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Case Report / Olgu Sunumu

Basal cell adenocarcinoma of the minor salivary gland: a case report

Minör tükürük bezinde bazal hücreli adenokarsinom: Olgu sunumu

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Basal cell adenocarcinoma (BCA) is an uncommon salivary gland neoplasm. The majority of salivary gland tumors are located in the parotid gland, while only a few involves the minor salivary gland of the oral cavity. Pathological diagnosis is important to differentiate BCA from other neoplasms, as the prognosis of the diseases is significantly different. In this article, we present a 41-year-old-male with BCA involving the upper labial mucosa.

Key Words: Basal cell adenocarcinoma; minor salivary glands; prognosis.

Bazal hücreli adenokarsinom (BHA), minör tükürük bezinde nadir rastlanan bir tümördür. Tükürük bezi tümörlerinin büyük bir çoğunluğu parotis bezinde; çok az bir kısmı ise oral kavitenin minör tükürük bezinde görülür. Hastalıkların prognozu anlamlı düzeyde farklı olduğundan, BHA'nın diğer tümörlerden ayırt edilmesinde patolojik tanı önem taşır. Bu makalede, üst dudak mukozasında yerleşmiş BHA'sı olan 41 yaşında erkek hasta sunuldu.

Anahtar Sözcükler: Bazal hücreli adenokarsinom; minör tükürük bezi; prognoz.

Basal cell adenocarcinoma (BCA) is a rare salivary gland tumor which is considered to be the malignant counterpart of basal cell adenoma. Most BCAs have been reported to occur in major salivary glands, most commonly the parotid gland. In minor salivary glands BCAs are extremely rare.[1] The histopathology of salivary gland tumors is complex and may be problematic for even the most experienced pathologist.[2] The major pathologic differential diagnostic considerations for BCA are basal cell adenoma and adenoid cystic carcinoma. Distinguishing BCA from adenoid cystic carcinoma is important due to poorer prognosis and higher prevalence of latter disease.[3] There are very few reports of lesions in minor glands. Basal cell adenocarcinoma is considered a low-grade malignancy but clinicopathologic behavior of this tumor is still unclear.[4]

In this report, we present a case of BCA involving the upper labial mucosa and discuss the treatment and differential diagnosis of BCA according to the literature.

CASE REPORT

A 41-year-old male was admitted to our clinic with a complaint of swelling involving the upper labial mucosa. Intraoral examination revealed a painless 3 cm diameter mass lesion, with well

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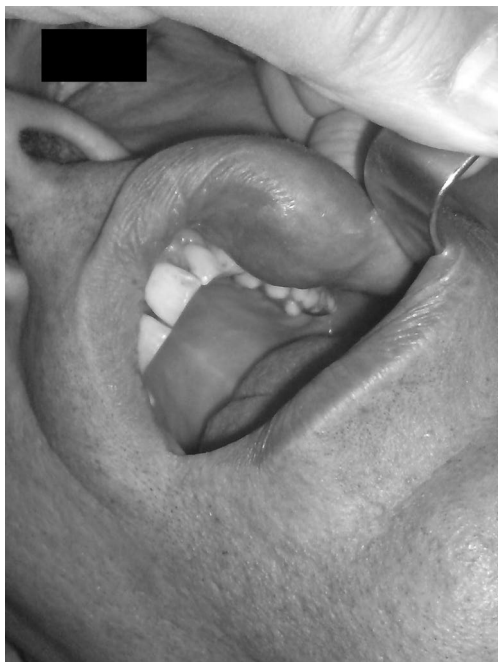

Figure 1. *The mass lesion involving the upper labial mucosa.*

defined borders and normal overlying mucosa (Figure 1). No palpable lymph node was noticed in the neck. No clinical signs of malignancy and no pathologic activity were observed on positron-emission tomography (PET) preoperatively. Informed consent was obtained from the patient.

