Myofibroma of the Oral Mucosa: A Case Report

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Key Words

Myofibromatosis • Myofibroma • Oral soft tissue • Irritation fibroma

Abstract

Objective: To report a case of coexisting irritation fibroma and myofibroma in oral mucosa. **Clinical Presentation and Intervention:** One case with two painless, nodular masses, adjacent to each other in the buccal mucosa, was clinically examined with a provisional diagnosis of irritation fibroma, salivary gland tumors, neurofibroma and schwannoma. Histological examination of the smaller swelling showed features of irritation fibroma, while the features of the other mass were compatible with myofibroma or leiomyoma. Additional immunohistochemical examination established the diagnosis of myofibroma. **Conclusion:** This was a case of a myofibroma that was clinically similar to an adjacent irritation fibroma, which highlights the possibility of misdiagnosis of a myofibroblastic tumor and underlines the importance of histologic examination together with immunohistochemical and/or histochemical analysis if necessary to establish the accurate diagnosis.

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Introduction

The terms myofibroma and myofibromatosis refer to myofibroblastic benign neoplasms that occur as solitary and multiple tumors, respectively [1]. The histopathologic features of both myofibroma and myofibromatosis are identical [2]. The most common site of myofibroma is in the head and neck region followed by the trunk, extremities and viscera [2]. On the other hand, this neoplasm is rare in the oral soft tissues [3]. The purpose of this article was to report a case of myofibroma of oral soft tissues located adjacent to an irritation fibroma.

Case Report

A 56-year-old female was referred to the Department of Oral Medicine/Pathology, School of Dentistry, Aristotle University, Thessaloniki, Greece, with two asymptomatic nodular masses in the right buccal mucosa of approximately 2 months' duration. Clinical examination showed two nodular lesions, one small: 1 × 0.5 cm, and the larger 1 × 1 cm. The lesions were well demarcated, sessile, nontender to palpation and covered by normal mucosa (fig. 1). No significant changes in size of the lesions were reported by the patient during the period of 2 months. The patient's medical and dental history was unremarkable. There was no history of similar lesions elsewhere in the patient's body or in her relatives.

Fig. 1. Nodular buccal masses representing a myofibroma (larger) and an irritation fibroma (smaller), respectively.

The initial clinical differential diagnosis included irritation fibroma, salivary gland tumors, neurofibroma and schwannoma. Under local anesthesia, excisional biopsies were performed.

Histological examination of the small swelling showed features of irritation fibroma, whereas that of the large swelling showed a poorly demarcated lesion of mesenchymal origin composed of short intersecting bundles and whorls of spindle and round cells (fig. 2a). The spindle cells had tapering nuclei and eosinophilic cytoplasm whereas the round cells had vesicular basophilic nuclei and small amounts of eosinophilic cytoplasm (fig. 2b). Many hemangiopericytoma-like blood vessels were present in central and peripheral regions of the lesion (fig. 2a, b). Mitotic figures were only rarely observed. Based on these findings a diagnosis of myofibroma or leiomyoma was suggested. Additional serial sections were stained for histochemistry and immunohistochemistry. Masson's trichrome stain revealed the presence of thick fibrous bundles with random, irregularly intersecting angles (fig. 3a). Furthermore, using the automated Envision/HRP immunohistochemical technique (DakoCytomation A/S, Glostrup, Denmark), as described previously [4], the neoplastic cells were found to be positive for vimentin, specific muscle actin, α -smooth muscle actin (α -SMA) and negative for desmin, S-100 and α_1 -antitrypsin (fig. 3b). The neoplastic cells were negative for CD34. Based on the combined findings of histopathology, histochemistry and immunohistochemistry, the diagnosis of a myofibroma of oral soft tissues was made. After 10 months' follow-up, no evidence of recurrence was observed.

