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REVIEW ARTICLE

Acinic Cell Carcinoma of the Minor Salivary Glands of the Tongue: A Case Study with Review of Literature

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ABSTRACT

Background: Acinic cell carcinoma (ACC) is a relatively rare and slowly growing tumor of the major salivary glands, usually arising from the parotid and rarely from the submandibular and sublingual salivary glands. ACC accounts for 3 to 4% of parotid tumors, 2 to 6% of all salivary gland tumors and 10 to 17% of all malignant salivary gland tumors.

Case report: A 40-year-old woman reported a slow growing mass in the oral cavity. The transoral aspiration biopsy was reported as pleomorphic adenoma arising from the minor salivary glands of the junctional zone of base of tongue. Rest of the neck examination was normal and magnetic resonance imaging (MRI) revealed the mass confined to the superficial layers of the tongue without invasion.

Intervention: The tumor was resected transorally with adequate margins. The tumor histology was typical of ACC with the margins free and wide on all the sides. The patient was kept on monthly follow-up where palpable nodes developed during the course of 6 months. The neck was addressed with supraomohyoid dissection and followed up without irradiation.

Conclusion: ACC is a low-grade malignant salivary neoplasm rarely diagnosed in minor salivary glands of the oral cavity. The overall prognosis after surgical resection depends on the extent of lesion and the adequacy of the initial resection. Hence, keeping in mind the malignant potential of the disease careful long-term follow-up is advised.

Keywords: Acinic cell carcinoma, Minor salivary gland, Wide excision, Neck dissection, Radiotherapy.

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INTRODUCTION

Acinic cell carcinoma (ACC) is a relatively rare and slowly growing tumor of the major salivary glands, usually arising from the parotid and rarely from the submandibular and sublingual salivary glands.[1,2] ACC accounts for 3 to 4% of parotid tumors, 2 to 6% of all salivary gland tumors and 10 to 17% of all malignant salivary gland tumors.[3,4]

The incidence of malignant tumors arising from the minor salivary glands is around 2 to 3% of all malignant neoplasms of upper aerodigestive tract.[5] In minor salivary glands palate is most common site, buccal mucosa, lips, tongue, retromolar trigone and paranasal sinuses are rarely involved.[5]

ACC of the minor salivary glands usually develops beneath the epithelium, the most common presenting symptom is a slow growing mass or swelling under a normal covering mucosa.[1] These tumors are usually single, soft masses that, grossly, may be round or lobulated and frequently appear encapsulated and has postulated that these tumors are less aggressive than that originating in the parotid gland.[6] The origin of these tumors from minor salivary glands is rare, and tumor arising from the junctional tongue zone, i.e. between the junction of the anterior two-third and posterior one-third is not reported in literature.[5]

We report a case of ACC of the junctional part of the posterior tongue and our approach to its management.

CASE REPORT

A 40-year-old female patient presented to us with a history of a mass in the oral cavity of 6 months duration, insidious in onset and progressive in nature.

History of change in voice and mild discomfort for swallowing was also noted. On examination of the oral cavity and oropharynx, the patient had a firm to hard exophytic tumor with a broad peduncle measuring 4 × 4 cm arising from the junctional tongue; tongue mobility was normal (Fig. 1). The rest of head and neck examination was normal.

A preoperative FNAC of the tongue mass was suggestive of pleomorphic adenoma. The tongue movements were

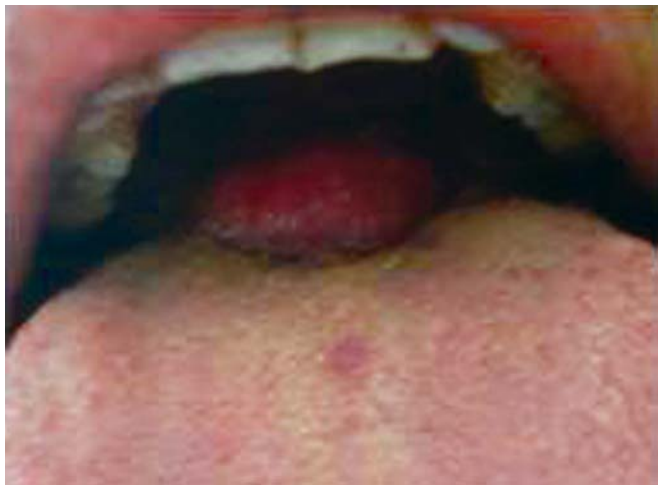

Fig.1: Clinical appearance of tumor in the junctional tongue

normal and the mass was felt to move relative to the protrusion of the tongue. Contrast magnetic resonance imaging (MRI) of the head and neck revealed the mass to be 4 × 4 cm without invasion of the deeper layers confined to the base of the tongue.

