Palatal Basal Cell Adenocarcinoma From Misdiagnosis to Cure: A Unique Case Report and Review

Ramzey Tursun, DDS, * *Dhafer Alsuwied, DDS,* † *J. Marshall Green III, DDS,* ‡ *and James Melville, DDS* §

We describe a 66-year-old woman who received a misdiagnosis of benign neoplasms twice before receiving the appropriate diagnosis of basal cell adenocarcinoma. At the time of recurrence, her care was assumed by the senior author, who made the appropriate diagnosis and administered treatment including transoral microvascular reconstruction, as well as adjuvant radiation therapy, which achieved a cure. A review of the current literature on this disease entity also is included.

J Oral Maxillofac Surg 77:100-108, 2019

Basal cell adenocarcinoma (BCAC) is a rare malignant lesion of the salivary gland, accounting for fewer than 2% of all salivary neoplasms.[1] In 2005 the World Health Organization defined BCAC as "an epithelial neoplasm that has cytological characteristics of basal cell adenoma (BCA), but a morphologic growth pattern indicative of malignancy."[2] We describe a 66-year-old woman treated by the University of Miami Oral/Head and Neck Oncology Service who received a misdiagnosis of benign neoplasms twice before receiving the appropriate diagnosis of BCAC. At the time of recurrence, her care was assumed by the senior author, who made the appropriate diagnosis and administered treatment including transoral microvascular reconstruction, as well as adjuvant radiation therapy, which achieved a cure. A review of the current literature on this disease entity also is included.

Case Report

A 66-year-old woman originally presented to the Oral/Head and Neck Oncology Service at the University of Miami for evaluation of a swelling in the right posterior palate. In August 2011 she had undergone a biopsy and definitive resection of the area. On histopathologic evaluation, the lesion was definitively described as a canalicular adenoma. The patient did not undergo follow-up as scheduled. After this period, she returned for evaluation, after a biopsy was performed by an oral and maxillofacial surgeon at an outside facility, with concerns of swelling in the area

*Associate Professor of Clinical Surgery and Director, Head & Neck Oncologic and Microvascular Reconstructive Surgery, Miller School of Medicine, University of Miami, Miami, FL.

†Fellow, Oral, Head & Neck Oncology and Microvascular Reconstructive Surgery, University of Texas Health Science Center at Houston, Houston, TX.

‡Lieutenant Commander and Associate Program Director, Naval Medical Center Portsmouth, Portsmouth, VA; Assistant Professor of Surgery, Uniformed Services University of the Health Sciences, Bethesda, MD; and Adjunct Assistant Professor of Surgery, Miller School of Medicine, University of Miami, Miami, FL.

§Assistant Professor of Surgery, Oral, Head & Neck Oncology and Microvascular Reconstructive Surgery, University of Texas Health Science Center at Houston, Houston, TX.

The views expressed in this article are those of the authors and do not necessarily reflect the official policy or position of the Department of the Navy, Department of Defense, or the United States government.

Conflict of Interest Disclosures: Dr Tursun serves as a consultant to AxoGen.

Address correspondence and reprint requests to Dr Tursun: Division of Oral and Maxillofacial Surgery, Miller School of Medicine, University of Miami, Deering Medical Plaza, 9380 SW 152nd St, Miami, FL 33157; e-mail: r.tursun@med.miami.edu

Received May 4 2018

Accepted August 7 2018

0278-2391/18/30878-4

https://doi.org/10.1016/j.joms.2018.08.003

FIGURE 1. Patient at presentation with noted mild fullness on right side.

Tursun et al. Palatal Basal Cell Adenocarcinoma. J Oral Maxillofac Surg 2019.

of the right hard and soft palate. The senior author took over her care and, on evaluation, noted a mass in the right posterior palate with mild facial swelling on the patient's right side (Fig 1). In the right posterior palate, a boggy lesion, approximately 2.5 × 2.5 cm, was palpated. No ulceration was noted over the lesion. The mucosa over the lesion was noted to be pink but

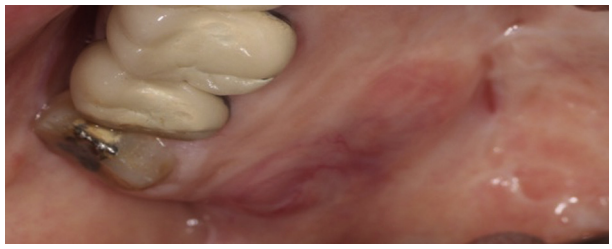


FIGURE 2. Preoperative clinical photograph of lesion and right posterior palate.

Tursun et al. Palatal Basal Cell Adenocarcinoma. J Oral Maxillofac Surg 2019.

thinned, with mild erythema and no pulsation (Fig 2). The biopsy that had been performed before arrival yielded a histopathologic diagnosis of ameloblastoma, and the findings were then reviewed by 2 general pathologists as well as an oral-maxillofacial pathologist on consultation who agreed with the diagnosis.

