World Journal of Advanced Research and Reviews
eISSN: 2581-9615 CODEN (USA): WJARAI
Cross Ref DOI: 10.30574/wjarr
Journal homepage: https://wjarr.com/

(CASE REPORT)

Peripheral Ameloblastoma: A rare case of soft tissue tumor (Case Study)

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World Journal of Advanced Research and Reviews, 2024, 23(03), 3055–3061

Publication history: Received on 17 August 2024; revised on 25 September 2024; accepted on 27 September 2024

Article DOI: https://doi.org/10.30574/wjarr.2024.23.3.2971

Abstract

A rare case of Peripheral ameloblastoma (PA) is reported. This rare type of tumor, classified as benign, locally invasive odontogenic tumor which histologically resembles an intraosseous ameloblastoma but develops in the soft tissues of the gingiva and mucosa. The prevalence of PA ranging from 2-10% compared to the other types of ameloblastoma. A case of PA in a 56-years-old male with an exophytic painless mass in the gingiva of the anterior mandible provisionally diagnosed as irritation fibroma is reported. Histopathological result of surgical treatment was concluded as acanthomatous peripheral ameloblastoma. Based on this experience clinical manifestation of gingival tumor a careful diagnosis should be taken by included the PA in differential diagnosis, in order to manage an adequate treatment and to ovoid of tumor recurrent.

Keywords: Peripheral ameloblastoma (PA); Soft tissue tumor; Gingival mass; Acanthomatous; Extraosseous

1. Introduction

Peripheral ameloblastoma (PA) is defined as an extraosseus variation of conventional ameloblastoma[1,2]. Ameloblastoma is a benign neoplasm originated from odontogenic epithelium with varied histopathologic features. Generally, ameloblastoma are characterized by their locally aggressiveness properties. Latest genetic researches have revealed a genetic mutation in MAPK tracts of multiple samples in ameloblastoma specimens. The most common mutation is in BRAFV600E[3].

Ameloblastoma are the most common benign neoplasm arises from odontogenic epithelium[4]. Ameloblastoma frequently found in mandible, maxilla or their surrounding tissues. In 2017, WHO classified ameloblastoma into 4 categories, which are conventional ameloblastoma, peripheral or extraosseus ameloblastoma, unicystic ameloblastoma or metastasized ameloblastoma.

Extraosseus type of ameloblastoma named peripheral ameloblastoma (PA) first repoerted by Kuru at 1911[5]. Then on 1959, Stanley and Krogh defined clinical and histopathological character of the ameloblastoma[6]. These lesions are usually painless and frequently grows as asymptomatic mass (Figure 1). Although they usually cofined in gigiva or oral mucosa, these lession could also cause depression on the underlying bone and resulting in saucer-like-impression due to bone resorption as the impact of growing mass' pressure. In 2016, Kandagal, *et al*. reported a unique case with peripheral ameloblastoma (PA) growing on maxillary gingiva with non-specific clinical view[7,8].

The objective of this case study is to present a rare case of peripheral ameloblastoma case and to describe their pathogenesis which subsequently leads to better diagnosis and proper management of a gingival mass.

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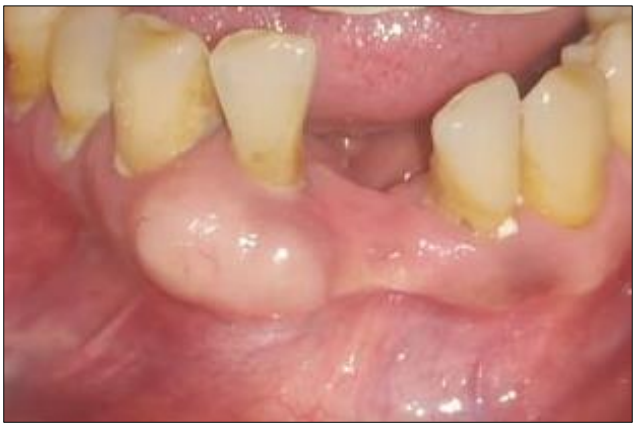

