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Case report

Myofibroblastoma of the tongue: A case report with immunohistochemical findings

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ABSTRACT

Myofibroblastoma is a rare benign stromal neoplasm with myofibroblastic differentiation most often seen in the breast. In the oral region, only one case of myofibroblastoma has been reported so far; therefore, the diagnostic criteria of this tumor are still controversial in the field of oral pathology. Herein, the authors present a case of myofibroblastoma of the tongue and demonstrate the usefulness of immunohistochemistry in diagnosing this tumor. Histologically, the tumor was composed of interlacing bundles of spindle-shaped or oval cells with hyalinized stroma and amianthoid fibers. Immunohistochemically, the tumor cells showed characteristic features of mammary type myofibroblastoma: diffuse immunoreactivity for desmin, vimentin, and Bcl-2; focal immunoreactivity for α-smooth muscle actin, CD34, and cyclin D1; and no immunoreactivity for h-caldesmon. The pathological diagnosis of myofibroblastoma was made. The authors propose myofibroblastoma as a distinctive entity in the oral region, which should not be confused with other spindle-cell tumors or hamartomatous proliferation with myofibroblastic differentiation, and recommend the inclusion of extramammary myofibroblastoma in the differential diagnosis of soft tissue tumors of the oral cavity.

1. Introduction

Myofibroblastoma is a rare benign stromal neoplasm with myofibroblastic differentiation most often seen in the breast. In 1987, Wargotz et al. [1] described cases of mammary stromal tumor and first used the term "myofibroblastoma," reflecting the cellular composition. This tumor is mainly a circumscribed, nonencapsulated proliferation of spindle or oval cells arranged in short fascicles separated by collagen bundles and sharing a variable degree of myofibroblastic differentiation, and less frequently, myoid differentiation [1–3]. Further, mast cells are conspicuous, multinucleated giant cells are sparsely distributed, and fat trapping may be seen [1–3]. Although myofibroblastoma may exhibit a great variety of morphological and immunohistochemical features, the myofibroblastic nature of spindle-shaped tumor cells can be demonstrated by characteristic immunohistochemical findings: typically, positivity for desmin, vimentin, and CD34, and negativity for cytokeratins, epithelial membrane antigen (EMA), S-100 protein, and h-caldesmon [1–4].

Extramammary myofibroblastoma is extremely rare and strictly resembles its mammary counterpart [5]. To date, only one case of "myofibroblastoma" in the oral cavity has been reported [6]; however, the presented tumor did not show immunoreactivity for desmin, which is an important characteristic of mammary type myofibroblastoma. Here, the authors describe a case of myofibroblastoma of the tongue that showed the typical immunohistochemical features of mammary type myofibroblastoma, and demonstrate the usefulness of immunohistochemistry in diagnosing this tumor.

2. Case report

A 65-year-old woman complained of a painless lingual swelling, which had been present for several weeks. Intraoral examination revealed a solitary, circumscribed, elastic hard, nontender nodule on the left lateral aspect of the tongue. The tumor was diagnosed as benign tongue tumor and removed under local anesthesia. The patients showed no sign of recurrence during the 5-year follow-up.

Macroscopically, the excised mass (Fig. 1A), measuring 17 mm × 10 mm × 10 mm, showed a tan-white cut surface and was covered by intact, nonulcerated mucosa. Neither hemorrhage nor necrosis was observed.

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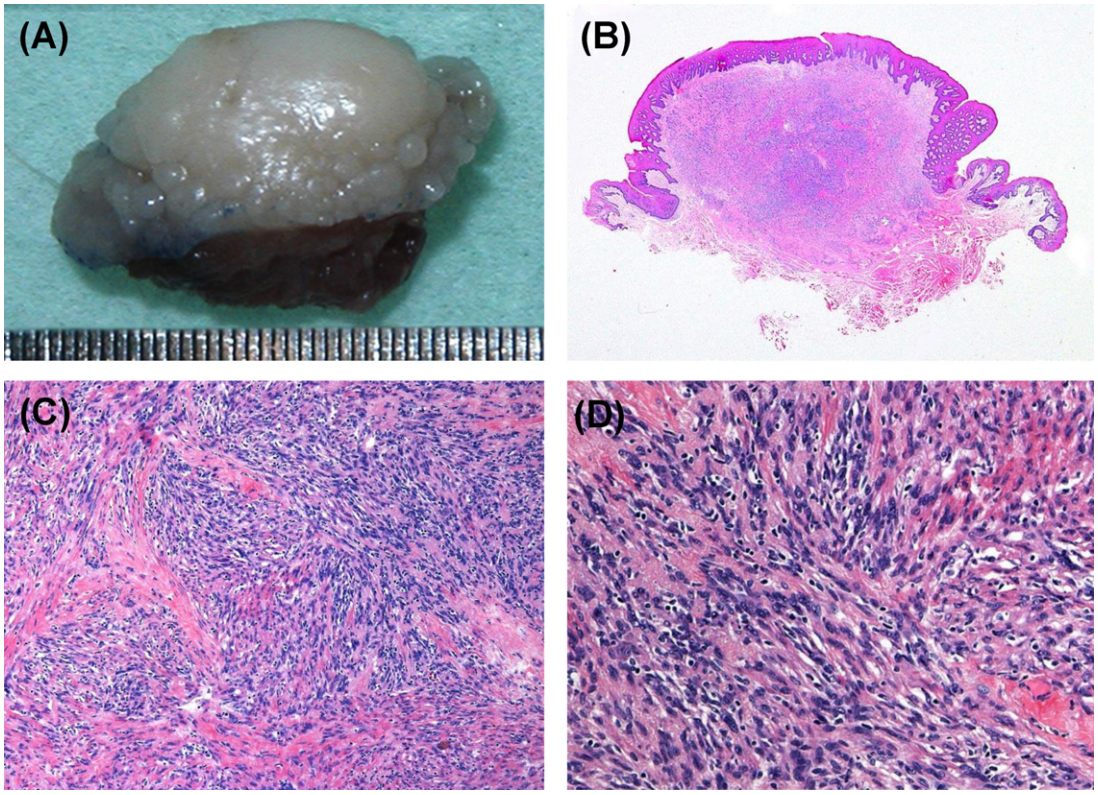