A computed tomography scan showed a radiolucent lesion in the right posterior maxilla extending to the palate with elevation of the right nasal floor and a small perforation through bone. Osseous erosion of the alveolus also was noted in this region (Figs 3, 4). A magnetic resonance image was obtained to mitigate the computed tomography scan artifact and to better visualize the lesion boundaries. The axial view showed a hyperdense lesion of the right maxilla and

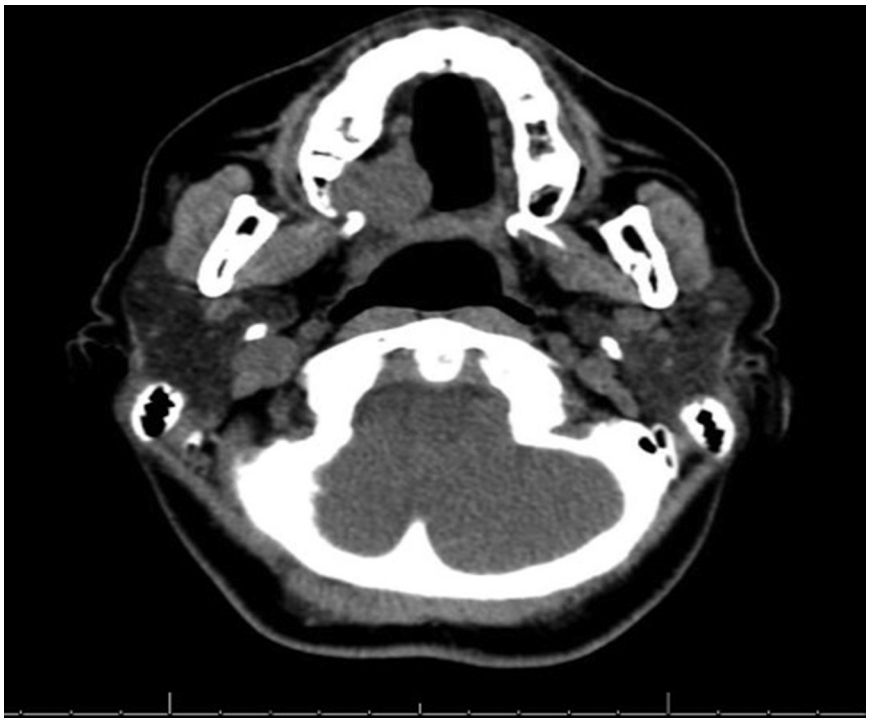


FIGURE 3. Axial view of soft tissue window of facial computed tomography scan showing right posterior maxilla and palate soft tissue mass.

Tursun et al. Palatal Basal Cell Adenocarcinoma. J Oral Maxillofac Surg 2019.

