



Basal cell adenoma of the salivary gland: Cribriform type, a rare case with review of literature

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ABSTRACT

Basal cell adenoma (BCA) of the salivary glands is a rare benign tumor resembling pleomorphic adenoma, but with a prominent basaloid cell layer. The majority of these tumors arise in the parotid glands and account for only 1% of all salivary gland epithelial tumors. We report one such case of a swelling in the floor of the mouth in a 55-year-old female where BCA is the most likely diagnosis; however, histological variation does show a similarity to malignant adenoid cystic carcinoma, thereby making the diagnosis difficult. The incidence of malignancy is relatively higher in the submandibular, sublingual and minor salivary glands. Approximately, 85% of sublingual gland tumors are malignant. Thus, we should be more careful when making a diagnosis in minor salivary gland tumors.

Key words: Adenoid cystic carcinoma, adenoma, basal cell, clinicopathological feature, cribriform, excision, immunohistochemistry, minor salivary gland, oral cavity

Introduction

Basal cell adenomas (BCAs) are rare occurrences and most of them usually arise from the parotid glands (80%)[1]. Other sites, such as the lips, palate and nasal septum have also been encountered. This condition usually affects people in the fifth decade of life, with a higher prevalence in women than men. Histologically, a basaloid cell layer surrounded by hyaline substance is noted. The differentiating factor for this tumor, when compared to other benign mixed tumors and salivary gland neoplasms is the absence of myoepithelial cells. A strong suspicion of other diagnoses, such as pleo-

morphic adenoma and malignant adenoid cystic carcinoma, should be present as these tumors often have histological similarities, especially if relying on cytology alone. Here, we present a case of one such diagnostic uncertainty in a 55-year-old woman who presented with an oral tumor, which was found to be a BCA, most likely.

Case Report

A 55-year-old woman non-smoker, presented to the surgical department with 2 years history of a painless swelling in the floor of the mouth. The swelling was slowly growing in size. She reported no associated

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Figure 1. An oval swelling measuring 4 cm × 3 cm was seen on the floor of the mouth on the right side, not crossing the midline.

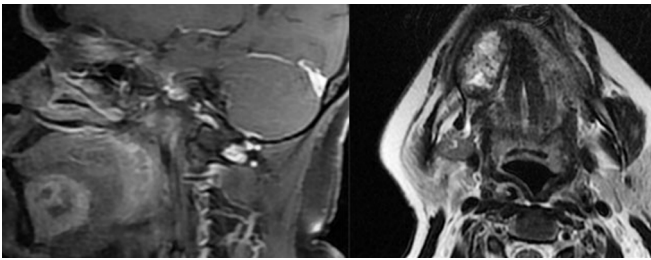


Figure 2. Magnetic resonance imaging of the head and neck showed a well-defined, lobulated heterogeneously enhancing T1 iso-hypointense and T2 hyperintense lesion measuring 3.5 cm × 2.9 cm × 3.2 cm in the right sublingual space.

features such as xerostomia, sialorrhea, or pain. There was no difficulty in the movement of the tongue. Her general well-being was otherwise unremarkable, and oral hygiene was good. Her background included hypertension and type 2 diabetes mellitus, as well as clinical depression.

On examination, all her vitals were stable. An oval swelling measuring 4 cm × 3 cm was seen on the floor of the mouth on the right side, not crossing the midline (Figure 1). The mucosa over the swelling appeared normal, and the trans-illumination test was negative. There was no tenderness over the swelling and no local rise of temperature. It was firm to hard in consistency and was non-fluctuant, but was mobile in all directions. No local lymph nodes were palpable.

Her blood results were all within normal limits (full blood count, urea, and electrolytes). Subsequently, she underwent an ultrasound scan of the neck, which showed a well-defined hypoechoic lobulated lesion with smooth margins in the right submandibular region deep to the muscles. Magnetic resonance imaging of the head and neck showed a well-defined, lobulated heterogeneously enhancing T1 iso-hypointense and T2 hyperintense lesion measuring 3.5 cm × 2.9 cm × 3.2 cm in the right sublingual space (Figure 2). Non-enhancing necrotic areas were seen within the tumor.

