PERIPHERAL CEMENTO-OSSIFYING FIBROMA OF THE ANTERIOR MAXILLARY GINGIVA (PERIPHERAL CEMENTO-OSSIFYING FIBROMA PADA GINGIVA MAKSILA ANTERIOR)

Teja Koswara*1

¹Department of Anatomical Pathology, Faculty of Medicine, Universitas Jenderal Achmad Yani, Cimahi Indonesia

JHDS.unjani.ac.id/jite Doi: 10.54052/jhds.

Article History Received: 26/06/2024 Accepted: 03/07/2024 *Corresponding author tejakoswr@yahoo.com

ABSTRACT

Peripheral cemento-ossifying fibroma (PCOF) is a reactive gingival growth due to trauma or local chronic irritation. It occurs in about 2% of all intraoral lesions or about 9.6% of all gingival lesions. Clinical differential diagnoses of PCOF are Peripheral giant cell granuloma, pyogenic granuloma or epulis granulomatosa, irritation fibroma, and osteoid osteoma. This report describes a case of a 39-year-old female with sessile growth measuring 2x2x1 cm on the anterior maxillary gingiva at 11 and 21 region. Clinical diagnosis was epulis An excisional biopsy was performed, and a granulomatosa. histopathological diagnosis was made of peripheral cemento-ossifying fibroma. PCOF is more common in females in the second decade of life and is usually seen in the anterior Maxilla. Lesions are sessile or polypoid growth with normal overlying surface or reddish—generally less than 2 cm. Radiology examinations are usually within normal limits, but sometimes, there are foci of calcification seen within the lesion, superficial bone erosion, or loss of interdental bone with the treatment with excisional biopsy and curettage for periosteum and periodontal ligament tissue. Also, it is essential to eliminate some

irritating factors to prevent recurrence. Recurrence rate is high, about 20% of cases. Clinical features of PCOF were similar to other more common gingival lesions such as epulis or fibroma. Histopathological examination was critical to make a definitive diagnosis.

Keywords: gingiva; maxilla; peripheral cemento-ossifying fibroma

ABSTRAK

Peripheral cemento-ossifying fibroma (PCOF) merupakan pertumbuhan reaktif pada gingiva yang disebabkan oleh trauma atau iritasi kronik lokal. Angka kejadiannya jarang, yaitu hanya sekitar 2 % dari seluruh lesi intraoral atau sekitar 9,6% dari seluruh lesi di gingiva. Diagnosis diferensial dari kelainan ini secara klinis antara lain: Peripheral giant cell granuloma, pyogenic granuloma atau epulis granulomatosa, irritation fibroma, dan osteoid osteoma. Tulisan ini melaporkan tentang seorang wanita berusia 39 tahun dengan massa sessile berukuran 2x1x1 cm pada gingiva maksila anterior di regio gigi 11 dan 21. Diagnosis awal adalah epulis granulomatosa. Pada pasien ini dilakukan eksisi bedah dan pemeriksaan patologi anatomi dengan diagnosis histopatologi peripheral cemento-ossifying fibroma. PCOF lebih sering ditemukan pada wanita, usia dekade kedua dengan lokasi yang paling sering pada gingiva maksila anterior. Lesi berupa pertumbuhan sessile atau polypoid dengan warna yang sama dengan jaringan sekitar atau sedikit hiperemik. Umumnya berukuran kurang dari 2 cm tetapi dapat berukuran besar mencapai 10 cm. Pada pemeriksaan radiologi biasanya tidak didapatkan kelainan, tetapi pada sebagian kecil kasus dapat ditemukan fokus-fokus kalsifikasi pada lesi, erosi pada bagian superfisial tulang atau hilangnya tulang interdental. Penatalaksanaan dengan eksisi bedah, kuretase jaringan periosteum dan ligamentum periodontal serta menghilangkan faktor-faktor yang dapat menyebabkan iritasi lokal untuk mencegah rekurensi. Kejadian

rekurensi pada PCOF cukup sering yaitu mencapai 20 % kasus. Gambaran klinis dari PCOF sukar untuk dibedakan dengan massa gingiva lainnya yang lebih umum seperti epulis atau fibroma. Diagnosis pasti hanya dapat ditegakkan melalui pemeriksaan histopatologi.