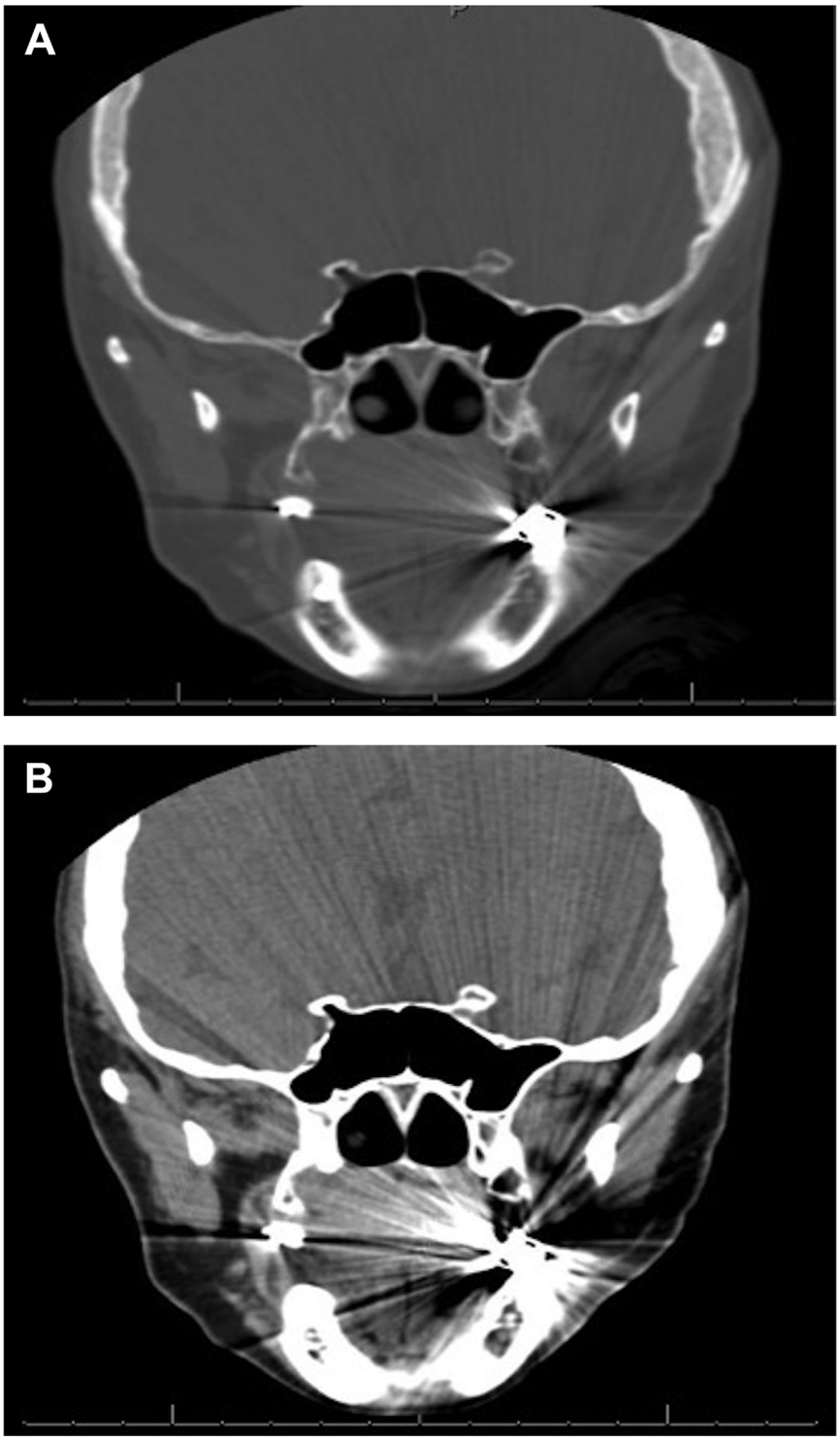


FIGURE 4. *A*, Coronal view of soft tissue and bone window of facial computed tomography. *B*, Multiple windows of the computed tomography scan are limited because of the dental prosthesis artifact.

Tursun et al. Palatal Basal Cell Adenocarcinoma. J Oral Maxillofac Surg 2019.

palate and further elucidated the location and extension (Fig 5).

