CASE REPORT

Collagenous fibroma (desmoplastic fibroblastoma) of the oral cavity

Sanjay Bhagalia, Manish Jain, Nilesh Pardhe, Sundaragiri Krishna Sireesha
Department of Oral Pathology, NIMS Dental College, NH 8, Jaipur, Rajasthan, India

Address for correspondence:
Dr. Sanjay R Bhagalia,
A-47, Model Town, Malviya Nagar,
Jaipur - 302 017, Rajasthan, India.
E-mail: buggydoc@hotmail.com

ABSTRACT

Collagenous fibroma (desmoplastic fibroblastoma) is a distinctive yet uncommon fibrous soft tissue tumor. These tumors are rather nondescript in their morphological appearance and have been diagnosed as fibromas or some other benign mesenchymal lesions for years. The most common sites are the upper extremities, followed by the lower extremities. Rare lesions arise in the head and neck region. We report a rare case in the oral cavity and present its unique histopathological features (central fat entrapment) besides others, and diffusely strong vimentin immunopositivity.

Key words: Central fat entrapment, collagenous fibroma, desmoplastic fibroblastoma, vimentin immunopositivity

INTRODUCTION

The collagenous fibroma (CF), is a slow-growing, benign, soft tissue lesion that was first described as recent as 1995 by Evans,[1] under the rubric, "desmoplastic fibroblastoma." As the term "fibroblastoma" may be construed as an immature or potentially malignant neoplasm, Nielsen *et al*[2] proposed the name "collagenous fibroma" as being not only more descriptive of its histological composition but also more reflective of its benign prognosis. Approximately 78 additional cases have been reported since then, describing the lesion better.[2-7] Miettinen and Fetsch[3] have till date discussed 63 cases, the largest series ever. The results obtained from all reports indicate clearly that CF has a propensity to occur in males in the fifth to sixth decades of life, presenting as subcutaneous or intramuscular lesions, having a wide anatomic distribution.[1,3,4] Mesquita *et al*[8] have recently claimed that their case was the first such CF seen intraorally. The CF presents as a slow-growing neoplasm of more than 6 months duration, measuring anywhere between 1 cm and 20 cm.[1,3,4] Clinically, the lesion is seen as a firm, well-circumscribed, round to oval, or lobulated mass, which on cut surface appears fibrous with a glistening gray-to-white aspect.[4] Microscopically, the overlying surface epithelium of CF is normal, although fat or skeletal muscle may be seen either entrapped between the rete pegs or in the center of the lesion. The neoplasm consists of sparsely distributed, medium to large, spindle- to stellate-shaped fibroblastic cells, interspersed among abundant intercellular collagen fibers. In dense fibrotic areas, cellularity is scarce and the cells do not exhibit any mitotic activity, nor is there any necrosis. Binucleated and multinucleated giant cells may be seen occasionally. Blood vessels are not a usual feature and inflammatory cells are almost absent. Immunohistochemically, the fibroblastic cells are intensely positive for vimentin. A few cells may show a positive immunoreactivity for muscle-specific and alpha-smooth muscle actin.[1,3,4] Ultrastructurally, the cells have characteristics of fibroblasts or myofibroblasts.[2] Sciot *et al*[9] studied two CFs and found that both exhibited an abnormality of the same band of the long arm of chromosome 11, more specifically, a rearrangement of 11 q 12. Because the fibroma of the tendon sheath showed the same chromosomal aberration as that of the oral lesion, they suggested that the two lesions may be related.