Kata kunci: gingiva; maksila; peripheral cemento-ossifying fibroma

INTRODUCTION

Peripheral cemento-ossifying fibroma (PCOF) is a reactive growth on the gingiva due to various factors that can cause local irritation, such as Calculus, plaque, microorganisms, tools used for orthodontic treatment or poor dental restoration. ^{1,2} This disorder is different from central cemento-ossifying fibroma, which is an intraosseous tumour (neoplasm). ³ The abnormal tissue in PCOF is thought to originate from the periodontal ligament, where pluripotent cells from the ligament can differentiate into osteoblast, cementoblast or fibroblast cells. ^{3,4}

PCOF generally does not cause damage to the underlying bone structure, but in a small number of cases, erosion can be found in the superficial part of the bone.

5,6,7 The most common location is the interdental papilla of the maxillary gingiva anterior to the molar teeth.

8,9 Approximately 60% of cases are located in the Maxilla, and more than 50% of cases are located in the region between the incisor

and canine teeth. ¹⁰ The incidence of PCOF is approximately 2% of all intraoral lesions biopsied. 11 PCOF is more common in women, children and young adults, with the highest incidence in the second decade. 8,9,10,11,12 The incidence rate decreases after the third decade. 10 Lesions can be sessile or pedunculated growths. Most measurements are less than 2 cm, but larger sizes can also be found. The colour is generally the same as the surrounding gingival tissue or slightly reddish. 11,12,13,14 Ulceration can be found on the surface of the lesion. 12,13 The incidence of recurrence is relatively high, reaching 20% of cases. ¹⁵ Recurrence is often caused by incomplete excision, repeated trauma or persistent local irritant factors. 15

Management of PCOF is surgical excision, curettage of the involved periosteum and periodontal ligament tissue and removal of various local etiological factors that irritate to prevent recurrence. Histopathological examination is necessary to establish a definite diagnosis. 6,8,17,18

CASE REPORT

A 39-year-old woman came to the dental and oral clinic complaining of a lump on the gums at the front of the upper jaw, which had been felt for approximately three months. The lump is felt to be getting bigger, painless, and bleeds easily. On physical examination, vital signs and generalist status were within normal limits. No abnormalities were seen during the examination. On extra-oral intraoral examination in the area of teeth 11 and 21, a stemless lump on the gingiva was seen measuring 2x1x1 cm with the same colour as the surrounding tissue: flat surface, no visible ulcers. Calculus is visible on the surrounding teeth. The mass is not fixed in the underlying bone tissue. There were no occlusion disorders or complaints of trismus. The clinical diagnosis based on the history and physical examination results was granulomatous epulis. It is planned to carry out an excisional biopsy. The results of laboratory examinations (routine blood, BT, CT, liver function tests and glucose were within normal limits. No radiological examination was carried out, either periapical or panoramic photographs. In this excisional patient, an biopsy performed, and the tissue specimen was sent to the anatomical pathology laboratory

for histopathological examination. The specimens are two tissues measuring 0.8x 0.7 x 0.5 cm and 0.5 x 0.5 x 0.5 cm, brownish in colour and springy in consistency (Figure 1).



Figure 1. The tissue specimen measures 0.8x 0.7 x0.5 cm and 0.5x0.5 x0.5 cm, brownish with a smooth surface.

