



ORIGINAL RESEARCH PAPER

Pathology

**UNUSUALLY LARGE UNUSUAL LESION
COLLAGENOUS FIBROMA: CASE REPORT.**

KEY WORDS:

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ABSTRACT Collagenous fibroma is a benign lesion that affects mainly the subcutaneous and muscle tissues. We describe a case of a collagenous fibroma that appeared in a white 28-year-old female, who presented with a well-circumscribed and firm lump in her palatal and gingival mucosa, measuring 70mm in diameter. Histopathology revealed a paucicellular fibroblastic proliferation within a highly collagenous matrix. These findings satisfied the diagnostic criteria for collagenous fibroma (desmoplastic fibroblastoma).

Introduction

Collagenous fibroma (CF) is also known as desmoplastic fibroma/fibroblastoma or fibroblastic/myofibroblastic soft tissue. It was first described by Evans in 1995 in a series of 7 cases [1]. The lesion has a predilection for men in the fifth and sixth decades of life and affects a wide variety of anatomical sites, though it is very rare in the oral cavity. The abnormal tissue usually develops slowly and painlessly within the subcutaneous and intramuscular layers of tissue, often accompanying a duration history of 6 months or more. Tumor size varies from 1.0 to 20.0 cm and, as well as the classic well-circumscribed and firm features reported, disk-shaped and lobulated growths patterns have also been reported. Clinically, it has a fibrous appearance with a glistening, gray-to-white superficial aspect [2,3].

Despite the fact that relatively few cases have been reported, CF has a good prognosis. The treatment consists in total surgical excision and no recurrence is the norm following such an approach [4]. The purpose of this article is to describe the clinical and histopathological findings of a case of CF in order raise awareness of such disease entity to oral pathologists.

Case report

A 28-year-old white female was referred to the Oral Medicine service of the São Leopoldo Mandic Dental School and Research Institute due to her complaint of a growing mass in the right buccal mucosa. On extra-oral examination nothing abnormal was detected. Intra-oral examination revealed a well-circumscribed nodular lesion on the right side of her palate, crossing the midline on one end and extending through to the buccal aspect of her posterior alveolar ridge, including the gingival mucosa. The lesion was firm on palpation, measured approximately 70-mm in

diameter and was covered by a pink-colored healthy-looking oral mucosa (Fig 1). The patient reported a 12-month duration of the growth since she first noticed it. No associated tooth mobility was noted and no evidence of bone involvement was detected radiographically. No habit or traumatic events were associated to such lesion. The patient was otherwise generally healthy and her medical history did not contribute to the current findings. Considering that the aforementioned description was strongly suggestive of a benign lesion, a surgical excision was therefore performed. The specimen was fixed in 10% buffered formalin solution and embedded in paraffin. Five µm sections were obtained and stained using hematoxylin and eosin.

Histological evaluation revealed a well-defined, non-encapsulated, fibroblastic proliferative lesion, characterized by paucicellularity within a highly collagenous matrix and spindle-shaped fibroblasts (Fig 2).

Postoperative evaluation at 8 months revealed no recurrence (Fig 3) though 24 months after surgery, a relapse of the palatal lesion was noticed, though much milder than the original growth (Fig 4). The patient decided against a new surgical intervention at this stage and preferred to continue with 6-monthly follow-up visits



Fig 1: Nodular lesion on the right side of the palate and buccal mucosa.



Fig 2: Highly collagenous matrix and spindle-shaped fibroblasts (Hematoxylin and Eosin, Original magnification 30 X).

Discussion

The case presented herein shows clinical and histopathological features that are compatible with the diagnosis of CF. CF is a rare benign soft tissue tumor, frequently found in several locations, such as the arm, the posterior portion of the neck and the upper back [4], yet the mouth is seldom a primary site [5].

The clinical features of the case described herein met the clinical criteria established in the literature for the diagnosis of a collagenous fibroma, namely solitary, firm, well-circumscribed, painless, large growth of long duration (nearly 18 months) [2,3], similar to our case, which together with all clinical features described, had a duration history of 12 months.

On gross examination, the lesion appears as an oval mass with a resilient, fibroelastic consistency on palpation and a homogeneous fibrous cut surface [2]. Histopathologically, stellate-shaped or spindle-shaped cells are observed in a paucicellular and highly collagenous matrix that included adjacent adipose tissue [6].

There is some controversy regarding the etiology of DF, especially as to whether it is an actual neoplastic growth or simply a reactive lesion. The literature overall shows a trend towards DF representing a neoplastic growth, as no evidence of an inciting event has been clinically associated with it nor has a specific cause to such a reactive fibrous proliferation been microscopically identified [2,7]. This was corroborated by the case presented herein as no habit or traumatic event could be related to the lesion.

In the oral cavity, the clinical differential diagnosis of DF should include a wide range of soft tissue lesions, especially inflammatory fibrous hyperplasia, traumatic fibroma, giant cell fibroma, neurofibromatosis, gingival fibromatosis and peripheral ossifying fibroma [4].

Inflammatory fibrous hyperplasia is a reactive lesion of fibrous connective tissue that develops in association with an ill-fitting full or partial denture and is histopathologically characterized by increased collagen deposition in a fibrous connective tissue [8]. In addition, a variable chronic inflammatory infiltrate is present. Traumatic fibroma is the most common tumor of the oral cavity, though in most instances it represents a reactive hyperplasia of fibrous connective tissue in response to local irritation or trauma [2]. Such lesions could be similar to CF in terms of size, color and consistency.

