of ulceration or secondary infection. The patient reported no pain, paresthesia, or mobility of adjacent teeth.

To evaluate the lesion's size, extent, and relationship to surrounding structures, CBCT was performed using a Giano HR unit (NewTom, Italy) with a mandibular field of view. Multiplanar reconstructions (axial, coronal, sagittal, and cross-sectional views) were analyzed with NNT Viewer software. Imaging revealed a localized, well-defined, homogeneous soft tissue

Key point

Myofibroma is a rare benign tumor of myofibroblastic origin, seldom found in the oral cavity. This case describes a solitary buccal soft tissue myofibroma in an adolescent, confirmed by histopathology and immunohistochemistry (IHC). Complete surgical excision with nerve preservation led to uneventful healing and no recurrence after six months.

prominence extending from the buccal vestibule toward the outer cortical plate, without evidence of cortical erosion, thinning, periosteal reaction, or displacement of adjacent teeth (Figure 1). The lesion was located close to the terminal branches of the mental nerve, which appeared preserved radiographically.

Under local anesthesia, complete surgical excision of the lesion was performed with meticulous preservation of adjacent neurovascular structures, particularly the terminal branches of the mental nerve (Figure 2). The surgical site was closed using 3-0 absorbable Vicryl sutures to achieve primary healing. Postoperatively, the patient was prescribed amoxicillin for antibiotic prophylaxis, along with nonsteroidal anti-inflammatory agents (ibuprofen and ketorolac) and dexamethasone to control pain, inflammation, and swelling. The postoperative course was uneventful, and the patient demonstrated satisfactory healing without neurological deficits. The excised specimen consisted of a firm, well-circumscribed nodular mass, tan to creamy in color, measuring 1.5×1×1 cm. On cut section, the lesion appeared white, homogeneous, and solid, with no evidence of hemorrhage or necrosis (Figure 3). Microscopically, the sections showed a well-demarcated nodular lesion exhibiting a characteristic biphasic growth pattern. The central area was composed of immature, plump, spindle-shaped cells arranged concentrically around branching, hemangiopericytoma-like blood vessels. In contrast, the peripheral region displayed fascicles and nodules of more differentiated,

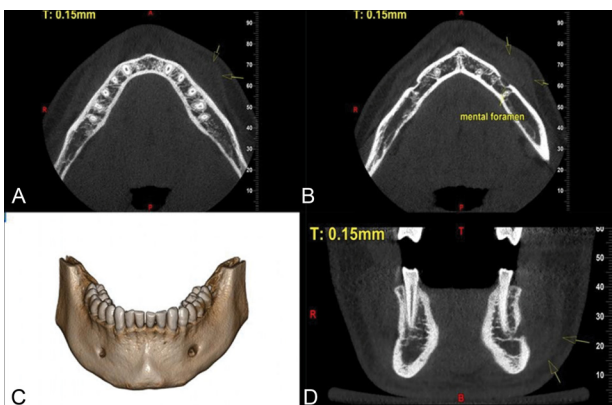


Figure 1. Cone-beam computed tomography demonstrates a well-defined, homogeneous soft tissue mass in the left buccal vestibule adjacent to the premolar–molar region of the mandible (A, B, D). three-dimensional reconstruction, highlighting the intact outer cortical boundaries and confirming the absence of bone involvement (C).

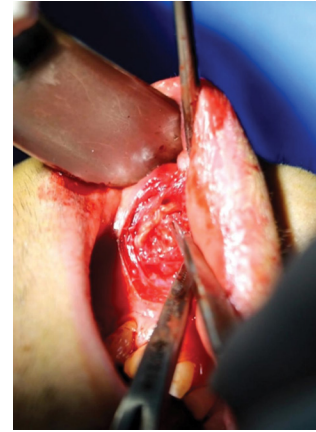


Figure 2. Intraoperative view of the excised lesion showing complete removal with careful preservation of the terminal branches of the mental nerve.

