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Adenoid Cystic Carcinoma of Tonsillar Fossa - A Rare Case report

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Abstract

Adenoid cystic carcinoma is a malignant epithelial tumor of the salivary glands. It represents less than 1% of all head and neck cancers and less than 10% of all salivary gland cancers. It shows a slow and indolent growth rate, a low probability of regional lymph node metastases, a high propensity for perineural invasion, multiple and / or late recurrences, and a high incidence of distant metastases. Surgical excision with wide margins is the treatment of choice, if it metastasizes to lymph nodules, post-surgical radiotherapy is recommended. In this article, a case report of adenoid cystic carcinoma of the oral cavity located in the left palatine tonsil and right peritonsillar tissue is discussed, which was treated with surgery and radiotherapy as an additional treatment.

Keywords: Adenoid cystic carcinoma, palatine tonsil

Introduction

Adenocystic carcinoma is a slow-growing but highly invasive epithelial neoplasia with a high recurrence rate, delineated by proliferation of ductal and myoepithelial cells in cribriform (classic), tubular, solid (basaloid), and cystic types. [1] It represents less than 1% of all head and neck cancers and less than 10% of all salivary gland cancers. It is the most common malignancy of the minor salivary glands. The oral cavity and oropharynx are the most frequent sites, followed by the parotid, submandibular gland, paranasal sinuses, larynx, trachea, and pharynx. [2] Unconventional sites include the external auditory canal, nasopharynx, lacrimal glands, breast, vulva, esophagus, cervix, and Cowper's glands. [3-8] In this article, a case report of adenoid cystic carcinoma of the oral cavity located in the left palatine tonsil and right peritonsillar tissue is discussed, which was treated with surgery and radiotherapy as an additional treatment.

Case Report

An 82-year-old male patient presented with left ear pain lasting 1 month. Oropharyngeal examination revealed a solid, pale, proliferative, bilobed, soft, and painless growth measuring 4 x 5 cm, arising from the left tonsillar fossa (Fig. 1). There were lymphadenopathies of pathological size whose nature was undistinguishable even on palpation. Axial T1-weighted magnetic resonance imaging (MRI) revealed a soft tissue lesion affecting the left tonsillar fossa that crossed the midline; extending upward to the left palatopharyngeal arch and uvula. The left tonsillar and lingual sulcus and the left vallecula were also infiltrated (Fig. 2). No peripheral or regional invasion and lymphadenopathy was detected. Whole-body positron emission tomography with fluorine-18 fluorodeoxyglucose computed tomography did not detect a delineate site for primary and distant metastasis. The lesion was extensively excised (in its entirety) under general anesthesia. Histological interpretation revealed adenoid cystic carcinoma in the left tonsillar fossa. Control punch biopsies of multiple margins of the tongue base, tonsillar housing, anterior and posterior tonsillar pillars, and bilateral uvular base were revealed histologically free of neoplasm. The TNM stage was determined as pT3 pNx MX. The histological pattern revealed a mixed cribriform pattern (70%) and a solid variant (30%). High-grade transformation was detected, but perineural invasion was observed, no lymphovascular emboli were observed (Fig. 3). The patient underwent adjuvant postoperative radiotherapy followed by regular follow-up for 12 months with no signs of new foci, recurrence, and distant metastases. The informed consent of the patient was taken. Institutional review board approval was procured from the HCG Manavata Cancer Institute ethics committee.

