Case Report: Subsequent hypopharyngeal adenoid cystic carcinoma accompanied by synchronous esophageal carcinoma following previous squamous cell carcinoma in hypopharynx

Ning Jia[1], Yu Chen[2], Boju Pan[3], Chunmei Bai[1], and Yingying Zhu[4]

[1]Chinese Academy of Medical Sciences & Peking Union Medical College Fuwai Hospital
[2]Chinese Academy of Medical Sciences & Peking Union Medical College Fuwai Hospital Department of Radiology and Imaging
[3]Peking Union Medical College Hospital Department of Pathology
[4]Peking Union Medical College Hospital Department of Otolaryngology

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Abstract

Background: The adenoid cystic carcinoma (ACC) barely arises in sites other than the salivary glands. Its presentation in the hypopharynx previously diagnosed as squamous cell carcinoma (SCC) is entirely unexpected. **Case Presentation**: A man was presented with a hypopharyngeal mass revealed by electronic laryngoscopy. Multiple biopsies pathologically showed well to moderately differentiated SCC. Six months after definitive concurrent chemoradiation, his endoscopy showed a recurrent mass in the same region before and pathology of gastroscopic biopsy showed moderately differentiated SCC of esophagus. The mass was removed after radical dissection. Histologic examination showed sheets and cords of tumor cells with basophilic matrix in hypopharynx known as ACC and moderately differentiated esophageal SCC. **Conclusion**: The documentation of metachronous ACC arising in hypopharynx following previous SCC would further supports the concern about multicomponent tumors in hypopharynx which is crucial in clinical practice.

Title page

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Running title: Hypopharyngeal adenoid cystic carcinoma following previous squamous cell carcinoma

Ning Jia, MD[1,2], Yu Chen, MD[3], Boju Pan, MD[4], Chunmei Bai, MD[1] and Yingying Zhu, MD[5]

[1]Department of Medical Oncology, Peking Union Medical College Hospital, Peking Union Medical College and Chinese Academy of Medical Sciences, Beijing, China

[2]Gansu Cancer Hospital, Lanzhou, Gansu, China

[3]Department of Radiology, Peking Union Medical College Hospital, Peking Union Medical College and Chinese Academy of Medical Sciences, Beijing, China

[4]Molecular Pathology Research Center, Department of Pathology, Peking Union Medical College Hospital, Peking Union Medical College and Chinese Academy of Medical Sciences, Beijing, China

[5]Department of Otolaryngology-Head and Neck Surgery, Peking Union Medical College Hospital, Peking Union Medical College and Chinese Academy of Medical Sciences, Beijing, China

Corresponding Author:

Yingying Zhu, MD, Department of Otolaryngology-Head and Neck Surgery, Peking Union Medical College Hospital, Peking Union Medical College and Chinese Academy of Medical Sciences, 1 Shuaifuyuan, Dongcheng District, Beijing 100730, China.

Email: *zhuyyent@163.com*

AUTHOR CONTRIBUTIONS

Ning Jia , conception, acquisition, interpretation, drafting, revision, approval, accountable; **Yu Chen** , acquisition, interpretation, revision, approval, accountable; **Boju Pan** , acquisition, interpretation, revision, approval, accountable;**Chunmei Bai** , revision, approval, accountable; **Yingying Zhu** , conception, revision, approval, accountable.

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CONFLICT OF INTEREST STATEMENT

There are no conflicts of interest.

DATA AVAILABILITY STATEMENT

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

ETHICS STATEMENT

This observation was approved by the ethics committee (Peking Union Medical College Hospital Institutional Review Board, No. K23C2283).

Abstract

Background : The adenoid cystic carcinoma (ACC) barely arises in sites other than the salivary glands. Its presentation in the hypopharynx previously diagnosed as squamous cell carcinoma (SCC) is entirely unexpected.

Case Presentation :

A man was presented with a hypopharyngeal mass revealed by electronic laryngoscopy. Multiple biopsies pathologically showed well to moderately differentiated SCC. Six months after definitive concurrent chemoradiation, his endoscopy showed a recurrent mass in the same region before and pathology of gastroscopic biopsy showed moderately differentiated SCC of esophagus. The mass was removed after radical dissection. Histologic examination showed sheets and cords of tumor cells with basophilic matrix in hypopharynx known as ACC and moderately differentiated esophageal SCC.

Conclusion : The documentation of metachronous ACC arising in hypopharynx following previous SCC would further supports the concern about multicomponent tumors in hypopharynx which is crucial in clinical practice.

