KBB Uygulamaları 2023;11(1):29-32
doi: 10.5606/kbbu.2023.43815

Case Report / *Olgu Sunumu*

OPEN ACCESS

High-grade cystadenocarcinoma of the parotid gland: A rare case report

Parotis bezinin yüksek dereceli kistadenokarsinomu: Nadir bir olgu

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ABSTRACT

Cystadenocarcinoma is a rare malignant tumor of the salivary glands. It has a favorable prognosis, and surgery is the primary therapeutic option. Diagnosis is based on pathologic findings, which indicate a mainly cystic growth pattern with intraluminal papillary proliferation. Currently, there are few studies reporting cystadenocarcinoma of the salivary gland in the literature. This case report presents a 75-year-old male patient with cystadenocarcinoma originating from the parotid gland with clinical, radiological, and pathological features. In this patient, radiological images suggested benign lesions, while fine-needle aspiration biopsy was reported as suspicious for malignant cytology and subtyping could not be performed. Total parotidectomy option could be used in salivary gland masses with suspected malignancy.

Keywords: Cystadenocarcinoma, high grade, parotid gland, salivary glands, tumor.

ÖZ

Kistadenokarsinom, tükürük bezlerinin nadir görülen malign bir tümörüdür. İyi prognoza sahiptir ve tedavide ilk seçenek cerrahidir. Teşhis, intraluminal papiller proliferasyonla birlikte esas olarak kistik bir büyüme paternini gösteren patolojik bulgulara dayanır. Güncel literatürde tükürük bezi kistadenokarsinomunu bildiren az sayıda çalışma bulunmaktadır. Bu olgu sunumunda, klinik, radyolojik ve patolojik özellikleri ile parotis bezinden köken alan kistadenokarsinomlu 75 yaşında bir erkek hasta sunuldu. Bu hastada radyolojik görüntüler benign lezyonları düşündürürken ince iğne aspirasyon biyopsisi malign sitoloji açısından şüpheli olarak raporlandı ve alt tiplendirme yapılamadı. Tükürük bezinin malignite şüpheli kitlelerinde total parotidektomi seçeneğinden faydalanılabilir.

Anahtar sözcükler: Kistadenokarsinom, yüksek dereceli, parotis bezi, tükürük bezleri, tümör.

Salivary gland tumors were divided into more than 30 benign and malignant histologic categories in the World Health Organization's (WHO) most recent classification in 2017.[1] Among these tumors, cystadenocarcinoma (CAC) was first described in 1991 and is a very rare malignant subtype.[2] Previously, CACs were included under the term CAC in the WHO classification of head and neck cancers, but they are now included under the term "Adenocarcinoma, Not Otherwise Specified" after revisions by WHO in the 2017 edition.[3] They are thought to constitute approximately 0.4% of all salivary gland malignancies. They most commonly involve the parotid gland among the major salivary glands. Cystadenocarcinomas are most common in the sixth and seventh decades of life, and there is no predominance between sexes. Cystadenocarcinomas, which are generally in the group of low-grade tumors, have a good prognosis.[4,5] The main treatment for CACs of salivary glands is radical surgical excision.[4] Depending on lymph node involvement in the neck, neck dissection can be added to surgical resection.[6]

Received: July 14, 2022
Accepted: January 02, 2023
Published online: February 20, 2023

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Citation:
Kumbul YÇ, Okur N, Okur E, Akın V, Kıran M. High-grade cystadenocarcinoma of the parotid gland: A rare case report. KBB Uygulamaları 2023;11(1):29-32. doi: 10.5606/kbbu.2023.43815

In this study, a case of high-grade CAC is presented in light of the literature.