Figure 1 Clinical View of Peripheral Ameloblastoma on the Labial Portion of tooth 42

Peripheral Ameloblastoma originated from dental lamina or from surface epithelium base of gingiva or oral mucosa soft tissues[2, 6, 9, 10]. Clinically, peripheral ameloblastoma presented as slow-growing, exophytic, asymptomatic mass[11]. Peripheral ameloblastoma mass could be pedunculated or sessile. Peripheral ameloblastoma generally have smooth surface with papilated variation and the color resembles adjacent tissues[1, 2, 5, 6]. The size of peripheral ameloblastoma lesions ranging from 0,2 to 4,5 cm in diameter. Panoramic radiograph usually do not reveal any bony involvement, because the mass is confined in the gingiva or alveolar mucosa without invasion to the underlying jaw bone.[1, 3, 5, 8] Most prevalent site of peripheral ameloblastoma are mandibular anterior to premolar gingival region. Several extra-gingival site of peripheral ameloblastoma are labial mucosa and oral base[1, 3, 4].

Peripheral or extraosseus ameloblastoma is ameloblastoma subtype categorized as very-rare type. The incidence of peripheral ameloblastoma is 1,3 – 10 % compared to all types of ameloblastoma[4, 5, 8]. Since first described by Kuru in year 1911, less than 200 case of peripheral ameloblastoma has been reported in scientific articles. Peripheral ameloblastoma have slight male predilection, with 50 to 60 years-old of age range[1, 3, 6]

Mandibular premolar region is the most common site of peripheral ameloblastoma followed by anterior region of the mandible. Peripheral ameloblastoma in maxilla are considerably rare, but a study has reported peripheral ameloblastoma in palatal soft tissue adjacent to maxillary tuberosity[2, 4, 6]. Incidence ratio of mandibular peripheral ameloblastoma is 2,4% and 1% of maxilla. Important considerations of that extraosseus ameloblastoma are they frequently found on edentulous ridge with wide varieties of differential diagnosis range from chronic inflammations to neoplasms, both benign or malignant[2, 3]. Most times, peripheral ameloblastoma initially diagnose as peripheral odontogenic fibroma. Thus it is the most common differential diagnosis of this tumor. Possible differential diagnosis of peripheral ameloblastoma are fibromatous epulis, irritation fibroma, giant cell granuloma, peripheral ossifying fibroma, papiloma, pyogenic granuloma, even basal cell carcinoma of the oral cavity[7, 11].

In most cases, peripheral ameloblastoma mass is entirely located in gingival connective tissue with extension or adjacent to overlying epithelium[1-3]. Conventional intraosseus ameloblastoma is locally aggressive, in contrast, peripheral extraosseus ameloblastoma have non-invasive characteristic. Therefore, they rarely cause bone resorption under the lesion.[4, 5, 11, 12].

Treatment recommendation of the peripheral ameloblastoma is surgical excision, and an adequate surgical safety margin in 1-2 cm radius from the lesion should be included. In a large lesions of peripheral ameloblastoma, can be found with more than 2 centimeters in diameter[12, 13]. Surgical decortication or more invasive surgical treatment should be considered as the possibility of tumor infiltration in the underlying bone. Wide excision and *en-bloc* resection with sufficient surgical safety margin is the safest surgical treatment. Periodically control is needed to observe the possible recurrence of the tumor up to 8-years period. Malignant transformation reported by Ide *et al.* (2009) with one case[12] .