On the basis of the clinical impression, surgical history, and detailed radiographic examination findings, the patient was offered treatment with surgical resection and immediate reconstruction with vascularized free tissue transfer. Subsequently, she was taken to the operating room, the lesion was outlined, and the resection was performed. Because of the history of recurrence and differing previous diagnoses, frozen sections were

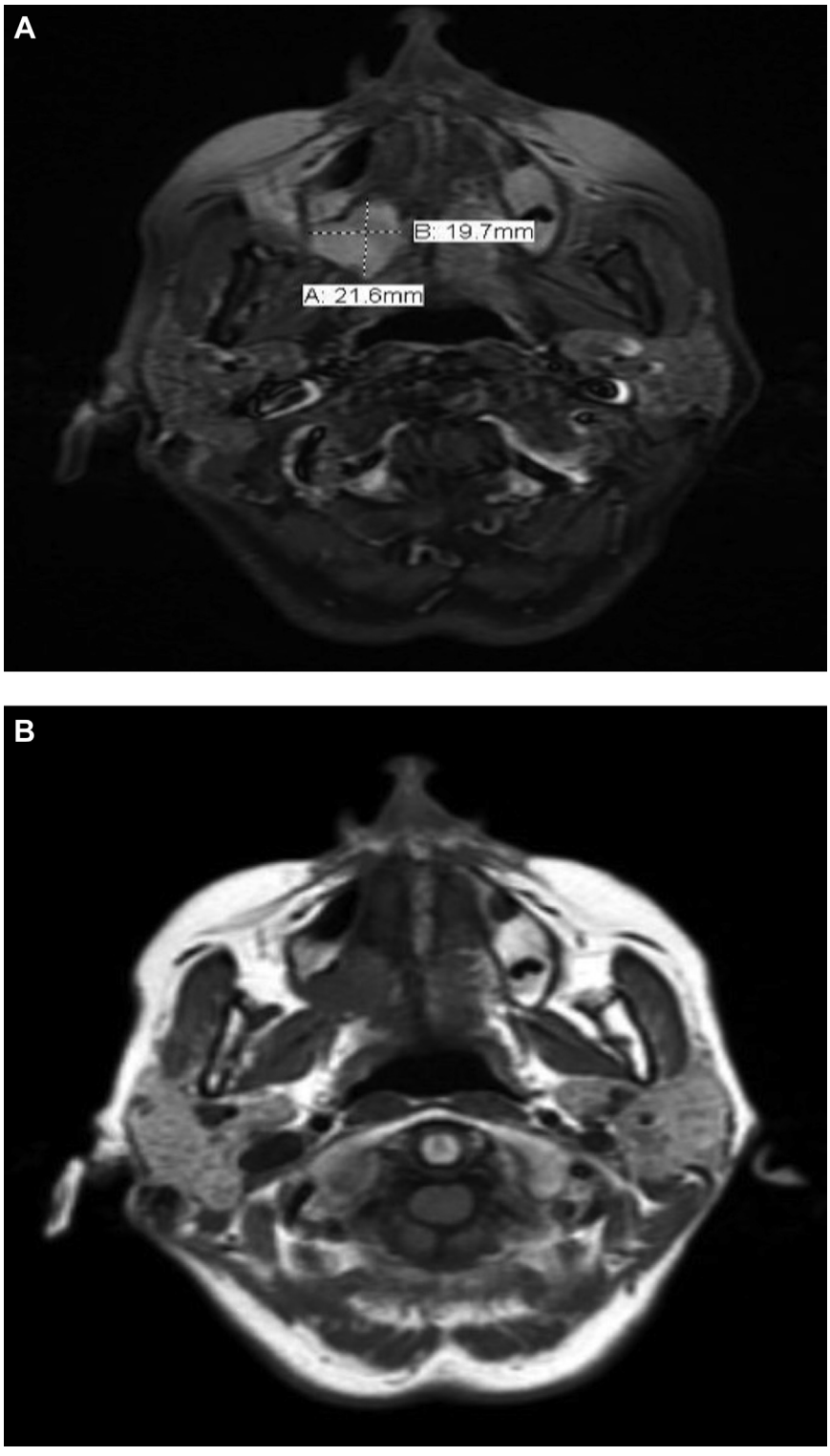
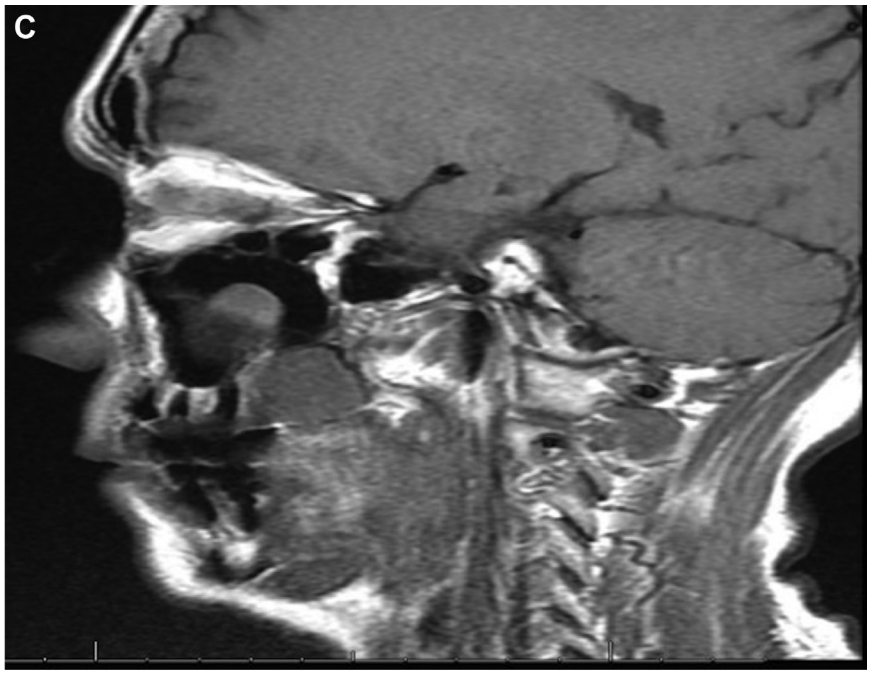