On microscopic examination, it appears that both tissues are lined with hyperplastic squamous epithelium (Figure 2), and some have experienced erosion in contact with neutrophil inflammatory cells mononuclear inflammatory (Figure 2). In the underlying stroma, there is visible proliferation of fibroblast cells and fibrocollagen connective tissue accompanied by deposits of basophilic amorphous masses that resemble cementum / "cementum-like calcification" (Figure 3). In one section, lamellar bone tissue is visible (Figure 4). Cell nuclei were within normal limits. No mitoses were found. Based on these images, the conclusion of histopathological examination was

peripheral cemento-ossifying fibroma.

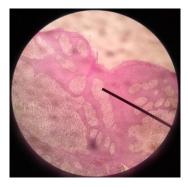


Figure 2. The squamous epithelium lining the gingival tissue appears to be hyperplastic, characterized by thickening of the stratum spinosum and elongation of the rete ridges. This picture is also called 'pseudoepitheliomatous hyperplasia' and is often found in peripheral cementoossifying fibromas. (H&E, Obj, 10x).

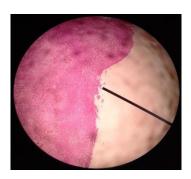


Figure 3. Some of the gingival squamous epithelial layers have experienced erosion accompanied by an inflammatory reaction characterized by neutrophil inflammatory cells and mononuclear inflammatory cells.

Erosion of the surface squamous epithelium is sometimes found in peripheral cemento-ossifying fibromas indicating tissue damage due to local irritation. (H&E, Obj, 10x).

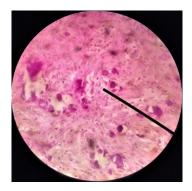


Figure 4. Locally in the connective tissue stroma, basophilic amorphous mass deposits with calcifications that resemble cementum masses are visible, namely a calcified mesenchymal tissue that forms the outer layer of tooth roots. In the stroma, the proliferation of fibroblast cells was seen with cell nuclei within normal limits (H&E, Obj, 10x).

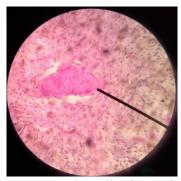


Figure 5. Mature lamellar bone tissue can be seen with osteocyte cells between the bone matrix in the connective tissue stroma. No osteoclasts cells or osteoid masses were seen. This strengthening occurs through the process of membranous ossification. (H&E, Obj, 10x).

DISCUSSION

Peripheral cemento-ossifying fibroma is a reactive (non-neoplastic) growth on the gingiva which is thought to originate from the gingival connective tissue, periodontal ligament or periosteum tissue of the alveolar bone. ^{3,4}. Many different diagnostic terminologies are used

for this disorder. Shepherd, in 1844, first mentioned this disorder as "alveolar exostosis". Bhasker et al. (1984) called this disorder "peripheral fibroma calcification". Some of the terminology used for this disease: Peripheral ossifying fibroma, peripheral cemento-ossifying fibroma, peripheral cementifying fibroma, peripheral fibroma with calcification, ossifying fibro-epithelial polyp, peripheral fibroma with cement genesis, peripheral fibroma with osteogenesis, calcifying or ossifying fibrous epulis and calcifying fibroblastic granuloma. ³ The incidence rate is rare, namely around 9.6% of all gingival masses. ¹¹ The clinical differential diagnosis of this disorder includes peripheral giant cell granuloma, pyogenic granuloma or epulis granulomatosa, irritation fibroma, fibrous hyperplasia and osteoid osteoma. ^{2,3,6} The pathogenesis of this disorder is still unclear. Some say it may be a secondary development (fibrosis) of granulation tissue because, clinically, it resembles pyogenic granuloma. There is also an opinion that links it to hormonal factors because of its predilection, which is more common in women and is often found in the second decade of life. The most widely accepted theory regarding its pathogenesis is hyperplasia of cells in the periosteum and periodontal ligament due to inflammation.

