Case Report

Peripheral Cemento-ossifying Fibroma Associated with an Unerupted Tooth

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Abstract:

Peripheral cemento-ossifying fibroma (PCOF) is a relatively rare tumor classified between fibro-osseous lesions. This lesion is more common in the maxilla than the mandibula. About 60% of the tumors occur in the maxilla and also more than 50% of all lesions affect the anterior region of the maxilla. The management of a pediatric patient with PCOF requires early diagnosis and treatment. Appropriate treatment protocol with close post-operative follow-up is required for these patients. We report here, the clinical case of a 9-year-old girl, with the disease duration of 1 year and was followed-up for 2 years post-surgically showing gingival health, normal radio-opacity of bone without any recurrence.

Keywords: Fibro-osseous Lesion, Oral Tumors, Peripheral Cemento-ossifying Fibroma, Pyogenic Granuloma.

Introduction:

Peripheral cemento-ossifying fibroma (PCOF) is a common benign osteogenic neoplasm of odontogenic origin represented about 3.1% of all oral the tumours and 9.6% of the gingival lesions (1). The 1992 World Health Organization (WHO) classification included it in the group of a single designation of cemento-ossifying fibroma with two histologic types, which are difficult to distinguish both clinically and radiographically (2). Therefore, biopsy and histopathological examinations are necessary for definitive diagnosis (3). These lesions may form as a result of irritants such as microorganisms, plaque, trauma, calculus, dental orthodontic appliances, ill-adapted crowns and irregular restorations (4).

Several types of localized reactive lesions may develop on the gingiva, including pyogenic granuloma, focal fibrous hyperplasia, peripheral giant cell granuloma, and PCOF (5). The pathogenesis of this tumor is uncertain. The mineralized product of this lesion probably originates from the periodontal ligament or from the periosteal cells (6). PCOF appears as a pedicled or cerebellar nodular mass originating from

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interdental papilla in a clinical examination. The color of the lesion ranges from red to pink frequently but not always while the surface is ulcerated (7,8). There is a slightly higher predilection for the maxillary arch (60%) and the incisor cuspid region (50%) but it can also be found in the mandible (9).

Clinically and histopathologically, this lesion has two forms: generalized and juvenile (10). The generalized form is in the most adult individuals, and because of the slow growth, it causes the patient to complain at a later stage (11,12). In contrast to the generalized type, the juvenile form is more aggressive in most cases, exhibiting rapid growth and short-lived facial swelling. The juvenile type is seen in earlier ages and most commonly in the maxilla, and less commonly in the mandibular (10). This type lesion predominantly affects adolescents and young adults, with peak prevalence between 10 and 19 years (7,9,13).

Although PCOF affects both genders, it is reported that the incidence is higher in women. The lesion is generally asymptomatic until growth produces a noticeable swelling and mild deformity. Displacement of teeth is an early clinical feature. Recurrence rate is considered to be high (8 %- 20 %) (14).

Radiologic features of the lesion appears well circumscribed and demarcated from surrounding bone. It initially shows no calcification but as lesion matures, there is increasing calcification. The radioluscent area becomes flecked slowly with radio-opacities until lesion appears extremely radio-opaque.

The purpose of this case report is to present clinical and histological findings of a pediatric patient with PCOF.

Case presentation:

A 9-year-old female patient referred to our pediatric department clinic with a chief complaint of pain during mastication in her upper left incisors region since 1 year. There was no relevant medical and family history. No trauma or fracture of the jaws.

Intraoral examination showed a hemorrhagic mass extended from the buccal to the palatinal side, measuring approximately 2.5 cm mesiodistally and about 2 cm buccolingually located in the upper left central and lateral incisors (Fig. 1). The overlying mucosal surface appeared pinkish color with erythematous patches. The gingival overgrowth was not firm in consistency, nontender on palpation. Uneruption of upper left central incisor was probably because of excessive pressure from the gingival growth.

The panoramic radiograph revealed a soft tissue shadow extending from upper left central to lateral incisor (Fig. 2). Intraoral periapical radiograph showed bone tissue was within normal limits, with no finding of any lesion. A provisional diagnosis of pyogenic granuloma and peripheral giant cell granuloma was established.

The lesion was excised completely under local anesthesia. The excisional biopsy was submitted for histopathologic analysis, which revealed that it was PCOF. Patient was followed up at regular intervals. The

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eruption of the related tooth was followed clinically and radiographically. No recurrence was noted on the second year follow-up visit (Fig. 3-4).

