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Olgu Sunumu / Case Report

Peripheral Ameloblastoma with Mixed Histological Patterns

Kompleks Histolojik Paternler İçeren Periferal Ameloblastoma

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ABSTRACT

Peripheral ameloblastoma is a relatively uncommon neoplasm of odontogenic origin histologically resembles the classic ameloblastoma and derived from epithelial and/or mesenchymal elements being part of the tooth forming apparatus. It can be originated in the soft tissue of oral cavity namely alveolar mucosa or gingiva. Among histopathological pattern the most common is acanthomatous followed by plexiform and follicular. Granular cell ameloblastoma once considered as the most aggressive variant of ameloblastoma but according to the recent studies it is believed that the earlier belief was a myth. We report a case of peripheral ameloblastoma in a 44 year old lady, histopathologicaly showing combined features of granular cell, plexiform, acanthomatous, follicular and desmoplastic ameloblastoma. As per our best knowledge this is the first case of its type.

Key words: Peripheral ameloblastoma, granular cell, acanthomatous, follicular, plexiform, desmoplastic

ÖZET

Periferal ameloblastoma, diş dokusu kaynaklı, klasik ameloblastomaya benzeyen, nispeten yaygın olmayan bir tümör olup dişi oluşturan yapıların bir parçası olarak epitel ve/veya mezenşimal elementlerden çoğalır. Alveolar mukoza veya gingiva olarak adlandırılan ağız boşluğunun yumuşak dokusundan köken alabilir. Histopatholojik sekiller arasında en yaygını, pleksiform ve foliküler tarafından takip edilen acanthomatousdur. Granüler hücre ameloblastoma'nın bir zamanlar ameloblastoma'nın en agressif formu olduğu düşünülürdü, ancak son dönemlerde yapılan çalışmalara göre önceki inanışların bir mit oluğu görüşü yaygınlaşmaya başladı. Kırk dört yaşındaki bir kadında, histopatolojik olarak granüler hücre, pleksiform, acanthomatous, foliküler ve desmoplastik ameloblastomanın kombinlenmiş özelliğini gösteren bir periferal ameloblastoma olgusu bildirilmiştir.

Anahtar kelimeler: Periferal Ameloblastoma, Granüler hücre, Acanthomatous, Foliküler, pleksiform, desmoplastik

INTRODUCTION

Peripheral ameloblastoma (P.A) is a relatively uncommon odontogenic tumor that is histologically identical to the classic intraosseous ameloblastoma[1]. It originates in the soft tissue of oral cavity namely alveolar mucosa or gingiva. It accounts for 1 – 5 % of all ameloblastomas. The first case of PA was reported by Kuru in 1911. However, Philipsen and coworkers stated that what Kuru described was not a peripheral but rather an intraosseous ameloblastoma having penetrated through the alveolar bone and fused with the oral epithelium[2] The granular cell ameloblastoma. (GCA) is one of the rarest entities and accounts for only 5% of all classic intraosseous ameloblastomas[3].

PA is rarely the initial presenting diagnosis to be made[4]. The differential diagnosis must be made with fibrous nodule, gingival tumors, peripheral odontogenic fibroma, peripheral ossifying fibroma, pyogenic granuloma, peripheral giant-cell

granuloma, papilloma, peripheral squamous odontogenic tumor, basal cell carcinoma and other hyperplastic swellings superficial to the alveolar ridge[5]. We present a case of gingival swelling located on the buccal aspect of right maxillary second molar.

CASE

A 44 year old lady reported to our department with the chief complaint of localized gingival growth with relation to maxillary right second molar. The growth had been present from last one year and was slowly increasing in size. The intraoral examination revealed a non tender mass on the buccal surface of maxillary right second molar. The growth was ovoid, sessile, reddish grey in color, firm and fixed to the underlying structures and measured 2 × 1 cm^2 approximately. The underlying mucosa was normal in color without showing any sign of ulceration. The radiographic examination revealed a slight inter – dental bone loss between maxillary right first molar and second molar. (Figure 1) After the correlation of radiographic and histopathological features provisional diagnosis of peripheral giant cell granuloma was given.

