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Canalicular adenoma of minor salivary gland. Report of a case and a brief review of the literature

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SUMMARY

We refer a case report of a canalicular adenoma of minor salivary gland, located at the submucosal region of the upper lip. The patients' clinical state was thoroughly studied, along with the histopathological findings. The surgical excision was the treatment of choice. Numerous histogenesis theories and the appropriate tumor treatment are mentioned, being always in accordance with the relative literature. Canalicular adenoma of minor salivary gland is a rare oral mucosa lesion. The differential diagnosis among many oral swellings is interesting. The surgical excision is the treatment of choice. The histological examination confirms the clinical diagnosis.

Key words: benign salivary gland neoplasm, canalicular adenoma, minor salivary gland.

INTRODUCTION

Canalicular adenoma (CA) constitutes a rare benign tumor which derives from minor salivary gland, although it was initially suggested that this lesion has a terminal duct origin (1, 2). McFarland was the first who described this lesion in 1942 as canalicular tumor, whereas some decades later other authors suggested the term monomorphic adenoma (3, 4). According to the latter term, CA and Basal cell adenoma (BCA) thought to be the same entity1. Few authors claimed that CA was merely a variant of BCA5. Nonetheless, the World Health Organization in 1991 (WHO) set a salivary gland tumours classification scheme where CA was recognized as separated tumor (1, 6).

A prevalence in adult females between the fifth and seventh decade of life with a ratio 1.8:1 from female to male is reported (6, 7). CA represents 2-6.5% of all head and neck benign lesions as well as 1–3% of all salivary tumors (5, 6). Albeit, it is referred as the second or the third most frequent neoplasm of minor salivary glands (6). In addition, geographic and racial factors may influence the incidence of CA (8, 9). In some studies a higher frequency of CA is stated in North American and European populations compared to Asian populations (9).

The most frequent location of CA reported is upper lip (80%), followed by buccal mucosa, palate, tongue, floor of mouth, larynx, parotid gland, maxillary sinus, mandible, oesophagus, retromolar area (1, 6, 7, 9).

Clinically, the tumor appears to be a multifocal (13%) or solitary (87%), painless, mobile, slowly growing, firm or slightly fluctuant on palpation, submucosal swelling (7, 8, 10), that does not usually cause ulceration of the overlying mucosa (1, 2). The aforementioned mucosa appears usually pinkish while in some cases its hue may also be bluish mimicing a mucocele (6-8). The majority of CA range between 0.5-2 cm in diameter (6, 8). As regards pain, most lesions are reported as asymptomatic until the size of the swelling becomes visible (1).

CASE REPORT

An 84-years-old male patient was referred to our Clinic by his dentist, who detected a submucosal swelling, located at the upper labial mucosa, close to the central frenum (Figure 1). His medical history included Parkinson's disease, depression and a moderate dementia, well-controlled by the appropriate medication. The intraoral examination revealed

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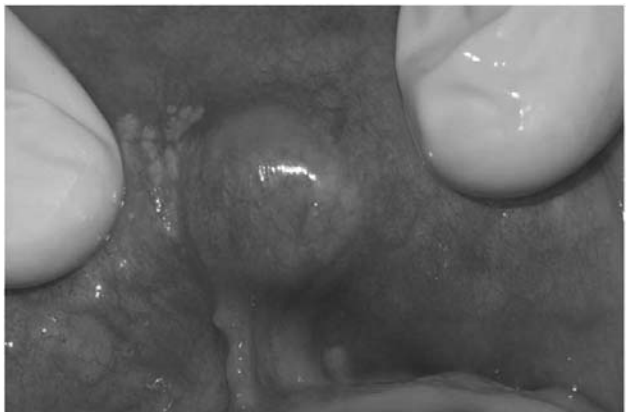

Fig. 1. Preoperative appearance of the lesion

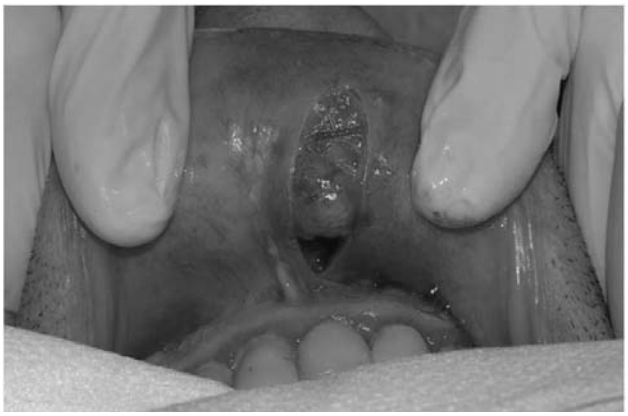

Fig. 2. Interoperative appearance. The well defined lesion is surgically detached from the epithelium which covered it.

