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Case Report

Peripheral Odontogenic Fibroma: Case Report and Literature Review

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Abstract: Odontogenic fibroma is a rare benign tumor of mature fibrous connective tissue, with varying amounts of inactive-appearing odontogenic epithelium, with or without evidence of calcification. There are two types ; central odontogenic fibroma and peripheral odontogenic fibroma. Peripheral odontogenic fibroma is a rare, non-encapsulated benign tumor of the oral mucosa often mistaken for a reactive lesion. Peripheral odontogenic fibroma is very often underdiagnosed or simply misdiagnosed, and therefore not much is known about this lesion. Through this work, we tried to bring out the sociodemographic, clinical, paraclinical and therapeutic characteristics of this lesion.

Keywords: Odontogenic tumour, peripheral odontogenic fibroma, management.

INTRODUCTION

The oral mucosa is constantly exposed to various stimuli and, therefore, manifests a wide spectrum of lesions ranging from reactive lesions to tumoral lesions [1]. Odontogenic fibroma is a rare benign tumor of mature fibrous connective tissue, with varying amounts of inactive-appearing odontogenic epithelium, with or without evidence of calcification. There are two types depending on the anatomical sites involved; an intraosseous fibroma also called central fibroma and extraosseous odontogenic fibroma called peripheral odontogenic fibroma [2].

Peripheral odontogenic fibroma, due to its fairly common clinical presentation, is often mistaken for an inflammatory or reactive gingival lesion such as pyogenic granuloma, peripheral ossifying fibroma and peripheral giant cell granuloma [2, 3]. In 1992, despite the persistent gray areas concerning it, the World Health Organization included it in its classification among odontogenic tumors [4]. Numerous studies have explored the subject over the years, changing our knowledge of this lesion, however, without succeeding in fully lifting the veil on all of these gray areas.

This article reports a case of peripheral odontogenic fibroma of a 45-year-old woman who came for consultation for a mass located in the posterior mandibular sector and reviews the relevant literature concerning the sociodemographic, clinical, paraclinical and therapeutic data of this lesion.

OBSERVATION

A 45-year-old woman was referred to the *Centre médical du palais*, in Douala, Cameroon for treatment of a right mandibular mass that had been evolving for approximately three years. His personal history was unremarkable. On extra-oral examination, a slight facial asymmetry was noted due to lower right cheek swelling. The swelling left the integuments free. Intraoral examination revealed the presence of a reddish, bumpy, sessile mass. The mass was located at the avulsion site of the mandibular right first molar, occupying the vestibule. It measured about four centimeters along its longest axis (Figure 1). The mass was painless and firm on palpation. She did not bleed on contact with the examination probe. The adjacent teeth were slightly mobile.

The radiographic examination revealed a small alveolar lysis between the second premolar and the second molar, precisely at the level of the avulsion site of the left mandibular first molar (Figure 2). An incisional biopsy was performed based on the diagnostic hypotheses of pyogenic granuloma, peripheral ossifying fibroma and peripheral

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odontogenic fibroma. Histopathological examination revealed a distinctly cellular fibrous connective tissue stoma interspersed with islets and strands of odontogenic epithelium that appeared inactive. Budding of the basal layer of the surface epithelium and the presence of calcification were also observed. All this allowed us to confirm the diagnosis of peripheral odontogenic fibroma.

The excision of the lesion was performed under local anesthesia at the same time as the extraction of the remaining mobile molars. Careful curettage of the area was performed and primary closure performed using simple stitches with 3/0 absorbable sutures. Follow-up of the patient over one year revealed no recurrence (figure 3).