Giant cell fibroma is a fibrous tumor in which connective tissue cells, many of which are multinucleated, assume a stellate shape; clinically, it measures less than 1.0 cm in maximum diameter and does not show entrapment of adjacent tissue [2]. In the present case, such differential diagnosis, including giant cell fibroma, could have been almost excluded solely on size, as DF has been shown to reach large proportions.

Patients with neurofibromatosis present multiple neurofibromas that can occur anywhere in the body. A typical cutaneous manifestation of this condition is the so-called café au lait pigmentation. In the oral cavity, lesions present as painless slow growing soft nodules that vary in size from small lumps to large masses [9]. Insofar as a single lesion from both conditions would pose some difficulty to be differentiated intraorally, those from neurofibromatosis tend to occur as multiple nodules in association with other cutaneous and systemic manifestations.

Gingival fibromatosis is associated with genetic factors with a relatively early onset, namely individuals younger than 20 years and in association the eruption of either deciduous or permanent teeth. The most striking feature is gingival hyperplasia, which may affect a group of teeth and remain stable or later extend to other gnathic segments. The gingiva therein appears firm on palpation, with normal color and smooth or slightly dotted surface. Histologically, it is composed of hypocellular and hypovascular yet dense collagenous tissue [10]. The main differential aspect of the lesion presented in this case was the fact that no association with tooth eruption was detected.

Peripheral ossifying fibroma is a reactive lesion that occurs exclusively in the gingiva. It is additionally described as a pedunculated or sessile nodular growth, which usually develops in the interdental papilla with color varying from red to pink. It generally measures less than 2 cm in diameter and affects mostly young adults [11]. Once again, the criterion size was crucial to differentiate DF from peripheral ossifying fibroma from the clinical point of view, not to mention that, histopathologically, one would be dealing with completely different morphological entities.

The current treatment of choice for DF is total surgical excision and the prognosis is good. No patient to date had reportedly experienced recurrence. Our patient was treated by complete surgical excision and she had no recurrence or new growths in first 8 months postoperatively, though an initial relapse of the palatal lesions was noticed 24 months after surgery, which contradicts cases reported elsewhere and reinforces the importance of long-term follow-up.

Declaration of Conflicting Interests

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References

1. Evans HL. Desmoplastic fibroblastoma: a report of seven cases. *Am J Surg Pathol* 1995;19:1077-81.
2. Mesquita RA, Okuda E, Jorge WA, de Araújo VC. Collagenous fibroma (desmoplastic fibroblastoma) of the palate: a case report. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 2001 Jan;91(1):80-4.
3. Miettinen M, Fetsch JF. Collagenous fibroma (desmoplastic fibroblastoma): a clinicopathologic analysis of 63 cases of a distinctive soft tissue lesion with stellate-shaped fibroblasts. *Hum Pathol.* 1998 Jul;29(7):676-82.
4. de Sousa SF, Caldeira PC, Grossmann Sde M, de Aguiar MC, Mesquita RA. Desmoplastic fibroblastoma (collagenous fibroma): a case identified in the buccal mucosa. *Head Neck Pathol.* 2011 Jun;5(2):175-9.
5. Nonaka CF, Carvalho Mde V, de Moraes M, de Medeiros AM, Freitas Rde A. Desmoplastic fibroblastoma (collagenous fibroma) of the tongue. *J Cutan Pathol.* 2010 Aug;37(8):911-4.
6. CAZAL, C. et al. Collagenous fibroma (desmoplastic fibroblastoma) of alveolar bone: a case report. *J Bras Patol Med Lab.* 2005 Jun; 41(3): 185-8.
7. Bernal K, Nelson M, Neff JR, Nielsen SM, Bridge JA. Translocation (2;11)(q31;q12) is recurrent in collagenous fibroma (desmoplastic fibroblastoma). *Cancer Genet Cytogenet.* 2004 Mar;149(2):161-3.
8. Pereira TD, de Lacerda JC, Porto-Matias MD, de Jesus AO, Gomez RS, Mesquita RA. Desmoplastic fibroblastoma (collagenous fibroma) of the oral cavity. *J Clin Exp Dent.* 2016 Feb 1;8(1):e89-92.
9. García de Marcos JA, Dean Ferrer A, Alamillos Granados F, Ruiz Masera JJ, García de Marcos MJ, Vidal Jiménez A, Valenzuela Salas B, García Lainez A. Gingival neurofibroma in a neurofibromatosis type 1 patient. *Med Oral Patol Oral Cir Bucal.* 2007 Aug 1;12(4):E287-91.
10. He L, Ping FY. Gingival fibromatosis with multiple unusual findings: report of a rare case. *Int J Oral Sci.* 2012 Dec;4(4):221-5.
11. Franco-Barrera MJ, Zavala-Cerna MG, Fernández-Tamayo R, Vivanco-Pérez I, Fernández-Tamayo NM, Torres-Bugarín O. An update on peripheral ossifying fibroma: case report and literature review. *Oral Maxillofac Surg.* 2016 Mar;20(1):1-7.