Giant Cell Fibroma in an Elderly Woman: A Report of a Rare, Late and Unusually Large Presentation.

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ABSTRACT

Background

Giant cell fibroma (GCF) is a rare benign oral fibrous lesion which is predominantly found in the mandible of Caucasians but rarely in the maxilla and black population above the third decade of life.

Objective: *To draw the attention of clinicians to a rare, late and unusually large presentation of Giant Cell Fibroma*

Case report*: The index case was a 68-year-old fisherwoman with a ten-year-old slow-growing painless swelling occupying the left anterior maxilla. The lesion crossed the midline to the right causing incompetent lips and teeth displacement leading to aesthetics and functional problems.*
Upon clinical examination, a provisional diagnosis of peripheral giant cell granuloma was made. Radiological essentially revealed a soft tissue mass, histopathological evaluations confirmed the diagnosis of GCF and the patient was prepared for surgical excision. A surgical excision of the soft tissue mass and peripheral ostectomy was performed.

Conclusion*: GCF has distinct histopathologic features occurring in any age, race, site and decades of life; growing very large causing aesthetics and functional problems. Immunohistochemistry if available is an additional resource to unequivocally establish the diagnosis.*

Keywords: Giant Cell, Fibroma, Maxilla, Histology, Nigerian

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Received: 16-Sept, 2022

Revision: 23 Nov, 2022

Accepted: 25 Nov, 2022

Citation: Oluwakuyide RT, Castano BO, Akinshipo AO, Ayodele AO. Giant cell fibroma in an elderly woman: a report of a rare, late and unusually Large Presentation. Nig J Dent Res. 2023; 8(1): 1-6. https://dx.doi.org/10.4314/njdr.v8i1.1

INTRODUCTION.

Giant cell fibromas (GCFs) are distinct benign fibrous hyperplastic lesions of the gingiva and oral mucosa. It was first described and named by Weathers and Callihan in 1974 for its characteristically large stellate-shaped, mononuclear and multinucleated giant cells when compared with the other benign fibrous hyperplastic lesions of the gingiva and oral mucosa.[1] It represents about 2%–5% of all oral fibrous proliferations.[2,3] Although the aetiology is unknown, the most accepted is the hypothesis that GCF is a response to minor trauma or recurrent chronic inflammation.[1,5] Gender predilection remains controversial. While some reported it to be commoner in females, others reported males and yet some said there is equal sex predilection, but it is commonly found in the young age of the first three decades of life. [1–5] The mandibular gingiva is the most frequent site of presentation, though it can occur at other sites such as the tongue, palate, and buccal mucosa.[1-4] GCF may present as an asymptomatic pedunculated or sessile swelling with a pebbly, bosselated or nodular surface, and fibrous or elastic nature which may be confused with a papilloma or fibroma on the provisional diagnosis.[1,4,5] The distinct histological features consist of uninflamed fibrous tissue in which there are numerous large spindle, and stellate-shaped cells with prominent basophilic cytoplasm distinguish GCF from other oral fibrous lesions and these giant cells may be multinucleated and dendritic.[1,5,6] Very few cases had been reported regarding this tumour of controversial origin.[4] Surgical excision is the treatment of choice and recurrence is rare.[6] Hence, this presentation of an abnormally large GCF in a 68-year-old fisherwoman along with a brief literature review.

CASE REPORT.

A 68-year-old fisherwoman presented at the Oral and Maxillofacial Surgery clinic of General Hospital Lagos, on account of an anterior maxillary swelling of 10 years duration. There was no history of tobacco and alcohol intake. The swelling was a painless slow growing lesion. A provisional clinical diagnosis of peripheral giant cell granuloma of the left maxilla was made.

On examination, the swelling was essentially on the left maxilla, measuring 9cm x 10cm in its widest diameter, extending from the maxillary left second premolar to the right maxillary lateral incisor, occupying half of the primary maxilla to the left commissure of the mouth (Figure A). It was lobulated and firm in consistency with areas of hyperemia. There was a displacement of the left central and lateral incisors along with the canine obliterating the buccal sulcus (Figure A). Oral hygiene was poor and the patient was not on any medication. Medical history was not contributory. CT scan describes an expansile mass. Routine blood tests and electrocardiogram (ECG) were within normal range. The differential diagnosis of peripheral giant cell granuloma, peripheral ameloblastoma and peripheral ossifying fibroma was made.