There was no evidence of infiltration of the adjacent muscles or median raphe. No hemorrhage or calcification was observed within the lesion. The right submandibular gland was normal in size and signal intensity. A few sub-centimeter lymph nodes were seen in the level 1 B region.

An excision biopsy of this lesion was planned. Intra-operatively there was no infiltration of the tumor to the surrounding muscles and a wide local excision with 0.5 cm margin was performed (Figure 3). She was then discharged post an uneventful recovery and is on regular follow-up.

The histopathology (HPE) reported an encapsulated tumor composed of small basaloid cells arranged in

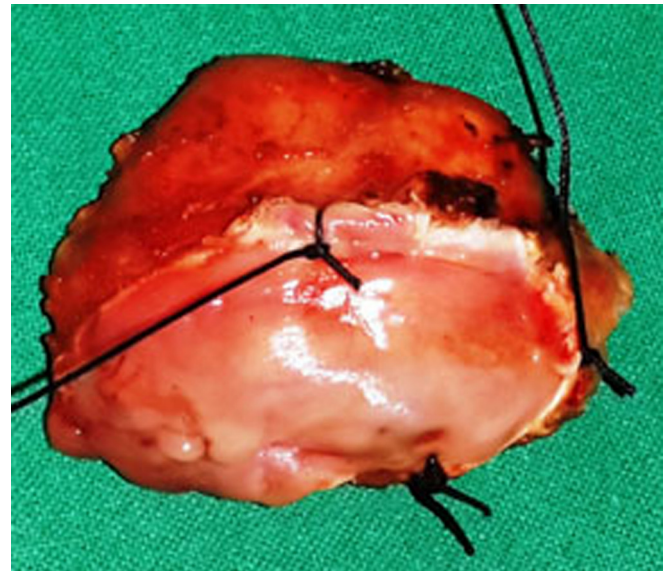


Figure 3. Wide local excision of the tumor with 0.5 cm margin.

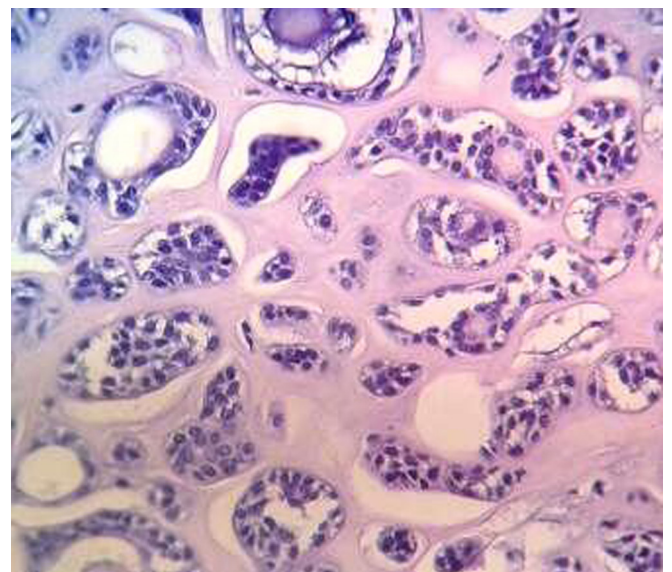


Figure 4. Histopathology revealed an encapsulated tumor composed of small basaloid cells arranged in tubules and a cribriform pattern.

tubules and a cribriform pattern. The cells were found to have basophilic nuclei and scant cytoplasm, containing densely eosinophilic basement membrane like material within the tubules. These were surrounded by hyalonized stroma with thin walled blood vessels and occasional lymphoplasmacytic infiltrate. No perineural invasion was seen (Figure 4). The conclusion from that report was of a high likelihood of the lesion being a BCA of the salivary gland. The absence of perineural invasion and good circumscription with a capsule ruled out adenoid cystic carcinoma in this case. However, immunohistochemistry (IHC) staining with C-kit favored a diagnosis of adenoid cystic carcinoma. In view of this, a regular follow-up of the patient was suggested.

Discussion

Salivary gland tumors account for <3% of all the neoplasms [2]. Approximately, 80% of these are of epithelial origin, and the majority of these being benign adenomas. As mentioned before, BCAs constitute about 1% of the salivary gland tumours. Kleinsasser and Klein first described monomorphic adenomas in 1967 and BCAs are a subtype of this entity, as classified by the World Health Organization [3].

