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Annals of Case Reports

Fujii T, et al. Ann Case Rep: ACRT-173.

Case Report

DOI: 10.29011/2574-7754/100073

A Rare Case of Granular Cell Type Peripheral Ameloblastoma with a Papilloma - like Appearance

Tomoko Fujii[1], Yuichi Ohnishi[1*], Masahiro Watanabe[2], Hiroyuki Hamada[1], Masahiro Nakajima[1]

[1]Second Department of Oral and Maxillofacial Surgery, Osaka Dental University, Chuo-ku, Japan

[2]Department of Dentistry and Oral Surgery, Osaka Red Cross Hospital, Osaka-shi, Japan

[*]**Corresponding author:** Yuichi Ohnishi, Second Department of Oral and Maxillofacial Surgery, Osaka Dental University, Chuo-ku, Japan. Tel: +81728562111; Email: onishi_yu_ku@msn.com

Citation: Fujii T, Ohnishi Y, Watanabe M, Hamada H, Nakajima M (2018) A Rare Case of Granular Cell Type Peripheral Ameloblastoma with a Papilloma - like Appearance. Ann Case Rep: ACRT-173. DOI: 10.29011/2574-7754/100073

Received Date: 31 March, 2018; **Accepted Date:** 07 April, 2018; **Published Date:** 17 April, 2018

Summary

Peripheral ameloblastoma is a tumor that develops in the gingival epithelium or the alveolar bone surface. The most common subtype is acanthomatous ameloblastoma, whilst the rarest is granular cell ameloblastoma, which has no known cases reported in the literature. The present study describes the case of a 31-year old male who was referred to the Second Department of Oral and Maxillofacial Surgery, Osaka Dental University (Osaka, Japan) in December 2003, presenting with gingival swelling at the labial surface of the lower left cuspid. The lesion was similar in appearance to a papilloma with a granular surface, and was clinically diagnosed as a benign gingival tumor. A biopsy was performed and subsequent histopathological examination suggested a diagnosis of ameloblastoma. In February 2004, marginal resection of the mandible was performed under general anesthesia. Histopathological analysis indicated that the mass was an alveolar lesion containing abundant granular cells, with no tumor invasion into the mandible noted. Therefore, the final diagnosis was confirmed as granular cell ameloblastoma. The post-operative course of the patient was uneventful, and there has been no recurrence for 14 years' post-surgery. A number of benign oral mucosal lesions have been identified to be associated with several types of human papillomavirus (HPV). The present study therefore utilized commercially available HPV DNA to confirm the presence of HPV.

Keywords: Granular Cell Type; Odontogenic Tumor; Peripheral Ameloblastoma

Introduction

Ameloblastoma is a tumor of tooth germ epithelial origin; it is generally known to be a tumor of the jawbone, and rarely occurs in the soft tissue [1,2]. Peripheral Ameloblastoma (PA) is a tumor that develops in the gingival epithelium or the alveolar bone surface [3,4]. The most common is subtype is acanthomatous ameloblastoma [4], whilst granular cell ameloblastoma is the rarest, with no known reported cases in the literature until now.

It has been demonstrated that a number of benign oral mucosal lesions are associated with several types of Human Papillomavirus (HPV) [5-8]. The current study utilized commercially available HPV DNA to confirm the presence of HPV in a rare case of granular cell ameloblastoma. The primary aim of the study was to present a rare case of PA and determine the type(s) of HPV that are associated with this granular cell subtype, and to discuss the implications with regard to etiology if the disease.

Clinical Case Report

A 31-year-old male was referred to the Second Department of Oral and Maxillofacial Surgery, Osaka Dental University (Osaka, Japan) in December 2004, presenting with gingival swelling at the labial surface of the lower left cuspid. The lesion, measuring 5 x 7 mm in size, was similar in appearance to a papilloma, and was clinically diagnosed as a benign tumor (Figure 1). Radiographically examination revealed bone resorption in the tumor regions (Figure 2). A biopsy was performed and subsequent histopathological examination resulted in the updated diagnosis of ameloblastoma (Figure 3). In February 2004, a marginal resection of the mandible was performed under general anesthesia. Histopathological analysis of the resected tissue indicated that the mass was an alveolar lesion containing abundant granular cells, with no tumor infiltration into the mandible noted. Therefore, the final diagnosis was confirmed as granular cell ameloblastoma (Figure 4). The post-operative course

of the patient was uneventful, and there has been no recurrence for 14 years' post-surgery.