Discussion

Myofibroma of oral soft tissues is rare. A review from 1960 to 2007 described the clinicopathological features of 44 cases [3]. Since then, 6 additional cases have been re-

ported [5–8]. Data analysis of 51 cases, including our case, shows that myofibroma of oral soft tissue may arise at any age (birth to 70 years), but mainly in the first and second decades of life (birth to 20 years: 57%, adults ≥ 20 years: 42%). However, in our case it occurred during the 5th decade. Overall, the lesion shows a slight predilection for women (female:male ratio 1.6:1) as in this case except for the child/teenager subgroup (11 months up to 20 years) with a reverse male:female ratio 1.8:1. The tongue is most commonly involved (30%) followed by buccal mucosa (20%) as in this case. Clinically, myofibroma of oral soft tissues appears as a slow-growing painless, firm, and rubbery swelling covered by normal mucosa or ulcerated due to irritation. The size of lesion varies from 0.4 to 8 cm [3, 5–8].

Histologically myofibroma exhibits a biphasic pattern of light and dark-stained areas. The light area mainly consists of spindle cells with eosinophilic cytoplasm and tapering or cigar-shaped nuclei, arranging in short fascicles or whorls and nodules, at the periphery of the lesion. However, sometimes these cells are distributed haphazardly throughout the lesion. In contrast, the dark-stained area located more centrally, consisting of round cells or small spindle cells arranged around thin-walled, irregularly branching, hemangiopericytoma-like blood vessels. These cells have basophilic nuclei, small eosinophilic cytoplasm and indistinct cell margins [2]. In some cases the light and dark areas are not separate and the two cell subpopulations are intermixed [3]. Mitotic figures are only rarely observed but lesions deeply located are often ill-defined and focally tend to infiltrate the surrounded tissue [9]. Masson's trichrome stain shows the presence of thick fibrous bundles that are distributed with irregular intersecting angles [6]. Immunohistochemically, myofibroma cells express α -SMA, specific muscle actin, vimentin and are negative for desmin S-100 and CD34 [2, 3, 9, 10]. In our case, histological, histochemical and immunohistochemical findings fulfill the criteria for a diagnosis of myofibroma of oral soft tissues.

Histopathologic differential diagnosis of myofibroma of oral soft tissues includes leiomyoma, schwannoma, nodular fasciitis, benign fibrous histiocytoma and solitary fibrous tumor, desmoid type of fibromatosis and infantile fibrosarcoma [2, 3]. The neoplastic cells of vascular leiomyoma are positive for desmin in contrast to negative myofibroma. Furthermore, Masson's trichrome stain in vascular leiomyoma shows the presence of delicate fibrous tissue that surrounds smooth muscle cells and in the septa between masses of neoplastic cells. This distribution is different from that of the thick fibrous

Fig. 2. a Intersecting bundles and whorls of spindle and round cells. Angular hemangiopericytoma-like blood vessels (arrows). **b** Intermixed spindle and round cells and hemangiopericytoma-like vessel (arrows).

Fig. 3. a Thick bundles of collagen (green color) in the stroma. Masson's trichrome stain. **b** α-SMA in neoplastic cells. Envision/HRP technique.

bundles, with random, irregularly intersecting angles prominently observed in myofibroma [10]. Schwannoma and neurofibroma do not contain hemangiopericytoma-like blood vessels and the neoplastic cells are positive for S-100 protein. Nodular fasciitis demonstrates extravasated red blood cells, myxoid stroma, and chronic inflammatory cells, features that are absent in myofibroma [3]. Benign fibrous histiocytoma consists of fibroblast-like spindle cells and histiocytic-like cells arranged in a storiform

pattern. These cells express factor XIIIa and α₁-antitrypsin [2]. Solitary fibrous tumor may be differentiated from myofibroma because its neoplastic cells express CD34 [11]. Unlike myofibroma, desmoid-type fibromatosis is not characterized by biphasic cellular populations. Finally, infantile fibrosarcoma is characterized by the presence of uniform spindle cells that form fascicles, focal necrosis and hemorrhages, features that are not normally seen in myofibroma [2].

Surgical excision is the treatment of choice for myofibroma of oral soft tissues. Recurrence rate appears to be low. Follow-up data were reported for 33 cases, ranging from 6 months to 13 years. Three cases (9%) recurred possibly due to inadequate surgical excision [3, 5–8].

Conclusion

This was a case of a myofibroma clinically similar to an adjacent irritation fibroma, which highlights the possibility of misdiagnosis of a myofibroblastic tumor and underlines the importance of immunohistochemical and histochemical examination to establish the accurate diagnosis.

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