CASE REPORT
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ISTANBUL UNIVERSITY CERRAHPASA

Salivary Adenocarcinoma, Not Otherwise in Palate: Clinical, Radiological, and Histopathological Findings of a Rare Case

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Abstract

The objective of this report was to summarize the clinical, radiological, and histopathological features of an adenocarcinoma, not otherwise specified, case. A 28-year-old female patient was administered to the Department of Oral and Maxillofacial Radiology with pain and swelling on the palate. Initially, the patient was evaluated clinically and then radiologically examined with panoramic radiography, intraoral ultrasonography, computed tomography, and magnetic resonance imaging. After that, the patient was referred to the Department of Oral and Maxillofacial Surgery with an initial diagnosis of a salivary gland tumor. A biopsy specimen was taken and sent to the Department of Medical Pathology. Histopathological examination revealed a diagnosis of adenocarcinoma, not otherwise specified. Salivary adenocarcinoma, not otherwise specified, is a rare and highly aggressive tumor. Intraoral ultrasonography may be used for the preliminary diagnosis of palatal tumors such as adenocarcinoma, not otherwise specified. Computed tomography and magnetic resonance imaging are important imaging modalities to evaluate adenocarcinoma, not otherwise specified.

Keywords: Adenocarcinoma, computed tomography, magnetic resonance imaging, salivary glands, ultrasonography

INTRODUCTION

Salivary gland malignancies represent a wide variety of pathologies because they contain many secretory cells and there are many types of cells in their ductal structures. These malignancies make up about 5% of all head and neck cancers.[1] Mucoepidermoid carcinoma and adenoid cystic carcinoma are the 2 most common salivary gland malignancies. Adenocarcinoma is the third most common malignancy of the salivary gland.[2,3] Adenocarcinoma, not otherwise specified (ANOS), is a malignant salivary gland tumor that exhibits ductal differentiation but lacks any of the histomorphologic features that characterize the other defined types of salivary carcinoma. The modifying term "not otherwise specified" should be included because most other epithelial salivary gland malignancies are also adenocarcinomas. Adenocarcinoma, not otherwise specified, constitutes 5.3% of malignant salivary gland tumors.[1-4]

Two-dimensional plain radiographs are the starting point part of imaging salivary gland pathologies. Panoramic and posteroanterior head radiographs, along with clinical findings, can distinguish between bone lesions that resemble the appearance of salivary gland pathologies. Plain films also enable assessment of the adjacent bone structure in the presence of malignant or benign lesions of the salivary glands. Computed tomography (CT), magnetic resonance imaging (MRI), and ultrasonography (USG) are imaging techniques used to view salivary glands. Ultrasonography is an important assistant imaging technique used in the diagnosis of maxillofacial pathologies, lymph nodes, facial bone fractures, and salivary gland

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disorders. In addition, USG is used in fine needle-guided aspirations.[5-7] In a study, they concluded that intraoral USG is very important for an initial evaluation of palatal salivary gland tumors. With this non-invasive, fast, and easy-to-apply imaging technique, initial evaluation can be made about the location and size of palatal salivary gland tumors. Magnetic resonance imaging and CT are the gold standard in imaging salivary gland pathologies.[7,8] These techniques help to accurately and fully visualize the salivary glands. Magnetic resonance imaging shows the degree of the lesion and perineural/meningeal spread better than CT due to its multiplanar ability and higher soft-tissue resolution, whereas CT shows bone lesions, extension, and calcification better than MRI.[8]

Adenocarcinoma, not otherwise specified, is a pathology that is difficult to diagnose and should be examined in detail in histopathological, morphological, and immunophenotypically.[9] The objective of this report was to summarize the clinical, radiological, and histopathological features of an ANOS case and review the findings in light of the literature.

