Kim et al., J Clin Case Rep 2017, 7:11
DOI: 10.4172/2165-7920.10001041

Journal of Clinical Case Reports

Case Report Open Access

Canalicular Adenoma of the Parotid Gland: A Rare Case Report and Review of Literature

Do-Hyoung Kim, Jun-Young Paeng, Sung-Tak Lee and So-Young Choi*

Department of Oral and Maxillofacial Surgery, School of Dentistry, Kyungpook National University, Daegu, South Korea

Abstract

Canalicular Adenoma (CA) is an uncommon, benign salivary gland tumor that was described by the World Health Organization classification in 1991. CA has a significant predilection for occurrence in the minor salivary glands, with most cases occurring in the upper lip, followed by the buccal mucosa and palate. Rarely, CA can involve the major salivary glands, for example the parotid gland. A small number of cases of CA of the parotid gland have been reported in the literature. We report a rare case of CA on the left parotid gland of an 81-year-old man with review of literature.

Keywords: Canalicular adenoma; Parotid gland; Fine needle aspiration

Introduction

Canalicular adenoma (CA) is an uncommon, benign salivary gland tumor that was described by the World Health Organization classification in 1991 [1]. CA has a significant predilection for occurrence in the minor salivary glands, with most cases occurring in the upper lip, followed by the buccal mucosa and palate [1-5]. Rarely, CA can involve the major salivary glands, for example the parotid gland [2,3,6-9]. CA usually occurs as a single entity, but multi-focal CAs have also been reported [10,11]. Clinically, CAs are grossly well-circumscribed nodular lesions, and are not usually accompanied by clinical symptoms, such as pain or paresthesia. Treatment of CA usually consists of local surgical removal of the swollen area [1,2,11]. The prognosis is good, and recurrences are not common [1-3,10,11]. We report a rare case of CA of the parotid gland of an elderly man, whose histologic examination results were consistent with CA.

Case Report

An 81-year-old man arrived at the Kyungpook national university dental hospital on October 14, 2015, presenting with nodular swelling at the left preauricular area. The patient had no other specific disease or traumatic history. The swelling began about one month previously and had slowly enlarged. The swollen area, measuring around 1.5 cm in diameter, was not evident in the resting position, but became evident during mouth opening. Clinical examinations revealed no specific symptoms, such as pain, paresthesia, or motor disturbance of the temporomandibular joint, except for induration. To enable diagnosis, we performed fine needle aspiration (FNA) and computed tomography (CT) of the paranasal sinuses using an intravenous contrast agent. The FNA results suggested that the swelling was caused by Warthin`s tumor. On the CT image, we saw a well-circumscribed, oval mass in the left parotid gland (Figures 1A-1C). This radiologic finding also suggested Warthin`s tumor. The lesion, which was found to be a well-capsulated, nodular solid mass, was excised under general anesthesia (Figure 1D). The specimen was about 1.5 cm in diameter and was surrounded by a thin, fibrous capsule (Figures 2A and 2B). The results of the histopathologic examination revealed that the mass consisted of single-layered cords of uniform, cuboidal epithelial cells with basophilic nuclei.

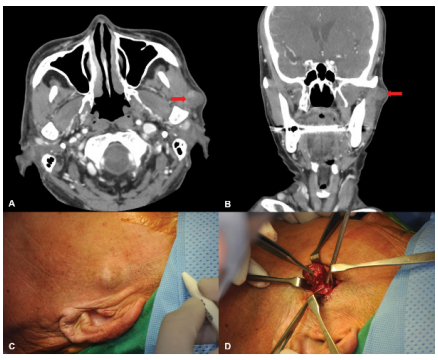

Figure 1: A and B. The axial and coronal views of the enhanced computed tomography scan. A well-demarcated mass is seen in the parotid gland of the left side. C. The appearance of the nodular mass of the left preauricular area. D. Removal of the mass, which was well-encapsulated by fibrous tissue.

