Peripheral Ossifying Fibroma: A Case Report

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SOMMAIRE

Cet article décrit un cas de fibrome ossifiant périphérique chez une jeune fille de 12 ans. On y discute des caractéristiques cliniques, radiologiques et histologiques et formule des recommandations sur le diagnostic différentiel, le traitement et le suivi. On insiste aussi sur l'importance de maintenir une excellente communication avec le patient.

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Many types of localized reactive lesions may occur on the gingiva, including focal fibrous hyperplasia, pyogenic granuloma, peripheral giant cell granuloma and peripheral ossifying fibroma (POF).[1–3] These lesions may arise as a result of such irritants as trauma, microorganisms, plaque, calculus, restorations and dental appliances.[2,3] The purpose of this article is to present a case of POF, briefly review the current literature on this condition and emphasize the importance of discussion of a reasonable differential diagnosis with the patient or a parent.

Case Report

A healthy 12-year-old girl presented to the pediatric dental unit at the IWK Health Centre with a "lump behind her front teeth." She had been referred by her physician to the ear, nose and throat department, but was subsequently referred to the pediatric dentistry department. According to the patient, the "reddish purple lump" had been present for approximately 4 months and her mother stated that it had just recently become visible between the front teeth. As reported by the patient, the lump was interfering with her bite and felt uncomfortable, "similar to a canker sore." Occasionally, bleeding occurred when she brushed her teeth. During the consultation, it became apparent that the patient's mother was very concerned about the pathogenesis of the lesion. According to the mother, their family physician had discussed the possibility of the lesion being a carcinoma. This had raised the mother's anxiety level considerably.

Clinical Examination

Clinical examination revealed an erythematous maxillary central papilla visible from the facial aspect (**Fig. 1**). Palatally, the lesion appeared exophytic and nodular with an irregular surface (**Fig. 2**). It measured approximately 10 mm laterally, 8 mm in the anterior–posterior direction and 6 mm thick. It extended from 2 mm to the left of the palatal midline to 8 mm to the right of the midline. The lesion appeared reddish-pink with areas of white. It was slightly pedunculated with what appeared to be a broad-based attachment. The lesion was not fluctuant, nor did it blanch with pressure, but had a rubbery consistency. It was tender to firm pressure, but not to light palpation.

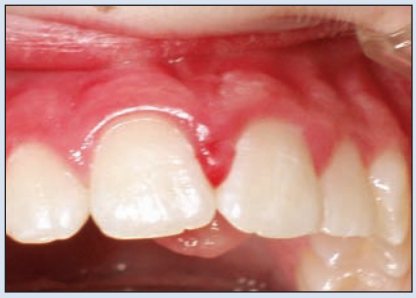

Figure 1: Facial view of an erythematous maxillary papilla just visible below incisors.

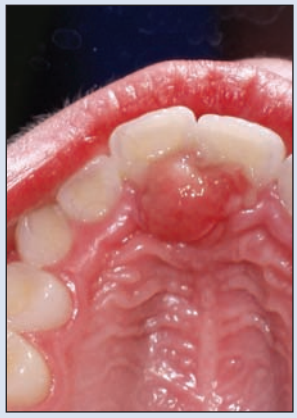

Figure 2: Palatal view of the lesion.

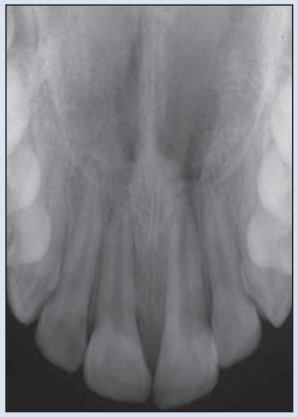

Figure 3: Maxillary occlusal radiograph showing normal aspect.

Radiographic Examination

Panoramic and maxillary occlusal radiographs were obtained. The radiographic examination was within normal limits, with no findings pertaining to the maxillary exophytic lesion (**Fig. 3**).

Diagnosis

The differential diagnosis consisted of irritation fibroma, pyogenic granuloma and peripheral giant cell granuloma (PGCG). This differential diagnosis was discussed with the patient and her mother in an attempt to alleviate fears of squamous cell carcinoma.

Treatment

Under general anesthesia, the lump was excised completely using both a scalpel and an electrocautery device. The removed tissue and adjacent periosteum measured 15 mm × 15 mm × 3 mm. The tissue was submitted to the oral pathology division for histopathologic diagnosis. Adjacent teeth were scaled to remove any local irritants.

Microscopic examination of the excised tissue revealed a gingival nodule that was partly ulcerated and partly lined with hyperparakeratinized stratified squamous epithelium with a normal maturation pattern. Much of the nodule consisted of hypercellular, well-vascularized fibrous connective tissue containing plump mesenchymal cells as well as numerous multinucleated giant cells. The specimen also exhibited a fairly large area of immature bone formation but no evidence of malignancy.

The histopathologic diagnosis was peripheral cemento-ossifying fibroma. The oral pathologist was contacted by the author to confirm that the terms POF and peripheral cemento-ossifying fibroma could be used interchangeably. The pathology report stated that portions of the fibroma showed typical areas of pyogenic granuloma as well as smaller areas of PGCG.