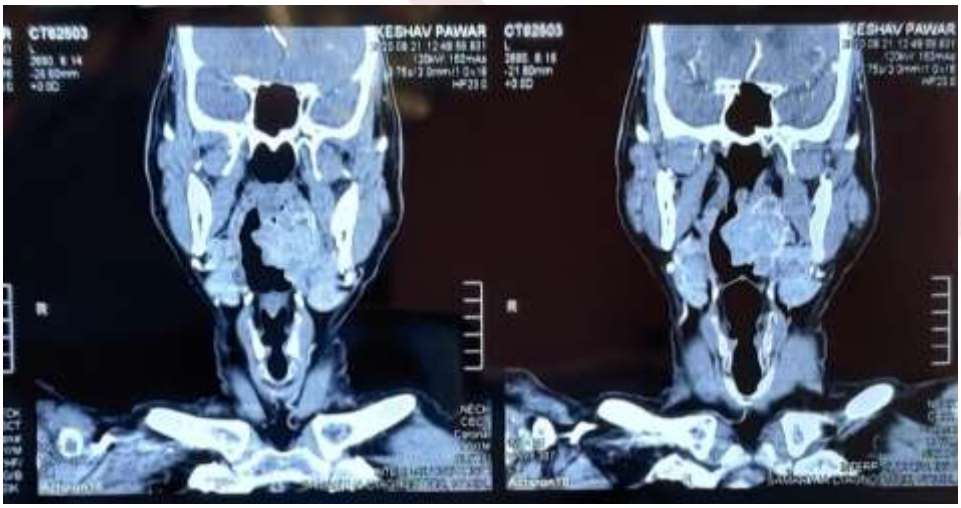

Figure 1

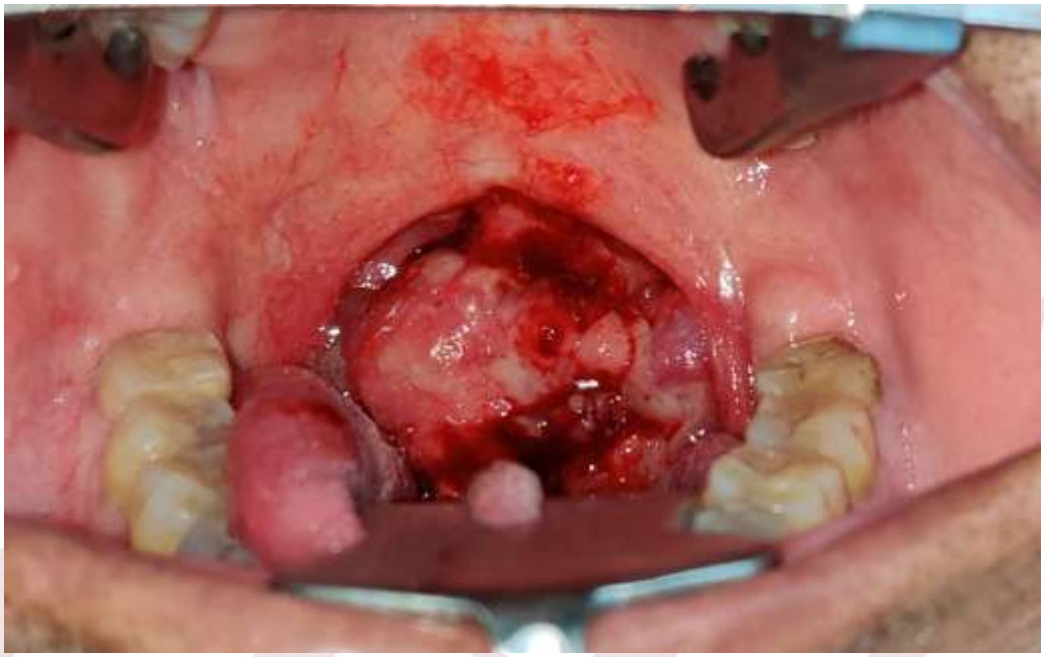

Figure 2

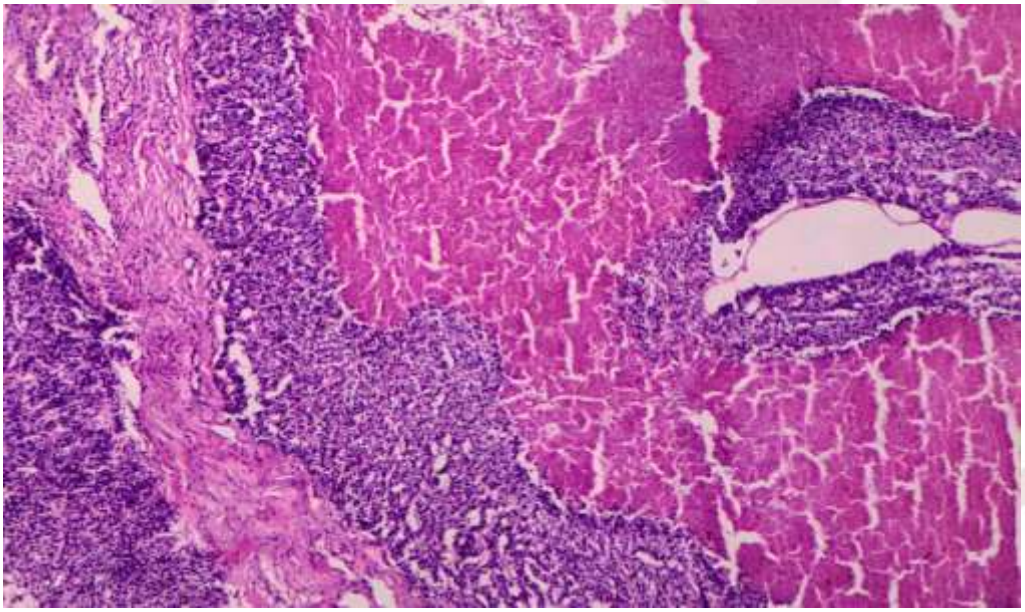

Figure 3

Discussion

Adenocystic carcinoma is outlined by a slow and indolent growth rate, a low contingency of regional lymph node metastases, a high susceptibility for perineural invasion, multiple and / or late recurrences, and a high incidence of distant metastases. [1]

First described as "cylindroma" by Billroth, it is usually classified with salivary gland neoplasm; even though it can emerge from any site where mucous glands are present. [9] Half of these neoplasms occur in glandular areas other than the major salivary glands, such as the lacrimal glands, the ceruminous glands of the external auditory canal, the esophagus, the breast, the prostate, the uterus, the cervix, and the Bartholin glands. Unconventional sites include the external auditory canal, nasopharynx, lacrimal glands, breast, vulva, esophagus, cervix, and Cowper's glands. 3-8 Adenocystic carcinoma is composed of myoepithelial cells and ductal cells that have varied arrangements. Morphologically, 3 growth patterns have been described: cribriform (classic), tubular and solid (basaloid) with little tendency to the formation of ducts or cysts. The tubular and cribriform types have a better prognosis than the solid type. Perineural invasion is a characteristic feature of the tumor and usually invades well beyond clinically apparent limits. [1,10]

In our case, histologically, tumor infiltration was prominent in the left tonsil parenchyma including the surface epithelium (capsule) and the central lymphoid tissue together, whereas on the right side, tumor was localized superficially and adherent to the tonsillar capsule. Adenoid cystic carcinoma of the oral cavity may locally extend through perineural invasion. The biopsy specimens of the tongue base were clear; no tumoral infiltration was noted. On the other hand, simultaneous separate foci in the left and right palatine tonsils or metastasis to contralateral tonsil sparing the oral structures in between may be the possible mechanisms of the spread pattern.