Fine needle aspiration cytology revealed small epithelial cells observed around the hyaline stromal globule on the erythrocyte ground. The basoloid cells which sometimes form tridimensional groups with narrow cytoplasm which had thin granular nuclear chromatin were seen (Figure 2). In order

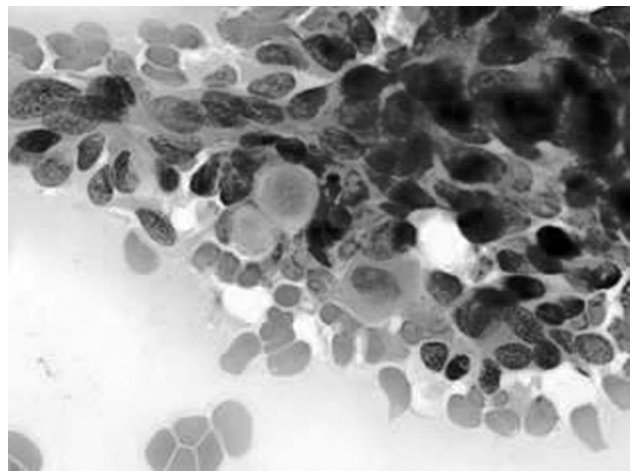

Figure 2. *The basoloid cells which form tridimensional group around hyaline globule (H-E x 1000).*

to arrive at a definite pathological diagnosis, wide transoral surgical excision with complete removal of the tumor was performed. Following wide local excision, macroscopically, the mass lesion measured 2.5x2x1.5 cm, was dirty white in colour and solid in consistency. On histopathological examination, tumor islands with palisade shape were observed. There were two types of tumor cells with variable sized solid and trabecular structures in the collagenous stroma. The tumor consisted of two types of cells: the narrow cytoplasmic small cells with dark nuclei and the cells with polygonal eosinophilic cytoplasm. Vascular invasion and soft tissue infiltration were observed (Figure 3 and 4). Immunohistochemical evaluation revealed pancytokeratin (+), vimentin (+), actin (-), CD117 (-), CD34 (-) and diagnosed as basal cell adenocarcinoma.

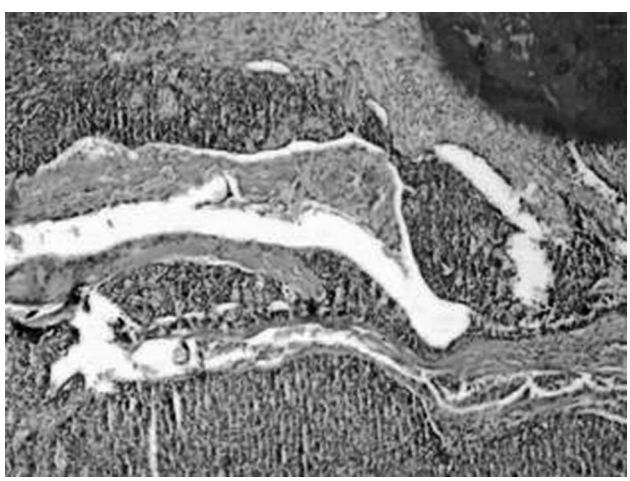

Figure 3. *The histopathological configuration of vascular invasion, (H-E x 200).*

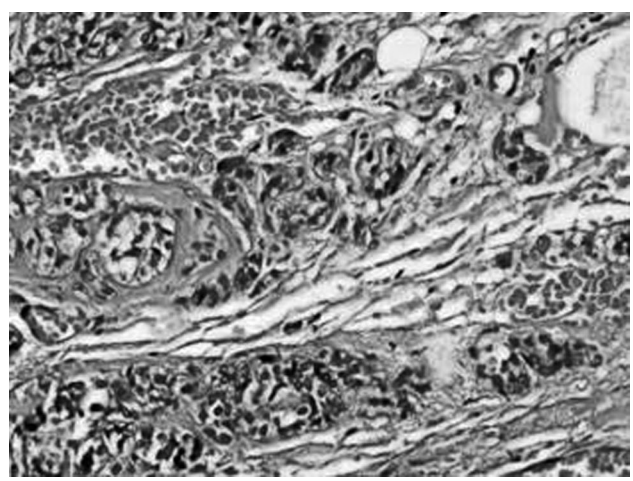

Figure 4. *Histopathological aspect of soft tissue infiltration, (H-E x 400).*

The postoperative period was uneventful, and there was no evidence of recurrence after 14 months of follow-up.