An incisional biopsy was done for histopathological evaluation. This revealed a tissue composed of hyperplastic parakeratinized stratified squamous epithelium with prominent rete pegs overlying a collagenized connective tissue stroma within which are large stellate-shaped fibroblasts. Also seen are multinucleated stellate cells and endothelial-lined vascular channels (Figures 1 &2). A diagnosis of giant cell fibroma was made. Surgical excision of the lesion was done under general anaesthesia (Figures B & C). There were neither complications nor recurrence after a year of follow-up

.CLINICAL AND HISTOPATHOLOGY PICTURES

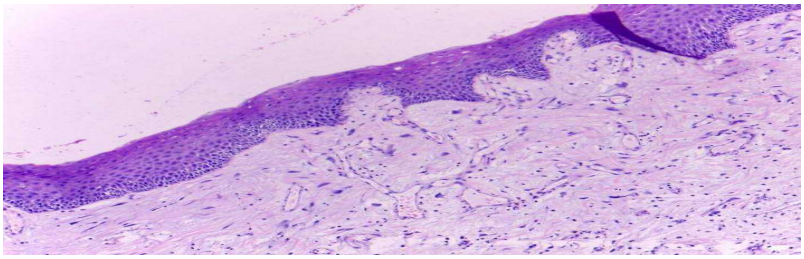

Figure 1: Photomicrograph of the Giant Cell Fibroma showing hyperplastic squamous epithelium (H&E; original magnification x 4)

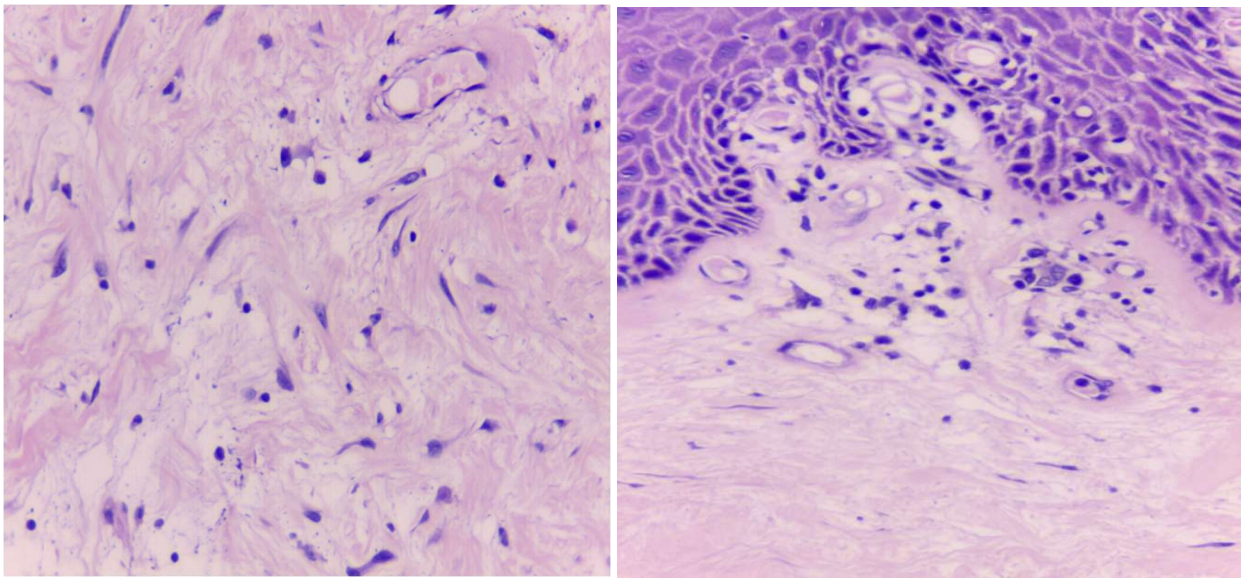

Figure 2: Photomicrograph of microscopic features revealing compact fibrous connective tissue with numerous large spindle-shaped mononuclear and multinucleated cells. (H&E, original magnification x 100).

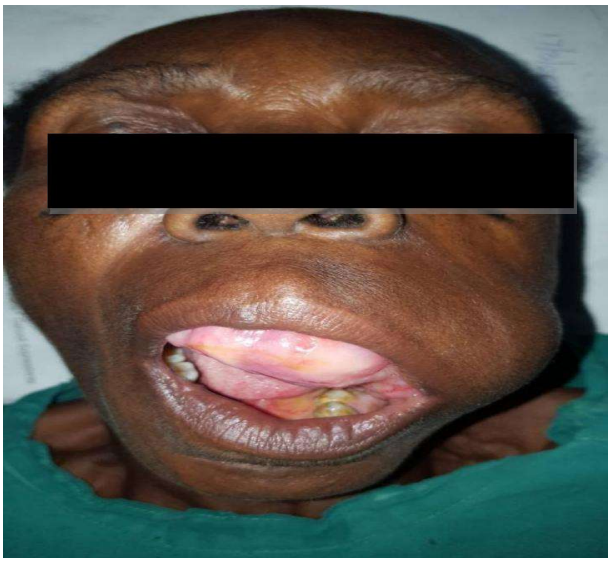

Figure A: Pre-operative picture of the large lesion transverse right central incisor to the left first premolar.