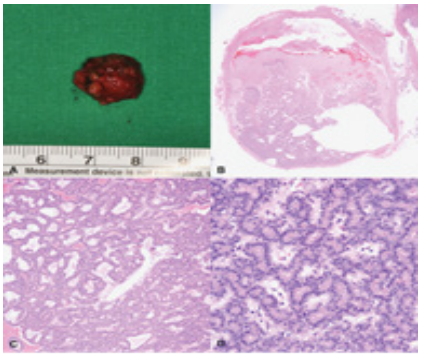

Figure 2: A. An image of the removed specimen, which was about 1.5 cm in diameter. B. The round mass was well-encapsulated by fibrous tissue (12.5x hematoxylin and eosin staining). C. Parallel rows of cord appeared to form ductal structures or long canals (100x hematoxylin and eosin staining). D. The mass was composed of single-layered cords of uniform cuboidal cells with basophilic nuclei (400x hematoxylin and eosin staining).

***Corresponding author:** So-Young Choi, Department of Oral and Maxillofacial Surgery, School of Dentistry, Kyungpook National University, Daegu, South Korea, Tel: +82 53 600 7576; Fax: +82 53 426 5365; E-mail: dentalchoi@knu.ac.kr

Received September 13, 2017; **Accepted** November 22, 2017; **Published** November 27, 2017

Citation: Kim D, Paeng J, Lee S, Choi S (2017) Canalicular Adenoma of the Parotid Gland: A Rare Case Report and Review of Literature. J Clin Case Rep 7: 1041. doi: 10.4172/2165-7920.10001041

Author	Year	Age	Gender	Symptom	Site	Size	Treatment
Rossiell et al. [9]	2002	52	Female	Swelling with pain	Left	2 cm	Superficial parotidectomy
Philpott et al. [8]	2005	71	Female	Swelling	Right	not available	Superficial parotidectomy
Liess et al. [7]	2006	26	Male	Swelling	Left and right	1 cm (left), 2 cm (right)	Superficial parotidectomy
Butler et al. [6]	2009	85	Female	Swelling	Not available	1.7 cm	Excisional biopsy
Current report	2017	81	Male	Swelling	Left	1.5 cm	Local excision

Table 1: An overview of clinicopathological features of CA of the parotid gland, obtained from the literature.

Parallel rows of cords appear to form ductal structures or long canals (Figures 2C and 2D). These findings were consistent with CA; thus, the final diagnosis was CA. The patient showed good prognosis, with no complications or recurrence at the time of reporting.

Discussion

Salivary gland tumors are unusual and less than 3% of all head and neck neoplasms [12]. CA represents less than 1% of all salivary gland tumors [13]. Most salivary gland tumors of the head and neck involve the major salivary gland, with benign, minor salivary gland tumors often involving the hard palate, buccal mucosa and tongue [14]. CA, however, has a significant predilection for occurrence in the minor salivary glands, with highest occurrence observed in the upper lip, followed by the buccal mucosa. CAs are sometimes multifocal. Despite this predilection for the upper lip, there are reports of CA occurring in the lower lip [15-17]. There have been no reports of CA of the submandibular gland or sublingual gland, and CA is rarely found in the parotid gland; to our knowledge, only 14 cases have been observed, based on the findings of 10 case reports. Reported examples of CA of the parotid gland are summarized in Table 1 [5-9,18-22]. Ansari et al. also reported monomorphic adenomas in the parotid gland, but provided no differentiation between CA and basal cell adenoma [23]. In addition, six of these reports did not provide specific information, such as clinical symptoms, histologic findings, or treatment methods [5,8-22].

Clinically, CAs are grossly well-circumscribed nodular lesions, which usually present with no clinical symptoms except for swelling, although painful CA has been reported by Rossiello et al. [9]. Commonly, CAs range from 0.5-2.0 cm in diameter and are grossly well-encapsulated [13]. However, in 1984, Daley et al. reported that approximately 10% of CAs were found in their non-encapsulated forms.5 Despite being uncommon, this histologic form could explain the recurrence tendency of CA.