CASE PRESENTATION

A 28-year-old female patient was applied to our clinic with pain and swelling on the palate (Figure 1). Informed consent was obtained from the patient. Initially, the patient was evaluated clinically, and then radiological examination was made with panoramic radiography. Panoramic radiography revealed a well-defined radiolucent area on the left maxillary molar region (Figure 2). Related teeth were vital on the pulp vitality test. For the initial evaluation of the lesion, intra-oral USG was applied to the swelling in the palate. Intra-oral sonographic examination revealed a homogenous hyperechoic solid lesion with internal vascularization. It had a solid appearance on USG images (Figure 3A and B). Then,

Figure 1. Swelling on the palate seen in the intraoral examination of the patient.

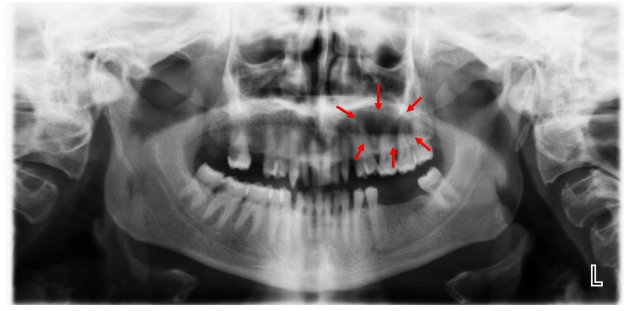

Figure 2. Panoramic radiography revealed a well-defined radiolucent area on the left maxillary molar region (red arrows).

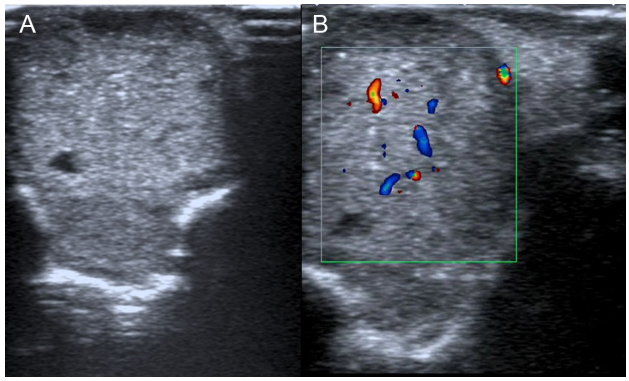

Figure 3. A, B. (A) Intraoral ultrasonography (USG) examination revealed an isoechoic lesion that has a solid appearance on USG. (B) Intraoral Doppler USG examination revealed internal vascularization within the lesion.

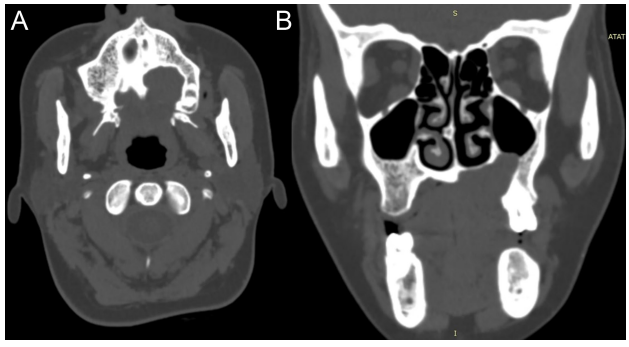

Figure 4. A, B. (A) Axial computed tomography (CT) image revealed a hypodense area in the posterior maxilla. (B) Coronal CT image showed perforation of palatal cortical bone and floor of the left maxillary sinus.