CASE REPORT

A 75-year-old male patient was admitted to the otorhinolaryngology clinic with the complaint of a painless, growing mass on the right angle of the mandible for about a year. The patient had no disease except benign prostatic hyperplasia in the history. On physical examination, a rigid, well-circumscribed, fixed mass of approximately 4 cm in diameter was palpated in the right parotid lobe. No lymphadenopathy was palpated on neck examination. Facial nerve functions were normal. The complete blood test and routine biochemistry tests were normal. The mass was evaluated with contrast-enhanced neck computed tomography. Atypical pleomorphic adenoma and Warthin tumor were considered in the preliminary radiological diagnosis (Figure 1). Fine needle aspiration biopsy was performed on the mass. The biopsy was reported as suspicious cytology for malignancy. The patient underwent total parotidectomy. Facial nerve functions were unaffected after surgery. Histopathological examination of the mass reported high-grade CAC. In the pathological examination, there was encapsulation in the tumor, and there was no tumor in the surgical margins. There was also no lymphovascular invasion and perineural invasion in the tumor. Immunohistochemical

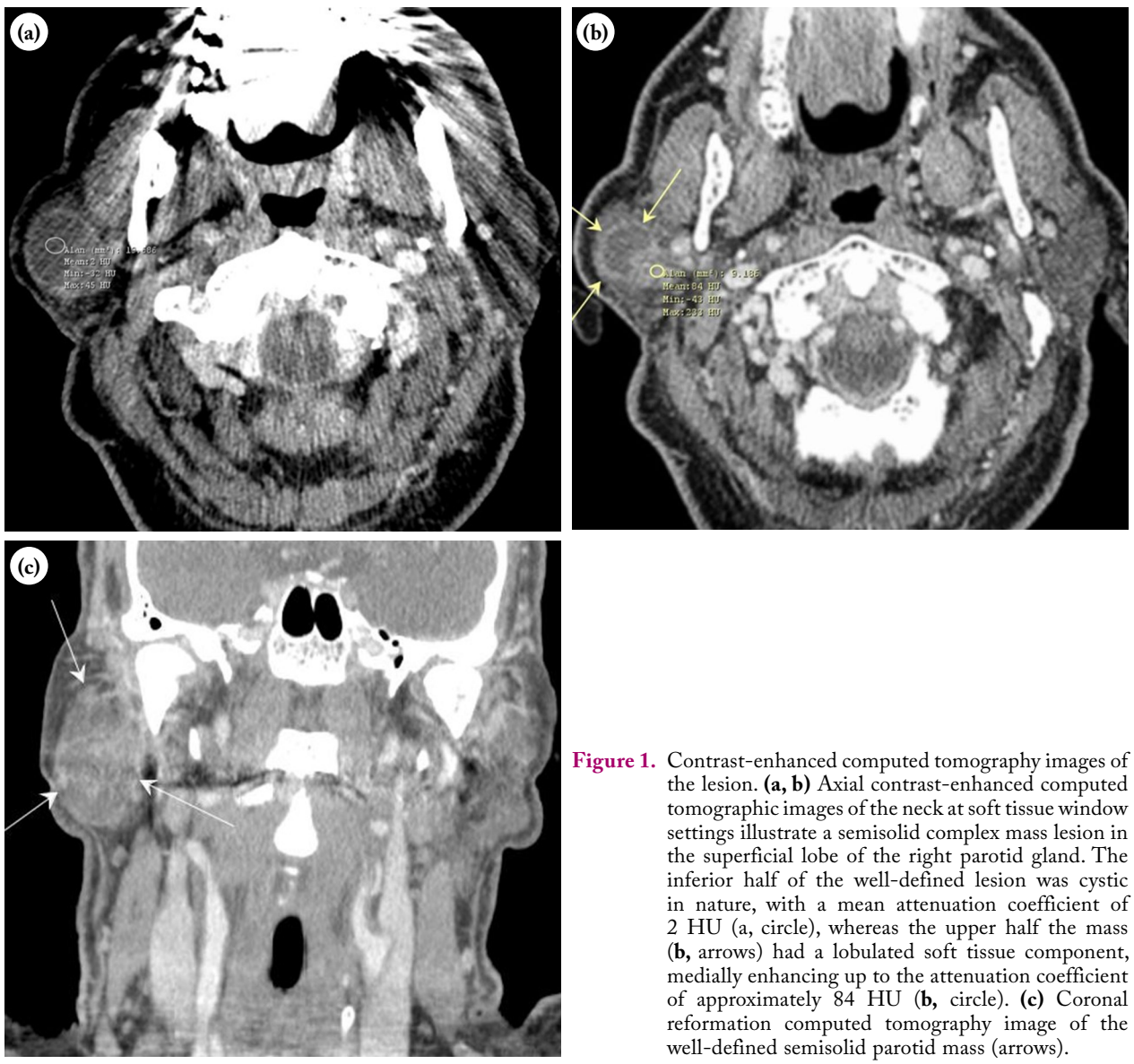

Figure 1. Contrast-enhanced computed tomography images of the lesion. **(a, b)** Axial contrast-enhanced computed tomographic images of the neck at soft tissue window settings illustrate a semisolid complex mass lesion in the superficial lobe of the right parotid gland. The inferior half of the well-defined lesion was cystic in nature, with a mean attenuation coefficient of 2 HU (a, circle), whereas the upper half the mass (**b,** arrows) had a lobulated soft tissue component, medially enhancing up to the attenuation coefficient of approximately 84 HU (**b,** circle). **(c)** Coronal reformation computed tomography image of the well-defined semisolid parotid mass (arrows).

examination of the tumor observed positive staining with pancytokeratin, epithelial membrane antigen (EMA), carcinoembryonic antigen (focal), and p63 (focal), while discovered on gastrointestinal stromal tumor 1 (DOG1) and S-100 were negative. Ki67 was 20% positive in the dense area (Figure 2). In terms of possible metastasis of the malignancy, positron emission tomography was performed on the patient, and fluorine-18 fluorodeoxyglucose uptake was detected at physiological limits in the whole body. The radiation oncology department did not identify an indication for adjuvant radiotherapy. The patient has been followed up for about six months, and no signs of recurrence have been observed.