Simultaneous bilateral tonsil involvement or metastasis to contralateral tonsil was not reported previously for the adenoid cystic carcinoma of the oral cavity. In cases of intraoral adenoid cystic carcinoma, multiple simultaneous foci may appear. The detailed evaluation of the oral cavity to reveal the site of involvement is important. In case of tonsillar involvement, bilateral tonsillectomy is more recommended and a comprehensive option both for definite detection of foci and treatment.

Conclusion

In conclusion, the case report of adenoid cystic carcinoma underscores the complex and challenging nature of this rare malignancy. The presented case not only highlights the diagnostic difficulties associated with this disease but also serves as a reminder of the potentially indolent yet relentlessly progressive course of adenoid cystic carcinoma. The multi-disciplinary approach to management, involving surgical resection, adjuvant radiation therapy, and close surveillance, reflects the current standard of care for this malignancy.

Furthermore, the case report underscores the need for ongoing research and clinical trials aimed at identifying novel therapeutic targets and treatment modalities for adenoid cystic carcinoma. The rarity of this disease presents a significant barrier to the development of evidence-based guidelines, making it imperative for clinicians to engage in collaborative efforts and knowledge-sharing to improve the outcomes for patients affected by this challenging malignancy.

Ultimately, this case report serves as a call to action for continued vigilance in the diagnosis and management of adenoid cystic carcinoma, as well as a testament to the resilience and determination of both patients and healthcare providers in confronting the complexities of rare cancers. By sharing experiences and insights, we can strive to enhance our understanding of adenoid cystic carcinoma and work towards improved outcomes and quality of life for those impacted by this disease.

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