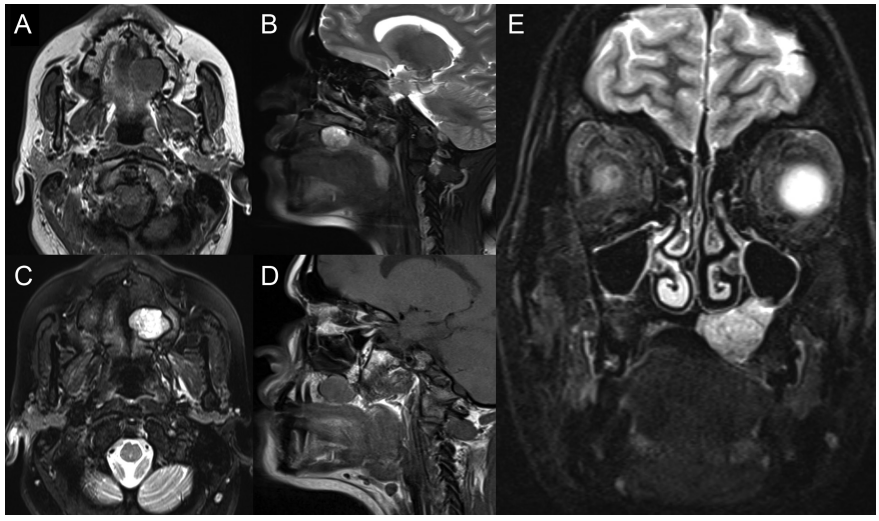

Figure 5. A-D. (A) T1-weighted axial magnetic resonance image (MRI) revealed a well-defined hypointense lesion on the posterior maxilla. (B) Fat-saturated T2-weighted sagittal MRI image revealed a hyperintense lesion on the posterior maxilla. (C) Fat-saturated T2-weighted axial MRI image revealed a hyperintense lesion on the posterior maxilla. (D) T1-weighted sagittal MRI image revealed a well-defined hypointense lesion on the posterior maxilla. (E) Fat-saturated T2-weighted coronal MRI image revealed a hyper-intense lesion on the posterior maxilla.

the lesion was evaluated with CT and MRI for detailed examination. Computed tomography showed a soft tissue density area in the posterior maxilla. The greater palatine foramen was adjacent to the lesion. It was caused by the perforation of palatal cortical bone. It invaded the maxillary sinus, and it caused perforation of the maxillary sinus floor (Figure 4A and B). T_1-weighted MRI images showed a well-defined hypointense lesion and fat-saturated T_2-weighted MRI images showed a well-defined heterogeneous hyperintense lesion (Figure 5A-E) After that, the patient was referred to the Department of Oral and Maxillofacial Surgery with an initial diagnosis of salivary gland tumor. The excisional biopsy specimen was taken and sent to the Department of Medical Pathology. Histological examination of hematoxylin and eosin-stained sections revealed delicately collagenous connective tissue infiltrated by numerous ducts like structures, nests, and cords. Tumor cells were showing atypia, hyperchromatism, pleomorphism, and few mitotic figures. Immunohistochemical examination showed that the tumor cells were positive for ytokeratin 7 (Figure 6A), vimentin (Figure 6D), AE1/AE3, and P63 (Figure 6C). The cells had intranuclear inclusions and abundant eosinophilic cytoplasm (Figure 6B). These tumor features did not meet the diagnostic criteria for any other known types of salivary gland carcinomas, and hence, the diagnosis of ANOS was confirmed, according to the World Health Organization classification of salivary tumors published in 2005.[10] Large local excision with negative resection margins was planned for the treatment. The patient was prepared for surgery under general anesthesia. Hemi-maxillectomy operation was performed on the posterior region of the left maxilla, starting from tooth region 21 and covering the tuber region, with an intraoral approach.

DISCUSSION

Malignant neoplasms of the salivary glands are rare pathologies. Asymptomatic swellings, which are not observed in the salivary glands, are lesions that should cause suspicion.

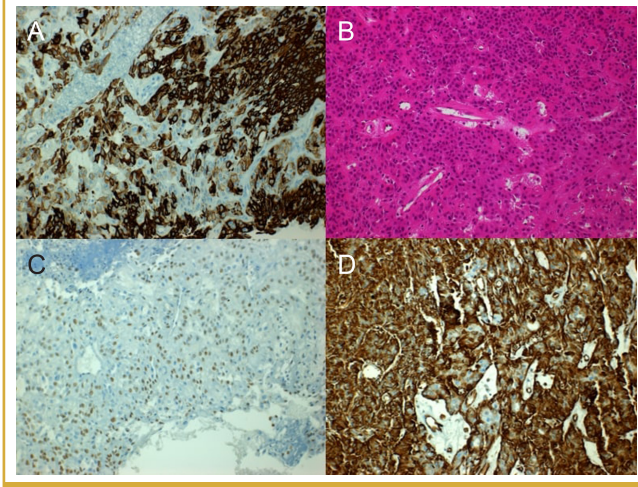

Figure 6. A-D. (A) Positivity for cytokeratin 7 (×20). (B) This example demonstrates an organoid arrangement of cells that have abundant eosinophilic cytoplasm (H&E ×10). (C) Nuclear positivity for P63 antibody (×20). (D) Positivity for vimentin (×20).