In our patient, we performed FNA and CT with intravenous contrast. FNA is considered a safe, soft, and cost-effective diagnostic modality that causes little discomfort to the patient and carries less risk than more invasive procedures in terms of specimen acquisition [24]. However, the effectiveness of FNA has been disputed [25,26]. Furthermore, when FNA is used during cytological examination of head and neck lesions, accuracy is lowest for lesions of the parotid gland [25,27]. This inaccuracy may be due to the complexity, diversity and relatively low incidence rate of parotid gland tumors [25,27,28]. Although this makes FNA an ancillary diagnostic modality regarding parotid gland tumor diagnosis, it remains effective at differentiating malignant and benign tumors [29]. Our FNA results were suggestive of Warthin`s tumor, therefore we suspected that the tumor was more likely to be benign than malignant. To our knowledge, only one literature report has described CT results as part of CA diagnosis. Yamada et al. reported that CA was observed in the CT image as a homogenously enhanced mass with a clear margin. Similarly, our CT image showed an enhanced oval mass on the left side of the parotid gland, which, like the FNA findings, indicated a benign tumor [30].

Histologically, we observed single-layered cords of uniform cuboidal cells with basophilic nuclei, which were well-encapsulated by fibrous tissue (Figures 2B and 2D). The nuclei were monomorphic rather than polymorphic. A mitotic figure was not observed, and nucleoli were not conspicuous. Parallel rows of cord appeared to form ductal structures or long canals, with a characteristic beaded appearance (Figure 2C). These features were in accordance with CA, as described by Barnes et al. in 2005 [13].

The most important lesions to consider in the differential diagnosis of CA are mucoceles, 31 pleomorphic adenomas, and basal cell adenomas. However, in the parotid area, Warthin's tumor and adenoid cystic carcinoma should be also differentiated from CA. Adenoid cystic carcinomas with cribriform, tubular patterns tend to be misdiagnosed as CA [3,9]. Historically, CA was considered a subgroup of basal cell adenoma or pleomorphic adenoma [5]. Pleomorphic adenomas can be differentiated from CAs by their chondromyxoid components, despite their similar clinical symptoms, such as swelling and free movement. Basal cell adenoma of the trabecular subtype could be similar to CA histopathologically; however, they can be differentiated from CAs by their specific ultrastructural features and their characteristic myoepithelial lineage [31,32]. Unlike CA, basal cell adenomas are composed of multilayered cords of polygonal or cuboidal cells and exhibit a scanty amount of basophilic or amphophilic cytoplasm [13]. Differential diagnosis between benign entities is not crucial, because treatment considerations among them do not differ. However, it is important to differentiate between CA and malignant entities, such as adenoid cystic carcinoma, because the treatment methods are drastically different.

In this case, the radiologic and histologic findings were consistent; they identified the lesion as well-encapsulated with hypervascularity. Based on the FNA, CT, and clinical results, the lesion was thought to be a benign tumor of the parotid gland. Although superficial parotidectomy could be considered as a surgical treatment option, considering the patient's old age, local surgical excision was adopted as the most appropriate strategy. This strategy was suitable for the patient, as there have been no reports about malignant formation, and this lesion has a relatively low recurrence tendency.

Conclusion

Although CA is an uncommon entity, especially when it is not located on the upper lip, it is important to consider this tumor type when differentiating between pleomorphic adenoma, basal cell adenoma, Warthin's tumor, and especially adenoid cystic carcinoma, when this occurs in the parotid gland. An appropriate biopsy is required for accurate diagnosis, because it may be difficult to differentiate CA from other diseases using FNA and CT. Thus, CA should be considered when evaluating salivary gland tumors of the parotid gland.

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