Fig. 1. Macroscopic appearance. (A) The tumor, measuring 17 mm × 10 mm × 10 mm, was covered by intact, nonulcerated mucosa. Neither hemorrhage nor necrosis was observed. (B) Histologically, the tumor is a well demarcated but nonencapsulated (H&E). (C, D) The tumor is composed of interlacing bundles of spindle-shaped or oval cells with hyalinized stroma and collagen fibers (H&E ×200, H&E ×400).

Histological examination showed a well-demarcated but nonencapsulated tumor (Fig. 1B) composed of interlacing bundles of spindle-shaped or oval cells with hyalinized stroma and collagen fibers (Fig. 1C and D). Trichrome staining, which is useful to confirm the histological features of myofibroblastoma [1], revealed blue-green staining of the broad collagenous bands and red staining of the myofibroblastic tumor cells. Adipose tissue and skeletal muscle fibers were trapped at the periphery, where multinucleated giant cells were also seen. Mast cells were scattered throughout the tumor. Mitotic figures were extremely rare and considerable atypia of the tumor cells was not noted. Zoning phenomenon and hemangiopericytoma-like area, which are the characteristic histological features of myofibroma, were not present in this specimen.

Immunohistochemically, the tumor cells showed diffuse intracytoplasmic positivity for desmin (Fig. 2A) and vimentin (Fig. 2B); a few cells showed not only diffuse intracytoplasmic staining but also nuclear positivity for Bcl-2 protein (Fig. 2C), a member of the apoptosis regulator protein family. Nuclear positivity for cyclin D1 was noted in some tumor cells (Fig. 2D). A few tumor cells, besides capillary vessels, were positive for α-smooth muscle actin (α-SMA) and CD34. H-caldesmon, a reliable marker in distinguishing smooth muscle versus myofibroblastic cellular differentiation [3], was negative for tumor cells. S-100 protein was negative for spindle-shaped myofibroblastic cells although adipose and muscle tissues at the periphery of tumor were positive. Further, D2-40, cytokeratins (CAM5.2 and AE1/AE3), and mucins (MUC1, MUC2, MUC4, MUC5AC, MUC6, and MUC16) were negative. The mean Ki67 labeling index of five individual regions was 8.7%. The pathological diagnosis of myofibroblastoma was made.

3. Discussion

It has been argued whether myofibroblastoma in the oral region differs from other spindle-cell tumors with myofibroblastic differentiation, such as myofibroma. Although myofibroblastoma has been included in some articles dealing with myofibromatosis, Lingen et al. [7] described myofibroblastoma as a distinct entity and suggested that it should not be confused with myofibroma because the former does not exhibit the zoning pattern or hemangiopericytoma-like areas present in the latter. Similarly, Montgomery et al. [8] indicated that myofibroblastoma differs from myofibroma by lacking hemangiopericytoma-like areas between lobules. In addition, myofibroblastoma immunohistochemically demonstrates strong intracytoplasmic staining for desmin [1–4], which myofibroma does not [7–9]. In the current case, histological examination revealed interlacing bundles of spindle cells with hyalinized collagen fibers but without zoning phenomenon or hemangiopericytoma-like areas, and immunohistochemistry showed diffuse immunoreactivity for desmin and vimentin, focal positivity for α-SMA and CD34, and negativity for h-caldesmon. These findings are consistent with the characteristic features of myofibroblastoma, and do not coincide with those of myofibroma or myofibromatosis.

In 1990, Sahin et al. [6] first reported a case of “myofibroblastoma” in the oral cavity; since this report, other authors have debated the diagnosis of this case. For example, Jones et al. [9] believed that the lesion described by Sahin et al. represents myofibroma rather than myofibroblastoma, considering the histological, immunohistochemical, and ultrastructural findings. On the other hand, Lingen et al. [7] did not include the Sahin et al. case in their series of myofibromas because the description did not