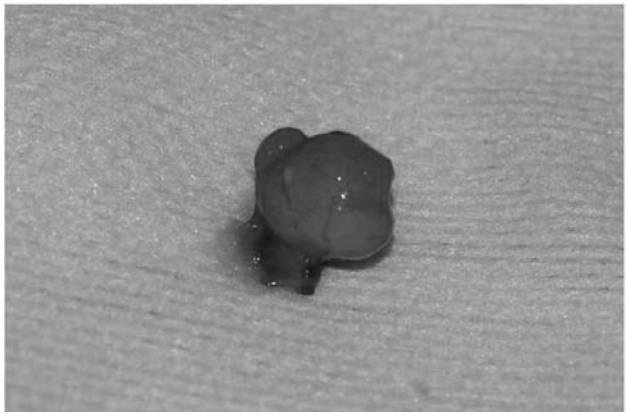

Fig. 3. The surgically excised lesion

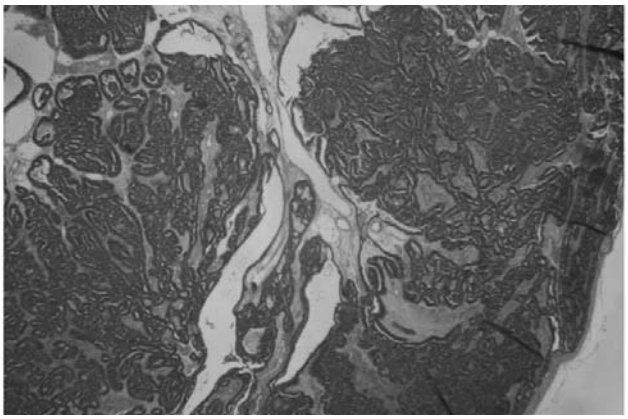

Fig. 4. Canalicular adenoma covered by thin fibrous capsule (H-E ×50)

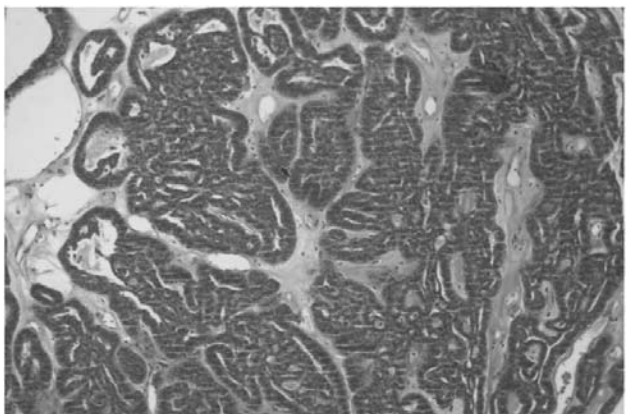

Fig. 5. The epithelial element of canalicular adenoma is composed of cylindrical cells, without atypia, while mesenchymal element is myxoid (H-E X100)

a well circumscribed submucosal tumor about 7 mm in diameter, covered by normal in color labial mucosa. The lesion was hard, painless and movable in palpation. There were no swollen cervical lymph nodes. No clear information was given concerning the time when the lesion was primarily observed.

The initial clinical diagnosis included oral benign neoplasm and chronic sialadenitis of minor salivary glands due to sialoliths of relevant glands. The tumor was surgically excised in toto under local anesthesia (Figures 2 and 3). The post-operative period was uneventful. Histological examination revealed a CA (Figures 4 and 5).

DISCUSSION

A case of a CA is described. The tumor was an asymptomatic submucosal swelling in the upper labial mucosa. Histological examination of the surgically excised swelling confirmed the diagnosis of CA.

The main histological appearance of CA comprises solid structures, trabeculae, tubules, and cribriform or membranous patterns (6). More specifically, CA appears to be often encapsulated and multifocal, and it is composed of isomorphic columns of columnar and cuboidal cells, which constitute strands or ducts in a loose highly vascular stroma with small cystic spaces (1, 5, 7). Moreover, the nuclei are monomorphic with scattered, stippled chromatin (5). Mitoses are infrequent as well as necrosis (5). A study detected intraluminal squamous balls in 61% of their cases and microliths in 50% (2).

Immunohistochemical markers have also been used for diagnosis of a CA. The most useful tools

include positivity for p16, CD15, cytokeratin AE1/AE3 and S-100 protein (7, 8) as well as negativity for vimentin, A-SMA, CK5/6 and p63 (1, 2, 7). However, the necessity of this examination remains quentionable (5).

Diagnosis is set with the use of excisional biopsy while fine needle aspiration and incisional biopsy bear contra-indications (8). In some cases, CA appears to be multifocal and either partially encapsulated or even unencapsulated, which may lead in a misdiagnosis (8). Extra caution is needed in order to distinguish CA from other benign or malignant tumors. Differential diagnosis includes BCA, polymorphous low grade adenocarcinoma, adenoid cystic carcinoma, papillary cystadenocarcinoma, mucoepidermoid carcinoma, benign and malignant neoplasias of glandular tissue, sialolithiasis with secondary sialadenitis, vascular anomalies, lipomas, mucocele, thrombosed vessel, ductal adenoma, reticulated myoepithelioma, ameloblastoma, adenomatoid odontogenic tumor, paraganglioma, and skin basal cell carcinoma (9-11).

The treatment of choice includes the thorough surgical excision of the lesion (1). In the rare case of a CA located at the parotid gland, the recommended treatment was parotiedectomy (5). Enucleation has been suggested as alternative treatment of CA as well (10). As regards incisional biopsy, it should not be indicated due to the possibility of histological artefacts and the low risk of fragmented specimens owing to the cystic or multifocal structure of CA (10).

CA shows a recurrence rate of 5% of all surgically excised lesions (5). Albeit, some authors claim that recurrence is related to the multifocality of tumor and as a result it quite difficult to distinguish persistence of the lesion from recurrence (5, 7). Therefore, it is recommended a long-term follow up (approximately 11 years) without any risk of malignancy (6, 11, 12). One more supportive element of the benignity of CA is the absence of aggressive features of the tumor, whereas multifocal and cribriform patterns should not be counted as evidence of malignant transformation (1, 6).

CONCLUSION

The CA is a rare minor salivary gland benign tumor, usually growing on the upper lip. The thorough surgical excision of the tumor constitutes the treatment of choice. The histological examination confirms the diagnosis and it should be conducted carefully, in order to avoid misdiagnosing the CA as other benign or malignant tumors.

CONFLICT OF INTEREST

The authors report no conflict of interest.

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