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Collagenous fibroma (desmoplastic fibroblastoma) of alveolar bone: a case report

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Fibroma colagenoso (fibroma desmoplásico) do osso alveolar: relato de caso

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key words

Collagenous fibroma

Desmoplastic fibroblastoma

Soft tissue tumors

Immunohistochemistry

abstract

Collagenous fibroma (desmoplastic fibroblastoma) is a rare benign soft tissue tumor with a fibroblastic origin. In oral mucosa only two cases have been described in the literature. We describe the case of a 42-year-old white woman whose complaint was a painless, slow-growing mass under the prosthesis. Histopathologic features included sparsely distributed stellate or spindle fibroblasts within a rich collagenous stroma. Tumor cells were diffusely stained for vimentin and rare cells stained for smooth muscle actin and factor XIIIa. Total surgical excision was performed and no recurrence is expected.

unitermos

Fibroma colagenoso

Fibroma desmoplásico

Tumores de tecido mole

Imuno-histoquímica

resumo

O fibroma colagenoso (fibroma desmoplásico) é um tumor de tecido mole raro de origem fibroblástica. Na mucosa oral apenas dois casos foram descritos pela literatura. Nós descrevemos o caso de uma mulher de 42 anos de idade, cuja queixa era uma lesão indolor, de crescimento lento sob sua prótese total superior. Os achados histológicos da lesão incluíram fibroblastos escassos, estrelados ou fusiformes, dispersos em um estroma rico em colágeno. As células lesionais foram positivas para vimentina, mas rara marcação foi observada para actina músculo liso e fator XIIIa. A excisão cirúrgica total foi realizada e a recidiva da lesão não é esperada.

Introduction

Collagenous fibroma (CF) is a benign soft tissue tumour first described by Evans in 1995. It is also known as desmoplastic fibroblastoma (DF) and is originated from cells with a fibroblast/myofibroblast phenotype. Those tumors exhibit an abundant collagen deposition and their true nature is discussed.

Up to the present, a small number of cases were described (85 approximately) and the largest series was presented by Miettinen and Fetsch[7]. Generally patients complain of a slowly growing and painless mass[5, 8]. Tumors are subcutaneous or intramuscular and involve different parts of the body including arms, shoulders, feet or ankles, legs and hands. However, few cases were reported in the head and neck region, being one in the thyroid gland and two in the oral cavity, specifically in palate and parotid gland[3, 6, 11].

Microscopic examination shows a hypocellular soft tissue mass predominantly composed by collagenous

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extracellular matrix. Tumor cells are spindle or stellate-shaped with large or oval nuclei that may contain eosinophilic nucleoli. Binucleated cells and multinucleated giant cells may be present and fat entrapment is also possible. Vessels are often imperceptible due to the surrounding collagenous stroma[1, 2, 7].

Differential diagnosis of soft tissue lesions can be done through the immunohistochemical analysis as many are the tumours with similar characteristics.

Presently we report a case of a collagenous fibroma and discuss its clinical, histologic and immunohistochemical findings.

Case report

A 42-year-old woman was referred to the Public Health Care Hospital of Guarulhos (São Paulo, Brazil) because of a painless mass on the maxillary alveolar bone. The lesion had been present for three months, affecting her upper prosthesis stability. Medical history was not contributory and she presented satisfactory health conditions. Intraoral examination reveled a 10-mm firm consistent mass, with an ulcerated reddish surface probably caused by the prosthesis trauma, located at the alveolar bone of maxilla. Periapical radiography showed no osseous involvement, but an osteogenic reaction of the subjacent bone could be seen (**Figure1**).

Macroscopic features

An excisional biopsy was performed and the material was sent to the Oral Pathology Department of Universidade de São Paulo for histological examination. The tumor was fixed in 10% formalin solution and was paraffin embedded. Sections were routinely processed and stained in hematoxylin-eosin.

Microscopic features

Microscopic examination showed that the lesion was nodular and partially surrounded by a fibrous pseudocapsule. It presented a fibrous benign aspect, was hypocellular and composed by fibroblast/myofibroblast cells dispersed in a collagenous-rich stroma (**Figure 2**). Those cells varied from spindle to stellate-shaped and were often binucleated with large elongated to oval nuclei (**Figure 3**). Nuclei exhibited delicate chromatin, and small eosinophilic nucleoli were present. Mitotic figures, areas of necrosis, metaplastic bone formation and calcification were not seen. Inflammatory cells were absent.

Immunohistochemical findings

Immunohistochemical staining was performed using a labeled streptavidin-biotin system. Three-µm tissue sections were prepared and the used antibodies included S100 protein (1:1000, Dako, Carpenteria, Ca), vimentin (V9 clone - 1:200, Biogenex, San Ramon, Ca, USA), factor XIIIa (1:1200, Dako) and α-smooth muscle actin (SMA) (1A4 clone - 1:200, Dako). Antigen retrieval for vimentin was achieved by microwave treatment (three times for 5 minutes at 700W in citric acid, 10mmol/L, pH 6.0). For factor XIIIa 0.04% trypsin (0.04g of pepsin and 0.134g of calcium chloride in 100mL of Tri-buffered saline solution at pH 7.6; 37°C) was used.