Approximately 15%-32% of tumors in the parotid are malignant, 41%-45% of submandibular tumors, and 70%-90% of sublingual tumors are malignant. Clinically in the salivary gland malignancies, rapid growth, pain, facial nerve involvement, cervical adenopathy, trismus, skin ulceration, and fistulas can be observed. Of minor salivary gland tumors, 50%are malignant. Symptoms depend on the localization and size of the tumor. It usually causes painless submucosal swelling, and a small, ulcerated area may appear on the mucosa. In the present case, there was a complaint of pain, and intraoral examination revealed swelling and ulcer of the mucosa area.[9-11]

Adenocarcinoma, not otherwise specified, is a highly malignant rare tumor with differentiation of glandular or ductal adenocarcinoma but without other specific histological features. It constitutes 1% of all head and neck carcinomas and it is most common in the parotid glands, followed by small salivary glands in the palate.[4] In the present case, it was localized in the palate. Adenocarcinoma, not otherwise specified, is most common in women. It is observed more frequently in the fifth and eighth decades of life.[11] The case presented in this study is a 28-year-old female patient.

As the initial imaging technique, USG is an important adjunct technique that can demonstrate the necessity of CT/MRI imaging, which is an advanced imaging method in salivary gland tumors. It provides information about tumor morphology, localization, local invasion, and dimensions.[12] Ultrasonography allows the physician to obtain real-time images without creating radiation exposure. Among its advantages are its non-invasiveness and easy and fast application.[13] Salivary glands are structures that can be easily monitored by USG due to their superficial location. It is an imaging technique that will guide the clinician when fine-needle aspiration or therapeutic aspiration is required.[14] Ultrasonography allows us to detect the presence of vascularization and tissue characterization with the Doppler technique.[15] In the color flow Doppler examination of salivary gland tumors, vascularity is increased.[8] Also, solid masses can be distinguished from cystic ones by USG.[6] In the present study, intra-oral USG examination revealed a homogeneous hyperechoic solid lesion with internal vascularization. Computed tomography and MRI are techniques used to evaluate suspicious salivary gland masses.[8] These are the most important methods used to evaluate tumor borders, local invasion, and perineural spread in pathologies with suspected malignancy. For parotid gland and minor salivary glands, MRI and CT are preferred methods. Magnetic resonance imaging is an excellent technique for soft tissue differentiation.[15] Postcontrast fat-suppressed images allow us to examine the perineural spread of malignancies in the salivary glands. In the present report, T_1-weighted MRI images detected well-defined hypointense lesions and fat-saturated T_2-weighted MRI images were well-defined heterogeneous hyperintense lesions. However, CT should be used to show changes in the bone. With CT, the presence of bone erosion in palatal minor salivary gland malignancies can be detected.[14] In the present case, we detected perforation in the palatal bone and invasion into the maxillary sinus with CT.

The most effective and accepted view for the treatment of ANOS is surgery. Large local excision with negative resection margins is an important positive survival effect. More aggressive local resection was proposed is in the T category exceeds T_1 because inadequate resection cause of the local recurrence.[4] In the present case, a hemi-maxillectomy operation was performed for treatment.

Histopathologically, ANOS does not carry the properties of other salivary gland adenocarcinomas. Therefore, it was categorized as a separate group. The neoplastic epithelium may be glandular, papillary, cystic, cribriform, solid, lobular, nested, or cords. Tumor cells can be morphological of various types, including cubic, column, polygonal, and oval. Adenocarcinoma, not otherwise specified, according to nuclear morphology, pleomorphism, hyperchromatism, necrosis, mitotic index, and ill-demarcated borders are categorized as low, intermediate, or high-grade.[9] In our study, in the histological examination of the part stained with hematoxylin and eosin, a delicate collagen connective tissue that has been leaked by numerous channels such as structures, nests, and cords has been revealed. Tumor cells showed atypia, hyperchromatism, pleomorphism, and few mitotic figures.

