Choriostoma of the Gingivae following Trauma: An Unusual Case in a Nigerian With Review of Relevant Literature

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ABSTRACT

Objective: Chondromas are benign intraosseous cartilaginous tumors composed of mature hyaline cartilage; they may however arise from the soft tissue in rare cases without bone or joint involvement and are called extra skeletal or soft tissue chondromas. Extra skeletal chondromas (ESC) are commonly seen in the hands and feet but rarely in the oral cavity. We report a rare case of extra skeletal chondroma of the gingiva in a 31-year-old male patient following trauma to the anterior maxillary soft and hard tissues.

Case Description: This is a case of a 31year old hypertensive patient with 3weeks history of gingival swelling in relation to the upper incisors which were fractured following an injury sustained from a motorbike fall. Clinical examination revealed a firm, non-tender localized gingival enlargement adjacent to the palatal surfaces of the maxillary incisors which had a lobulated appearance, with areas of inflammation that bled on gentle probing. The palatal gingival swelling was excised under local anesthesia, while gingivoplasty was performed on the labial gingiva of the same teeth. A periodontal dressing was placed on the surgical sites and removed at one-week review. Histopathologic examination of the palatal swelling revealed connective tissue that was densely infiltrated by mixed inflammatory cells. In some areas, there was focal chondroid tissue with chondrocytes of varying sizes in a chondromyxoid matrix.

Conclusion: A rare case of extra skeletal chondroma is reported, we encourage report of more cases with long term follow-up in multicenter study among Nigerians, to ascertain the prevalence, gender, site of ESC in this population.

Keywords: choriostoma , gingiva, swelling, surgery

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INTRODUCTION

Chondroma is a benign intraosseous cartilaginous tumor. It is usually asymptomatic and histologically composed of mature hyaline cartilage present in periosteal or endosteal locations of bone . It may however arise from the soft tissue in rare cases without bone or joint involvement. When this occurs it is called extra skeletal or soft tissue chondroma[1]. Extra skeletal chondroma(ESC) is often regarded as a choriostoma; which is an island of histologically normal tissue found in an abnormal location. Numerous types of tissues such as cartilage, bone, salivary or thyroid gland can occur as choriostoma[2].

ESC has a site and age group predilection for the limbs (specifically the hands and feet) and patients in the 3rd to 6th decades of life respectively . Although, orofacial extra skeletal chondroma is uncommon, cases in the tongue, masseter, preauricular region, gingiva, tonsil and nasal cavity have been reported[1-5]. As an update to existing data in scientific literature, we report a rare case of extra skeletal chondroma of the gingiva in a 31-year-old male patient following trauma to the anterior maxillary soft and hard tissues.

CASE DESCRIPTION

A 31-year-old male patient was referred to the Periodontology Unit of the Lagos University Teaching Hospital (LUTH), on account of a gum swelling of 3 weeks duration, in relation to the upper front teeth which were fractured following an injury sustained following a fall from a

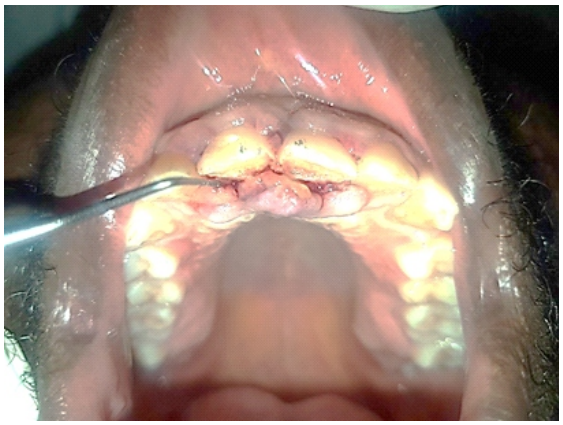

Figure 1: Clinical picture shows an enlarged gingival mass on the palatal surface of the maxillary central incisors.

The labial marginal gingivae in relation to the maxillary anterior incisors appeared slightly erythematous and enlarged with plaque accumulation. Although, the patient's oral hygiene status was fair with an oral hygiene index score of 1.3 (according to the Simplified Oral Hygiene Index of Green and Vermillion Index), there was moderate to severe plaque accumulation around the upper incisor teeth. There was false pocketing on the palatal gingiva of the maxillary incisors. All the teeth were present except the lower right second molar, which had been previously extracted. The maxillary central incisors had Ellis Class II fractures. Intraoral periapical radiographs revealed coronal radiolucency indicative of the fractures. (Figure 2) There was however no widening of periapical ligament space, no periapical pathology and no alveolar bone loss. A clinical diagnosis of localized gingival enlargement in relation to teeth #212, 11, 21 and 22 secondary to trauma, complicated by poor plaque control and Ellis Class II fractures of teeth #11 and 21 was made. The treatment administered included oral prophylaxis, followed by debridement of the gingival enlargement and irrigation with 0.2% chlorhexidine gluconate and normal saline. The fractured maxillary central teeth were restored using composite restorative material. One week later, the patient was reviewed and had gingivectomy done adjacent to teeth 22, 21, 11 and 12 under local anaesthesia in order to excise the palatal gingival enlargement , while gingivoplasty was performed on the labial gingiva in relation to the same teeth (Figure 3).