Histopathologic Examination:

In histopathological examination surface of the lesion was widely ulcerative (Fig. 5). Under the ulcerative surface the lesion composed of spindle shaped fibroblasts. These cells showed whorled, storiform pattern. Immature bone trabeculae and spheroid cementum-like material were also observed in the vascular rich lesion (Fig. 6).

Discussion:

PCOF is a reactive, non-neoplastic tumour-like growth of soft tissue lesion commonly arising from the region of the interdental papilla. It consists of fibrous tissue with variable amounts of calcific material resembling bone, cementum, or both (15). Although etiopathogenesis of PCOF is stil unclear, it has been suggested that these lesions originate from periodontal ligament cells (16,17). PCOF is often associated with local irritants, such as subgingival plaque, calculus, or trauma from dental appliances and poor-quality dental restorations. Lesions usually involve gingival soft tissue, very rare bone involvement can be seen (17). The incidence of the lesion is higher in females than in males (18), with a peak incidence between 2nd and 3rd decades (9,14,18). For this reason, it was thought that hormonal effects may also play a role in the growth of this tissue (19). It was reported that the prevalence of PCOF is 1.9% in children aged between 0 to 9 years, while Cuisia and Brannon reported a 10% prevalence of these lesions in children between 5 and 9 years of age (19). In addition to these, it is reported in the literature that there are 2 PCOF cases in the newborn (20).

Endo et al. has made an attempt that using immunohistochemical analysis to distinguish cementifying fibromas from fibrous dysplasias and ossifying fibromas for keratin sulfate and chondroitin-4-sulfate. As a result of the analysis, while fibrous dysplasia showed intense immunostaining for chondroitin-4-sulfate, cementifying fibromas showed significant immunereactivity for keratin sulfate (17).

About 60% of tumours occur in maxilla, with anterior gingiva being most commonly involved. More than 50% of all cases affect the region of the incisors and canines. Clinically, PCOF is a slow-growing asymptomatic gingival mass measuring less than 2 cm in diameter and located more commonly in interdental gingiva (18,21). As the size of the tumor increases, it causes pain, and functional changes (5). It may be sessile or pedunculated, identical in color to that of gingiva or slightly reddish, and may have an ulcerated surface. PCOF is more firm and less friable than other lesions and generally has a longer course, which explains the calcification and/or ossification (9). Radiographically, some, but not all lesions, may show foci of calcifications scattered in the central area of the lesion. Radiographs usually do not reveal underlying bone involvement but rarely superficial erosion of bone may be seen (22). In certain cases it is difficult to differentiate between pyogenic granuloma, fibrous hyperplasia, and peripheral giant cell

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granuloma (23). Histological examination is imperative for definitive diagnosis, which is based on focal presence of bone or other calcifications in cellular connective tissue (9,19). This emphasizes the need for histopathological examination of biopsy specimen for an accurate diagnosis because of difficulty in diagnosing PCOF based only on clinical and radiographical observations (24,25). Treatment of PCOF consists of removal of etiological factors, and aggressive surgical excision of the lesion including the periodontal ligament and the periosteum (19,25). PCOF reportedly has high recurrence rate of approximately 20%, hence long-term postoperative follow-up is imperative (19).

Conclusion

PCOF is a slowly progressing lesion and its growth is limited. Histopathological examination is essential for accurate diagnosis. Because the high recurrence rate of the lesion, post-operative follow-up is necessary.

References:

1. Delbem A, Cunha R, Silva J, Soubhia A. Peripheral cement-ossifying fibroma in child. A follow up of four years report of a case. Eur J Dent. 2008;2:134-37.
2. Gardner DG. The peripheral odontogenic fibroma: An attempt to classification. Oral Surg Oral Med Oral Pathol. 1982;54:40-48.
3. Kumar SKS, Ram S, Jorgensen MG, Shuler CFS, Edghizadeh PP. Multicentric peripheral ossifying fibroma. Journal of Oral Science. 2006;48(4): 239-243.
4. Mishra AK, Maru R, Dhodapkar SV, Jaiswal G, Kumar R, Punjabi H. Peripheral cement-ossifying fibroma: A case report with review of literature. World J Clin Cases. 2013;1(3): 128-133.
5. Bhaskar SN, Jacoway JR. Peripheral fibroma and peripheral fibroma with calcification: report of 376 cases. J Am Dent Assoc. 1966;73:1312–1320.
6. Yadav R, Gulati A. Peripheral ossifying fibroma: a case report. J Oral Sci. 2009;51(1): 151-4.
7. Ganji KK, Chakki AB, Negaral SC, Verma E. Peripheral cemento-ossifying fibroma: Case series. Hindawi Publishing Corporation Case Reports in Dentistry. 2013;2013:1-6.
8. Hannemann JAC, Pereira AAC, Ribeiro Junior NV, Oliveira DT. Peripheral ossifying fibroma in a child: report of case. J Clin Pediatr Dent. 2003;27:283-285.
9. Neville BW, Damm DD, Allen CM, Bouquot JE. Text book of oral and maxillofacial pathology. 2nd ed. Philadephia: W.B. Saunders Co; 2004. p. 451-452.
10. Hemalatha VT, Austin RD, Mathew P. Juvenile Ossifying Fibroma Masquerading as Maxillary Sinus Malignancy: Case Report: Turkiye Klinikleri J Dental Sci. 2012;16: 305-310.
11. Smith SF, Newman L, Walker DM. Papadopoulus H. Juvenile aggressive psammomatoid ossifying fibroma: an interesting, challenging, and unusual case report and review of the literature. J Oral Maxillofac Surg. 2009;67:200-206.
12. Speight PM, Carlos R. Maxillofacial fibro-osseous lesions. Current Diag Pathol. 2006; 12:1-10.

[DOR: 20.1001.1.23222913.2019.8.4.4.0]

13. Das S, Das AK. A review of pediatric oral biopsies from a surgical pathology service in a dental school. Pediatric Dentistry. 1993;15(3):208–211.
14. Farquhar T, Maclellan J, Dyment H, Anderson RD. Peripheral Ossifing Fibroma: A Case Report. JCDA. 2008;74: 9.
15. Guru SR, Singh SS, Guru RC. Peripheral cement-ossifying fibroma: A report of two cases. Journal of Health Sciences& Research. 2016;7(2): 71-75.
16. Zain RB, Fei YJ. Fibrous lesions of the gingiva: a histopathologic analysis of 204 cases. Oral Surg Oral Med Oral Pathol.1990; 70: 466-470.
17. Endo Y, Uzawa K, Mochida Y, Nakatsuru M, Shiiba M, Yokoe H et al. Differential distribution of glycosamino-glycans in human cementifying fibroma and fibro-osseous lesions. Oral Dis. 2003;9:73-76.
18. Zhang W, Chen Y, An Z, Geng N, Bao D. Reactive gingival lesions: a retrospective study of 2,439 cases. Quintessence Int. 2007;38(2):103-110.
19. Cuisia ZE, Brannon RB. Peripheral ossifying fibroma-a clinical evaluation of 134 pediatric cases. Pediatr Dent. 2001;23:245-248.
20. Kohli K, Christian A, Howell R. Peripheral ossifying fibroma associated with a neonatal tooth: case report. Pediatr Dent.1998;20(7): 428-9.
21. Kurdukar PA, Kurdukar AA, Chaudhari VV. Peripheral cement-ossifying Fibroma- A Clinical and Histomorphological Case Report. International Journal of Contemporary Medical Research. 2016;3:2020-2022.
22. Kendrick F, Waggoner WF. Managing a peripheral ossifying fibroma. J Dent Child. 1996;63:135–138.
23. Ashish Y, Mishra MB. Peripheral cement-ossifying fibroma of mandible: a case report. Indian J Stomatol. 2011;2(3):193-196.
24. Nagaraj T, Saxena S, Ghouse N, Sahu P, Biswas A. Peripheral cement-ossifying fibroma-A case report. Journal of Advanced Clinical&Research Insights. 2018;5(2):44-47.
25. Sachdeva SK, Mehta S, Sabir H, Rout P. Peripheral cement-ossifing fibroma with uncommon clinical presentation: A Case Report. Int J Dent Sci. 2018;20(1):17-23.

Tables and Charts:

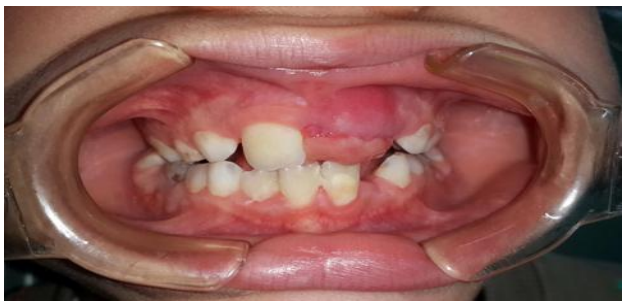

Figure 1: Localized gingival enlargement of the upper left maxilla.

