Clear Cell Myoepithelial Carcinoma of the Base Tongue Managed by the Mandible Preserving Pull-Through Oropharyngectomy Approach

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Abstract Myoepithelial carcinoma (MC) is rare disease that comprises of only about 2 % of all salivary gland carcinomas and MC that focally or predominantly displays clear cell-type tumor cells are considered as CCMC. We recently got to treat a rare case of a base tongue clear cell myoepithelial carcinoma. (CCMC) Our patient, to the best of our knowledge is the second case of base tongue CCMC and the first with metastatic involvement of the cervical lymph nodes. We successfully managed the tumor using the “mandible preserving pull-through oropharyngectomy approach” Knowledge of the different surgical approaches and techniques is thus vital for better oncologic, functional and aesthetic outcomes following surgery for tumors especially in challenging sub sites like the oropharynx.

Keywords Mandible preserving pull-through oropharyngectomy approach · Minor salivary gland tumors · Myoepithelial carcinoma · Clear cell myoepithelial carcinoma

Introduction

Myoepithelial carcinoma (MC) is rare disease that comprises of only about 2 % of all salivary gland carcinomas and MC that focally or predominantly displays clear cell-type tumor cells are considered as CCMC [1]. Minor Salivary gland involvement of CCMCs is extremely rare [2–7]. To date, only about 18 cases of CCMC have been reported worldwide. Among them, only four cases developed in the minor salivary gland of the oral cavity, only one of which developed in the

base of the tongue. Our patient is possibly the second case of base of tongue CCMC and the first with metastatic involvement of the cervical lymph nodes.

Case Report

A 56-year-old lady, with comorbidities of diabetes and hypertension, controlled on medications presented to us with a complaint of a foreign body sensation in her throat and a husky voice for 3 months. Her past and family histories were unremarkable. Clinical examination aided by a fiberoptic laryngoscopy revealed a large 5×4 cm well circumscribed polypoidal growth arising from the left side of the base tongue, which was displacing the epiglottis posteriorly and was causing a mechanical narrowing the laryngeal inlet.

Examination of the neck revealed a significant matted 2.5 cm ipsilateral left level II cervical lymph node. A punch biopsy of the lesion showed only mildly dysplastic squamous epithelium with subepithelial haemorrhage, however FNAC of the left level II nodes was suggestive of salivary gland neoplasm. A PET CT scan done for staging revealed an irregular 5×4×3 cm enhancing heterodense mass with necrotic area in the left side of the base tongue with uptake in the left level II cervical lymph nodal region. (SUV 2.7) (Fig. 1)

The patient was planned for a definitive surgery using the “mandible preserving pull-through oropharyngectomy approach.” The left neck was accessed by the horizontal limb of the conventional Crile’s incision and a left modified radical neck dissection was performed preserving the spinal accessory nerve. A lingual floor of the mouth incision was subsequently made from the left tonsillar pillar to just across the midline. The extrinsic tongue muscles on the left side were then cut to release the tongue after protecting the hypoglossal nerve. The lingual nerve and sublingual salivary gland were kept on the

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Fig. 1 A PET CT scan showing an irregular enhancing heterodense mass with necrotic area in the left side of the base tongue with the nodes along the left level II

mandible along with a cuff of tissue to facilitate closure. This facilitated the dropping the tongue, thus ensuring a complete visualization of the polypoidal tumor which was arising from the base tongue. The tumor was then excised with clear margins as confirmed by a frozen section (Fig. 2ab). A primary closure of the floor mouth was achieved, which was reinforced with a levator scapulae muscle and a platysma flap. A temporary tracheostomy was fashioned to ensure airway protection. The patient made an uneventful recovery and was decannulated and discharged on the 8th post-operative day. The patient was started on an intensive speech and swallowing rehabilitation program and regained her normal swallowing and speech function.