FIGURE 5. A T1 axial view (*A*) and T2 axial view (*B*) show a hyperdense lesion of the right maxilla and palate, and a sagittal view **(Fig 5 continued on next page.)**

Tursun et al. Palatal Basal Cell Adenocarcinoma. J Oral Maxillofac Surg 2019.

used during surgery to ensure disease control. Nonspecific, intraoperative frozen sections returned a finding of a malignant neoplasm of minor salivary gland origin. Wide local excision was completed, with the new malignant pathologic diagnosis being borne in mind, and immediate reconstruction was performed with an anteromedial thigh fasciocutaneous flap by an intraoral anastomosis technique (Figs 6-8).



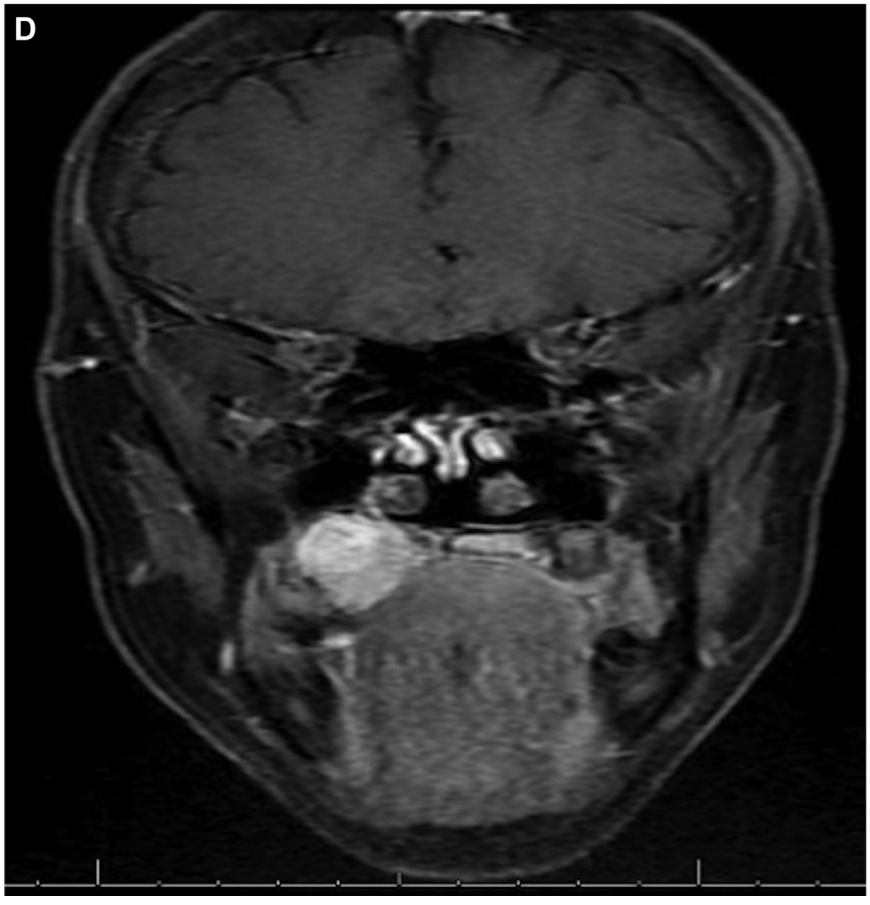


FIGURE 5 (cont'd). (*C*) and coronal view (*D*) elucidate the lesion location and extension.

Tursun et al. Palatal Basal Cell Adenocarcinoma. J Oral Maxillofac Surg 2019.

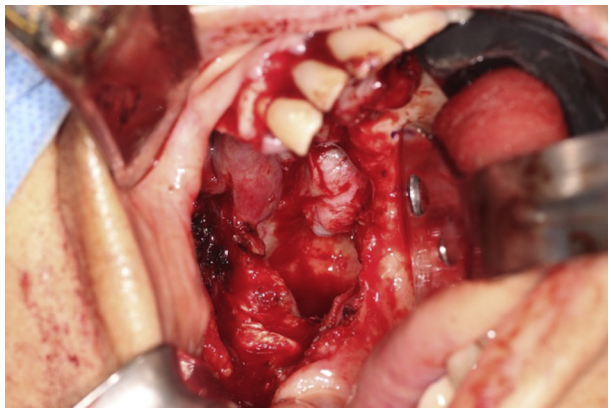


FIGURE 6. Ablative defect of wide excision after intraoperative finding of basal cell adenocarcinoma on frozen section analysis.

Tursun et al. Palatal Basal Cell Adenocarcinoma. J Oral Maxillofac Surg 2019.

The patient's postoperative course was uneventful, and she was discharged home with the appropriate care. The final pathologic results were reviewed and showed a finding of BCAC. Furthermore, despite the frozen margins being clear on completion, the final pathologic finding was interpreted as showing a close margin in 1 focal area. The case was reviewed by our multidisciplinary tumor board, which recommended adjuvant radiation therapy. Because of the finding of carcinoma, a workup for metastasis was performed and no distant disease was identified. The patient

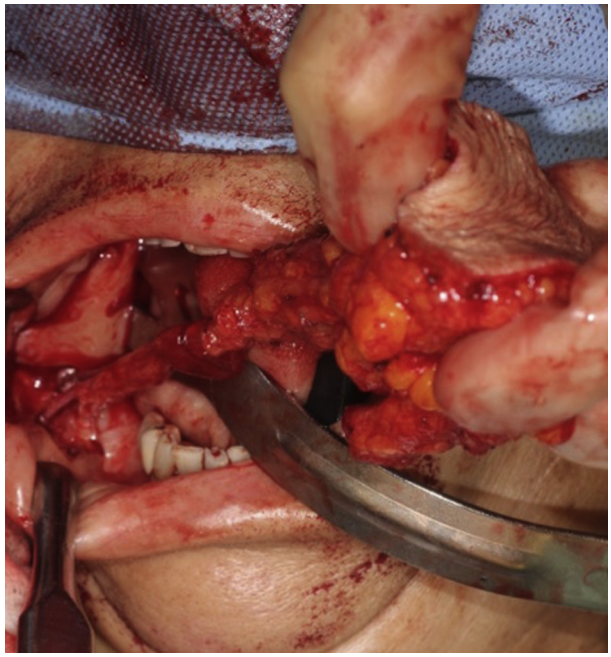


FIGURE 7. Intraoral anastomosis of anteromedial thigh flap before inset. No extraoral incision was used in preparing the facial vessels for microvascular anastomosis.

Tursun et al. Palatal Basal Cell Adenocarcinoma. J Oral Maxillofac Surg 2019.

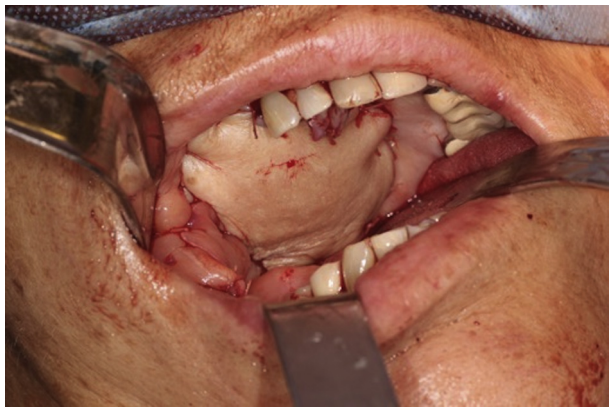


FIGURE 8. Inset of anteromedial thigh flap into ablative defect.

Tursun et al. Palatal Basal Cell Adenocarcinoma. J Oral Maxillofac Surg 2019.

then successfully underwent a course of postoperative radiation therapy without complications, and the most recent follow-up at 38 months showed no evidence of disease.