2. Material and methods

A 56-years old Asian male of Javanese descent visited our Oral and Maxillofacial Surgery Clinic of Dr. Mohammad Soewandhie General Hospital, Surabaya-Indonesia with a painless mass on the anterior mandibular gingiva. He started noticing the presence of mass about six years prior coming to the clinic. Gingival mass marked initially about 1 centimeter in diameter, gradually increased in size for the past year. The mass was reported never cease in size,

asymptomatic and no history of infection. The teeth surrounded the mass was found firm, no mobility nor loss of sensation around the gingival mass. Dental history of the patient revealed the use of removable dental prosthesis since 2016. From medical history, the patient was diagnosed with stage II hypertension treated with nifedipine (one tablet of 30 miligrams, three times a day) and candesartan cilexetil (one tablet of 8 miligrams, once a day), and does not comply to take the medicine regularly. Patient also had history treated for his tuberculosis, but the patient only followed the treatment for 3 months.

Intraoral examination revealed a well-circumscribed 10 mm x 5 mm x 5mm exophytic, non-pedunculated, non-ulcerated gingival mass on the labial gingiva of tooth 42 and lingual extension was not found. The color of the mass surface is similar to the surrounding tissues. The consistency of the gingival mass was solid and non-tender on palpation (Figure 1).

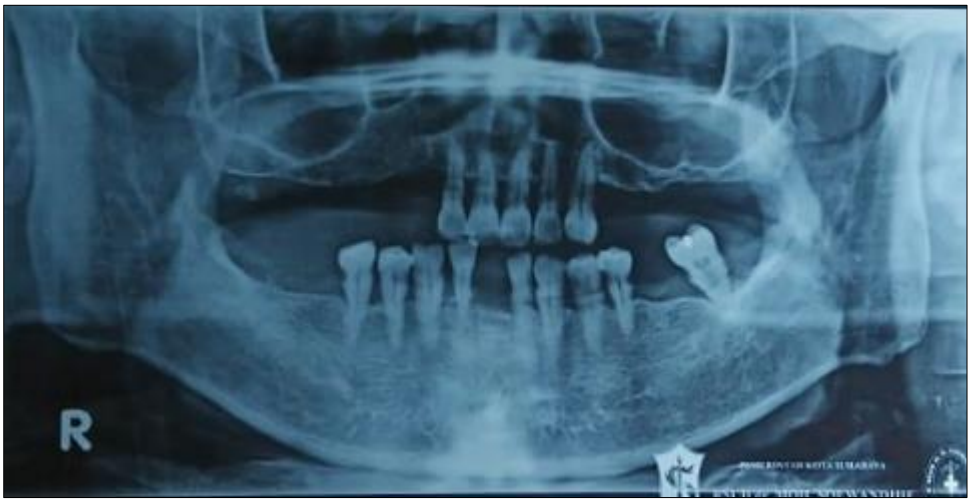

Figure 2 Panoramic Radiograph showing no underlying bone resorption nor bony involvement

Based on clinical, laboratory, and radiographic examination results, this mass was diagnosed as irritation fibroma with differential diagnosis included peripheral ameloblastoma and peripheral odontogenic fibroma. Treatment planning for this case was excisional biopsy.

A mucoperiosteal incision was made starting from distolabial aspect of tooth 43 through the marginal gingiva near the cervical part of tooth 43 annd followed by vertical-obliquely in the direction of mesiolabial region of tooth 42.

Full-thickness of mucoperiosteal flap was elevated (Figure 3a) and surgical excision of the mass including the safety margin was made within 1,5 cm radius from the gingival mass and underlying bones. (Figure 3b)