Access this article online

Quick Response Code:

Website: www.jomfp.in

DOI: 10.4103/0973-029X.99089

CASE REPORT

A 58-year-old female patient was referred to the Department of Oral and Maxillofacial Surgery, at NIMS Dental College, Jaipur, Rajasthan, due to a complaint of a firm, slow-growing, pedunculated lesion measuring 2 cm × 1 cm, in the lingual gingival region in relation to the permanent mandibular left 3rd molar since 3 months. The patient was not medically compromised. Intraoral examination showed a well-circumscribed, firm mass, covered by normal mucosa. The related teeth were not mobile, ruling out underlying bone involvement, and intraoral radiographs confirmed the same. Regional lymph nodes were normal and patient did not complain of any pain. The lesion was completely excised

under local anesthesia and the specimen was sent to the Department of Oral Pathology, NIMS Dental College, Jaipur, Rajasthan, for histopathological evaluation. The specimen was fixed in 10% formalin solution and subsequently embedded in paraffin. 5µm sections were made and stained with HandE, and 3 µm sections were made for immunohistochemical analysis, by subjecting the sections to the streptavidin method. The immunohistochemical analysis was performed by using primary antibodies against vimentin (1:200) (Biogenex, San Ramon, California). Antigen retrieval for vimentin was accomplished by microwave treatment (3 times for 5 minutes) at 700 W in citric acid (10 mmol/L, pH 6.0). The above-mentioned immunohistochemical analysis was performed in the Department of Histopathology at Santokba Durlabhji Memorial Hospital, Jaipur, Rajasthan.

GROSS FEATURES

The tissue received after the excisional biopsy was white in color, firm, shiny on cut surface with a soft yellowish area in the center of the cut surface.

HISTOPATHOLOGICAL FEATURES

Sections stained with HandE were observed under 4×, 10×, and 40× to reveal a surface mucosa which was normal, overlying a dense, homogenous, eosinophilic connective tissue. The connective tissue consisted of dense collagen bundles in which were seen scarcely distributed spindle to stellate fibroblastic cells [Figure 1]. Multinucleated giant cells were also observed. Blood vessels were few, as were inflammatory cells. A peculiar, yet unique feature, seen in our case, was an area in the center of the section, consisting of fat cells [Figure 2]. Similar fat cells were also observed between the rete pegs. Sections were also subjected to the Masson's Trichrome Stain which revealed areas positive for collagen (blue-green) and some areas positive for muscle (red). The cytoplasm of the lesional cells took up a red stain [Figure 3].

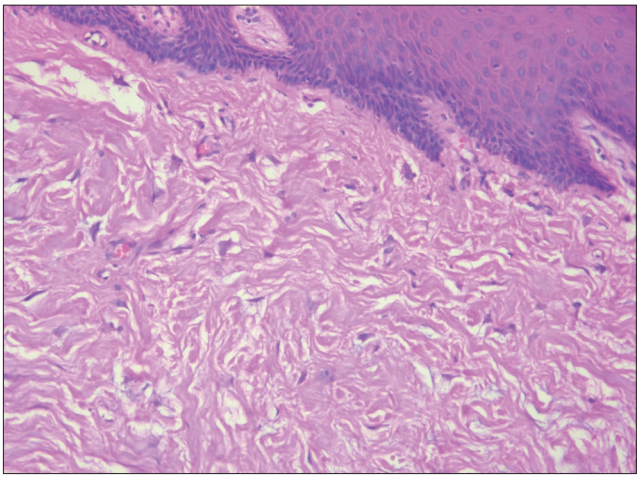

Figure 1: Scant distribution of lesional fibroblastic cells in dense collagenous matrix (H and E, 10×)

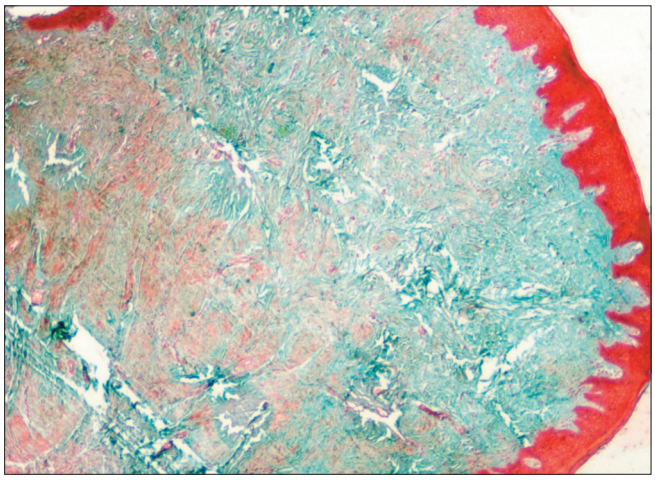

Figure 3: Section shows collagen (blue-green) and muscle (red) (Masson's Trichrome, 4×)