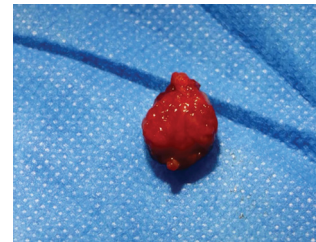


Figure 3. Excised lesion, well-circumscribed and firm. Cut surface is homogeneous and solid, with no hemorrhage or necrosis.

myoid-appearing cells with areas of variable hyalinization. Scattered mitotic figures were identified; however, necrosis and calcification were absent (Figure 4).

Histopathologic examination confirmed these findings, showing spindle-shaped myofibroblastic cells arranged in fascicular and whorled patterns within a collagen-rich stroma, consistent with myofibroma. Immunohistochemical analysis supported the diagnosis, showing strong cytoplasmic positivity for α -smooth muscle actin (α -SMA) with negative staining for desmin, S-100 (neural marker), and cytokeratin (epithelial marker). The Ki-67 proliferative index was low (approximately 1–2%), indicating limited mitotic activity (Figure 5).

The postoperative recovery was uneventful, with preserved neurosensory function in the distribution of the mental nerve and satisfactory mucosal healing at the surgical site. At the six-month follow-up, no clinical or radiographic evidence of recurrence was observed, confirming favorable short-term outcomes. However, given the potential for late recurrence in myofibromas, long-term clinical and radiographic monitoring was advised.

Discussion

Myofibroma is a rare benign mesenchymal neoplasm that primarily affects infants and young children, with

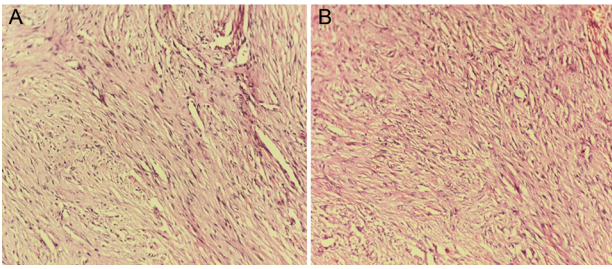


Figure 4. Microscopic description (A, B): Sections reveal a well-circumscribed nodular lesion exhibiting a characteristic biphasic growth pattern. The central area is composed of immature, plump spindle-shaped tumor cells arranged around numerous thin-walled, branching (“hemangiopericytoma-like”) blood vessels. The peripheral portion shows nodules and fascicles of variably hyalinized, myoid-appearing cells. Occasional mitotic figures are observed, while necrosis and calcification are absent (original magnification $\times 200$).

oral cavity involvement being particularly uncommon. Among intraoral sites, the buccal soft tissue is an especially rare location, with only a limited number of cases documented in the literature. This rarity contributes to significant diagnostic challenges, as the lesion may clinically mimic other spindle-cell neoplasms such as fibroma, neurofibroma, leiomyoma, or even lipoma (1, 2). In the present case, the integration of clinical examination, CBCT, and immunohistochemistry (IHC) proved essential for achieving an accurate diagnosis and guiding appropriate surgical management.

The patient presented with a solitary lesion, which is the most frequent clinical manifestation of myofibroma (5). The relatively rapid enlargement observed within two months was consistent with the typical biological behavior of this tumor. The absence of a relevant family history suggested a sporadic presentation.

Due to the tumor’s rarity and its histopathologic overlap with other benign mesenchymal lesions, the diagnosis of oral myofibroma is particularly challenging. Histopathological analysis in combination with IHC is considered indispensable (6). The principal histological differentials include leiomyoma, fibromatosis, myofibromatosis, and low-grade sarcomas (7). The IHC profile—characterized by strong positivity for smooth muscle actin (SMA) and negativity for desmin, S-100, and cytokeratin—is highly specific for myofibroma and was confirmed in our case. Furthermore, the identification of the characteristic biphasic pattern and a low Ki-67 proliferative index was critical to distinguish this benign entity from malignant spindle-cell tumors, thereby avoiding unnecessary overtreatment.