ICONOGRAPHY

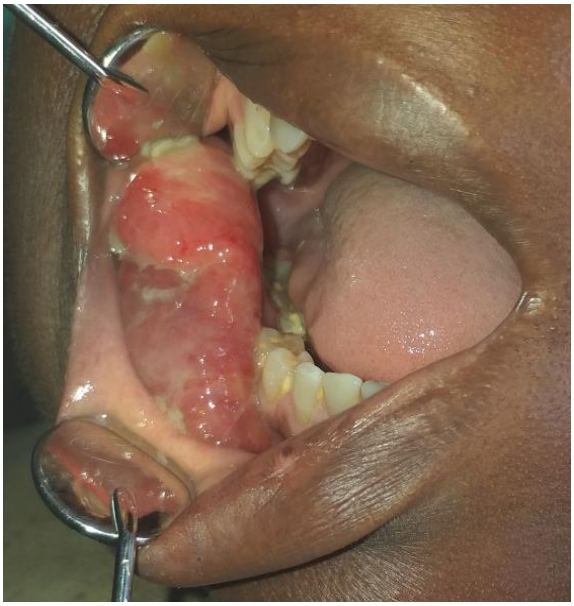

Figure-1: Intraoral image of the mass occupying the right vestibule

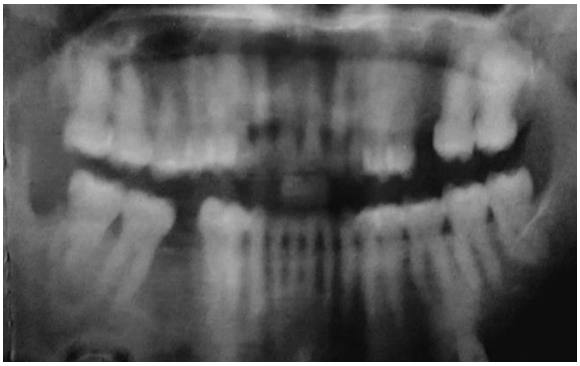

Figure-2: Dental panoramic x-ray of right peripheral odontogenic fibroma

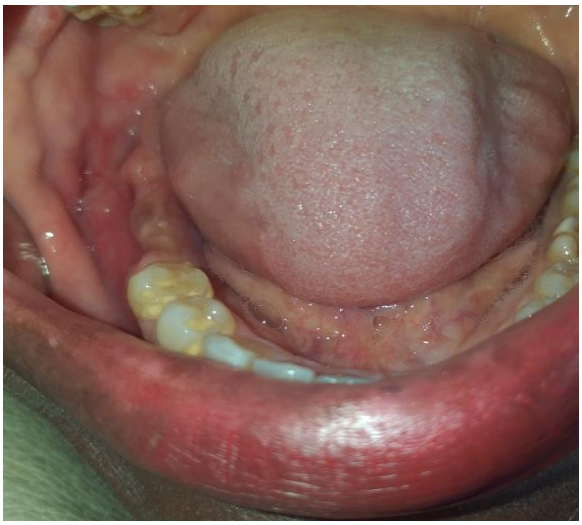

Figure-3: Intraoral image 01 year postoperative

DISCUSSION

Peripheral odontogenic fibroma is a rare benign tumor of mesenchymal origin and composed of fibrous connective tissue containing odontogenic epithelium [5, 6]. It represents only 1.2% to 4.7% of all odontogenic tumors [3, 7, 8].

Peripheral odontogenic fibroma was defined by the World Health Organization in 1971 as an extraosseous homolog of central odontogenic fibroma, a fibroblastic neoplasm containing dentin and/or cementum [9].However, some misunderstandings still remained. In 1982, Gardner distinguished and separated peripheral odontogenic fibroma from peripheral ossifying fibroma with which it was often confused. Then in 1992, the World Health Organization in its classification included it in odontogenic tumors [4, 10].

Peripheral odontogenic fibroma regarding its pathogenesis, is still controversial in the literature. The most accepted hypothesis is that the peripheral odontogenic fibroma comes from the ectomesenchyme, more precisely from the periodontal ligament, from the remains of the dental lamina or from the surface epithelium [1]. Farman, in 1975, suggested that the ectomesenchyme in the gingiva could induce secondary proliferation of the dental lamina remnants and also of the basal layer of the gingival epithelium [11]. Nevertheless, although this lesion is widely accepted as an odontogenic tumor of mesenchymal origin, its histogenesis has not yet been established. Some studies have even floated the idea of considering peripheral odontogenic fibroma as a mixed tumor, since both epithelium and mesenchymal components are necessary for histological diagnosis. [3, 12, 13].