DISCUSSION

Malignant tumors of the intraoral minor salivary glands constitute 2-3% of all malignant neoplasms of the upper aerodigestive tract and less than 25% of all salivary gland tumors. Most tumors arising in minor salivary glands are malignant.[5] The most common sites are the hard palate, nasal cavity and paranasal sinuses. Basal cell adenocarcinomas comprise 1.6% of all salivary gland neoplasms and 2.9% of malignant salivary gland neoplasms. This tumor most commonly involves the parotid gland (90%) and very rarely the minor salivary glands of the oral cavity. The average age of patients is 60 years with no sex predilection.[6] Yu et al.[7] reported that BCA was most frequently in the minor salivary glands, but Fonseca and Soares[8] described that BCA of salivary glands was an uncommon entity occurring almost exclusively in the major salivary glands. The differential diagnosis of the neoplasm includes its benign counterpart, the basal cell adenoma, a solid variant of adenoid cystic carcinoma, undifferentiated carcinoma, and basaloid squamous carcinoma. Diagnosis of basal cell adenocarcinoma hinges on the identification of invasion into adjacent tissue. Vascular and perineural invasion are often seen in basal cell adenocarcinoma.[8] In our case, the vascular invasion and soft tissue infiltration were observed on the histopathological examination.

Very few cases have been reported in the minor salivary gland. Gupta et al.[9] reported a case of BCA in the tongue. Peel and Seethala[10] reported a case of BCA in buccal mucosa and Parashar et al.[11] described BCA in the lip and tongue. Poulopoulos et al.[4] reported a BCA case involving the upper labial mucosa. Ward et al.[3] presented a case of BCA originating from a hard palate minor salivary gland. In our case, we presented a BCA case involving the upper labial mucosa. There was no evidence of metastasis in our case.

Most BCA cases are believed to develop de novo but as many as 25% of cases may arise from a pre-existing basal cell adenoma. Basal cell adenocarcinomas are low-grade carcinomas that rarely cause distant metastasis. They may be locally destructive with a propensity to recur.[12] The diagnosis of BCA may be difficult because clinical examination, computed tomography, ultrasonography and magnetic resonance imaging occasionally do not exclude other benign and malignant salivary gland tumors, and microscopic examination of biopsy specimens sometimes is unable to distinguish it from basal cell adenoma. If a tumor has a marked similarity to basal cell adenoma, further examination is necessary to determine the final diagnosis.[13]

Surgical excision with a wide margin to ensure complete removal has been suggested as the primary treatment for BCA. Enucleation and curettage is to be avoided.[9] Regional lymph node dissection is recommended only if there is evidence of metastatic disease.[4] In minor glands, wide excision with postoperative radiotherapy has been proposed on the basis of a higher likelihood of neural and vascular invasion.[13] But Parashar et al.[11] stated that BCA is an exceptionally rare salivary gland tumor that demands complete surgical removal with adequate margins despite its low-grade behavior. As with minor salivary gland malignancies in general, postoperative radiation is recommended for close surgical margins or following surgical excision of recurrent disease.[3] Furthermore, it has been reported in some studies that the conservative surgery for BCA in minor salivary glands may be a viable option.[4,9] Therefore, we performed wide surgical excision in our presented case and observed no recurrence or any trouble postoperatively. We did not perform regional lymph node dissection because there was no evidence of metastatic disease in our case. Scholtz et al.[14] reported that the recurrence rate of BCA was about 25-30% and 10% metastatize to regional lymph nodes or distant organs.

In conclusion, BCA is a rare entity occurring in minor salivary glands and BCA of the labial mucosa is a very rare presentation. It is important to distinguish BCA from other neoplasms like adenoid cystic carcinoma and basal cell adenoma because of the differences for prognosis and treatment.

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