CASE REPORT

Cribriform Adenocarcinoma of Minor Salivary Gland in a 47-Year-Old Non-Smoker Man: An Educational Case Report

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Abstract

Introduction: Salivary gland tumor is a rare disease which consists less than 5% of head and neck neoplasms. In this case-report we have described a patient with rare immunohistopathological findings that is known as cribriform adenocarcinoma.

Case presentation: A 47-years-old non-smoker man suffering right cervical mass was evaluated by ultra-sonography and computed tomography (CT).Although USG findings were normal, however, CT-scan revealed necrotic, hypodense lymphadenopathy in carotid and right submandibular area. Right medial mass of mandibular ridge measuring 30×14 mm with destroying mandible was seen. The mass was growing through medial to lateral part of sublingual region.

Management and prognosis: Lesion dissection and pathologic evaluation showed polymorphous low-grade adenocarcinoma in gingiva and right mandible, infiltration of malignant cells into the lymph nodes with cribriform. Creatine kinase, especially CK 5/6, Vimentin, Cytokeratin, KI-67, S-100, Bcl2 and Epithelial Membrane Antigen were positive in immunohistochemical evaluation confirming myoepithelial carcinoma and cribriform adenocarcinoma. No infiltration was seen in essential soft tissues surrounding the mass origin.

Conclusion: Precise and early diagnosis are necessary for prevention of tumor invasion and differentiation from low grade adenocarcinoma and low-grade adenocarcinoma that have similarity to cribriform adenocarcinoma.

Keywords: Adenocarcinoma, Case Report, Head and Neck Neoplasms, Immunohistochemistry, Salivary Glands

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Introduction

Salivary gland tumor (SGT) is a rare disease which consists of less than 5% of tumors in head and neck.[1] Different types of tumors have been reported in salivary glands. Nowadays, approximately 20 types of malignant tumors were seen in salivary glands.[2] The most reported of SGT are not malignant, and adenoma is the most common ones, while mucoepidermoid and cystic carcinoma are the most malignant mass which was seen in salivary glands.[1] Epidemiologic surveys have been declared the effects of demographic and geographic regions on the prevalence of the SGTs and their histopathologic findings. Elder patients and males are more susceptible to overtake SGT.[3,4] Recent studies have been reported that malignant tumors consist of approximately ones in SGTs and adenocarcinoma had the least prevalence.[5,6]

A retrospective study from 1971 to 2009 was done on 165 patients who suffered from salivary gland tumors in Mashhad, Iran and a reevaluation done on histopathologic specimen according to world health organization. This study declared that the most common malignant SGT is mucoepidermoird carcinoma. They stated that the palate is the most probable region to find salivary tumors, while similar studies mentioned Parotid gland as the most probable one. The researchers concluded that focusing on palate and other sites of minor salivary glands are as important as Parotid gland.[7]

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According to the previous studies, this case was reported for its rare immunohistopathological findings which was reported as cribriform adenocarcinoma.

Case Presentation

A 47-years-old non-smoker man referred to Golestan hospital, Ahvaz, Iran suffering from right cervical mass from 2 months ago.

There was no erythema, tenderness and warmness on the mass region. The tumor was not mobile and the patient did not have any past medical or familial history. Hoarseness and snoring were negative. There was not any significant findings in laboratory data. Screening tests including upper and lower Gastrointestinal endoscopy, and parenchyma of lungs were normal, too.

In the ultra-sonographic evaluation, lobulated hypoechoic area measuring 30×14 mm suggestive of infiltrated nodal recurrence was seen at the right side of neck deep to Sternocleidomastoid muscle. Both thyroid lobes had normal size and homogenous echo pattern. Salivary glands were symmetric and normal and other cervical structures were symmetric and normal.

Brain MRI with Gadolinium declared that all parts of gray and white matter in central nervous system (CNS) were normal. There was not any involvement in sella, skull base, upper cervical spine, paranasal sinus and mastoid air cell, vascular system, Calvarium and bone marrow (Figure 1).

Intravenous contrast computed tomography (CT) was done and cervical vascular enhancement was normal. Necrotic, hypodense lymphadenopathy was reported in carotid region and right submandibular area. The CT findings were suggestive for Lymphoma or metastasis. Right medial mass of mandibular ridge measuring 30×14 with destroying mandible was seen. The mass was growing through medial to lateral part of sublingual region. Other probable origins for metastasis like lung, liver and far lymph nodes were all normal in evaluation.