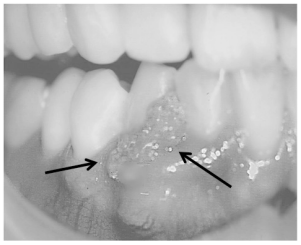

Figure 1

Figure 1: Image of the 5 x 7mm lesion (arrows), which was similar in appearance to a papilloma, at the initial referral.

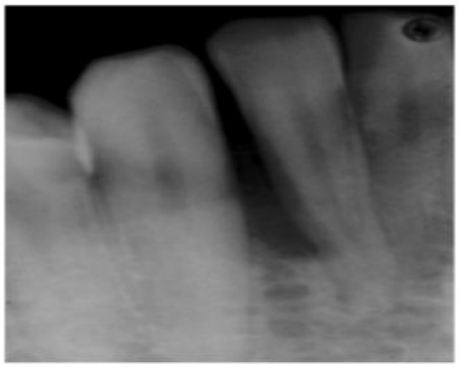

Figure 2: Radiographic examination result revealing bone resorption in the tumor region.

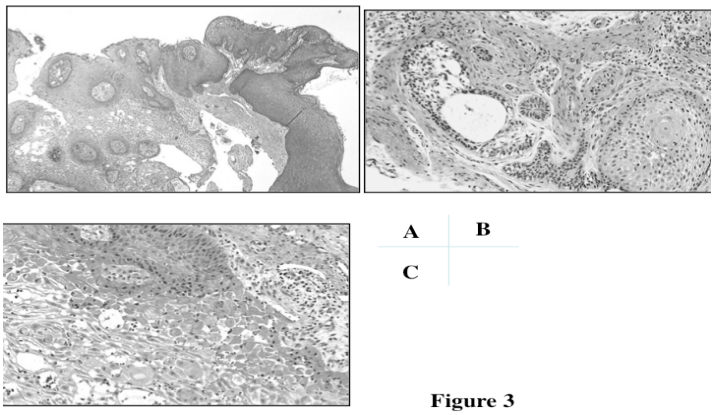

Figure 3

Figure 3: Histopathological analysis of the biopsy specimen showing ameloblastoma at magnification of (A)×40 and (B and C) ×100. Staining hematoxylin and eosin.

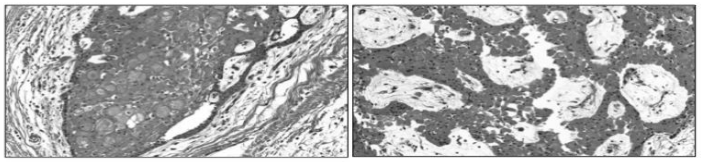

Figure 4: Histopathological examination of the resected tissue confirming the diagnosis of granular cell ameloblastoma. Staining, hematoxylin and eosin; magnification, ×100.

DNA Preparation and Polymerase Chain Reaction (PCR)

Tissue specimens were frozen in liquid nitrogen and stored at -70°C until use. DNA was isolated from tissues using TRIzoL○Rreagent (Invitrogen; Thermos Fisher Scientific, Inc., Waltham, MA, USA) according to the manufacturer's protocols. The primers used in the current study are presented in (Figure 5). The PCR conditions were as follows: 40 cycles of denaturation at 94°C for 1 min, annealing at 52°C for 2 min, and chain extension with Taq polymerase at 72°C for 1 min, followed by a final extension step at 72°C for 20 min. Subsequent to amplification, the final PCR mixture was separated by 2% agarose gel electrophoresis and stained with ethidium bromide. We used the Hela cell for the positive controls. Hela cells is known to widely an infection HPV18.

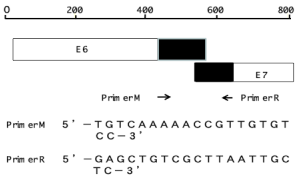

Figure 5

Figure 5: Location of consensus primers in the HPV gene. The consensus primers pair yielded 230-270 bop PCR polymerase chain products containing HPV-16, -18, -31, -33, -52b, and -58 DNAs. Open boxes indicate the open reading frame.

Application of Consensus Primers in PCR

PCR was performed on the DNA under the aforementioned conditions. The consensus primer pair yielded 230 to 270 bop of PCR products containing DNA that corresponded to HPV subtype16, 18, 31,33, 52b, and 58 (Figure 5) [9]. Result indicated 268bp and this was in agreement with HPV-18 (Figure 6).