Discussion

BCAC is a rare malignant lesion of the salivary glands, accounting for fewer than 2% of all salivary neoplasms.[1] In 2005 the World Health Organization defined BCAC as "an epithelial neoplasm that has cytological characteristics of basal cell adenoma (BCA), but a morphologic growth pattern indicative of malignancy."[2-4] It is considered the malignant counterpart of BCA. Histologic differentiation between the two is difficult, and frequently, they can only be separated by identification of the invasion of local structures or by perineural or vascular invasion.[2] The present case is a perfect example of the difficulties in the pathologic differentiation, in which the patient twice received a diagnosis of a benign lesion before the malignant BCAC diagnosis was rendered. Most BCACs arise de novo, but approximately 20% arise from a pre-existing BCA. Approximately 90% of BCACs are found in the parotid gland, and the remainder occur in the submandibular gland.[5] Lo et al[6] were the first to report BCAC of the palate, but the clinicopathologic behavior of this tumor, which originates from the minor salivary gland, is still unclear.[7]

It is necessary to differentiate BCAC from other basaloid cell tumors of the minor salivary gland, such as canalicular adenoma, BCA, adenoid cystic carcinoma, polymorphous low-grade adenocarcinoma, myoepithelial tumor, epithelial-myoepithelial carcinoma, and basaloid squamous cell carcinoma, because of the prognosis and potential differences in treatment.[8] BCAs with capsular invasion share several

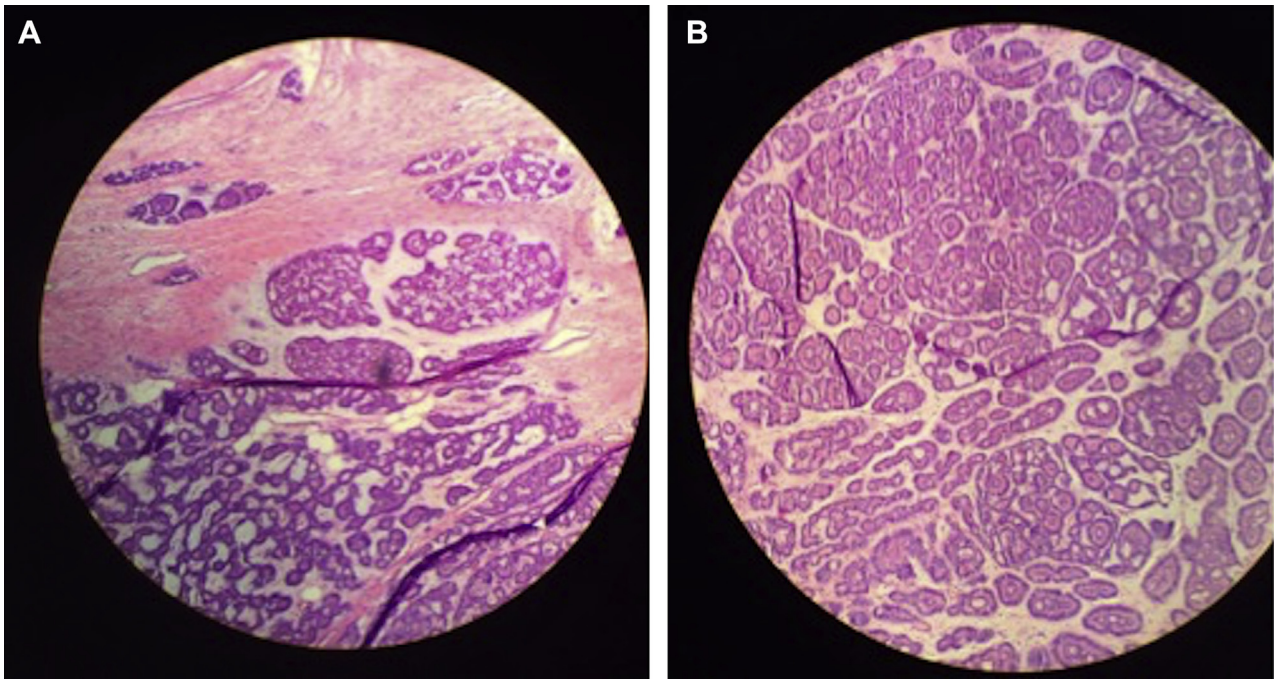


FIGURE 9. *A*, Low-power view of histologic examination with signs of invasion, with the absence of a capsule, and with evidence of mitotic figures (hematoxylin and eosin stain, magnification ×200). *B*, Higher-power view showing evidence of mitotic figures (hematoxylin and eosin stain, magnification ×400).