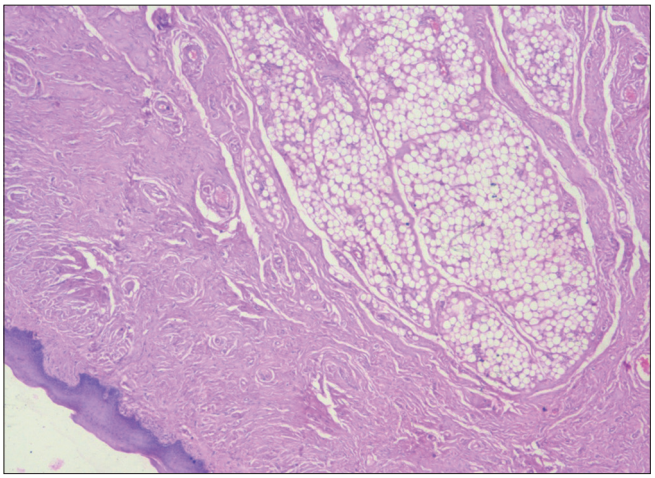

Figure 2: Fat cells in centre of section (H and E, 4 ×)

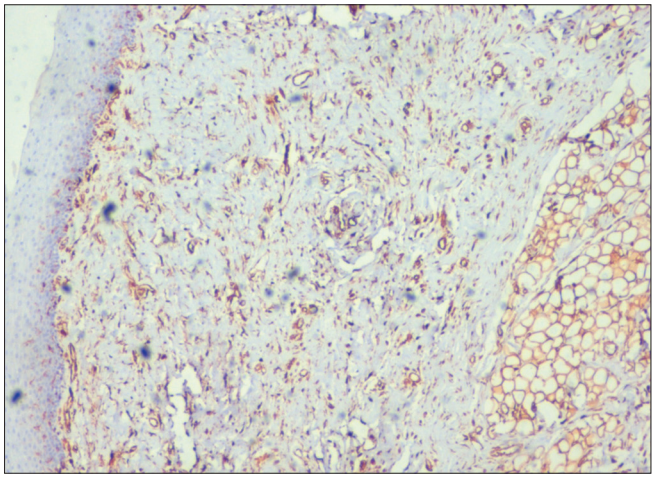

Figure 4: Vimentin positive lesional fibroblastic cells (4 ×)

IMMUNOHISTOCHEMISTRY

Immunohistochemically, the lesional fibroblastic cells were strongly positive for vimentin [Figure 4]. The collagen bundles were seen as long and circular thick bundles, distributed in a random manner throughout the section.

DIFFERENTIAL DIAGNOSIS

In the oral cavity, the differential diagnosis for CF should include a wide range of soft tissue lesions, especially inflammatory fibrous hyperplasia, traumatic fibroma, and giant cell fibroma. Other oral soft tissue lesions that are seen less frequently, e.g., soft tissue myofibroma, nodular fasciitis, fibromatosis, and neurofibroma, can also be considered in the differential diagnosis for oral CF.[8]

DISCUSSION

The clinical and histopathological features of our case comply with the criteria for the diagnosis of CF reported by Evans[1] and Nielsen *et al*[2] who delineated desmoplastic fibroblastoma and CF, respectively. Clinically, our case appeared somewhat similar to the previously described cases.[3] On gross examination, the lesion was seen as a firm mass, which on cut surface appeared shiny and included a central area of soft, yellowish tissue, which on histopathological evaluation turned out to be fatty tissue. A similar feature has been described by Miettinen and Fetsch[3] who also observed entrapment of fat in 51% of their series of 63 cases. Our case was also immunohistochemically as reactive to vimentin as were other cases. A doubt still persists as to the true nature of CF. According to Evans[1] and Nielsen *et al*,[2] CF is a neoplasm, as it does not have a preceding traumatic or inciting cause. Our patient also did not give any history of trauma or any other triggering event. The nature of the neoplastic cells in CF has been described by Evans[1] and Nielsen *et al*,[2] which have features of both fibroblasts and myofibroblasts.