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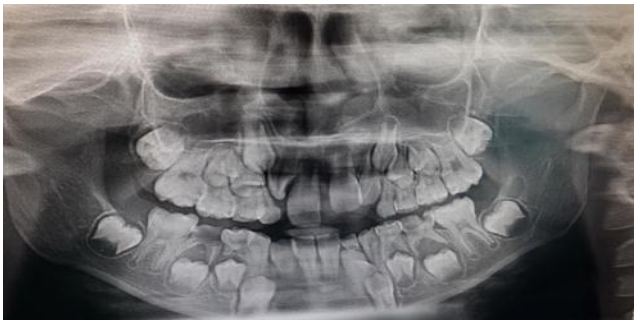

Figure 2: Panoramic x-ray revealed delayed eruption of upper left central incisor**.**

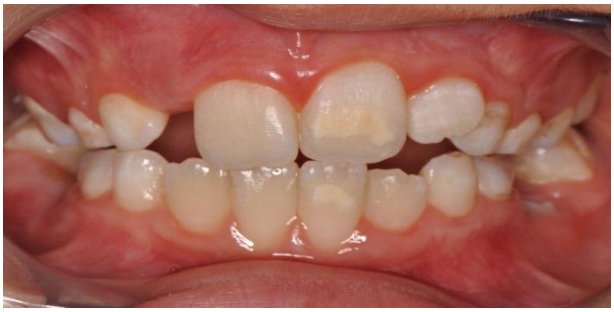

Figure 3: Upper left central and lateral incisors normally erupted after a 2-year follow-up**.**

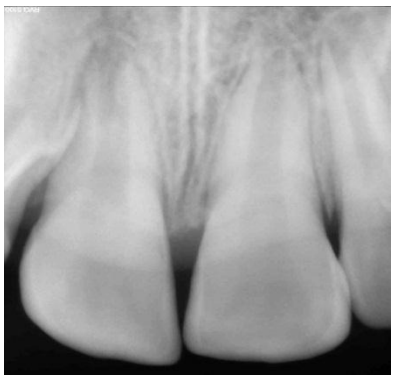

Figure 4: At 2-year follow-up, the periapical area of the left central incisor has remained normal**.**

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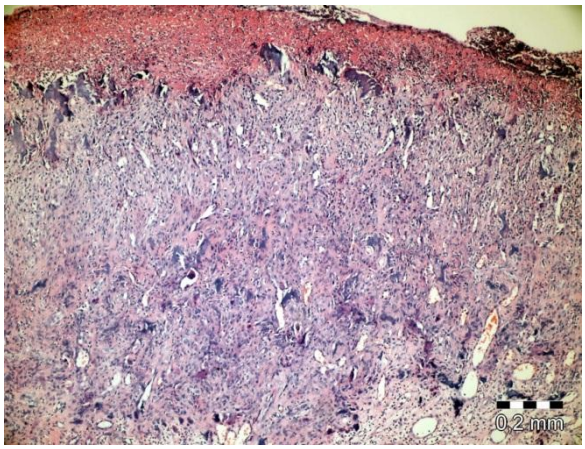

Figure 5: Ulcerated oral mucosa and well-circumscribed solid tumor mass composed of fibroblastic stroma H&E X100**.**

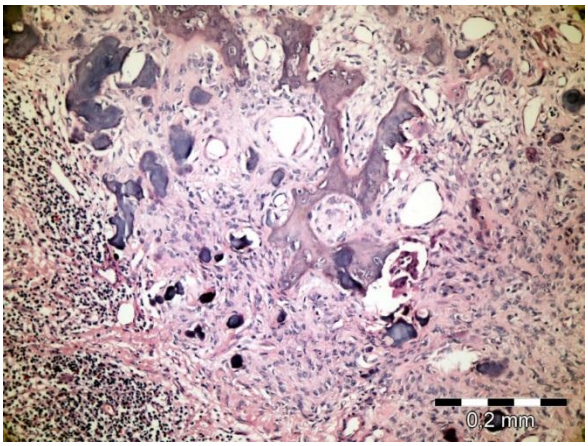

Figure 6: Trabeculae of bone and basophilic acellular spherical cementum-like material in fibroblastic matrix H&E 200**.**

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