The growth was surgically excised under local anesthesia and the excised specimen was sent to the department of oral and maxillofacial pathology for the histopathological examination. (Figure 2) The mass could be easily separated from the underlying bone but there was profuse bleeding associated with it. The surgical wound healed uneventfully.

Microscopic examination revealed odontogenic epithelium arranged in the form of follicles, strands and anastomosing cords. (Figure 3) Stellate reticulum with in follicles shows transformation to granular cells. (Figure 4) Intervening stroma is dense collagenous and hyalinzed at places. Some follicles exhibit acanthomatous changes with keratin formation and areas of cystic degeneration. (Figure 5) After the correlation of overall clinical and histopathological features final diagnosis of Granular cell - Peripheral ameloblastoma was given.

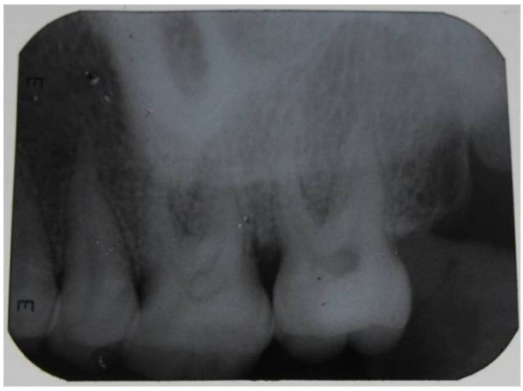

Figure 1. Intra oral periapical radiograph reveals that the lesion is not associated with bone

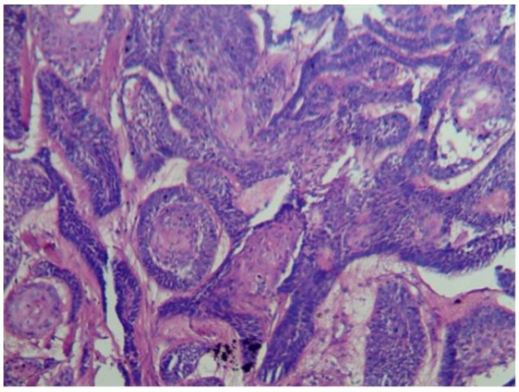

Figure 2. Odontogenic epithelial islands arranged in anastomosing cords and showing plexiform pattern Some follicles showing squamous metaplasia.

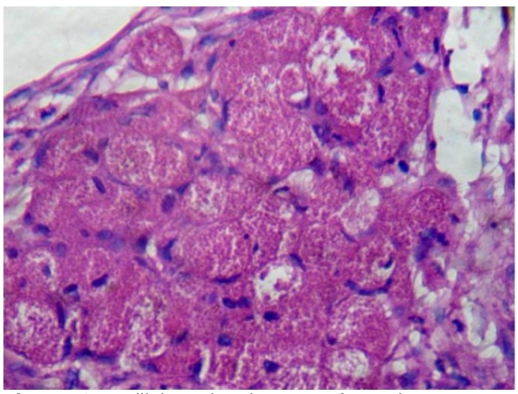

Figure 3. Follicles showing transformation to granular cells (H & E 40 X)

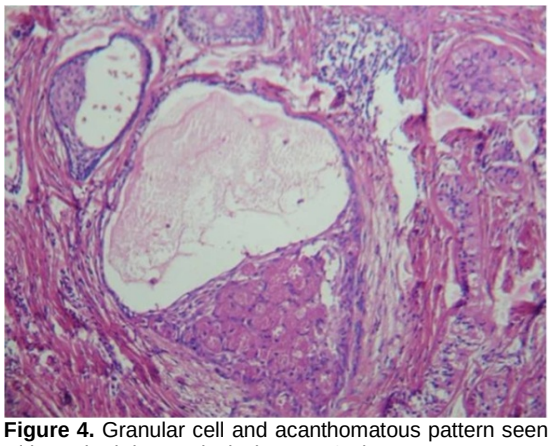

Figure 4. Granular cell and acanthomatous pattern seen with marked desmoplasia (H& E 10 X).