Tursun et al. Palatal Basal Cell Adenocarcinoma. J Oral Maxillofac Surg 2019.

pathologic features with BCACs, including large size and frequent cribriform patterns, but the malignant potential of these tumors seems highly limited and should be re-examined (Fig 9). In addition, β-catenin immunostaining may aid in the differentiation between a basal cell neoplasm and adenoid cystic carcinoma.[9]

Overall, BCAC has a more favorable outcome than other common malignant salivary gland tumors and is considered a low-grade malignancy.[10,11] The most common location of minor salivary gland tumors is the palate (42% to 54%), followed by the lips, but any other oral site harboring minor salivary glands may be affected.[12] The diagnosis of BCAC depends

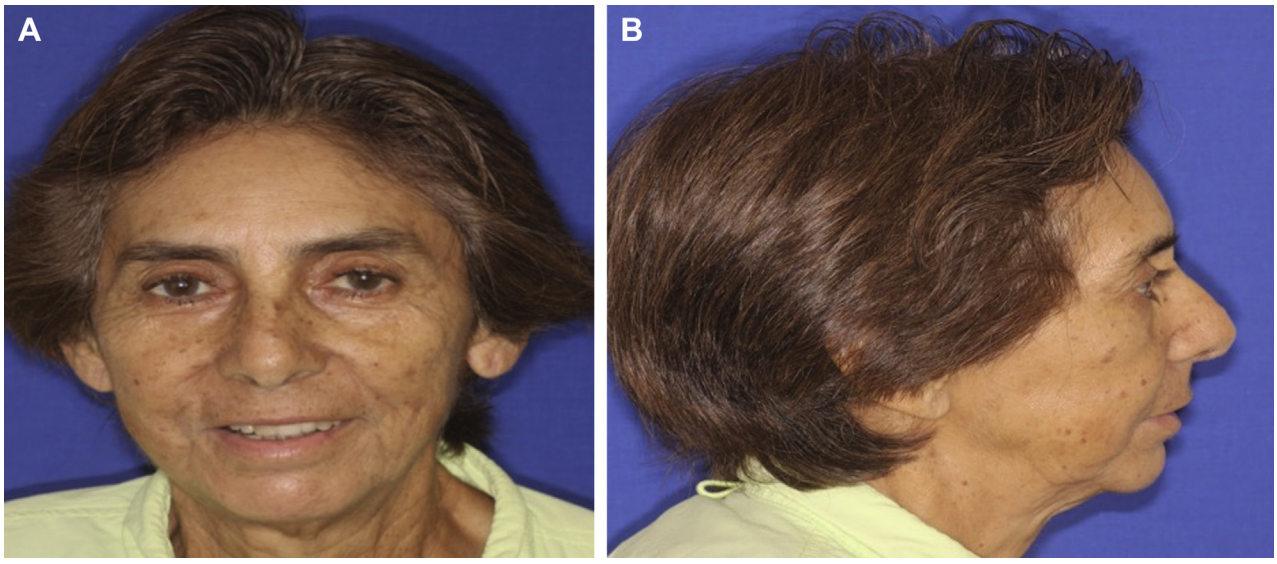


FIGURE 10. *A*, Frontal view of patient 3 years after surgery well-healed with no facial deformity. *B*, Lateral view with no scarring of neck or face owing to intraoral approach for vascular anastomosis to facial vessels.

Tursun et al. Palatal Basal Cell Adenocarcinoma. J Oral Maxillofac Surg 2019.

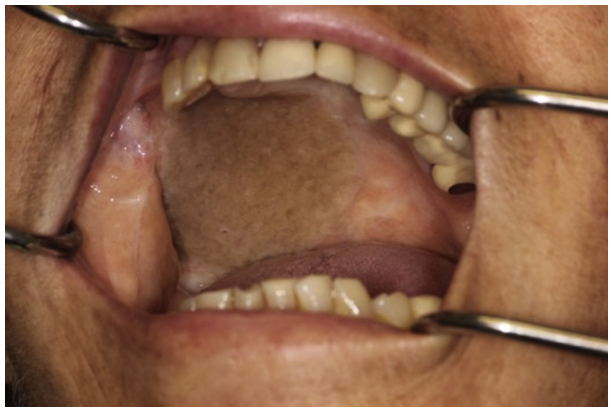


FIGURE 11. Intraoral view of well-healed flap with no signs of clinical recurrence.

Tursun et al. Palatal Basal Cell Adenocarcinoma. J Oral Maxillofac Surg 2019.

on the presence of features similar to adenoma but with an infiltrative growth pattern. Furthermore, adenoid cystic carcinoma, sialoblastoma, and basaloid carcinoma must be excluded.[13] The widely accepted treatment for BCAC is complete surgical excision.[14] Ellis and Wiscovitch[3] reported that among 29 patients with BCAC of the major salivary gland, 7 (28%) had at least 1 incident of local tumor recurrence and 3 (12%) had metastasis to the lung or cervical lymph nodes. An extensive literature review performed by Cuthbertson et al[7] included international and non–English-language literature; including their reported case, they identified a total of 72 cases of minor salivary gland BCAC.[15] Regional lymph node dissection is recommended only if there is evidence of metastatic disease to the cervical chain, and adjuvant radiation therapy has been used for invasive tumors and local recurrences.[16] It is unclear whether radiation therapy plays a major role in the management of this tumor, but it can be effective if the margin of the surgical specimen is close or the lesion shows local recurrence.[17]

A discussion also could be considered with regard to the plan for the immediate reconstruction of the maxillectomy defect. A review of the literature shows that this topic remains controversial. It has been our experience and has been the finding of other authors, as discussed by Moreno et al,[18] that in maxillectomy defects consisting of resection of the hard palate by greater than 50%, immediate free flap reconstruction results in superior functional outcomes when compared with obturation. Furthermore, it is often posited that immediate reconstruction leads to a delay in the diagnosis of recurrence when compared with obturation, but this assertion has not been supported by our experience or the findings of Moreno et al. In this case, an aggressive resection and clear frozen margins led the surgical team toward an immediate reconstruction. In retrospect, the permanent specimen finding of a close surgical margin could be used in making a case for delayed reconstruction.