Chronic irritation of the periosteum and periodontal ligament causes metaplasia of the connective tissue, dystrophic calcification and ossification.^{3,4,11}

Lesions in PCOF can be sessile or pedunculated growths on the gingiva with the same colour as the surrounding tissue or more hyperemic, and they can be accompanied by ulceration on the surface. The consistency can be chewy or hard if it contains a lot of bone components. The most frequent location is in the anterior Maxilla between the incisors and canines. ^{11,12,13,14} PCOF can experience rapid growth due to the influence of various growth factors such as Fibroblast growth factor and vascular endothelial growth factors, as well as other factors such as nitric oxide synthetase.17,18 In this case, the lesion was a sessile growth with the same colour as the surrounding tissue without any ulceration on the maxillary anterior gingiva in the tooth region ^{11,21}, namely in the middle part between the upper right and left incisors.

PCOF is more common in women (2-4 times more often than men) in the second decade of life, when the incidence rate decreases with increasing age. PCOF can occur at any age but rarely occurs under ten years of age. Two cases of PCOF were reported in a newborn baby and a 3-month-old baby. ¹⁶ In this case, the patient is a 39-

year-old woman whose age is slightly older than the age of most PCOF patients in general, namely in the second decade.

Generally, PCOF is less than 2 cm in size, but it has been reported as large as 10 cm. It can cause facial deformity, shifting teeth or occlusion disorders. ¹⁹ In this case, the lesion was measured at 2x1x1 cm without extra-oral abnormalities or occlusion and trismus. On post-biopsy intraoral examination, a central diastema was seen on teeth ^{11,21}, which indicates a shift of the incisors at the location of the lesion.

On radiological examination, no abnormalities are usually found. Sometimes, intra-lesion calcification may also be seen depending on the degree of mineralization. Other features that can be found are erosion of the superficial part of the bone and loss of interdental bone. ^{18,19} In this case, no radiological examination was carried out because the initial clinical diagnosis was granulomatous epulis.

The definitive diagnosis of PCOF is through histopathological examination. The microscopic picture of PCOF is a proliferation of fibroblast cells and connective tissue accompanied by basophilic masses that resemble cementum ("cementum-like calcifications") and bone tissue (ossification). The surface squamous epithelial layer of the gingiva often

experiences hyperplasia and can be accompanied by ulceration. ^{3,6,17,18} In this case, the histopathological picture is in accordance with the microscopic picture of PCOF; namely, there is a proliferation of fibroblast cells and connective tissue, "cementum-like calcification", and bone tissue.

Management of PCOF is surgical excision of the lesion using a scalpel or electrosurgery technique accompanied by curettage of the tissue around the lesion, including the periosteum and periodontal ligament, to prevent recurrence. Besides that, it is important to eliminate various etiological factors that can cause local irritation and educate patients to maintain oral hygiene. ^{15,16,20} With adequate surgical treatment, the prognosis is generally good. ²⁰ The presence of lesion tissue remaining after surgery can cause recurrence. The incidence of recurrence in PCOF ranges from 8-20%. ¹⁵

CONCLUSION

Peripheral cemento-ossifying fibroma is a reactive growth on the gingiva due to trauma or chronic irritation. This disorder is rarely found and has frequent recurrences. The clinical picture is difficult to differentiate from other more common gingival masses, such as epulis or fibroma,

so a histopathological examination is required to establish a definite diagnosis.

CONFLICT OF INTEREST

The authors declare no potential conflicts of interest.

ACKNOWLEDGEMENT

Our thanks go to the professionals who assisted in the research and preparation of the paper.

REFERENCES

- 1. Kapoor H, Arora R. A Massive peripheral ossifying-fibroma: Uncommon presentation of a common lesion. Oral Health and Dental Management 2014; 13 (4): 940-44.
- 2. Peralles PG, Viana APB, Azevedo ALR, Pires FR. Gingival and alveolar hyperplastic reactive lesions: Clinic-pathological study of 90 cases. Brazilian Journal of Oral Sciences 2006; 5(18): 1085-89.
- 3. Lazare H, Peteiro A, Sayáns MP, Gándara V, Caneiro J, Garcia A, Anton I, et al. Clinicopathological features of peripheral ossifying fibroma in a series of 41 patients. British Journal of Oral and Maxillofacial Surgery 2019; 57: 1081–85.