The gross specimen revealed a grey white firm $5 \times 4 \times 2$ cm polypoidal tumor arising from the at base of tongue. Microscopy revealed a tumor composed of cells that were seen in sheets, alveolar masses and trabeculae separated by hyalinized fibrous septae. Further the cells were large round, oval to polyhedral with moderate to abundant clear cytoplasm, small round to oval, bland nuclei, some of which

were eccentrically placed. The final histo-pathology with immunohistochemistry correlation [Immunopositive for CK7, HMW Keratin, Epithelial membrane antigen (EMA), Keratin, S-100 p and negative for Muscle Actin, SMA and GFAP] was suggestive of clear cell myoepithelial carcinoma resected with clear margins (Figs. 3 and 4) 2 left level II nodes of the 16 lymph nodes dissected showed metastatic carcinomatous deposits. The patient was considered and received 60 Gray of postoperative external beam conformal radiotherapy and presently disease free and is on follow up for the past 14 months (Fig. 5).

Discussion

Although myoepithelial differentiation is common among salivary gland tumors, cases in which the tumors themselves are primarily formed by myoepithelial cells are very rare. Furthermore, among salivary gland myoepithelial carcinomas, CCMC are extremely rare.

Fig. 2 a Intra operative photograph showing the complete tumor visualization which was excised with clear margins

Fig. 3 H&E $\times 20$: Microscopy revealed a tumor composed of acini, strands and tiny nests. The cells were small, round with clear cytoplasm and hyperchromatic nuclei suggestive of clear cell myoepithelial carcinoma

A diagnosis of CCMC is a challenge and this entity needs be differentiated from the other salivary gland tumors that contain clear cell components and from odontogenic and metastatic tumors, which include clear cell oncocytomas, acinic cell carcinoma, high-grade mucoepidermoid carcinomas, sebaceous carcinomas and metastatic clear cell renal cell carcinomas.

The treatment of CCMC is extrapolated from the treatment of myoepithelial carcinomas, which generally includes a wide surgical excision of the primary tumor site and a lymph node dissection only in the presence of metastatic involvement [9]. The challenge in managing our patient of CCMC was largely due to its location in the oropharynx, which is generally

considered to be a relatively inaccessible region, and many different open and minimally invasive surgical approaches have been described, each with their own advantages and disadvantages [8–10].

Oropharyngeal tumors can be accessed through many different surgical approaches, which can be broadly divided into the transoral, transpharyngeal, transmandibular and the combined transoral- transcervical, approaches. The choice of approach is largely dictated by the size and location of the tumor and if a concomitant neck dissection is being planned. The tumor in our patient was surgically accessed and resected using a mandible preserving pull-through oropharyngectomy approach which is essentially a modification of the **combined**

Fig. 4 Tumor cells showing immunopositivity to high molecular weight keratin (HMW Keratin), S-100p, CK7 and Epithelial membrane antigen (EMA)

Fig. 5 Postoperative clinical photograph of the patient depicting an excellent functional and aesthetic outcome

transcervical transoral approach. The potential advantages of this approach include a reasonably good exposure, a reliable postoperative function and an excellent cosmetic result, largely all of which was achieved in our patient (Fig. 4). Oncologically safe resections of the base tongue tumors can thus be performed by avoiding the potential morbidities associated with a mandibulotomy, a lip-split, a tongue split or the classical pull through approach. The potential disadvantages include loss of sensation over the floor of mouth and tongue and the possible need for additional approaches to improve visualization.

Post-surgical radiotherapy has been selectively used in CCMC, especially in cases of adjacent structure involvement and in cases of margin positivity. Adjuvant radiotherapy was considered in our patient in view of the cervical lymph node positivity [3]. Chemotherapy has been used as a form of systemic therapy in the management of metastatic disease. The prognosis of CCMC remains unclear due to the small number of reported cases. However, a few studies on CCMC report a 50 % metastatic rate suggesting an aggressive clinical behavior pattern [7].

In conclusion, this report highlights two issues i.e., the unusual occurrence of a minor salivary gland CCMC and the use of the mandible preserving pull-through oropharyngectomy approach to access and excise the base tongue tumor with an excellent form and function. Knowledge of the different surgical approaches and

techniques is thus vital for better oncologic, functional and aesthetic outcomes following surgery for tumors especially in challenging sub sites like the oropharynx.

Conflict of Interest None declared

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