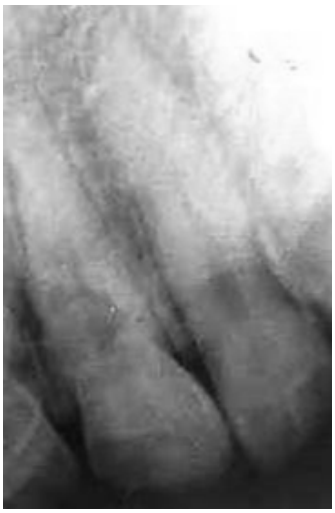

Figure 2: Periapical radiograph of the maxillary centrals showing coronal radiolucency due to Ellis class 1 fracture

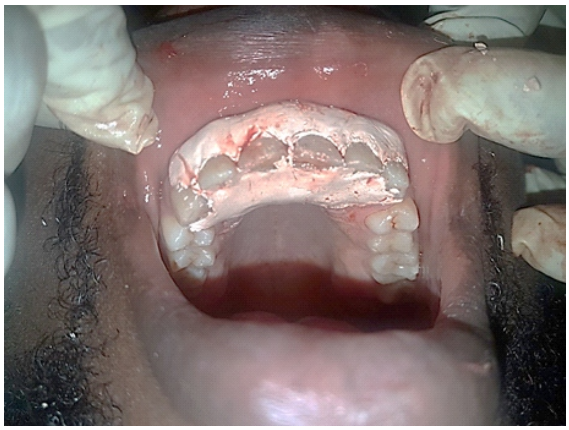

Figure 3: Clinical picture shows periodontal dressing (Coepak) in place immediately after the excision of the lesion.

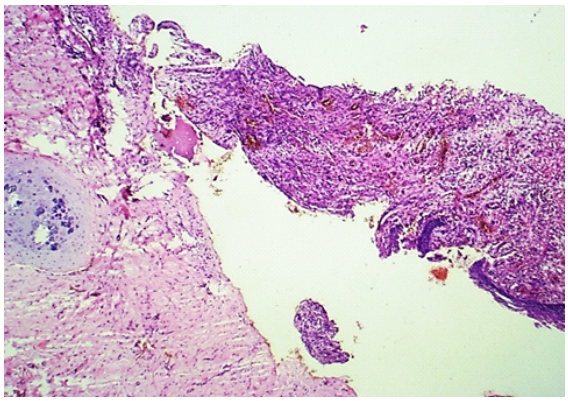

Figure 4: Photomicrograph (H&E X 10) of the gingiva composed of stratified squamous epithelium overlying fibrous connective tissue that contains inflammatory cells (Green arrow) and chondromyxoid connective tissue area that contains island of chondrocytes (Black arrow).

Hemostasis was achieved and a non-eugenol periodontal dressing (Coepak) was placed on the surgical site (Figure 4). The excised lesion was sent for histopathology. Histopathologic examination revealed a hyperplastic stratified squamous surface epithelium and connective tissue densely infiltrated by mixed inflammatory cell infiltrate consisting of neutrophils, plasma cells and lymphocytes. The connective tissue in some areas appeared hyaline-like and poorly cellular and in some other areas there was focal chondroid tissue with chondrocytes of varying sizes in a chondromyxoid matrix. (Figure 5). Periodontal dressing was removed 1 week post- surgery with surgical site healing satisfactorily. Though the patient has been reviewed and the surgical site was observed to be clinically and radiologically normal, he would be closely followed up on 6 monthly review.

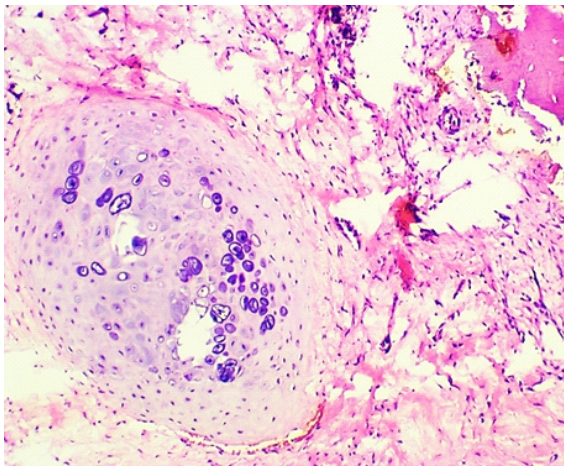

Figure 5. Photomicrograph of the gingiva showing area of a focal island of chondroid tissue with island of residual bone, in chondromyxoid connective tissue (H & E X 40)

DISCUSSION

Extra skeletal chondroma (ESC) is a benign cartilaginous tumor that occurs in soft tissue without bone or joint involvement. It usually present clinically as a slow-growing indolent expanding mass, occasionally associated with tenderness and pain and commonly affects the soft tissues of the hands and feet. It has also been reported to occur in other sites such as the head and neck, abdominal wall, ovaries and the lungs[1-5].

