Case Discussion

Endoscopic trans-pterygoid resection of a low-grade cribriform cystadenocarcinoma of the infratemporal fossa

Vikram G. Ramjee a, Landon J. Massoth a, c, John P. Richards II b, Kibwei A. McKinney a, *

a Department of Otolaryngology — Head & Neck Surgery, University of Oklahoma Health Sciences Center, Oklahoma City, OK, USA
b Department of Otolaryngology - Head and Neck Surgery, University of Arizona, Tucson, AZ, USA
c Department of Otolaryngology - Head and Neck Surgery, University of Washington, Seattle, WA, USA

Received 19 August 2019; accepted 19 January 2020
Available online 18 March 2020

Abstract This article presents a case of low-grade cribriform cystadenocarcinomas (LGCCC), a rare salivary gland tumor manifesting in the infratemporal fossa (ITF). The lesion in this case is unique in its location, histopathology, and management in that the tumor resection was performed using an exclusively endoscopic, endonasal approach. This case highlights the expanding application of endoscopic skull base techniques to address an indolent, slow-growing malignancy of the ITF.

Copyright © 2020 Chinese Medical Association. Production and hosting by Elsevier B.V. on behalf of KeAi Communications Co., Ltd. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

Introduction

Low-grade cribriform cystadenocarcinoma (LGCCC) is a recently described rare salivary gland neoplasm that largely arises in the parotid gland and exhibits an indolent course. In 1996, Delgado et al. originally defined this tumor as a low-grade salivary duct carcinoma (LGSDC). In the third edition of the World Health Organization (WHO) Classification of Head and Neck Tumors, it was initially categorized as a variant of cystadenocarcinoma. However, the most
recent edition of the WHO Classification has reclassified this entity as a low-grade intraductal carcinoma.2 As a rule, LGCCC is a parotid tumor but rare cases have been reported to arise from the submandibular gland, accessory parotid gland, infra-parotid lymph nodes, and palate.3–6 In this report, we detail the only case in the English literature of primary LGCCC arising from tissue within the infratemporal fossa (ITF) and its surgical management using an exclusively endoscopic, endonasal approach.

Case description

A 46-year-old woman presented with an incidental finding of an ITF mass on magnetic resonance imaging (MRI) that was performed in preparation for spinal surgery. She complained of mild right-sided facial pain and headache, but was otherwise asymptomatic. Physical examination and sinonasal endoscopy were unremarkable. An MRI of the head and neck revealed a well-circumscribed, lobulated mass of the pre-styloid parapharyngeal space, measuring 2.5 cm × 2 cm (Fig. 1). Computed tomography angiography (CTA) demonstrated an enhancing mass that displaced the right internal carotid artery posteriorly without invasion, and branches of the right external carotid artery coursing along its lateral and postero-lateral margins. A benign tumor or low-grade malignancy of the parapharyngeal space was suspected. Surgery was offered, including endoscopic medial maxillectomy and trans-pterygoid approach under stereotactic, computer-assisted navigation.

A nasoseptal flap was elevated at the outset of the case in preparation of reconstruction of the ITF defect. A posterior septectomy was performed, providing binarial access. A modified Denker’s approach, transecting the nasolacrimal duct, and medial maxillectomy were then performed. A trans-pterigoid approach to the infratemporal fossa was then performed using a 2-surgeon, 3-handed technique. The tumor was visualized and found to be well-encapsulated. The lesion was then dissected circumferentially from its soft tissue attachments and removed en bloc. Because of the large size of the defect and its proximity to the internal carotid artery, an abdominal fat graft was harvested and used in conjunction with the nasoseptal flap to reconstruct the ITF defect.

The histopathologic and immunohistochemistry studies identified the tumor as a low-grade cribriform cystadenocarcinoma, likely arising from the deep lobe of the parotid gland. The lesion was resected in its entirety, with clear tumor margins. At 8 months postoperatively, follow-up imaging, including MRI and positron emission tomography/computed tomography (PET/CT), showed no sign of recurrent disease or distant metastases.

Discussion

LGCCC is a rare neoplasm that has recently been reclassified by the WHO as a low-grade intraductal carcinoma.2 Originally, the malignancy was considered to be a low-grade variant of salivary ductal carcinoma. However, both its clinical behavior and histopathologic features have shown that the tumor differs from other salivary duct carcinomas in its pattern of growth and the absence of pathologic characteristics such as nuclear atypia, invasiveness into surrounding tissue, or metastasis to regional lymph nodes. Since its initial description in 1996, 47 cases have been described in the literature, including the present case. Although the number of cases with long-term follow up is low, none have shown evidence of recurrence, metastasis, or mortality from the tumor.7–9 The majority of tumors has arisen within the parotid gland, but has also infrequently involved the submandibular gland, palate and minor salivary glands. To the best of our knowledge, there have been no reported cases of LGCCC (or LGSDC) arising in the infratemporal fossa.