Because of highly suspicious to malignant lesions, the patient candidate for neck exploration and dissection. The specimen including an encapsulated piece of 6×3×1.5-centimeter cream-brownish rubbery tissue with hemorrhagic foci was prepared for pathologic reviews.

In pathological records, polymorphous low-grade adenocarcinoma was seen in Gingiva and right mandibular area. Right Palatine tonsils and nasopharynx were free of tumor. Microscopic evaluation showed a lymph node tissue infiltrated by malignant cells showing diverse growth patterns including cribriform, solid and microcytic spaces which were filled with mucoid material. No significant nuclear atypia was seen but nuclei showed irregular membrane and frequent nuclear grooves. Mitotic activity was easily seen (Figure 2). Immunohistological findings showed that Creatine kinase (CK) especially CK 5/6, Vimentin, Cytokeratin, KI-67, S-100, Bcl2 and Epithelial Membrane Antigen (EMA) were positive, while TTF1, Napsin, CD10 and Calponin were negative. They declared that the lymph nodes were involved due to myoepithelial carcinoma. Figure 3 shows three samples of immunohistochemistry results for S100 (A: positive reaction), Bcl2 (B: positive reaction) and Calponine (C:

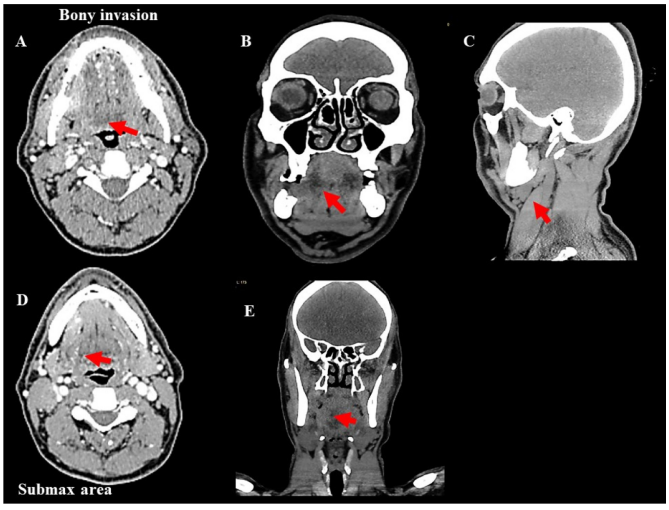

Figure 1. Selected views of Gadolinium-enhanced MRI of presented case. Necrotic, hypodense lymphadenopathy lesions are presented in carotid region and right submandibular area.

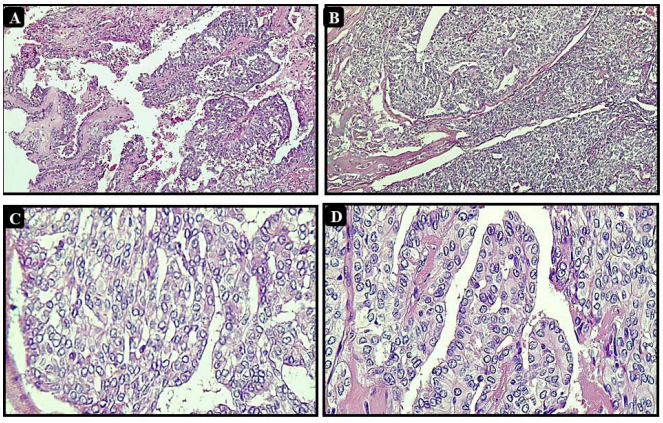

Figure 2. Cytopathologic presentation of polymorphous low-grade adenocarcinoma. After biopsy, the samples were fixed and stained using H&E staining method. A and B: represents the involved tissue with ×40 magnification; C and D: represents the involved tissue with ×400 magnification. Malignant cells are infiltrated into the lymph node and made a diverse growth pattern, including cribriform, solid and microcytic spaces which were filled with mucoid material. Irregular membrane and frequent nuclear grooves with active mitotic cells are evident.