In the head and neck region, the oral cavity (specifically the tongue) has been reported to be its most common site of occurrence[5,7,8]. A previous review of 34 cases which occurred in the head and neck region showed that 24 cases occurred in the tongue[1]. Documented patient age of occurrence of ESC varies over a wide age range of between 10-80 years with more prevalence among the 4th and 7th decades of life[9]. Whilst some investigators report a male gender prevalence [4,9], others report a slight female gender prevalence [1,10]. Gingival occurrence of ESC have rarely been reported in the literature[3,6]. Presently, we report a case located within the palatal gingival tissue in a Nigerian male patient. As far as we know this is the first reported case of oral ESC among Nigerians. However, the age, gender or location prevalence s of oral ESC among Nigerians cannot be determined from a single case study. Determination of such prevalence's among Nigerians can be achieved from lager sample size studies.

The aetiology of ESC remains unclear, though 3 major hypotheses have been postulated. These hypothesis include: the differentiation of pluripotent mesenchymal progenitor cells, the formation of cartilaginous materials due to chronic inflammation and aberrant embryonic cartilage proliferation. In addition, the growth of multipotent mesenchymal cells have been implied to be stimulated by factors such as inflammation, trauma, or irritation[1,5,10,11]. A probable aetiopathogenesis for the present case may be stimulation of pluripotent mesenchymal cells by trauma with irritation from plaque and calculus playing a secondary role. Cytogenetic studies have in addition revealed mutation of the HMGA2 gene on chromosome 12q15[1,9] in patients with ESC.

Computed tomography (CT Scan) imaging reports from previous studies have revealed well-demarcated lobulated soft tissue tumour mass containing peripheral or central curvilinear calcifications without involvement of the underlying bone in 33-70% of cases[1,4,13]. In the present case, a pre-operative CT Scan was not done.. However, with histopathologic examination we did not observe the presence of distinct calcifications within the tumour mass. Drug induced gingival enlargement was a clinical differential made in this case, the patient being a hypertensive patient managed on amilodipine. However the clinical absence of a generalized pattern of gingival enlargement (a localized gingival enlargement was present in this case) coupled with the specific histologic presentation of the lesion resulted in a definitive diagnosis consistent with ESC.

Oral ESC is usually diagnosed with histological evaluation[13]. Microscopically it is composed of lobules of mature, adult hyaline cartilage, with chondrocytes seen growing in groups/islands.

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Oral ESC is usually diagnosed with histological evaluation[13]. Microscopically it is composed of lobules of mature, adult hyaline cartilage, with chondrocytes seen growing in groups/islands. One-third of cases may display widespread calcification, particularly at the centre of the lobules and abundant myxoid matrix with immature cells may sometimes be observed[12-14]. Similarly, histologic presentation of the present case revealed the presence of focal chondroid tissue areas composed of chondrocytes of varying sizes in a chondromyxoid connective tissue matrix. Previous immunohistochemical studies on ESC have shown positive reaction of tumour with S-100, CD 57 and glial fibrillary acidic protein.[1,13] Our diagnosis was based on comprehensive clinical, radiologic and histologic evaluation of the tumour.

It is important to note that oral ESC differs from cartilaginous metaplasia (CM). CM occurs in oral soft tissues areas located beneath ill-fitting dentures. It histologically presents as a diffuse zone/s of dystrophic calcification with single or clustered cartilage cells at different phases of maturatio.n.[5,12] The present case occurred in the palatal gingiva of a patient with full complement of natural teeth.

Intra-oral ESC is usually treated with surgical excision as was done in this case. A total of approximately 10-15% of cases have been reported to recur after surgical excision and so far,, there has not been any report of malignant transformation in the literature.[3,4,13]. We have followed up the patient for only a period of 6 months and so far, the site of tumour excision appears normal with no clinical and radiological evidence of tumour regrowth. However, a comprehensive report on tumour recurrence cannot be provided at this time of our report due to short follow up period. Patient would be followed up on a 6-monthly basis for an initial period of 3 consecutive years.

CONCLUSION

ESC is a rare tumour among Nigerians and Worldwide. Though benign, its aetiology and exact biologic nature remains unknown.

Recognition of its exact clinicopathological and histological presentation is therefore necessary for proper identification and management. A rare case of extra skeletal chondroma is reported, we recommend a large study and long term follow up in a multicenter study among Nigerians to ascertain the prevalence, gender, and site of ESC in this population.

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