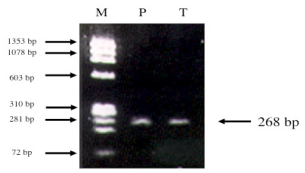

Figure 6

Figure 6: Polymerase chain reaction analysis of the consensus primers. HPV was detected in the tumor. M: maker, P: positive control Hela cell, T: present case.

Discussion

PA is a rare odontogenic lesion that primarily develops in gingiva. PA is similar histologically to intraosseous ameloblastoma, but it does not exhibit the same aggressive and invasive behavior [10]. PA is considered to originate from one of two possible sources: 1) Extra osseous remnants of the dental lamina [11]; or 2) the basal cell layer of the epithelium, which is regarded as having odontogenic potential [4].

It is well documented that PA occurs in an older population compared with intraosseous ameloblastoma, with patients ranging from 23 to 92 years of age [11,12]. Buchner et al. [3] reviewed 32 reported cases of PA, and determined that the mean patients age was 52 years, whilst El-Mofty et al. [1] reviewed 11 cases and reported that the mean age of incidence of PA was 47 years,8 years older than the mean age of incidence of intraosseous lesion. The bone over PA is not usually affected upon radiographic examination. However, in certain cases, superficial cupping, (or "saucerization") of the bone has been reported. This is widely considered to occur as a result of resorption opposed to neoplastic invasion by PA.

In the majority of cases, PA does not exhibit aggressive behavior and is much less invasive than intraosseous lesion. Therefore, with regard to treatment, less radical surgery is required. However, in case described in a study by Lee et al. [13], a small focus of ameloblastoma was identified on the surface of tissue removed from the buccal gingiva, which had originally appeared to be unassociated with the tumor; thus, the study concluded that a requirement existed for aggressive surgical management in certain cases [12]. If bony resorption is suspected or found, the lesion with periosteal perforation. Treatment for such cases ranges from a marginal resection of the mandible, leaving the lower border of the jaw intact, or a simple local excision. The patient in the present case underwent a marginal resection of the mandible due to the detection of bone resorption. The most common histological types of ameloblastoma are the plexi form and follicular types, whilst the granular cell type only accounts for 5% of allameloblastomacases [14,15]. Clinically, acanthomatous is the most prevalent form of PA in literature. The granular cells observed in the granular cell type ameloblastoma are relatively large and polygonal or circular in morphology. Nuclei contain granular of the eosinophilic cytoplasm, were strongly stained exists around the cell. Certain studies have reported that granular cell type tissues exhibit a positive reaction to keratin [16,17], and E-cadherin with epithelial specificity, in addition to containing to no filaments and desmosomes [18,19]; thus, granular cells are generally understood to originate from epithelial tissue [20].

Previously, certain HPV types were detected (via DNA hybridization or immune peroxidase staining techniques) in a number of malignant and benign tumors of the upper aero digestive tract, including the oral cavity [5-8]. It has been reported that HPV may serve a role in the development of oral mucosal tumors. Such tumors including squamous cell carcinoma, squamous papilloma and specimens of intraoral leukoplakia, all of which have been previously identified to contain HPV. The distinct HPV subtypes detected in the mucosa include types 1, 2, 4, 6, 7, 11, 13, 16, 18, 32, and 57. In particular type 16 has been associated with squamous cell carcinoma and dysplastic specimens. Kahn et al. [21] investigated by means of an immune histochemical staining technique for the detection of HPV genus-specific structural antigen in formalin-fixed, paraffin-embedded tissue. One case positive for HPV antigen, whereas none of randomly selected ameloblastomas in adults was positive. The present case was demonstrated to contain HPV-18 DNA. Previous reports have noted the frequent association of type 6 and 11 with benign lesions, whilst types 16 and 18 have been identified in oral squamous cell carcinomas and cervical intraepithelial neoplasia and carcinoma. However, following the completion of an increased number of studies, the patterns of association between HPV types and form of lesion have become less clear. For example, HPV-16 has now been detected in malignant and benign oral lesions.

Conclusion

The results of the present study provide evidence for inclusion of PA in the category of HPV associated benign oral mucosal lesions. Debate remains as to whether HPV exerts a causal role in the development of oral mucosal tumors. The probability that HPV functions alone is not likely; however, it appears to be a reasonable assumption that it may serve a role as an initiator or cofactor. HPV may be the stimulus necessary to cause the basal cell or dental lamina rests to proliferate with subsequent formation of PA. The confirmation of the presence of HPV and its further typing (HPV 16 o 18) in PA reinforces the theory that not only is HPV associated with numerous malignant and benign oral lesions, but that it may also be directly implicated in their development.

No Conflict of Interest

Financing by the same authors.

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