- 4. Chhina S, Rathore AS, Ahuj P. Peripheral ossifying fibroma of gingiva: A case report. International Journal of Case Reports and Images 2011; 2(11): 21-4.
- 5. Sah K, Kale A, Hallikerimath S, Chandra S. Peripheral cemento-ossifying fibroma: Report of a recurrence case. *Contemporary Clinical Dentistry* 2012; 3(5): 23-6.
- 6. Priyanka A, Abhishek A, Vishakha V. Peripheral cemento-ossifying fibroma A clinical and histomorphological case report. International Journal of Contemporary Medical Research 2016; 3(7): 2020-22.
- 7. Rangil JS, Silvestre FJ, Bernal JR. Cemento-ossifying fibroma of the mandible: Presentation of a case and review of the literature. J Clin Exp Dent. 2011; 3(1): 66-9.
- 8. Joseph J , Maheshwari T. An unusual clinical presentation of multifocal peripheral cemento-ossifying fibroma : A case report. Journal of Oral Medicine, Oral Surgery, Oral Pathology and Oral Radiology 2015; 1(2): 99-102.
- Aroumougam A , Vezhavendhan
 N, Suganya R , Umamaheswari G.
 Peripheral cemento-ossifying fibroma of

- Maxilla. Journal of Scientific Dentistry 2017;7(1): 28-30.
- 10. Uppal J, Gupta ND, Trivedi H. Peripheral cemento-ossifying fibroma in an adolescent: A case report. Mod Res Dent 2018; 3(1): 200-02.
- Shah S, Patel M, 2, Shah K, Vyas S, Anchalia S. Peripheral cemento-ossifying fibroma: a case report.
 Journal of Government Dental College and Hospital 2014, 01(01): 42-5.
- 12. Anugraha G, Sumarta NP. Peripheral ossifying fibroma of the anterior maxillary gingiva. Dent. J. (Majalah Kedokteran Gigi) 2019; 52(4): 204–08.
- 13. Birant S, Kasimoglu Y, Cankaya AB, Aksakalli N, Seymen F. Peripheral cemento-ossifying fibroma associated with an unerupted tooth. Int J Med Invest 2019; 8(4): 78-84.
- 14. <u>Chatterjee</u> A, <u>Ajmera</u> N, <u>Singh</u> A. Peripheral cemento-ossifying fibroma of Maxilla. J Indian Soc Periodontol 2010; 14(3): 186-89.
- Sah K, Kale AD, Hallikerimath S,
 Chandra S. Peripheral cementoossifying fibroma: Report of a reccurence case. Contemp Clin Dent 2012; 14(3): 23-5.

- 16. Ravindranath A, Yaseen SM, Satish Y. Peripheral ossifying-fibroma in Infant: A Case Report. J Dent Probl Solut 2015; 2(2): 38-40.
- 17. Dubey KG, Shekhar S. An unusual case of peripheral ossifying-fibroma of the mandible in an adult male: Case report. Kronicle of Dental Science 2017; 1 (2): 66-71.
- 18. Raj P, Nausheen E, Rawther N, James J. Peripheral ossifying fibroma of the posterior Maxilla: A rare case report. International Journal of Scientific Study 2015; 3 (6): 217-20.
- 19. Trasad V A, Devarsa GM, Subba Reddy VV, Shashikiran N D. Peripheral ossifying fibroma in the maxillary arch. J Indian Soc Pedod Prev Dent 2011; 29(3):255-59.
- 20. N Singh, V Kumar, S Savita. Electro-surgical management of a peripheral ossifying fibroma. S. Afr. dent. j. 2017; 72(5):