Because of good mouth opening and excellent exposure of the tumor by traction of anterior two-third of the tongue we could successfully resect the whole tumor with clear margins by transoral approach. A clear plane was identified between the tumor and the normal tongue tissue which offered no difficulty in dissecting the tumor from the tongue bed. The surgical defect of the tongue was closed primarily, the postoperative period was uneventful. Tongue mobility, swallowing and speech were satisfactory.

The postoperative histopathological report revealed ACC. On microscopy the tumor cells were arranged in sheets with a microcystic pattern (Figs 2 and 3). The individual cells had predominantly clear cytoplasm which was PAS-positive and diastase resistant (Fig. 4). Nuclei were small, vesicular and uniform. There was no necrosis or perineural invasion.

Immunohistochemical studies showed positivity for cytokeratin (CK) and epithelial membrane antigen (EMA) in tumor cells (Fig. 5). Smooth muscle actin and S100 were negative ruling out myoepithelial origin of cells. Because of the satisfactory excision of the tumor substantiated by margin free resection by pathology, postoperative adjuvant radiotherapy was not advised and the patient was advised regular follow-up.

After 6 weeks on follow-up the patient presented with a left level II lymph node 1 × 2 cm, firm in consistency. Rest of the neck and primary was normal; aspiration biopsy of the node was suggestive of metastatic carcinoma of salivary gland origin.

Because of the midline location of primary tumor bilateral supraomohyoid neck dissection was performed.

Fig. 2: Tumor seen beneath lining epithelium of tongue, also seen is a circumvallate papillae (H&E; ×100)

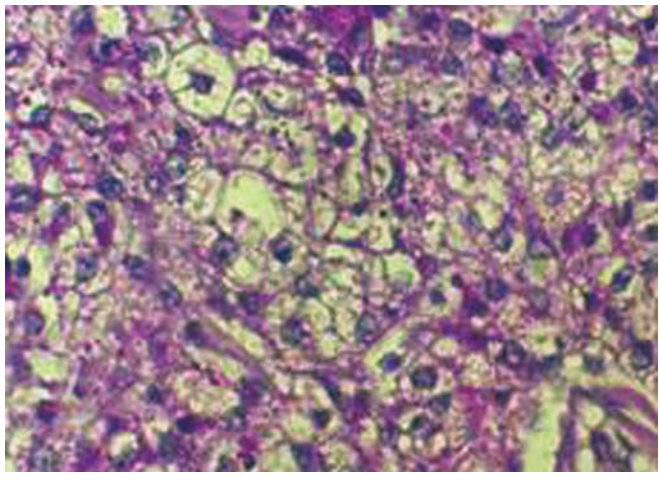

Fig. 4: PAS positive cytoplasmic granules resistant to diastase digestion (PAS; ×200)

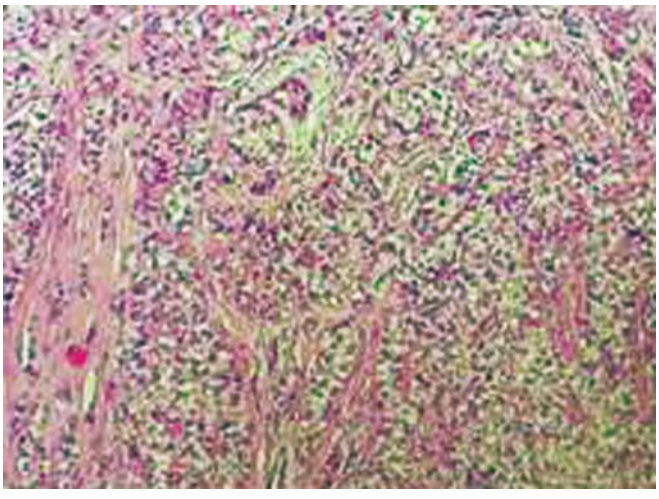

Fig. 3: Neoplastic cells having mildly pleomorphic nuclei and abundant optically clear cytoplasm with distinct cell borders arranged in sheets, nests and microcystic pattern (H&E; ×200)