Cells stained diffusely for vimentin (**Figure 4**) and rare cells stained for SMA (**Figure 5**) and factor XIIIa (**Figure 6**). S100 reactivity was not observed.

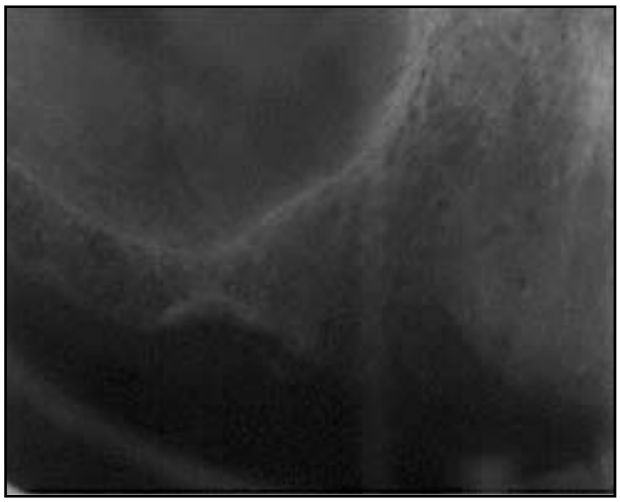

Figure 1 – *Periapical radiography exhibiting no osseous involvement and an osteogenic reaction of the alveolar bone*

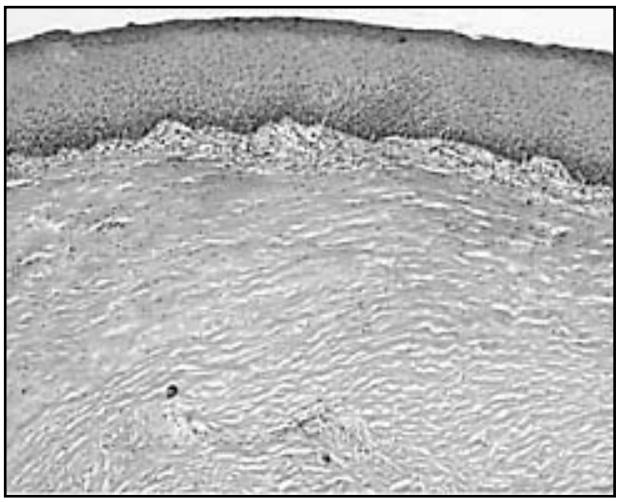

Figure 2 – *Photomicrography of CF showing a lesion partially surrounded by a fibrous pseudocapsule, hypocellular with cells dispersed in a collagenous rich stroma (H&E 25x)*

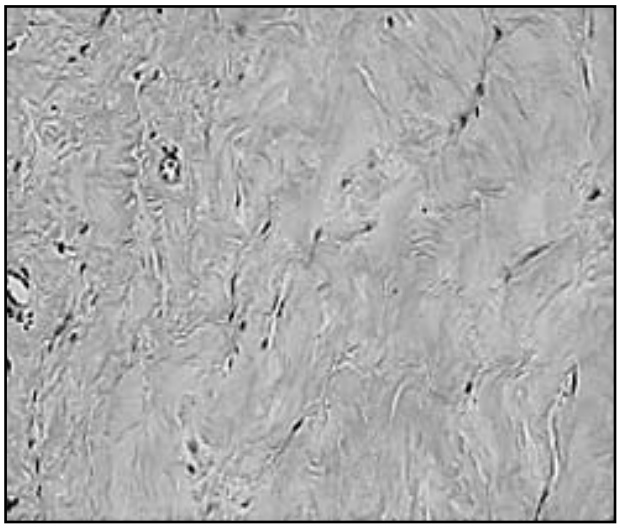

Figure 3 – *Photomicrography of CF presenting spindle to stellate-shaped cells. The cells were often binucleated with large elongated to oval nuclei (H&E 200x)*

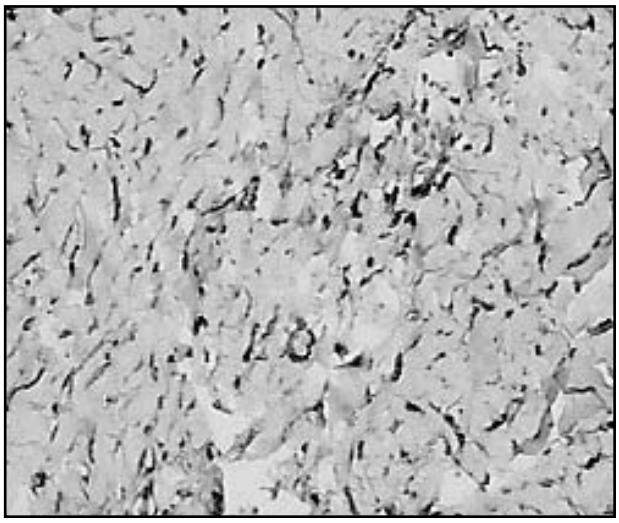

Figure 4 – *Immunohistochemistry detection for vimentin. Cells were diffusely positive (200x)*