Follow-up

The patient presented for a follow-up examination 20 days postoperatively. The surgical site appeared to be healing well (**Fig. 4**). There was no evidence of recurrence of the lesion, and the child was asymptomatic.

Discussion

Intraoral ossifying fibromas have been described in the literature since the late 1940s. Many names have been given to similar lesions, such as epulis,[1] peripheral fibroma with calcification,[1] peripheral ossifying fibroma,[2,3] calcifying fibroblastic granuloma,[4] peripheral cementifying fibroma, peripheral fibroma with cementogenesis[5] and peripheral cemento-ossifying fibroma.[6] The sheer number of names used for fibroblastic gingival lesions indicates that there is much controversy surrounding the classification of these lesions.[5,7]

It has been suggested that the POF represents a separate clinical entity rather than a transitional form of pyogenic granuloma, PGCG or irritation fibroma.[1] Eversole and Rovin[2] stated that, with the similar sex and site predilection of pyogenic granuloma, PGCG and POF, as well as similar clinical and histologic features, these lesions may simply be varied histologic responses to irritation. Gardner[3] stated that POF cellular connective tissue is so characteristic that a histologic diagnosis can be made with confidence, regardless of the presence or absence of calcification. Buchner and Hansen[8] hypothesized that early POF presents as ulcerated nodules with little calcification, allowing easy misdiagnosis as a pyogenic granuloma. Several publications[2,3,7–9] address

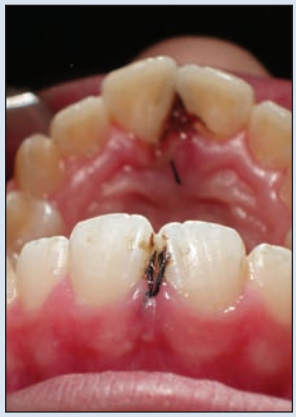

Figure 4: Palatal view of surgical site showing satisfactory healing 20 days after surgery.

the issue of histologic differentiation in depth, but this is beyond the scope of this article.

When presented clinically with a gingival lesion, it is important to establish a differential diagnosis. In this case, the clinical features led to a differential diagnosis of irritation fibroma, pyogenic granuloma or PGCG. Although it is also important to maintain a high index of suspicion, discussion with family members should be tactful to prevent undue distress during the waiting period between differential diagnosis and definitive histopathologic diagnosis.

Because the clinical appearance of these various lesions can be remarkably similar, classification is based on their distinct histologic differences. The POF must be differentiated from the peripheral odontogenic fibroma (PODF) described by the World Health Organization.[3,8] Histologically, the PODF has been defined as a fibroblastic neoplasm containing odontogenic epithelium.[9] Despite a preponderance of literature supporting differentiation, some authors continue to argue that the POF (or peripheral cemento-ossifying fibroma) is the peripheral counterpart of the central cemento-ossifying fibroma.[6]

The POF, as discovered in this case, is a focal, reactive, non-neoplastic tumour-like growth of soft tissue often arising from the interdental papilla.[1,3] It is a fairly common lesion, comprising nearly 3% of oral lesions biopsied in 1 study,[1] approximately 1%–2% in other studies.[9–11] In 1993, Das and Das[12] obtained similar results, with 1.6% POFs among 2,370 intraoral biopsies.

POF may present as a pedunculated nodule, or it may have a broad attachment base.[1,11,13] These lesions can be red to pink with areas of ulceration, and their surface may be smooth or irregular. Although they are generally < 2 cm in diameter,[8,13] size can vary; reports range from 0.2–3.0 cm[8,11] to 4 mm–8 cm[1,14] and some lesions may be as large as 9 cm in diameter.[15] Cases of tooth migration and bone destruction have been reported, but these are not common.[15]

The female to male ratio reported in the literature varies from 1.22:1[16] and 1.7:1[8–10] to 4.3:1.[2] By most reports, the majority of the lesions occur in the second decade, with a declining incidence in later years.[1,2,8–10] There are 2 reported cases of POF present at birth, presenting clinically as congenital epuli.[17,18] In a 2001 study, Cuisia and Brannon[11] reported that only 134 out of 657 diagnosed POFs (20%) were in the pediatric population (0–19 years), with 8% in the first decade. In a retrospective study of 431 cases in the Chinese population by Zhang and others,[16] the mean age of incidence of POF was found to be 44 years, which is contradictory to previously published literature. POF appears to be more common among white people than black[11] and slightly less common among those of Hispanic origin.[12]

The lesion may be present for a number of months to years before excision, depending on the degree of ulceration, discomfort and interference with function.[1,8] Approximately 60% of POFs occur in the maxilla,[8,9,16] and they occur more often in the anterior than the posterior area,[9,12,16] with 55%–60% presenting in the incisor-cuspid region.[1,2,8,11,16]