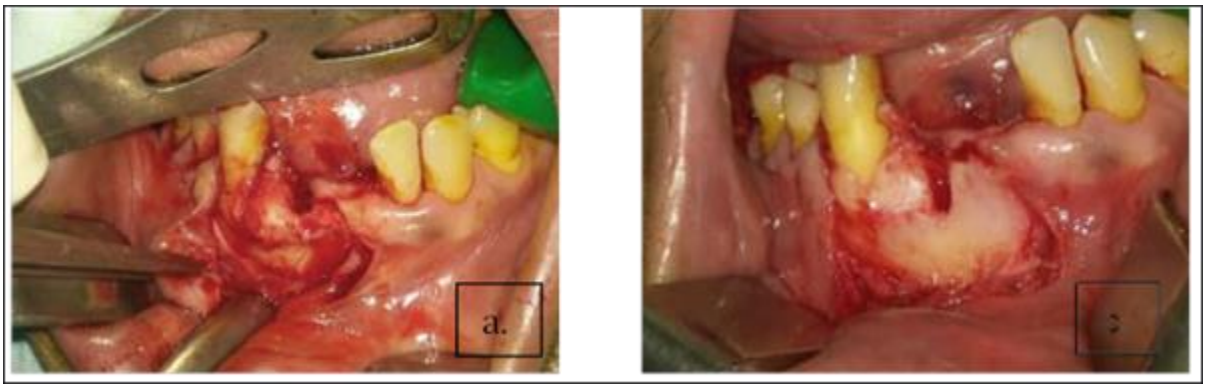

Figure 3 a. Full-Thickness Mucoperiosteal Flap Elevation ; b. Clinical View of the Surgical Site Post Excisional Biopsy

Bone freshening and undermining of the remaining gingiva and mucosa around the surgical site was done for releases the soft tissue to prevent soft tissue tension during wound closure and to achieved the flap closing for primary intention healing. The flap was returned to position and sutured with absorbable polyglycoic acid suture, size 4.0.

The mass and bone specimen was sent to Pathology Anatomy lab for further histopathologicaly examination. (Figure 4 a,b)

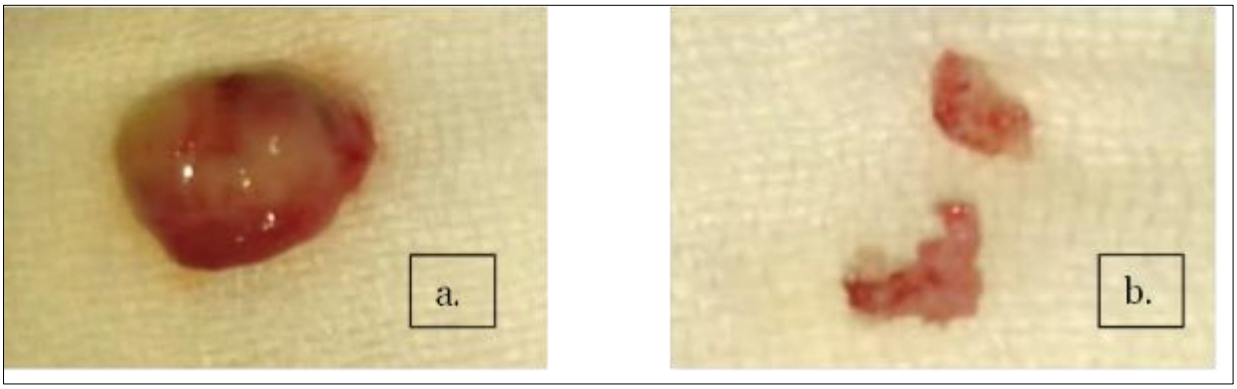

Figure 4 a. Gingival Mass Specimen; b. underlying bone specimens

The result of pathologic examination of the gingival mass specimen revealed as non-infiltrative. The exposed bony margins appeared uninvolved. No neoplastic invasion or marrow infiltration was found.

Histopathological examination demonstrated presence of a squamous epithelium at the outermost part of the specimen. There were proliferation activity in the stroma with soft chromatin pallisading cells were arranged marginally, and differentiated squamous cells in the middle. No signs of malignancy was found.

The clinical presentation and the histological findings were consistent as acanthomatous peripheral ameloblastoma.