CONCLUSION

In the oral cavity, the differential diagnosis should include giant cell fibroma as well as a whole range of soft tissue lesions such as, inflammatory fibrous hyperplasia and the simple fibroma. The last mentioned lesion, i.e., the simple fibroma is of great significance, as we feel, that a large number of CF may have been diagnosed in the past as the simple fibroma. In CF, the collagen presents as long, thick, and wavy bundles with a random distribution. The fibroma also displays thick and wavy bundles of collagen with a random distribution, but the bundles are shorter than those seen in CF. Moreover, entrapment of adjacent fat and muscle appears to be a unique feature of CF.[8]

Immunohistochemical analysis should be carried out on all such lesions. Our aim was to draw the attention of all oral pathologists to review cases in the past diagnosed as fibromas, which could possibly be CF/desmoplastic fibroblastomas instead.

The current treatment of CF is complete surgical excision as the prognosis is good thereafter. Our patient, examined after 4 months, showed complete recovery with no recurrence.

ACKNOWLEDGEMENT

We thank Dr. Vikas Singh and Dr. Ruchika Tiwari, Oral and Maxillofacial Surgeons, NIMS Dental College, Jaipur, Rajasthan, for their surgical expertise, and Dr Shubha Gupta, Head, Department of Histopathology, Santokba Durlabhji Memorial Hospital, Jaipur, Rajasthan, for providing immunohistochemistry laboratory facility.

REFERENCES

1. Evans HL. Desmoplastic fibroblastoma. A report of seven cases. Am J Surg Pathol 1995;19:1077-81.
2. Nielson GP, O'Connel JX, Dickersin GR, Rosenberg AE. Collagenous fibroma (desmoplastic fibroblastoma): A report of seven cases. Mod Pathol 1996;9:781-5.
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;29:676-82.
4. Hasegawa T, Shimoda T, Hirohashi S, Hizawa K, Sano T. Collagenous fibroma (desmoplastic fibroblastoma). Report of four cases and review of literature. Arch Pathol Lab Med 1998;122:455-60.
5. Weisberg NK, DiCaudo DJ, Meland NB. Collagenous fibroma (desmoplastic fibroblastoma). J Am Acad Dermatol 1999;41:292-4.
6. Jang JG, Jung HH, Suh KS, Kim ST. Desmoplastic fibroblastoma (collagenous fibroma). Am J Dermatopathol 1999;21:256-8.
7. Ide F, Shimoyama T, Horie N, Tanaka H. Collagenous fibroma (desmoplastic fibroblastoma) presenting as a parotid mass presenting as a parotid mass. J Oral Pathol Med 1999;28: 465-8.
8. 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;91:80-4.
9. Sciot R, Samson I, van den Berghe H, Van Damme B, Dal Cin P. Collagenous fibroma (desmoplastic fibroblastoma): Genetic link with fibroma of tendon sheath. Mod Pathol 1999;12:565-8.

How to cite this article: Bhagalia S, Jain M, Pardhe N, Sireesha SK. Collagenous fibroma (desmoplastic fibroblastoma) of the oral cavity. J Oral Maxillofac Pathol 2012;16:277-9.

Source of Support: Department of Oral & Maxillofacial Surgery, NIMS Dental College, Jaipur; and, Department of Histopathology, Santokba Durlabhji Memorial Hospital, Jaipur. **Conflict of Interest:** None declared.