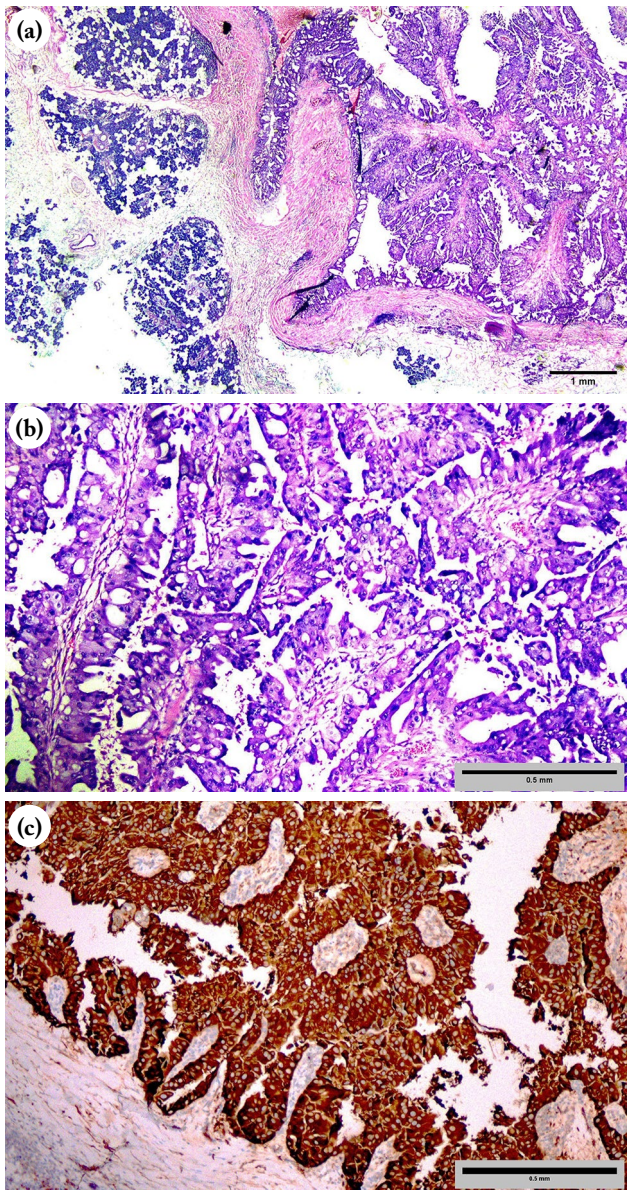

Figure 2. Images of the histopathological examination of the lesion. **(a)** The lesion consisted of large cuboidal and columnar cells, which had cystic development, formed papillary structures, and was separated from the adjacent salivary gland tissue by fibrous connective tissue (H&E, ×20). **(b)** Papillary structures and nuclear atypia are more prominent (H&E, ×100). **(c)** Diffuse, potent, cytoplasmic positivity was observed with epithelial membrane antigen staining at ×100 magnification.

DISCUSSION

Few cases of salivary gland cyst adenocarcinoma were reported in the literature, and data about this subject is limited. In the study by Cai et al.,[4] 16,923 patients diagnosed with salivary gland malignant tumors over a 21-year period were examined, and only 65 of them were CACs. In the same study, no difference was found between males and females, the median age was 62 years, and CAC most frequently affected the parotid gland. In the patient we presented, the parotid gland was involved, and the patient's age was higher than in the literature.

For the histopathological diagnosis of CAC, it is important to demonstrate intraluminal papillary proliferation and a cystic growth pattern.[3] In addition, CACs are divided into three grades, and Grade III is the least common.[4] In our case, the tumoral mass showed cystic development and consisted of large cuboidal and columnar cells forming papillary structures. Contrary to most CAC cases in the literature, histopathological examination results indicated a high grade (Grade III) in our case.

The absence of specific clinical and radiological findings of CAC may lead to the diagnosis of CAC only after histopathological examination.[7] Although fine needle aspiration biopsy is recommended for diagnosis in the preoperative period, it is difficult to diagnose CAC with fine needle aspiration biopsy.[8] We also initially performed a fine needle aspiration biopsy for the diagnosis in the preoperative period. The biopsy result was suspicious for malignancy. We think that this is the most challenging pathology result for otolaryngologists during the treatment of parotid gland masses. If the mass is in the superficial parotid gland and is benign, superficial parotidectomy is sufficient, whereas superficial parotidectomy is insufficient for a malignant parotid mass. Another issue that needs to be taken into account is that the patient's risk of facial paralysis increases with recurrent parotid surgeries on the same side. Therefore, we did not take any risk for this patient and performed total parotidectomy in a single session.

After surgery for CAC, recurrences usually occur in the cervical lymph nodes, and recurrence rates

are lower at the primary tumor site than at possible metastatic sites.[4] It was reported that the presence of lymph node metastasis is the main factor that increases the risk of recurrence after surgery.[7] In the study by Cai et al.,[4] 25 of 65 patients with CAC underwent lymphadenectomy, and positive lymph nodes were detected in 16% of these patients. In the study by Guo et al.,[7] lymph node metastasis was detected in 18.5% of the patients. In another study, cervical lymph node metastases were detected in three of five CAC cases, at the earliest two years and at the latest 21 years later.[9] The recurrence rate of CAC after surgery was reported to be approximately 34%, and two lung metastases were detected in a series of 27 patients.[7] In the case we presented, there was no sign of cervical lymph node metastasis on positron emission tomography; thus, we chose the wait-and-see approach instead of lymph node dissection. In addition, radiotherapy was not applied because the capsule integrity of the tumor was not impaired and there was no tumor in the surgical margins. The presence of late recurrences in CAC cases implies that CAC is an insidious tumor. Therefore, we recommend that CAC patients be checked at regular intervals.