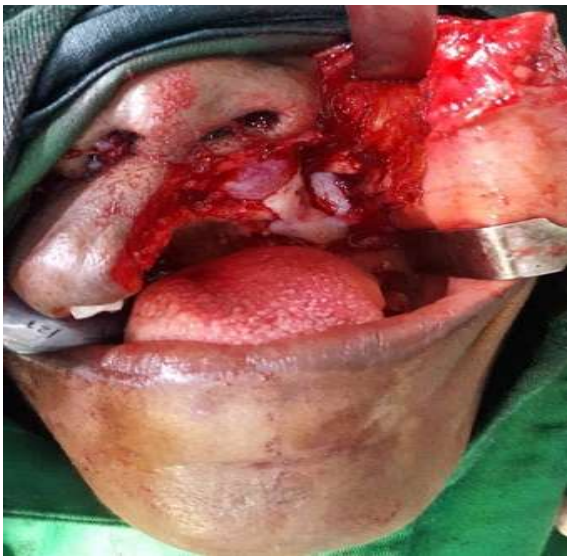

Figure B: Intra-operative picture during excision and peripheral ostectomy.

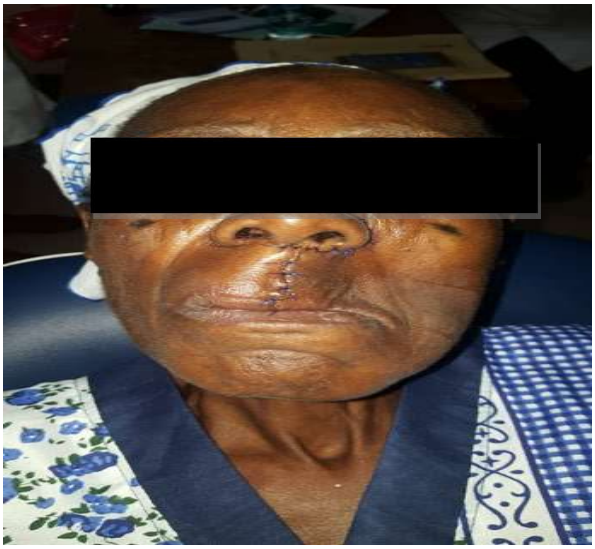

Figure C: Post-operative picture during routine wound dressing

DISCUSSION

Giant cell fibroma (GCF) of oral cavity is a fibrous lesion containing stellate and multinucleated giant cells, described in 1974 as an entity by Weather and Callihan, who applied the term "giant cell fibroma" to this oral mucosal tumour. GCF had been previously categorized as pyogenic granuloma, peripheral gingival fibroma, peripheral giant cell granuloma and peripheral ossifying fibroma by Eversole and Rovin.[1,2,4] The classification of GCF as a distinct entity within fibrous hyperplastic lesions was based on its distinctive clinical and histological features though they may share common characteristics.[5'6] This lesion is most frequently seen in Caucasians.[4,7]

GCF usually presents as asymptomatic small pedunculated or sessile exophytic growth, firm in consistency with pebbly or bosselated overlying mucosa with the occasional ulcerated surface due to acute trauma.[2,4,7] It is frequently found twice on the gingivae of the mandible than other parts of the oral cavity, followed by the tongue, buccal mucosa or palate with a reported average size of 0.5-1.0 cm diameter.[2,4,7] In the present case, the lesion involved the gingiva of the left maxilla and it was about 10 cm in its widest diameter extending to the adjoining palatal mucosa. It was firm in consistency with a smooth and lobulated surface which contrasted the observations from other studies that reported that this lesion is frequently found on the mandible, making this present case rare and unique.[4,7,8] Furthermore, the size was about 10cm in its widest diameter and found in the Nigerian population which contrasted reports from other studies that it is frequently seen in Caucasians and not more than 1.7cm in its greatest diameter.[1,4–8] Although GCF is frequently seen below the third decade of life, Sabarinath et al[4] in their study reported cases occurring in the seventh decade of life which is consistent with the observation in this present case.

To the best of our knowledge, this case is the first and oldest known reported case of GCF in literature in our environment. Plausible explanation for this late presentation may be related to ignorance and poor motivation in seeking medical assistance, which may be pervasive in this patient's low socio-economic status depicted by the picture of poor oral hygiene (Figure A), and not until the lesion constituted an emotional and functional embarrassment that the patient sought for medical assistance.