Keywords: hypopharyngeal cancer, adenoid cystic carcinoma, squamous cell carcinoma, basaloid squamous cell carcinoma with adenoid cystic-like features, collision tumor

1 | INTRODUCTION

Adenoid cystic carcinoma (ACC) is a rare primary neoplasm of salivary glands which is 1% of all head and neck cancers and 10% of all salivary gland neoplasms. ACC arises in both major and minor salivary

glands. The parotid gland accounts for 25% of origin in head and neck which is the most common site. [1] Occasionally it arises in the nose, the paranasal sinuses, the lachrymal glands, the ceruminous glands of the external auditory canal, the larynx, the nasopharynx, and the palate.

Anatomically, ACC of hypopharynx is an extremely rare disease. The most common histologic type of hypopharyngeal malignancy is squamous cell carcinoma (SCC), which originates very differently from ACC. Moreover, treatment backbone of ACC is surgery while the role of radiation and systemic therapy are still controversial, unlike the treatment strategy of SCC.

We report the case of a hypopharyngeal ACC presenting several months after a previous diagnosis of SCC. The rare incidence and a short time earlier history of SCC could pose a diagnostic challenge for ACC in hypopharynx.

2 | CASE PRESENTATION

A 51-year-old male heavy smoker was presented with a history of dysphagia. Electronic laryngoscopy showed a mass in right pyriform fossa and postcricoid region. Multiple biopsies were taken and examined pathologically showing well to moderately differentiated SCC. Then the patient received definitive concurrent chemoradiation in local hospital. Six months later, his symptoms reappeared and endoscopy showed a mass in right pyriform fossa. After he was admitted to our hospital, contrasted CT scanning demonstrated a heterogeneously enhancing 1.6 × 1.5 - cm laryngopharyngeal mass, mainly involving right pyriform fossa and right aryepiglottic folds (Figure 1). Pathology of gastroscopic biopsy showed moderately differentiated SCC of esophagus. The patient underwent total pharyngolaryngoesophagectomy. Pathology revealed sheets and cords of tumor cells with basophilic matrix in hypopharynx. The tumor cells showed scant cytoplasm and had typical small angulated and hyperchromatic nuclei (Figure 2). Immunohistochemistry revealed the tumor cells consist of epithelial or myoepithelial elements, i.e. CD117 positive in epithelial elements, P63, SMA and S100 positive in myoepithelial elements, supporting the diagnosis of ACC. And moderately differentiated esophageal SCC was also identified. The patient remained disease free on the date of last follow up.

3 | DISCUSSION

SCC accounts for 90% of tumors in hypopharynx. Other histological types are relatively uncommon. As for ACC, it is extremely rare. Dysphagia is one of the common symptoms related to ACC, especially for solids, which are similar to SCC. Imaging studies, e.g. CT or MRI, contribute to provide anatomic details that are useful for surgical planning, but not to make differentiation from SCC. A tissue diagnosis is required to make the diagnosis. Only few authors reported cases of ACC in hypopharynx.[2,3]

Furthermore, the pathologic features of basaloid squamous cell carcinoma (BSCC) can be similar to that of ACC. BSCC generally displays certain histological features, such as no presentation of bi-layered structures and basophilic matrix, and diffuse immunopositivity for p63 and p40 in tumuor cells revealed the absence of myoepithelial elements.[4] Although it is known that BSCC with adenoid cystic-like features (BSCC-AdC) occurs more commonly in esophagus[5], BSCC-AdC also sometimes occurs in hypopharynx.[6]

Bicomponent cancer of SCC and ACC also should be concerned for the potential limitation of tissue biopsy. Unlike overlapping malignancy of head and neck SCC and esophageal SCC, collision tumor of head and neck was barely seen. But there was collision tumor of SCC and ACC in larynx or hypopharynx reported before, even with synchronous esophageal carcinoma like this case. [7,8] Complete histological investigation of a neoplasm and affection on multicomponent tumors are crucial in the successful diagnosis of a collision tumor.

To our knowledge, hypopharyngeal subsequent ACC following SCC is a rare tumor not previously described. Pathology is needed to make the correct diagnosis, and complete excision is currently the standard treatment approach. Our case demonstrates that clinicians should be aware of the possible ACC, even if a diagnosis of SCC was made before, because different therapy strategy and oncological follow-up planning need to be considered for these two tumor entities.

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Figure legends

Figure 1. Transverse contrasted CT – a hypopharyngeal mass.

Figure 2. Sheets of tumor cells with basophilic matrix. The tumor cells showing scant cytoplasm and typically having small angulated and hyperchromatic nuclei. Immunohistochemistry revealing the tumor cells consist of epithelial or myoepithelial elements (not shown).100x