In the sixteenth day of postsurgical day, the patient was recalled for further follow up and surgical site showed with optimum healing conditions, there were no signs of swelling, slight hyperemia, and non-tender on palpation. No nerve disturbancies as parasthesia of the lower lip, oral mucosa, and gingival region in the region of anterior mandible nor in the tounge

3. Results and Discussion

A well-circumscribed gingival mass of a male patient in his fifth to sixth decades of life of the patient presented in this case study is in accordance to the description of peripheral ameloblastoma[3, 4, 11]. Peripheral ameloblastoma is a rare, benign, extraosseous odontogenic soft tissue tumor, clinically presented as a painless exophytic mass on tooth bearing areas with a smooth, or papillary surface. Peripheral ameloblastoma also known as extraosseous ameloblastoma[14].

The prevalence of peripheral ameloblastoma ranging between 1,3 – 10 % of all ameloblastoma cases, and 1% of all oral cavity tumor, 11% of all odontogenic tumor[15, 16]. Peripheral ameloblastoma cases that have been reported happened in patients around 16-92 years of age, with mean age 50,2 years-old. About 50% of the lesion occur in 40-60 years-old patients. Diameter of the mass is aroung 1-2 centimeters. Peripheral ameloblastoma commonly happened on the buccal mucosa of the mandible, especially anterior to premolar region[17].

In irritation fibroma is gingival overgrowth as reactions to chronic irritation. Ill-fitted dentures of the patient could be the source of irritation [2, 12]. As mentioned above that the differential diagnosis of a gingival mass can be varies from chronic reaction inflamed gingiva to gingival tumor and it can be benign or malignant. Irritation fibroma is the most common gingival lesions found due to chronic reaction and can be presented with irritation fibroma, peripheral giant cell granuloma, peripheral ossifying fibroma, papiloma, pyogenic granuloma and fibromatous epulis[18]. One of consideration important of this lesion is to differentiate with a peripheral ameloblastoma. The histologic feature of irritation fibroma showed with a structures resembled with interwoven fibrous tissue.

Most cases of peripheral ameloblastoma presented as exophytic mass which grows locally in soft tissue of edentulous or tooth bearing area. Macroscopically, peripheral ameloblastoma is presented with a solid mass, sessile or pedunculated, with pink to reddish color, variations on lesion surface is common ; smooth, granular or wart-like[13, 18]. The definitive diagnosis of peripheral ameloblastoma cannot be taken by relying only on clinical presentation, because this benign gingival mass resembled as any other gingival enlargement lesion[14, 15, 19, 20].

Based on progresivity, peripheral ameloblastoma is less-invasive compared to intraosseus ameloblastoma[19]. This benign tumour have the potential to infiltrate underlying bone or causing the underlying bone resorb due to growing pressure of the soft-tissue mass. The rarity of bony infiltration is attributed to the existance of fibrous barrier around the lesion which is produced by the gingival tissue or the periosteum[8, 16, 20].

The definitive diagnosis of Achantomathous peripheral ameloblastoma was made by the presence of squamous metaplasia, which is seen in the red-circle area. (Figure 5) This result was marked as importance histopathological findings for the key to make definitive diagnosis[11, 12, 14].