Most cases of GCF are clinically misdiagnosed as fibroma, papilloma, pyogenic granuloma or peripheral giant cell granuloma because the clinical presentation and epidemiology of these lesions are similar.[2,7,8] However, its distinctive histopathologic characteristics and ultra-structure confirm the diagnosis of this lesion.[4,5] The origin of GCF may be controversial, however, most authorities believe that it is a separate entity while others are of the opinion that there are no therapeutic or prognostic differences between GCF and other epulis.[4,9] Clinicopathologic characteristics of GCF

demonstrate a bosselated, pedunculated exophytic mass largely composed of fibrous connective tissue loosely arranged with a prominent vascular element, especially in the subepithelial zone.[5] GCF has abundant dense collagen fibres that is responsible for its firm clinical appearance and it shares similar histopathologic features with other common fibrous hyperplastic tissues like fibromas, and fibroepithelial polyps; although it is differentiated from these similar lesions by the presence of multiple large stellate shaped fibroblast and multinucleated giant fibroblast in a loose collagenous stroma (figures 1&2).[4] Electron microscopic and immunohistochemical studies have suggested that these cells are unusual, atypical and giant fibroblasts which are formed by the fusion of mononuclear cells or macrophages with intracellular microfilament.[4,5,7] Histopathologic evaluation of this present case correlates with the findings of previous studies (Figures 1 &2).

The origin of these stellate giant cells was initially thought to be different but was later refuted when these cells showed negative reactions for antibodies against S-100 protein and the absence of Birbeck granules in the ultrastructural studies ruled the possibility of Langerhans cells representing these giant cells.[5] The stellate giant cells are mostly concentrated in the lamina propria and typically located subjacent to the epithelium, but are less common or absent in the central portion of the lesion.[5] Thus, their presence is largely dependent on the collagen pattern in the lamina propria. These cells often present with well-defined borders, large nuclei, and slender elongated cytoplasmic processes often arranged in loose immature avascular fibrous connective tissue and may contain brown pigment characteristics of melanin.[9]

These pathognomonic cells are often smudged, never hyperchromatic (compared with truly dysplastic fibroblasts), with hyperplastic overlying epithelium that has thin elongated interconnecting rete ridges (Pseudoepitheliomatous hyperplasia), but without chronic inflammatory infiltrate.[8] The histology of this present case showed stellate and giant cells subjacent to the epithelium in a loosely arranged avascular connective tissue which is consistent with observations from the literature.[3,4,7,8] (Figure 2).

Galvao et al[5] indicated that immunohistochemical expression of mast cell tryptase in GCF of the oral mucosa, the possible interaction of these cells with stellate giant cells and their role in fibrosis and tumour progression exhibited a significant interaction with stellate giant cells present in GCF and thus suggesting the involvement of mast cells in the induction of fibrosis and modulation of endothelial cell function in GCF. Fibrous hyperplasias are considered reactive proliferations of fibroblastic tissue rather than neoplastic proliferations mostly resulting from chronic injury, irritation or a stimulus whose source cannot always be determined.[9,10]

Immunohistochemical studies done to determine the origin of these fibroblasts have shown them negative staining for cytokeratin, S-100 protein, CD 68, HLA DR, neurofilament, HHF, CD 68, and tryptase but positive staining only for vimentin and prolyl - 4 – hydrolase, suggesting that GCF stellate and multinucleate cells have a fibroblast phenotype origin.[1,2] However a negative reactivity of giant cells for desmin with the negative reaction for alpha-smooth muscle actin (HHF-35), CD 68, and factor XIIIa, eliminated a myofibroblastic origin of stellate cells.[3,5] However a minor subset of giant cells in a few cases showed a positive reaction to factor XIIIa, indicating that stellate cells may be of fibroblastic lineage with a variable mixture of cells from mucosal dendrocytes.[3,4,7] According to some studies, based on a variable expression of PCNA, the most plausible hypothesis is that these cells are atypical fibroblasts that originate from differentiated mononuclear cells.[5,6,10]

Histochemical and immunohistochemical studies have also revealed the distinct difference in the extracellular matrix between GCF and fibroepithelial polyps which is the presence of elastin in fibroepithelial polyps.[3,4] In this present study, immunohistochemistry was not done being a resource-scarce environment.

The treatment of choice for GCF is simple surgical excision in adults while electrosurgery or laser excision is preferred in children, although spontaneous regression has been reported and recurrence is rare.[4,5,9,10]

In this present case, the Oral and maxillofacial surgeon carried out excision of the soft tissue and peripheral ostectomy due to pressure indentation on the bone by the expansile fibrous lesion. However,

there had been no recurrence after a 4-year postoperative follow-up.

CONCLUSION

GCF is a rare oral lesion that could occur in any age, race, jaw, or gender and may grow to a very large size if left untreated. It is thus, pertinent that dental surgeons do a thorough clinical assessment by proper treatment planning, histopathological evaluation and where available immunohistochemical analysis when encountering oral fibrous lesions. In a resource scarce environment as ours, the distinct histopathologic features of loose fibrous collagenous stroma with multiple large stellate shaped fibroblast and multinucleated giant fibroblast helped confirm diagnosis.

Source of support
Nil.
Conflict of interest
None declared

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