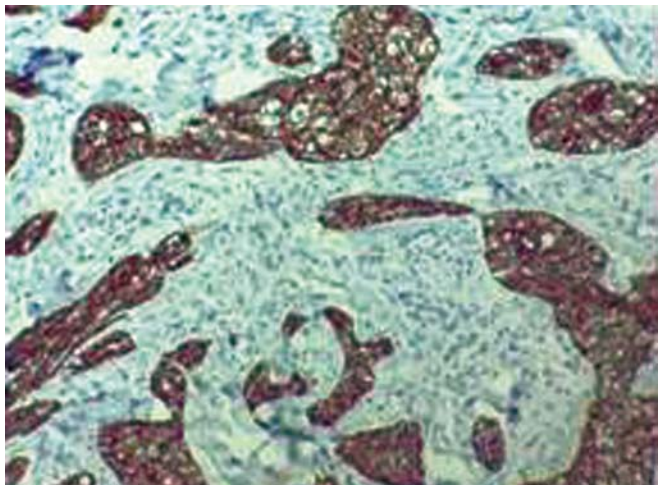

Fig. 5: Tumor cells expressing cytokeratin (cytokeratin AE1/AE3; ×100)

The histopathological examination showed metastatic carcinoma of level I, IIa and IIb lymph nodes on the left side and reactive changes of all lymph nodes on the right side (Fig. 6). Because of the presence of multiple positive neck metastasis particularly on left side, postoperative adjuvant radiotherapy to the primary as well as neck was given. Patient is on regular follow-up with disease free survival of 3 years.

DISCUSSION

Acinic cell tumor of the salivary gland has been first described by Nasse in 1892 as a benign tumor.[7] Later in 1950s it was reported as malignant, originating from reserve cells of salivary gland ducti and besides local recurrence, pulmonary and bone metastasis was observed.[8] The incidences are more seen in the fourth and fifth decades and in females reports in other age groups are also reported.[3,4] In the oral cavity it presents as a nontender swelling is recognized incidentally or during dental examination.[9] They may present as ulcerated mucosal lesions of various sizes, and sometimes appear as solid, microcystic, follicular and papillary-cystic patterns.[4] The biologic behavior of tumor is not influenced by the histopathological appearance, size of tumor, degree of differentiation and infiltrative margins.[4]

Batsakis et al studied that ACC's are derived from the intercalated ducts of the salivary duct and tubular system.[10] Rare metastasis to the cervical lymph nodes and hematogenously spread to the lungs or bones are reported in 7 to 29% of cases.[8] Local tumor excision transorally by conventional method or by LASER is the treatment of choice for early lesions.[2] No protocols are reported in literature about the multimodality treatment of these tumors are reported like the need for lymph node dissection and the need for postoperative neck dissection.[11] The need for postoperative radiotherapy are validated in clinically aggressive tumors and found not essential in small tumors in which resection margins are free of tumor.[12,13]

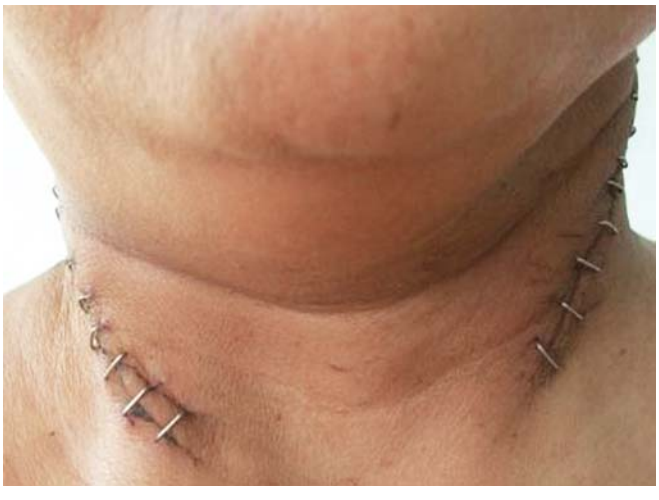

Fig. 6: Postoperative photo showing bilateral supraomohyoid sutured wound

Hiratsuka et al reviewed 84 cases of intraoral ACC of the minor salivary glands and found more in the superior regions like the palate and lesser in the inferior anatomic regions like the retromolar trigone and the floor of mouth.[14] They did subsite stratification analysis and reported lesion in the palate in 28 cases, buccal mucosa in 20, lip in 17, tongue in eight, retromolar trigone in seven and floor of the mouth and gingival in four cases.[14]