For solitary intraoral lesions, incisional biopsy is generally recommended to establish a diagnosis, followed by complete surgical excision, which remains the treatment of choice (6). Local recurrence has been reported (5, 7), but it is most often attributed to anatomical constraints that limit complete excision, particularly near vital neurovascular structures (8). In rare circumstances where the tumor is unresectable or demonstrates repeated

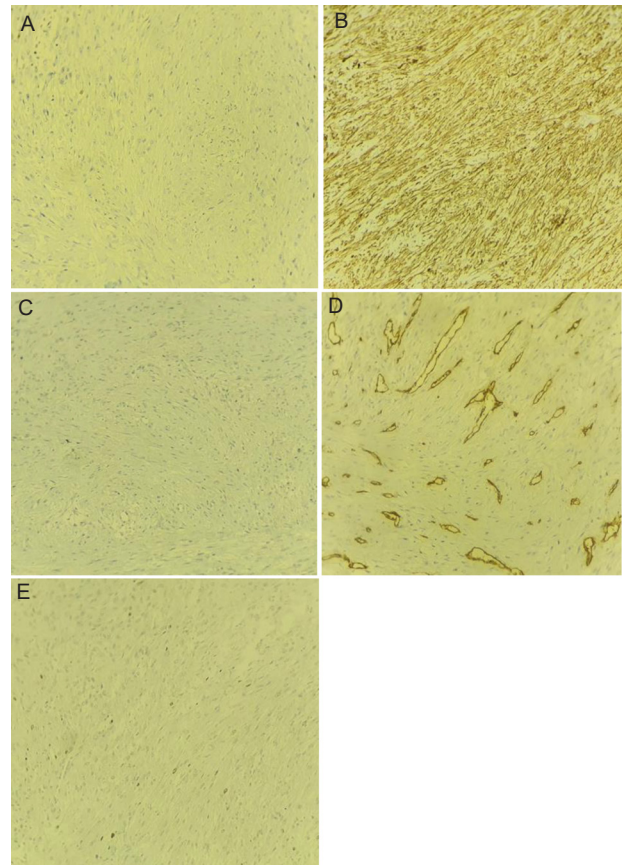


Figure 5. Immunohistochemical staining profile of the lesion: (A) Tumor cells negative for S-100 protein, excluding neural differentiation; (B) Strong cytoplasmic positivity for smooth muscle actin (SMA), confirming myofibroblastic nature; (C) Desmin negativity in tumor cells, ruling out smooth muscle origin; (D) CD34 highlights the vascular channels; (E) Ki-67 immunostaining shows low proliferative activity (approximately 1–2% of tumor cells), (original magnification $\times 200$).

recurrences, adjuvant modalities such as chemotherapy or radiotherapy may be considered, although these approaches are uncommon and not routinely required (9).

The prognosis of solitary myofibroma is excellent following adequate surgical removal. Conversely, multicentric visceral involvement tends to display a more aggressive clinical course and may even prove fatal (6, 7). Importantly, myofibroma demonstrates no evidence of malignant transformation or metastatic potential (10).

Conclusion

Solitary myofibroma of the oral cavity is an exceptionally rare benign tumor that can clinically mimic other spindle-cell lesions, posing diagnostic challenges. Accurate diagnosis requires a multidisciplinary approach integrating clinical assessment, advanced imaging such as CBCT, and histopathologic confirmation with IHC. Complete surgical excision with preservation of adjacent neurovascular structures remains the treatment of choice, providing excellent prognosis with minimal risk of recurrence. Although malignant transformation is exceedingly rare, long-term follow-up is recommended

to monitor for late recurrences. Greater awareness among clinicians and pathologists is essential for timely recognition and optimal management.

Authors' contribution

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Conflicts of interest

The authors declare that they have no competing interests.

Ethical issues

This case report was conducted in accordance with the World Medical Association Declaration of Helsinki. The patient provided written informed consent for publication of this case report. Ethical issues (including plagiarism, data fabrication, double publication) have been completely observed by the authors.

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