In conclusion, in cases where the fine needle biopsy result is suspicious for malignancy, total parotidectomy can be performed instead of superficial parotidectomy to reduce the risk of facial paralysis. Patients with lymph node metastases are candidates for advanced treatment options such as neck dissection. The wait-and-see option may be preferred in patients with clinically undetectable lymph node metastases.

Patient Consent for Publication: A written informed consent was obtained from the patient.

Data Sharing Statement: The data that support the findings of this study are available from the corresponding author upon reasonable request.

Author Contributions: Idea/concept, design, control/supervision, analysis and/or interpretation, literature review, writing the article, critical review, references and fundings: Y.Ç.K.; Idea/concept, design, control/supervision, data collection and/or processing, analysis and/or interpretation, writing the article, critical review: N.O.; Idea/concept, design, control/supervision, analysis and/or interpretation, literature review, writing the article, critical review: E.O.; Idea/concept, design, control/supervision, analysis and/or interpretation, literature review, writing the article, references and fundings: V.A.; Idea/concept, design, control/supervision, data collection and/or processing, analysis and/or interpretation, writing the article: M.K.

Conflict of Interest: The authors declared no conflicts of interest with respect to the authorship and/or publication of this article.

Funding: The authors received no financial support for the research and/or authorship of this article.

REFERENCES

1. Mengi E, Kara CO, Tumkaya F, Ardic FN, Topuz B, Bir F. Salivary gland tumors: A 15-year experience of a university hospital in Turkey. North Clin Istanb 2020;7:366-71. doi: 10.14744/nci.2020.57767.
2. Seifert G, Sobin LH. The World Health Organization's Histological Classification of Salivary Gland Tumors. A commentary on the second edition. Cancer 1992;70:379-85. doi: 10.1002/1097-0142(19920715)70:2<379::aid-cncr2820700202>3.0.co;2-c.
3. de Lima-Souza RA, Leonel ACLDS, de Almeida OP, de Castro JFL, Carvalho EJA, Perez DEDC. Salivary gland cystadenocarcinoma: Rare neoplasm in buccal mucosa. Oral Oncol 2021;118:105314. doi: 10.1016/j.oraloncology.2021.105314.
4. Cai CF, Sun JY, He ZY, Lin HX, Liu FR, Wang Y. Clinicopathological characteristics, treatment, and survival outcomes of cystadenocarcinoma of the salivary gland: A population-based study. Onco Targets Ther 2016;9:6569-72. doi: 10.2147/OTT.S111247.
5. Foss RD, Ellis GL, Auclair PL. Salivary gland cystadenocarcinomas. A clinicopathologic study of 57 cases. Am J Surg Pathol 1996;20:1440-7. doi: 10.1097/00000478-199612000-00002.
6. Mukaigawa T, Hayashi R, Miyazaki M, Shinozaki T, Tomioka T, Fujii S. Cystadenocarcinoma of the salivary glands with potential lymph node metastasis. Auris Nasus Larynx 2016;43:340-4. doi: 10.1016/j.anl.2015.08.012.
7. Guo B, Cao YA, Qin X, Ma C. The rare entity of cystadenocarcinoma (CAC) in parotid gland: A single-center experience. J Craniomaxillofac Surg 2019;47:826-31. doi: 10.1016/j.jcms.2019.02.005.
8. Aloudah NM, Raddaoui E, Aldhahri S, Al-Abbadi MA. Low-grade papillary cystadenocarcinoma of the parotid gland: Presentation of a case with cytological, histopathological, and immunohistochemical features and pertinent literature review. Diagn Cytopathol 2009;37:128-31. doi: 10.1002/dc.20971.
9. Mills SE, Garland TA, Allen MS Jr. Low-grade papillary adenocarcinoma of palatal salivary gland origin. Am J Surg Pathol 1984;8:367-74. doi: 10.1097/00000478-198405000-00005.