Several studies have focused on peripheral odontogenic fibroma in the literature. In general, their results were similar with respect to gender, age, and location. Daley and Wysocki, in 1994, found on a database resulting from their study and the literature,

107 cases for which the sex of the affected patient was known, 48 cases were male subjects and 59 cases were female subjects. They also found, out of 103 cases from their study and the literature, a wide age range ranging from two to 80 years, with a peak incidence in the third decade. Although all sites were affected. There was a predilection for the mandible, more precisely at the level of the mandibular premolar region. In the maxilla, the incisor region was the most expected [12]. Siar and Ng, in 2000, reported 46 cases and indicated a slight female preponderance (male/female ratio, 1:1.3), peak incidence in the second decade of life, with a wide range of age ranging from 05 months to 64 years, and a preferential location in the mandible (52.2%) [8]. In the study made by Ritwik and Brannon, in 2010, 151 closed cases were collected from the archives. The age range of patients with peripheral odontogenic fibroma in their case series was 05 years to 83 years with a mean of 37.3 (±17.8) years. The highest incidence of the lesion was between the second and fourth decades. The location of the lesion was reported for 147 cases. The occurrence of the lesion was similar to the other studies with a predilection for the mandibular arch (58.5%). An examination of both arches revealed that 110 cases or 74.5% of these lesions occurred in the incisor-cusp region, 17 that is 12.1% in the premolar region and 20 that is 12.5% in the molar region [13].

However, it happens to find in the literature some studies that had to find different data in their series of cases. Alaeddini and colleagues, in 2010; Lin HP and colleagues, in 2011, found in their case series that men were more affected than women [7, 14]. Lin CT and colleagues, in 2008, in the analysis of their case series revealed a slightly higher frequency of localized lesions in the maxilla [15]. Taken together, the data presented in our case compares favorably to the major previous case series. In our study, the female subject was 45 years old. The lesion was located in the posterior mandibular region.

Clinically, peripheral odontogenic fibroma manifests as a round or bumpy mass or nodule, asymptomatic, slowly growing, and generally varying in diameter from 0.5 to 3.5 cm [12, 13, 16]. However, there are exceptional cases reported in the literature up to 4.5 cm long [15, 17]. The color of the tumor is often normal but can also appear reddish or purplish. In rare cases, peripheral odontogenic fibroma can present as multiple or even diffuse lesions. In some studies, peripheral odontogenic fibroma bleeds on brushing and/or resembles a vascular lesion [8, 12, 13, 18, 19, 20]. Although infrequent, it can cause dental displacement and/or dental mobility [6, 15]. In our case, the mass, evolving for more than three years, was bumpy and reddish in color. It was firm on palpation and measured four centimeters along its longest axis. Dental mobility of the adjacent teeth was noted.

This clinical presentation described here, is not pathognomonic for peripheral odontogenic fibroma and the differential diagnosis must therefore be made with other types of reactive and/or peripheral neoplastic lesions that occur in the gums and/or oral mucosa. We can cite, among others, pyogenic granuloma, peripheral ossifying fibroma, peripheral giant cell granuloma, fibroepithelial hyperplasia and peripheral myxoma. [7, 8, 14, 15, 17]. In our case, we put forward as diagnostic hypotheses: pyogenic granuloma, peripheral ossifying fibroma and peripheral odontogenic fibroma.

Radiologically, it is found in the literature that peripheral odontogenic fibroma can cause alveolar bone resorption and dental displacements. Changes involving the underlying bone are not commonly seen in peripheral odontogenic fibroma. We can find the presence of calcification in some cases [3, 15, 20]. Ritwik and Brannon, based on data from their study, had found radiographic features for only 12 of the 151 cases recorded. The lesion lysed the alveolar bone in 06 of the 12 cases, that was 50% of cases found. In one of these cases, the presence of calcifications in the lesion could be demonstrated radiologically [13]. Lin CT and colleagues, found that 15 out of 25 cases in total, that is 60%, showed radiopaque images on radiographic examination in their case series [15]. In our case there was no presence of calcification. Nevertheless, a slight alveolar lysis was noted, which can be seen on the dental panoramic.