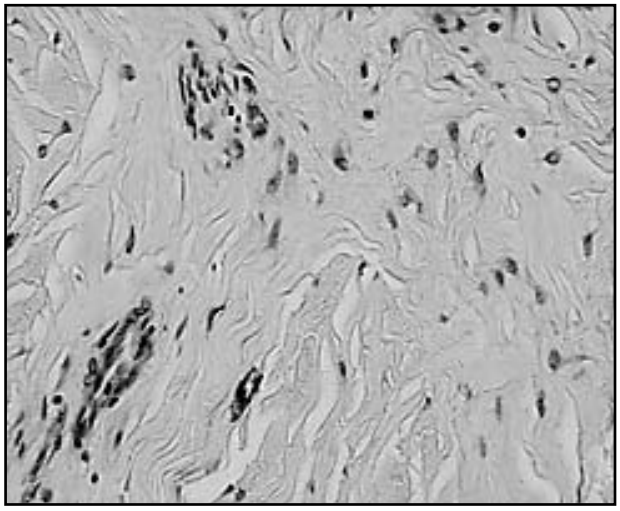

Figure 5 – *SMA were positive for vessel cells but only rare fibroblasts were reactive for this protein (200x)*

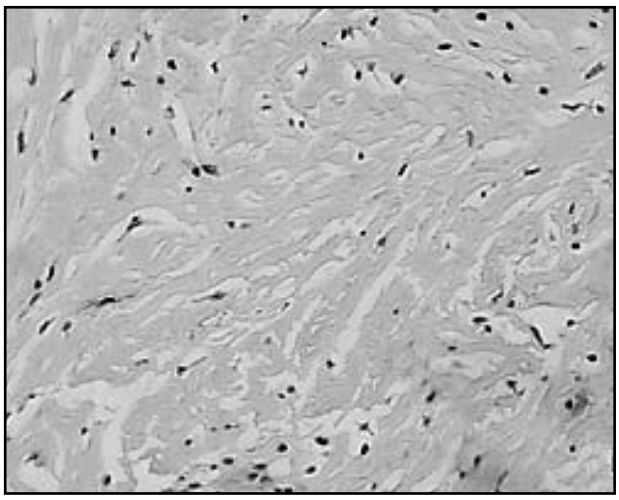

Figure 6 – *Photomicrography of immunohistochemistry reaction for factor XIIIa. Few cells were immunopositive for this protein (200x)*

Discussion

Evans was the first to introduce the term desmoplatic fibroblastoma, in 1995. Ever since, many other cases with CF-similar characteristics were published in the English literature. We believe that the case described here presents similar characteristics to the one described by Evans.

There are only two cases of CF reported in the oral region. Mesquita *et al.*[6] described a CF in hard palate of a 37-year-old woman and the other case was reported by Ide *et al.*[3] in the parotid gland region.

Soft tissue tumors are relatively common, but frequently represent a diagnostic problem for pathologists due to the similarity in histological aspects and an overlap in the staining profile among cells with fibroblastic and myofibroblastic differentiation. In addition, collagen-rich variants of nonfibroblastic or myofibroblastic neoplasms may demonstrate a similar profile that masks their true nature[9].

CF diagnosis is practically morphologic, since immunohistochemistry investigation is not very elucidative. However it is very useful to exclude other lesions in the differential diagnosis. We investigated immunohistochemical staining for S100, vimentin, factor XIIIa and SMA. Similarly to other authors we found that the cells were negative for S100 protein and strongly positive for vimentin. Nevertheless, no strong positivity was observed for factor XIIIa and SMA, contradicting Mesquita *et al.*[6] and Junkins-Hopkins and Johnson[4].

The differential diagnosis for CF in the oral cavity includes a wide variety of soft tissue lesions, especially

inflammatory fibrous hyperplasia, traumatic fibroma, giant cell fibroma and neurofibroma[9].

The origin of CF is still unclear. In the present case there was a history of local trauma caused by the prosthesis. But it is likely that the trauma came after the tumor growth, which was responsible for a superficial ulceration. Nevertheless, a traumatic origin is not discussed in the literature. It has been suggested that CF might originate from a chromosomal rearrangement (11q12) similar to the fibroma of the tendon sheath[10]. However, it is still unclear if this lesion is a reactive process or a true neoplasm.

Our case refers to a 42-year-old woman, although epidemiologic features from literature point out to male preference in the fifth decade of life[7, 9]. The treatment of choice was total surgical excision. Prognosis is excellent and no recurrence is expected.

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