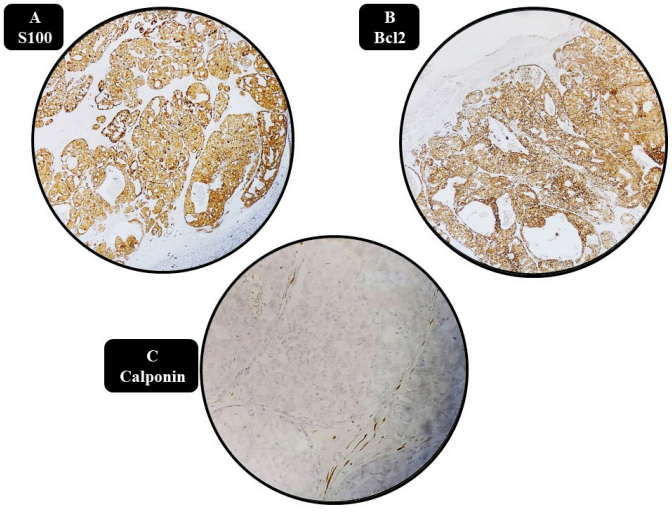

Figure 3. A sample of immunohistochemistry results from the minor salivary gland tumor in a 47-year-old non-smoker man involved with Cribriform adenocarcinoma. To evaluate the molecular pathology profile of tissue, the Creatine kinase (CK), especially CK 5/6, Vimentin, Cytokeratin, KI-67, S-100, Bcl2 and Epithelial Membrane Antigen (EMA), TTF1, Napsin and CD10 were evaluated in pathology department. From this list, TTF1, Napsin, CD10 and Calponin were negative and others represent a positive reaction. A: positive reaction of S100, and B: Bcl2. C: shows negative reaction of Calponine.

negative reaction).

According to pathologic and immunohistochemistry findings, gingiva and right submandibular biopsy was reported as cribriform adenocarcinoma of minor salivary gland which involved neck lymph nodes, too. Tonsillar tissue was involved with follicular hyperplasia and was free of tumor. Nasopharynx was not involved, too.

Because of margin free tumor resection and no far metastasis, the patient was followed by physical examination and IV contrast CT and fortunately no symptom or sign of recurrence were seen.

Discussion

This case was reported because of Cribriform adenocarcinoma of Minor Salivary Glands was considered in the WHO classification as a possible variant of polymorphous adenocarcinoma. This kind of malignant tumor has a high frequency of cervical lymph node metastasis (65%) at the presentation and the prognosis is highly favorable. Cribriform adenocarcinoma of the minor salivary gland is a tumor which was mostly in the sublingual site and other minor salivary glands are in the second place. Histopathological features of this kind of tumor can be misdiagnosed by papillary thyroid carcinoma. This malignant lesion usually is a solid-cystic and colloid lesion with eosinophilic particles. Cribriform adenocarcinoma composed of hybrid secretory-myoepithelial cells which makes the diagnosis precisely and rule out papillary carcinoma. All of the microscopic features were seen in our specimen and thyroid tumor markers including thyroglobulin and TTF-1 were negative. In contrast to polymorphous low – grade adenocarcinoma, Cribriform adenocarcinoma has significant different in location, cytology, histological architecture, metastasis and behavior. Cribriform subtypes have indolent behavior and early frequent metastasis.[8-11]

Our findings were confirmed by immunohistochemical findings, while no far metastasis was an exception in comparison to other similar studies. In ours, the lesion had a good and non-progressive prognosis like other similar reports.

Yamamoto et al. were published a case report in which presented two 43- and 48-years old men with involvement in cavernous sinus system. They found that both cases were suffered from mixture cribriform and solid tumor of cystic adenocarcinoma. These two cases were progressive maybe because of their delay in follow up, while our patient had a good prognosis without any far metastasis due to its early and correct diagnosis.[11]

Our case did not have any involvement in tongue or sublingual region, while some similar case reports were different. Suzuki et al. reported a 64-year-old woman with salivary carcinoma in the front part of the tongue, originating from a minor salivary gland. They confirmed the diagnosis by smears and partial glossectomy.[12] Fortunately, in our patient, there was not any infiltration in essential soft tissues surrounding the mass origin.

Conclusion

Early and precise diagnosis in cribriform adenocarcinoma of minor salivary glands can be important to prevent distal metastasis and improve the prognosis. Accurate Immunohistopathological findings should be reported by an expert pathologist because of its similarity to low grade adenocarcinoma and low-grade adenocarcinoma.

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