Histologically, peripheral odontogenic fibroma is characterized by varying amounts of inactive-appearing odontogenic epithelium embedded in a moderately cellular or collagen-like stroma. The odontogenic epithelium can vary from completely absent to abundant and usually occurs as small islands and strands [2, 8]. In rare cases, clear cell differentiation can be found in the epithelial component [7, 8]. Budding is often observed from the basal layer of the surface epithelium. Connective tissue stroma can vary from fibrous to myxoid, or fibromyxoid. Several studies have reported a higher frequency of a fibrous-like stroma in their case series [7, 13]. Hyalinization and/or calcification around or within or in close association with remnants of the odontogenic epithelium have been observed in peripheral odontogenic fibroma. Different types of calcifications can occur alone or in combination. These calcifications show characteristics of dysplastic dentin, amorphous ovoid cement-like calcifications or osteoid trabeculae [7, 13, 16]. Analysis of the activity of the surface epithelium, of the connective tissue stroma and the presence or absence of epithelial remains and/or calcifications within the lesion is essential for the histological diagnosis of peripheral odontogenic fibroma. These histological characteristics differ according to the studies [3, 7, 13, 14, 15]. In our case, histological examination revealed a distinctly cellular fibrous connective tissue stoma interspersed with islets and strands of odontogenic epithelium that

appeared inactive. There was also the presence of calcification within the lesion.

The treatment of choice for peripheral odontogenic fibroma is surgery, either excision of the lesion or excisional biopsy. [1, 3, 6]. Only the rate of recurrence of peripheral odontogenic fibroids varies according to the studies. Some studies have suggested a low recurrence rate of peripheral odontogenic fibroma. Lin CT and colleagues, in their study, founded no recurrence in their series of cases (25 cases), followed up after 1 year [15]. In the study of Slabbert and colleagues, one of the 30 cases assessed, that was 3%, had recurred after 14 months [20]. Similarly, only one out of 19 cases, that was 5%, of peripheral odontogenic fibroma had recurred after 1 year in the study of Alaeddini et al [7].

However, other studies reported a significant number of recurrences observed in their case series. Daley and Wysocki, in their study, found patient follow-up data for 18 of the 36 cases. In total, they had noted 07 cases of recurrence out of the 18 cases retained, that was 39%. These 07 cases had relapsed between one and four years after treatment [12]. Garcia and colleagues in their study, had found 03 cases of recurrence out of 17 cases in total, that was 18% [16]. The highest recurrence rate was presented by the Ritwik and Brannon study. In their study, there were 29 cases of recurrence out of the 58 cases followed over more than three months, that was 50% of cases of recurrence. The time between the first surgical excision and the recurrence was available for 19 cases. Among these 19 cases, 11 cases, that was 57.9%, had recurred during the first two years. Ritwik and Brannon found in their study that lesions with calcifications apposed to odontogenic remains were less likely to recur. In contrast, lesions without calcifications but with active surface epithelium in the form of budding from the basal cell layer of the rete ridges were associated with significantly higher recurrence. Nevertheless, they concluded by stipulating that complete surgical excision was the most important factor in preventing recurrence of peripheral odontogenic fibroma [13].

Given the inconclusive data regarding the prognosis of peripheral odontogenic fibroma, long-term follow-up of patients is mandatory. In the present case, no clinical signs of recurrence were observed one year after treatment.

CONCLUSION

Peripheral odontogenic fibroma is a rare benign odontogenic tumour. it is often clinically misdiagnosed as an inflammatory lesion. Surgical excision has been suggested as the treatment of choice. However, in view of the data collected in the literature, the possibility of recurrence of peripheral odontogenic fibroma must be considered even after treatment. Regular and long-term follow-up of the patient should be carried out.

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