The diagnosis of BCAC can be difficult to make with a simple biopsy, and the lesion may be easily mistaken for a benign basaloid tumor. Retrospectively, in the review of this case, the outside pathologist's new finding of ameloblastoma with a previous diagnosis of canalicular adenoma necessitated an oral-maxillofacial pathology consultation, which confirmed the diagnosis. In this case, it also would have been beneficial for an original slide request to be performed and a comparison completed of both sets of specimens despite the recurrence diagnosis having been confirmed by 2 general pathologists as well as an oral-maxillofacial pathologist. Owing to the many similar features, despite this review, inadequate sampling of the lesion may have left the pathologist without the ability to determine the infiltrative growth pattern that differentiates BCAC from similar lesions. In many such cases, we prefer to rebiopsy or personally review all outside pathologic diagnoses, and in our case, this may have proved beneficial. BCAC should be considered rare but nonetheless included in the differential diagnosis of lesions in this anatomic location when the lesions show similar clinical behavior. Furthermore, in this case, the patient underwent not only surgical ablation but also adjuvant radiation therapy because of the history of recurrence, as well as a close margin, and is disease free 3 years after surgery (Figs 10, 11).

References

1. Klima M, Wolfe K, Johnson PE: Basal cell tumors of the parotid gland. Arch Otolaryngol 104:111, 1978
2. Barnes L, Eveson JW, Reichart P, Sidransky D: Pathology and Genetics of Tumors of the Head and Neck: , *in* World Health Organization Classification of Tumors, Volume 9. Lyon, IARC Press, 2005
3. Ellis GL, Wiscovitch JG: Basal cell adenocarcinomas of the major salivary glands. Oral Surg Oral Med Oral Pathol 69:461, 1990
4. Franzen A, Koege lK, Knieriem HJ, Pfaltz M: [Basal cell adenocarcinoma of the parotid gland: A rare tumor entity: Case report and review of the literature]. HNO 46:821, 1998 (in German)
5. Jäkel KT, Löning T: Differential diagnosis of basaloid salivary gland tumors. Pathology 25:46, 2004
6. Lo AK, Topf JS, Jackson IT, et al: Minor salivary gland basal cell adenocarcinoma of the palate. J Oral Maxillofac Surg 50:531, 1992
7. Cuthbertson DW, Raol N, Hicks J, et al: Minor salivary gland basal cell adenocarcinoma: A systematic review and report of a new case. JAMA Otolaryngol Head Neck Surg 141:276, 2015
8. Barnes L, World Health Organization, International Agency for Research on Cancer. Pathology and Genetics of Head and Neck Tumors. Lyon, IARC Press, 2005
9. Jung MJ, Roh JL, Choi SH, et al: Basal cell adenocarcinoma of the salivary gland: A morphological and immunohistochemical comparison with basal cell adenoma with and without capsular invasion. Diagn Pathol 8:171, 2013
10. Ellis GL, Auclair PL: Atlas of Tumor Pathology. Tumors of the Salivary Glands. Washington, DC, Armed Forces Institute of Pathology, 1996

11. Yamagata K, Oka K, Yoshida H, et al: Basel cell adenocarcinoma arising from the minor salivary gland in the soft palate: A case report. Pathol Res Pract 202:475, 2006
12. Neville BW, Damm DD, Allen CM, Bouquot JE: Salivary gland pathology, *in* Neville BW, Damm DD, Allen CM, et al (eds): Oral and Maxillofacial Pathology (ed 2). Philadelphia, Saunders, 2002, pp 406–410
13. Ellis GL, Auclair PL: Malignant epithelial tumours, *in* Rosai LH, Sobin LH (eds): Atlas of Tumour Pathology, 3rd Series, Fascicle 17. Tumors of Salivary Glands. Washington, DC, Armed Forces Institute of Pathology, 1996, pp 257–267
14. Quddus MR, Henley JD, Affify AM, et al: Basal cell adenocarcinoma of the salivary gland. An ultrastructural and immunohistochemical study. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 87:485, 1999
15. Akiyama K, Karaki M, Hosikawa H, Mori N: A massive basal cell adenocarcinoma of the palatal minor salivary gland that progressed into the pterygopalatine fossa. Int J Oral Maxillofac Surg 41:444, 2012
16. Gross M, Maly B, Goldfarb A, Eliashar R: Basal cell adenocarcinoma in a buccal minor salivary gland. Acta Otolaryngol 124: 213, 2004
17. Wada T, Morita N, Sakamoto T, Nakamine H: Basal cell adenocarcinoma of the minor salivary gland: A case report. J Oral Maxillofac Surg 58:811, 2000
18. Moreno MA, Skoracki RJ, Hanna EY, Hanasono MM: Microvascular free flap reconstruction versus palatal obturation for maxillectomy defects. Head Neck 32:860, 2010