DISCUSSION

Defined as a tumor with the histologic characteristic of an intraosseous ameloblastoma but occurring in the soft tissue overlying the tooth bearing area of the maxilla and mandible[6], PA is also known as mucosal, extramedullary, extraosseous, soft tissue ameloblastoma or ameloblastoma of gingiva[7]. According to W.H.O classification (2005), the extraosseous/ PA is the extraosseous counterpart of the intraosseous solid/ multicystic ameloblastoma[8]. PA presents a painless, sessile firm and exophytic growth with relatively smooth sometimes granular, pebbly, papillary or warty surface[9]. The color varies between normal or pink and red or dark red[6]. PA is difficult to diagnose based only on clinical parameters and it can be confused with an epulis, fibroma, peripheral giant cell granuloma, pyogenic granuloma, peripheral ossifying fibromaand basal cell carcinoma[10]. Final diagnosis can be made on histological basis. (Table 1) In present case also diagnosis could not be established on clinical findings.Radiographically a PA is superficial to the cortical bone with no sign of bone involvement in majority of cases however: few cases have shown a slight underlying bone loss. Therefore it can be said that PAs are less aggressive than a classical intraosseous ameloblastoma which invades bone[11]. A few cases of PA are associated with minor bone resorption due to the depression resulting from the pressure of the tumor on bone[12]. The present case also showed a slight inter – dental bone resorption.

Histologically the tissue is composed of islands and strands of odontogenic epithelium, usually resembling the follicular pattern of intraosseous ameloblastoma[5,9]. The epithelium islands commonly exhibit the acanthomatous variant of this pattern with central areas of keratin formation or the cystic pattern. In some cases the islands of odontogenic epithelium often shows arranged in anastomosing cords and refer to the plexiform pattern[4]. The epithelial islands are usually surrounded by fibrous tissue[9]. Literature reveals only single case of PA with granular cell pattern[13]. In present case we found the combination of different patterns i.e. granular cell, plexiform, acanthomatous which makes the case very unique. The treatment of PA consists of surgical excision down through the periosteum. The recurrence rate of PA is much lower (19%) than that of IA (33%)[10].

The Granular cell ameloblastoma (G.C.A) once considered as the most aggressive variant of ameloblastoma but according to the recent studies it is believed that the earlier belief was a myth. [14] Numerous theories have been proposed on the origin and nature of these granular cells in ameloblastomas. The granular cells are epithelial in origin and several ultrastructural studies describe them as lysosome. [15] Nasu et al suggested that with age, the aged components progressively increased in the cytoplasm of some of the tumor cells, but the ability of lysosome to dispose these materials decreases, hence their cytoplasm packed with lysosomal granules[16]. Tandler and Rossi thought that these lysosomes might have been a result of some genetic alteration[17]. Recent Immunohistochemical studies suggest that the survival potential as well as the proliferative index of neoplastic granular cells in granular cell ameloblastoma is the least among all the variants of ameloblastoma[18].

On the basis of all these studies it can be said that the old belief that granular cell ameloblastoma is the most aggressive variant of ameloblastoma is just a myth, probably granular cells are just a transitional phase in the life cycle of ameloblastoma[19].

We conclude that present case was an extremely rare case, as per our best knowledge this is for the first time that a PA is showing various patterns.

Table 1. Differential diagnoses of PA with characteristic differentiating histopathological features[20,21].

Differential diagnosis	Characteristic histopathological features
Peripheral giant cell granuloma	Highly cellular stroma with proliferating plump fibroblasts and multinucleated giant cells.
Pyogenic granuloma	Large thin-walled vessels in a loose connective tissue stroma infiltrated throughout by leukocytes
Peripheral ossifying fibroma	Fibrocellular connective tissue stoma with numerous calcified masses
Fibroma	Dense fibrous stroma and atrophic epithelium
Basal cell carcinoma	Budding of epithelial islands into lamina propria and scattered mitotic figures. Peripheral palisading is present but lacks reverse polarity
Squamous papilloma	Numerous finger like projections lined by stratified squamous epithelium. Koilocytes may appear in connective tissue.

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