Salivary ANOS is a rare and overly aggressive tumor. Intraoral USG may be used for the preliminary diagnosis of palatal tumors such as ANOS. Computed tomography and MRI are important imaging modalities to evaluate ANOS. Dentists play a critical role in the early diagnosis of this type of pathology.

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REFERENCES

1. Huang AT, Tang C, Bell D, et al. Prognostic factors in adenocarcinoma of the salivary glands. *Oral Oncol*. 2015;51(6): 610-615. [CrossRef]

2. Oliveira LR, Soave DF, Oliveira-Costa JP, Zorgetto VA, Ribeiro-Silva A. Prognostic factors in patients with malignant salivary gland neoplasms in a Brazilian population. *Asian Pac J Cancer Prev*. 2011;12(2):363-368.
3. Adelstein DJ, Koyfman SA, El-Naggar AK, Hanna EY. Biology and management of salivary gland cancers. *Semin Radiat Oncol*. Amsterdam: Elsevier. 2012;22(3):245-253. [CrossRef]
4. Deng R, Tang E, Yang X, Huang X, Hu Q. Salivary adenocarcinoma, not otherwise specified: a clinicopathological study of 28 cases. *Oral Surg Oral Med Oral Pathol Oral Radiol*. 2012; 113(5):655-660. [CrossRef]
5. Bruneton J, Sicart M, Roux P, Pastaud P, Nicolau A, Delorme G. Indications for ultrasonography in parotid pathologies. *RöFo-Fortschritte auf dem Gebiet der Röntgenstrahlen und der bildgebenden Verfahren*. New York: Georg Thieme Verlag Stuttgart. 1983:22-24.
6. Yasumoto M, Shibuya H, Suzuki S, et al. Computed tomography and ultrasonography in submandibular tumours. *Clin Radiol*. 1992;46(2):114-120. [CrossRef]
7. Ishii J, Nagasawa H, Wadamori T, et al. Ultrasonography in the diagnosis of palatal tumors. *Oral Surg Oral Med Oral Pathol Oral Rad Endodontol*. 1999;87:39-43.
8. Rastogi R, Bhargava S, Mallarajapatna GJ, Singh SK. Pictorial essay: salivary gland imaging. *Indian J Radiol Imaging*. 2012; 22(4):325-333. [CrossRef]
9. Li J, Wang BY, Nelson M, et al. Salivary adenocarcinoma, not otherwise specified: a collection of orphans. *Arch Pathol Lab Med*. 2004;128(12):1385-1394. [CrossRef]
10. Barnes L, Eveson JW, Sidransky D, Reichart P. Pathology and genetics of head and neck tumors. *IARC*. 2005. p.w 238.
11. Santoro A, Laino L, Contaldo M, et al. Adenocarc NOS of the maxillary sinus: clinical and histopathological features with therapeutic considerations. *Otolaryngol*. 2011;1:2.
12. Bradley MJ, Durham LH, Lancer JM. The role of colour flow Doppler in the investigation of the salivary gland tumour. *Clin Radiol*. 2000;55(10):759-762. [CrossRef]
13. Diederich S, Wernecke K, Peters PE. Sialographic and sonographic diagnosis of salivary gland diseases. *Radiologe*. 1987; 27(6):255-261.
14. Bialek EJ, Jakubowski W, Zajkowski P, Szopinski KT, Osmolski A. US of the major salivary glands: anatomy and spatial relationships, pathologic conditions, and pitfalls. *RadioGraphics*. 2006; 26(3):745-763. [CrossRef]
15. Lee YY, Wong KT, King AD, Ahuja AT. Imaging of salivary gland tumours. *Eur J Radiol*. 2008;66(3):419-436. [CrossRef]