Neoadjuvant Chemotherapy as a viable treatment option for locally Advanced Adenoid Cystic Carcinoma of salivary gland: A case report

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Abstract

Adenoid cystic carcinoma (ACC) is a rare and aggressive variant of salivary gland neoplasm. Perineural invasion and resistance to present chemotherapeutic drugs makes treatment more challenging. Surgery remains the treatment of choice in resectable cases with postoperative radiotherapy in selected subset. In upfront technically unresectable cases neoadjuvant chemotherapy (NACT) can be used as an option to achieve R0 resection. Here we present a case of minor salivary gland ACC that was successfully down staged and underwent R0 resection after NACT.

Introduction

The salivary gland tumors are rare neoplasm of head and neck region, and only 20% are found to be malignant [1]. Adenoid cystic carcinoma (ACC) accounts for 10-20% of all malignant tumors of salivary glands [1–2]. Despite having slow growth kinetics ACC represents important therapeutic challenge because of its tendency of perineural invasion, thus posing a challenge to surgical resection and subsequent high chances of recurrence [1, 2]. For localized disease surgery remains the cornerstone of treatment. In the absence of long term data most of the clinicians recommend post operative radiotherapy to reduce the relapses. Primary treatment with radiation is considered when surgery is not feasible [2, 3].

Role of chemotherapy is still investigational and mainly remains limited to advanced and metastatic disease with poor outcomes [4]. Neoadjuvant chemotherapy (NACT) has shown promising results in various studies in locally advanced cancers of oral cavity [5]. Considering the dearth of NACT as treatment modality, the present case reports a locally advanced ACC that was down staged by NACT and further underwent a R0 surgical resection.

Case Presentation

A 50 year old gentleman presented with a three months history of recurrent swelling in left buccal mucosa and left neck region. He was operated twice at a private nursing home before presenting to our center. First excision was one year back via trans-oral route and second surgery was six months back via cheek incision; however detailed operative notes and histology reports were not available. On examination a healed scar was present over left cheek in pre-parotid region, intraoral examination showed only a small ulcer over left buccal mucosa near parotid duct opening. On bimanual palpation 3x3 cm submucosal mass was present in left buccal mucosa, densely adherent to overlying scar. Another 5x5 cm matted nodal mass was present in level Ib, II and III. Core needle biopsy from nodal mass was suggestive of adenoid cystic carcinoma

Contrast CT scan showed left buccal mucosa mass with matted nodal mass in level Ib, II and III and no lung metastasis [Figure 1]. In view of rapidly recurring primary and matted lymph nodes tumor was deemed technically unresectable and decision for neoadjuvant chemotherapy was taken in

multidisciplinary clinic. He received three cycles of paclitaxel (175mg/m²) and carboplatin (AUC-5) at three weekly intervals. After three cycles patient had partial response (approximately 60% regression in primary tumor and 80% regression in nodal mass). Post chemotherapy MRI showed near complete reduction in size of the nodal mass [Figure 1].

Patient underwent wide excision of primary tumor with superficial parotidectomy and modified type II neck dissection (level I- V). The skin and soft tissue defect was reconstructed with Bipodal pectoralis major myocutaneous flap (PMMC). Postoperative recovery was uneventful. Final histology showed adenoid cystic carcinoma of minor salivary gland with clear margins and 2 lymph nodes (both level IIb) showed metastatic tumor deposit out of 42 dissected nodes [Figure 2]. He received post operative 60 Gy radiotherapy to the primary and neck region. After two years of follow up patient is disease free.

Discussion

Adenoid cystic carcinoma (ACC) accounts for 1% of all head and neck cancer, majority arise from minor salivary glands and oral cavity is the most common sub site [1, 2]. Surgery remains the cornerstone of treatment whenever feasible in localized disease and radical radiation/ chemoradiation is only considered an option when tumor is deemed unresectable [1–3]. Despite achieving margin negative resection local recurrences and distant metastasis are quite common after surgery. Perineural invasion seen in 10-30% cases is peculiar feature of ACC and may be responsible for such high risk local recurrences [2, 3]. Adjuvant radiotherapy is an option to reduce the post operative recurrence in intermediate or high grade ACC or ACC resected with close or positive margin. Various retrospective studies have shown that radiotherapy seems to reduce local recurrences but the impact on survival is still not clear [6, 7]. Incidence of lymph node metastasis varies from 14-20% in salivary gland neoplasm [8]. In ACC chances of lymph node metastasis are low in compare to other salivary neoplasm; however in present case extensive nodal disease was present making it not suitable for upfront surgery.

In view of slow growth kinetics most of the ACC shows low response rate and do not benefit from chemotherapy. Chemotherapy and targeted therapy in ACC is mainly limited to recurrent and metastatic disease with poor outcomes [4, 6]. Few retrospective studies have shown the benefit of neoadjuvant chemotherapy (NACT) in very advanced, technically unresectable oral cancer patients [5]. In view of rarity of disease no good quality data exists regarding use of NACT in ACC or other salivary neoplasm. Only few case reports have shown role of NACT in locally advanced salivary neoplasm [9, 10].

In present case in view of advanced nodal disease we used NACT as an option as it was technically unresectable upfront and got a good response to chemotherapy. He underwent radical surgery with successful R0 resection. So in selected locally advanced/upfront unresectable cases NACT can be a practical therapeutic tool.

Conclusion

Surgery remains the definitive treatment for resectable ACC. There is scarcity of literature regarding use of neoadjuvant chemotherapy in salivary gland neoplasm. The Present case report suggests that neoadjuvant chemotherapy is a viable treatment option in selected locally advanced technically unresectable salivary neoplasms/ACC.

Declarations

1. Funding: No financial help received
2. Conflicts of interest: None to declare
3. Ethics approval: Obtained (Human research ethics committee – Geetanjali Hospital) Registration number: ECR/623/Inst/RJ/2014/RR-20
4. Consent to participate: Obtained
5. Consent for publication: Complete anonymity of study participant has been maintained and consent obtained as well for publication
6. Availability of data and material: complete transparency maintained and files uploaded in supplementary material
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8. Authors contribution: Separate contribution table attached

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Figures

Figure 1

[A and B] Prechemotherapy CT images showing primary buccal mucosa mass with matted nodal mass with necrosis in level II-IV. [C and D] Postchemotherapy MRI images show residual primary mass in buccal mucosa and almost resolved nodal mass in neck.

Figure 2

[A] Adenoid cystic carcinoma involving minor salivary gland of buccal mucosa.(HE;10x) [B] ACC showing nests of tumor cells interrupted by punched out spaces (HE; 10x) [C] ACC showing Cribriform pattern with basophilic matrix within punched out spaces. (HE;20x) [D] ACC cells having small angulated hyperchromatic nuclei with scant cytoplasm.

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