There have been only been a handful of reports describing the morphological findings of LGCCC. The findings included: (1) proliferation of ductal epithelial cells with tight junctions; (2) mild nuclear atypia and minimal size variation; and (3) cytoplasmic vacuoles and squamoid or metaplastic changes in tumor cells.7,9 The histology in this case revealed neoplastic epithelial cells with moderate atypia and nuclei that lacked any significant atypia or size variation. The tumor cells formed glands that varied significantly in size. In the larger glands, tumor cells demonstrated a cribriform growth pattern and signs of mucin production (Fig. 2). Moreover, there was no evidence of tumor invasion into the surrounding structures, tumor

Figure 1  A: Coronal T1-weighted MRI sequence demonstrating a lobulated, well circumscribed mass in the right infratemporal fossa. B: Axial T1 MRI demonstrating a T1-weighted hypointense lesion of the infratemporal fossa. C: Axial T2 MRI showing this lesion with T2-weighted hyperintensity.
necrosis, perineural extension, or vascular invasion, similar to other published reports of LGCCC. Immunohistochemistry revealed strong, diffuse, S100 positivity and the presence of myoepithelial markers (e.g., calponin) surrounding cystic spaces—consistent with previously reported cases of this tumor type.

Due to the favorable outcome of the diagnosis of LGCCC, a fine-needle aspiration (FNA) of the tumor is usually desirable for initial histopathologic examination. That was not possible in this particular case due to the location of the tumor within the ITF. Thus, the decision was made to proceed with an operative excisional biopsy for frozen histopathologic analysis and complete surgical removal of the tumor.

With respect to treatment, this case highlights the feasibility of complete surgical resection with an endoscopic, endonasal approach to the ITF. This approach provided a shorter recovery time, improved cosmesis, and reduced morbidity in comparison to more traditional sub-temporal, transtemporal and anterior transfacial approaches. We consider this a reasonable application of an endoscopic skull base technique to address an indolent, slow-growing malignancy of the ITF.

Declaration of Competing Interest

Kibwei A. McKinney is a part of the Speakers Bureau for Intersect ENT.

References

1. Delgado R, Klimstra D, Albores-Saavedra J. Low grade salivary duct carcinoma. A distinctive variant with a low grade histology and a predominant intraductal growth pattern. Cancer. 1996;78:958–967. http://www.ncbi.nlm.nih.gov/pubmed/8780532.
2. Seethala RR, Stenman G. Update from the 4th edition of the World Health organization classification of head and neck tumours: tumors of the salivary gland. Head Neck Pathol. 2017;11:55–67. http://www.ncbi.nlm.nih.gov/pubmed/28247227.
3. Weinreb I. Intraductal carcinoma of salivary gland (so-called low-grade cribriform cystadenocarcinoma) arising in an intra-parotid lymph node. Head Neck Pathol. 2011;5:321–325. http://www.ncbi.nlm.nih.gov/pubmed/21442195.
4. Nakatsu S, Harada H, Fujiyama H, et al. An invasive adenocarcinoma of the accessory parotid gland: a rare example developing from a low-grade cribriform cystadenocarcinoma. Diagn Pathol. 2011;6:122. http://www.ncbi.nlm.nih.gov/pubmed/22151879.
5. Brandwein-Gensler M, Hille J, Wang BY, et al. Low-grade salivary duct carcinoma: description of 16 cases. Am J Surg Pathol. 2004;28:1040–1044. http://www.ncbi.nlm.nih.gov/pubmed/15252310.
6. Tatemoto Y, Ohno A, Osaki T. Low malignant intraductal carcinoma on the hard palate: a variant of salivary duct carcinoma. Eur J Cancer B Oral Oncol. 1996;32B:275–277. http://www.ncbi.nlm.nih.gov/pubmed/8776426.
7. Wakabayashi N, Umezawa H, Matsumoto NM, Endo Y, Naito Z, Ogawa R. Low-grade cribriform cystadenocarcinoma: a review of the literature and case report. Plast Reconstr Surg Glob Open. 2017;5:e1306. http://www.ncbi.nlm.nih.gov/pubmed/28507867.
8. Nishijima T, Yamamoto H, Nakano T, et al. Low-grade intraductal carcinoma (low-grade cribriform cystadenocarcinoma) with tumor-associated lymphoid proliferation of parotid gland. Pathol Res Pract. 2017;213:706–709. http://www.ncbi.nlm.nih.gov/pubmed/28551385.
9. Kimura M, Mil S, Sugimoto S, Saida K, Morinaga S, Umemura M. Low-grade cribriform cystadenocarcinoma arising from a minor salivary gland: a case report. J Oral Sci. 2016;58:145–149. http://www.ncbi.nlm.nih.gov/pubmed/27021553.
10. Jeong JY, Ahn D, Park JY. Fine-needle aspiration cytology of low-grade cribriform cystadenocarcinoma with many psammoma bodies of the salivary gland. Korean J Pathol. 2013;47:481–485. http://www.ncbi.nlm.nih.gov/pubmed/24255638.