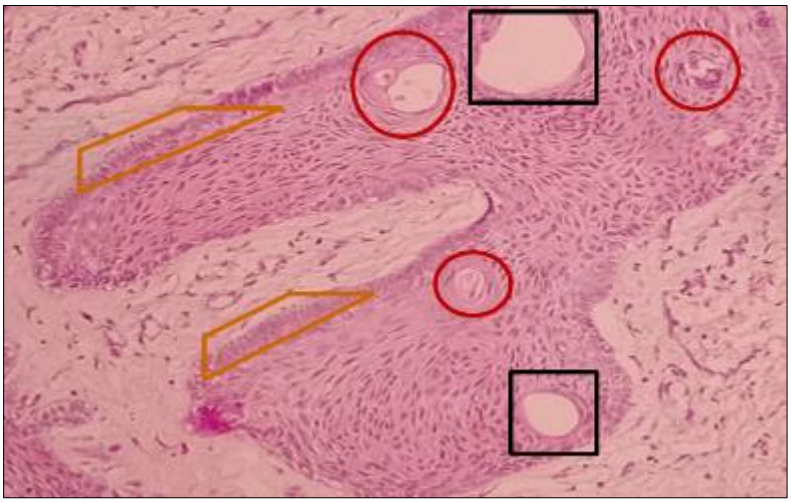

Figure 5 Histopathological view of Excisional Biopsy of Gingival Mass on tooth 42 Labial Region (hematoxylin-eosin stain, 40x magnification) demonstrating palisading squamous epithelium (brown-trapezium areas), angular cell resembling vacuole (black-box areas), and differentiation of centrally-squamous cell (red-circle area) are the characteristic of peripheral ameloblastoma, acanthomatous-type

Several researchers point out the importance of histopathological examinations of soft tissue tumors, because some of gingival enlargement that shown with abnormal mitotic activity has potential malignancy transformation. As seen in Basal Cell Carcinoma that is one of differential diagnosis listed in gingival enlargement lesion. It is originated from the proliferation and mutation of pluripotent basal cells within surface epithelium and adnexal structures, whereas peripheral amelobastoma arise from rest of Serres outside the jaw bone[12, 13, 17]. Those two can only be differentiated under immunohistochemistry staining,in which basal cell carcinoma is tested negative for cytokeratin. Peripheral ameloblastoma is derived from odontogenic epithelium, therefore their cells must have cytokeratin[8, 15, 20].

The peripheral ameloblastoma is a rare odontogenic tumor. It can be presented as a discrete gingival mass[4, 5, 9, 14]. This case highlights that a tumor mass in the gingiva regions should be considered as a soft tissue tumor such as peripheral ameloblastoma. The treatment planning of a gingival masses would be based on a comprehensive evaluation of prompt anamnesis and all diagnostic modalities available including but not limited to radiography examination, signs and symptoms, thorough clinical examination and obligatory biopsy examination of the mass tissue[1, 13, 18-20].

Proper management consisted of surgical excision with adequate surgical safety margins 1-2 cm around the lesion[18, 20]. Conservative management of peripheral ameloblastoma is not suggested due the higher recurence level[13, 21]. Surgical intervention of the cortex the underlying bone is necessary for definitive examination of the extension of the lesion, to ensure no bony involvement in this lesion. The finding of this case is in coherence with previous study mentioned that small mass of peripheral ameloblastoma with diameter less than 2cm is rarely causing bone destruction nor infiltration.[15, 18, 20]

Periodic follow-up is necessary as ths type of tumor has a possibility of late recurrence, which has been reported to occur 10 years or longer after initial surgery[1, 8, 13, 19]. The recorded recurrence rate of peripheral ameloblastoma is 16 to 20% and majority of the recurrence is found on the third-year periodic follow up[1, 12, 18, 20, 21].

4. Conclusion

This case study concluded that gingival mass found in the anterior region of mandibular alveolar process of the patient was peripheral ameloblastoma based on clinical, radiographic, and laboratory findings. Any of gingival enlargements, a

peripheral ameloblastoma as the differential diagnosis should be included, due to the possibility of ameloblastoma development from rests of the dental lamina in the gingiva of tooth bearing area nor edentulous region.

Prompt diagnosis also helps in deciding the proper surgical treatment to prevent recurrence

Compliance with ethical standards

Acknowledgments

The authors are very grateful to the Head and Staffs of Oral and Maxillofacial Surgery Department, Faculty of Dental Medicine, Universitas Airlangga and Oral and Maxillofacial Surgery Division, Dr. M. Soewandhie General Hospital Dental Clinic Surabaya, Indonesia who provided insight and expertise that greatly assisted the case study.

Disclosure of conflict of interest

All authors declare that they have no conflicts of interest relevant to the content of this manuscript. No financial, professional, or personal relationships or affiliations have influenced the work presented in this paper.

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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