BCA are benign and made up of basaloid cells with a definite basement membrane like structure and lacking a myoepithelial component, unlike pleomorphic adenomas [4]. Just-like in our patient, BCAs are characterized by their slowing growing nature and are generally asymptomatic. Our patient also fits into the average age range for developing this lesion, which is thought to be around 57 years [5]. BCA also tend to form cysts, although this was not observed in our case.

Histologically, there are several similarities between BCA and pleomorphic adenomas; however, the distinguishing feature of BCA is the absence of chondroid tissue and myxoid stroma [6]. Instead, BCA have numerous endothelial blood vessels with prominent small vessels; hence, they appear hemorrhagic on Computed tomography [7]. Adding weight to this diagnosis is the fact that pleomorphic adenomas tend to affect younger people more often than older patients, such as ours.

Another factor contributing to the diagnostic dilemma in such types of intraoral tumors is the near similarity between the benign and malignant neoplasm.

The common minor salivary gland tumors such as the pleomorphic adenoma, BCA, adenoid cystic carcinoma, and low grade adenocarcinoma present with similar morphological and immunohistochemical features to a large extent, thereby making a preoperative diagnosis difficult. An excision biopsy would be ideal to clinch the diagnosis [8]. The presence of the cribriform pattern in this case makes it a rare case and relatively difficult to distinguish from the adenoid cystic carcinoma. The cribriform type has a higher female predilection, affect the elderly, tend to be well-circumscribed and do not recur after a local excision [9]. IHC plays a key role in differentiating these types of tumors from the adenoid cystic carcinoma. These types of tumors are negative for MMP9, laminin, CD117, and myoepithelial marker (SMA, S100 and calponin) in the cells around the cribriform spaces [9]. If this is indeed the case, the patient will have a very good prognosis, as these tumors are benign and without any infiltrative features.

Conclusion

BCAs are rare benign neoplasms of the salivary gland and share a close resemblance to malignant adenoid cystic carcinoma, especially the cribriform type. A clear HPE report, along with immunohistochemistry is needed to distinguish between these two entities. Our case highlights the diagnostic challenge encountered when faced with such a lesion and clinicians should explore both possibilities prior to making final management decisions.

Conflict of interest statement

The authors have no conflicts of interest to declare.

References

1. Esteves AR, Dib LL, de Carvalho LV. Basal cell adenoma: A case report. *J Oral Maxillofac Surg* 1997;55:1323-5.
2. Klijanienko J, el-Naggar AK, Vielh P. Comparative cytologic and histologic study of fifteen salivary basal-cell tumors: Differential diagnostic considerations. *Diagn Cytopathol* 1999;21:30-4.
3. Jao W, Keh PC, Swerdlow MA. Ultrastructure of the basal cell adenoma of parotid gland. *Cancer* 1976;37:1322-33.
4. Jang M, Park D, Lee SR, Hahm CK, Kim Y, Kim Y, et al. Basal cell adenoma in the parotid gland: CT and MR findings. *AJNR Am J Neuroradiol*

- 2004;25:631-5.
5. Luna MA, Tortoledo ME, Allen M. Salivary dermal analogue tumors arising in lymph nodes. *Cancer* 1987;59:1165-9.
 6. Chawla AJ, Tan TY, Tan GJ. Basal cell adenomas of the parotid gland: CT scan features. *Eur J Radiol* 2006;58:260-5.
 7. Chiu NC, Wu HM, Chou YH, Li WY, Chiou YY, Guo WY, et al. Basal cell adenoma versus pleomorphic adenoma of the parotid gland: CT findings. *AJR Am J Roentgenol* 2007;189:W254-61.
 8. Turk AT, Wenig BM. Pitfalls in the biopsy diagnosis of intraoral minor salivary gland neoplasms: Diagnostic considerations and recommended approach. *Adv Anat Pathol* 2014;21:1-11.
 9. Li BB, Zhou CX, Jia SN. Basal cell adenoma of salivary glands with a focal cribriform pattern: Clinicopathologic and immunohistochemical study of 19 cases of a potential pitfall for diagnosis. *Ann Diagn Pathol* 2014;18:5-9.

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