Callender et al reported better disease free survival with adjuvant radiotherapy in high-grade tumors with close or invaded margins, perineural invasion and multiple-positive lymph nodes.[13] Fermont et al reported a 5 cm ACC involving the inferior pole of tonsil, posterior third of the tongue and the midline treated with radical radiotherapy alone.[15]

Ferlito et al reported three cases of ACC of minor salivary glands located in larynx, base of the tongue and right tonsil.[16] They defined the tumor of low-grade malignancy, which is rare and may recur after surgery with occasionally metastasis.[16] Anavi et al reported three cases of ACC of minor salivary glands of the oral cavity which were slow growing and asymptomatic.[17] Wide local excision was done in all the three cases with no radiotherapy.[17] Recurrence in one case was seen with other two having a good disease free survival.[17]

Omlie et al in their retrospective study on 21 cases of ACC of the minor salivary glands found that seven of them were located in the buccal mucosa, six in the palate, five in the upper lip, three in the retromolar trigone and the lower lip.[18] The tumor size varied from 0.6 to 1.6 cm and all were treated with wide local excision.[18] Recurrence or metastasis was seen in 10 patients.[18] They concluded it to be an indolent tumor, with occasional synchronous or metachronous association of malignant salivary gland tumors with other malignancies.[18]

Eneroth et al presented 10 cases of ACC of the parotid gland treated with surgery and radiotherapy.[19] They operated the parotid tumors in a salvage setting, 6 weeks after being irradiated with 2800 to 4200 rad.[19] They advised neck dissection bilaterally on a routine basis, while others disagree as adequately excised primary rarely metastasize to cervical lymph nodes.[20] They reported local recurrence of 8 to 59% and stressed the importance of wide local excision.[20] They found a regional lymph node metastasis in 5 to 16% and distant lung and bones in 15% with 5-year survival of 90%.[20]

Tanamoto et al reported an incidence of ACC in sublingual tumors of 4.5%.[21] Timon et al studied that large

tumors (e.g. 2.92 cm) carry a worse prognosis than do small tumors (e.g. 1.93 cm).[22] Holgar et al found lymphocytic reaction in 10% of fine needle aspirations of ACC leading to ambiguity in diagnosis.[23] Batsakis and others reported the ACC to be radiosensitive and found it to be useful in locally advanced tumors, late-stage disease and residual lesions.[10,24] They reported different overall survival with surgery and radical radiotherapy in parotid tumors.[10,24] Here a recurrence rate of 8 to 56% and lymph node and distant metastases were seen in 0 to 16%.[10,24] They reported ACC of the parotid that can be treated with superficial or total parotidectomy with sparing of the facial nerve.[10,24]

Tumors at the tongue base, floor of the mouth, retromolar trigone can be overlooked and are detected at advanced stages.[2] Management of these cases include histopathologic confirmation and evaluation of the performance status of the patient, size of the tumor, site of location and accessibility of the tumor.[2]

However, in our case which surprisingly presented with cervical lymph node metastasis within 6 weeks of follow-up for which she underwent neck dissection followed by postoperative radiotherapy. This shows the importance of concurrent management of both primary and neck in cases of ACC arising from minor salivary glands for a better disease control and management.

CONCLUSION

ACC is a low-grade malignant salivary neoplasm rarely diagnosed in minor salivary glands of the oral cavity. The overall prognosis after surgical resection depends on the extent of lesion and the adequacy of the initial resection. No evidence-based protocol suggests simultaneous neck dissection, adjuvant radiotherapy and chemotherapy. Hence, keeping in mind the malignant potential of the disease careful long-term follow-up is advised.

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