POFs are believed to arise from gingival fibres of the periodontal ligament as hyperplastic growth of tissue that is unique to the gingival mucosa.[1–3,19] This hypothesis is based on the fact that POFs arise exclusively on the gingiva, the subsequent proximity of the gingiva to the periodontal ligament and the inverse correlation between age distribution of patients presenting with POF and the number of missing teeth with associated periodontal ligament.[9,11,19] In a study of 134 pediatric patients with POF,[11] in only 2 cases was POF intimately associated with primary teeth, bringing into question the reactivity of the lesion. The exfoliation of primary teeth and eruption of their successors should result in an increased incidence of periodontal ligament-associated reactive lesions.[2,11]

Hormonal influences may play a role, given the higher incidence of POF among females, increasing occurrence in the second decade and declining incidence after the third decade.[9] In an isolated case of multicentric POF, Kumar and others[5] noted the presence of a lesion at an edentulous site in a 49-year-old woman, which once again raises questions regarding the pathogenesis of this type of lesion.

Histologically, the POF appears to be a nonencapsulated mass of cellular fibroblastic connective tissue[3] of mesenchymal origin, covered with stratified squamous epithelium, which is ulcerated in 23%–66% of cases.[1,8] Most ulcerated lesions occur in patients in the second decade.[2,8] POFs contain areas of fibrous connective tissue, endothelial proliferation and mineralization. Endothelial proliferation can be profuse in the areas of ulceration, which can be misleading in clinical diagnosis, as the lesion may appear to be a pyogenic granuloma.[1] The mineralized component of POF varies,

occurring in approximately 23%,[16] 35%[9] or 50%–75%[1,7,8] of cases according to published reports. Mineralization can vary between cementum-like material, bone (woven and lamellar) and dystrophic calcification.[1,7,8] The POF lesion is generally small and does not require imaging beyond radiographs.[3,20]

Treatment consists of conservative surgical excision[1,9] and scaling of adjacent teeth.[3] The rate of recurrence has been reported at 8.9%,[1] 9%,[11] 14%,[9] 16%[8] and 20%.[2] Therefore, regular follow-up is required.

Conclusions

POF is a slowly progressing lesion, the growth of which is generally limited. Many cases will progress for long periods before patients seek treatment because of the lack of symptoms associated with the lesion. A slowly growing pink soft tissue nodule in the anterior maxilla of an adolescent should raise suspicion of a POF. Discussion of the differential diagnosis should be done tactfully to prevent unnecessary distress to the patient and family. Zhang and others[16] noted that cancer was included in the differential diagnosis in only 2% of cases. In the current case, the family experienced distress related to the suggestion of squamous cell carcinoma before referral for treatment and definitive diagnosis. Treatment consists of surgical excision, including the periosteum, and scaling of adjacent teeth. Close postoperative follow-up is required because of the growth potential of incompletely removed lesions and the 8%–20% recurrence rate.

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Juvenile Idiopathic Arthritis: A Chronic Pediatric Musculoskeletal Condition with Significant Orofacial Manifestations

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SOMMAIRE

«Arthrite idiopathique juvénile» (AIJ) est une expression générale, utilisée pour décrire un groupe cliniquement hétérogène d'arthrites de cause inconnue se manifestant avant l'âge de 16 ans. Bien que l'AIJ se caractérise principalement par une inflammation chronique des articulations, cette expression englobe plusieurs catégories de maladies. L'étiologie de l'AIJ demeure mal comprise, et aucun des médicaments actuellement disponibles ne peut guérir la maladie. Le pronostic s'est toutefois grandement amélioré grâce aux progrès réalisés dans la classification et la prise en charge de la maladie. Les dentistes devraient se familiariser avec les symptômes et les manifestations buccales de l'AIJ pour aider à son traitement.

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Juvenile idiopathic arthritis (JIA) is the most common chronic rheumatic disease of childhood and an important cause of short- and long-term disability.[1] Patients with JIA experience a myriad of symptoms, including lethargy, reduced physical activity, poor appetite and flu-like symptoms. Although the initial manifestation of JIA is variable, the cardinal clinical features include persistent swelling of one or more joints, limited range of motion in the joints and pain during movement lasting at least 6 weeks. The age at onset of JIA is under 16 years of age.[1] In the worst expression of JIA in the face, these patients may exhibit severe retrognathia, open bite, microgenia and "bird-like" facies (**Figs. 1a** and **1b**).

Like other forms of arthritis, JIA is characterized by inflammation of the synovium of one or more joints. However, the term JIA has replaced previous terms such as *juvenile chronic arthritis* or *juvenile rheumatoid arthritis* to more accurately identify homogenous groups of children with distinct clinical features. The International League of Associations for Rheumatology (ILAR), which has provided the most recent classification, identifies 7 subtypes of JIA with specific exclusion and inclusion criteria[2] (**Tables 1** and **2**). Females are much more frequently affected with almost all types of JIA than males.[1,3,4] The worldwide prevalence of JIA varies between 16 and 150 per 100,000; the frequency of different subtypes of JIA vary with location and ethnicity.

Pathogenesis

Inflammation of the synovium is a key pathological feature of JIA. However, the exact trigger and factors that allow the inflammation