Giant Cell Fibroma of the Buccal Mucosa: A Case Report

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頸粘膜に発生した巨細胞線維腫の一例

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要約：口腔粘膜に発生する線維腫は通常、慢性的な刺激に対する反応性あるいは修復性の有茎性病変である。巨細胞線維腫は比較的稀な病変であり、既報告では歯肉に好発するとされている。この度、われわれは63歳、男性の頸粘膜に発生した巨細胞線維腫の一例を経験したので報告する。病理組織学的には粘膜下に線維性結節を含む脊椎様粘膜がみられ、結合織中に多数の呈状の多核巨細胞が認められた。術後6年であるが、再発なく経過良好である。

Key words: giant cell fibroma (巨細胞線維腫), buccal mucosa (頸粘膜), CD34 (CD34)

Introduction

Giant cell fibroma was first reported by Weathers and Callihan in 1974, and comprises 5-10% of all fibrous lesions. Giant cell fibroma is an asymptomatic, neoplastic oral mucosal tumor arising predominantly in the gingiva. Giant cell fibroma differs from other common irritative fibromas by the presence of stellate and multinucleated giant cells. The multinucleated giant cells are reportedly derived from fibroblasts, and synthesize collagens and proteins histochemically and ultrastructurally. Electron microscopy has revealed that the giant cells are formed by the fusion of mononuclear cells. The present article describes a case of giant cell fibroma of the buccal mucosa.

Case Report

A 63-year-old man was referred to our department with a mass of the right buccal mucosa. He was unaware of the lesion until it was pointed out by his dentist. At the time of the first visit, medical history included esophageal carcinoma for which he had undergone surgery 9 years ago.

Intraoral examination revealed a 1.0 × 0.8 × 0.4 cm, well-defined, elastic hard mass in the right buccal mucosa (Fig. 1). Coloration of mucosa over the growth was similar to normal buccal mucosa. Clinically, papilloma of the buccal mucosa was tentatively diagnosed.

Excision was performed under local anesthesia. During the operation, the tumor was excised together with overlying mucosa and surrounding normal tissue. No signs of recurrence have been seen at the time of writing, 6 years after surgery.

Histological examination revealed polyoid mucosa including a demarcated submucous fibrous nodule (Fig. 2). Numerous large stellate and multinucleated giant cells were scattered in the nodular fibrous tissue (Fig. 3 and 4). Density of mononuclear fibroblastic cells other than giant cells was moderately increased, but no atypia was apparent. Vascularization was not prominent, and collagen bundles were not arranged in any particular fashion, such as storiform and herring bone patterns. As a peculiar finding, mast cells were intermingled within the lesion. Immunohistochemical examination yielded negative results for EMA, S-100, desmin, CD68, CD31 and αSMA, and positive results for vimentin (Fig. 5A) and CD34.

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Fig. 1  The mass measuring 1.0 x 0.8 x 0.4 cm formed a well-defined, elastic hard mass in the right buccal mucosa.

Fig. 2  The lesion is a nodule of cellular fibrous connective tissue, relatively well demarcated in the submucosa. Hematoxylin and eosin.

Fig. 3  The tumor comprises numerous oval-to-spindle-shaped cells, polyhedral cells with large nuclei and scattered multinucleated giant cells in the fibrous connective tissue. Hematoxylin and eosin. ×100.

Fig. 4  Multinucleated giant cells and spindle cells associated with collagen bundles. Mast cells are also apparent in the tumor (arrows). Hematoxylin and eosin. ×400.

(Fig. 5B). The lesion was thus histopathologically diagnosed as giant cell fibroma.

Discussion

Fibromas are non-epithelial benign tumors that are seen frequently in the oral cavity. Most fibromas are reactive lesions and true tumors are extremely rare. In 1974, undescibed oral fibrous tumors with distinctive clinical and histological features were reported as giant cell fibroma by Weathers and Callihan. Clinically, giant cell fibroma is an asymptomatic, pedunculated and fibrous lesion. In a previous study, giant cell fibroma represented 4.7-5.0% of all biopsied fibrous lesions and 0.7-1.0% of total accessions. According to Weathers and Callihan, giant cell fibroma displays a peak incidence in the second decade of life, with about 60% of lesions occurring within the first 3 decades of life, a female-to-male ratio of 1.3:1, and a marked preponderance in Caucasians. A marked predilection for gingival mucosa is seen, with a ratio of about 2:1 for mandibular to maxillary lesions. Following in decreasing incidence are lesions of the tongue, palate, buccal mucosa and lip. As giant cell fibroma has particular predilections for location, age and race compared with the usual inflammatory fibrous hyperplastic
lesion, these 2 lesions may represent distinct entities⁴).

Histologically, the characteristic feature of giant cell fibroma is the presence of numerous large stellate cells and multinucleated giant cells⁵). Multinucleated cells of giant cell fibroma have been thought to probably derive from melanocytes⁶,⁷), undifferentiated mesenchymal cell⁸) or fibroblasts⁹,¹⁰). Recently, the most acceptable origin of giant cell fibroma has been fibroblastic, probably associated with myofibroblasts on the basis of histochemical and electron microscopy studies¹¹). In particular, examination of the cytoskeleton has suggested that giant cell fibroma multinucleated cells originate from myofibroblasts and/or undifferentiated mesenchymal cells, given the compatible actin filament characteristics¹²). In terms of immunoreactivity against proliferating cell nuclear antigen (PCNA) and Ki-67 in giant cell fibroma multinucleated cells, absence of Ki-67 immunoreactivity in multinucleated cells and the reported absence of mitoses in 108 cases of giant cell fibroma suggests that cell cycling in the absence of cytokinesis is unlikely to represent the mechanism of multinucleated giant cell formation, and differences in nuclear PCNA staining intensity within individual multinucleated giant cells may reflect sequential fusion of mononuclear fibroblasts with each multinucleated cell at different time points⁶).

In our case, immunoreactivities for vimentin and CD34 were found in tumor cells. Oral tumors showing positive immunoreactivities for both vimentin and CD34 are extremely rare¹³), and include solitary fibrous tumor¹⁴), giant cell fibroblastoma¹⁵,¹⁶) and giant cell angiofibroma⁷). Vimentin+/CD34+ cells in these tumors may be derived from fibrocytes that migrated into the submucosa. Fibrocytes are blood-borne cells with fibroblast-like properties and positivity for CD34¹⁷,¹⁸). Giant cell fibroma is thus considered to be a distinct entity from other tumor-like fibrous lesions, such as irritation fibroma and fibro-epithelial polyp, which comprise hyperplastic fibroblasts with over-production of collagen bundles.

Giant cell fibroma was treated in a conservative manner by simple surgical excision. Houston reported that 401 of 464 giant cell fibromas were treated by simple surgical excision, with recurrence noted in only 2 cases⁷). One lesion recurred once, and the other recurred twice⁷). Given the innocuous nature of giant cell fibroma, simple surgical excision is adequate as the treatment of choice⁷).

In conclusion, we reported a case of giant cell fibroma arising in the buccal mucosa of a Japanese man.

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Reference
