CASE REPORT

Squamous cell carcinoma of submandibular salivary gland: A rare case report

Vardendra Manvikar, Surekha Ramulu, Shilpa T Ravishanker, Chitra Chakravarthy
Department of Oral and Maxillofacial Pathology, 1Oral Medicine and Radiology and 2Oral and Maxillofacial Surgery, Navodaya Dental College, Raichur, Karnataka, India

Address for correspondence:
Dr. Vardendra Manvikar,
Senior Lecturer,
Department of Oral and Maxillofacial Pathology,
Navodaya Dental College, Raichur - 584 101, Karnataka, India.
E-mail: vsmanvikar79@yahoo.

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ABSTRACT
Malignant tumors of submandibular salivary gland are rare in occurrence. Squamous cell carcinoma of salivary glands accounts for about 0.9-4.7% of all salivary gland tumors with a predilection to occur in parotid gland due to perinodal involvement. Primary squamous cell carcinoma of submandibular salivary gland accounts to about 2% of the tumors and hence it is being represented for its rarity.

Key words: Lymphoepithelial carcinoma, mucoepidermoid carcinoma, squamous cell carcinoma

INTRODUCTION
Malignant salivary gland tumors are rather too infrequently observed. Mucoepidermoid carcinoma is the most common malignant salivary gland tumor. The squamous cell carcinoma of submandibular salivary gland is a malignant epithelial tumor of major salivary glands composed of epidermoid cells. There is no history or current evidence of a similar primary tumor elsewhere in the head and neck region. The terms primary squamous and primary epidermoid cell carcinoma are used frequently.

CASE REPORT
A 70-year-old female patient reported to the department of oral medicine and radiology with the chief complaint of swelling in the submandibular region since one and half months [Figure 8]. Similar swelling was present two years back, which regressed after taking medication. The swelling had been gradually increasing in size and not associated with constitutional symptoms nor related to food. A solitary spherical swelling of size 3 × 3 cm was tender, non-mobile and firm. No other swellings were palpable in the neck. The oropharyngeal cavity was normal. General examination was normal. No systemic diseases were detected. The provisional diagnosis was tubercular lymphadenitis. Routine blood tests were within the normal limits and fine needle aspiration cytology (FNAC) of the swelling was inconclusive. Wide surgical excision was done and biopsy was taken and sent to oral pathology department for further diagnosis. There was a diagnostic dilemma between lymphoepithelial carcinoma and mucoepidermoid carcinoma.

A salivary gland along with 3 lymph nodes and skin [Figure 1] were sent for histopathological examination. On grossing, there was thinning and color change in the specimen indicating the involvement of skin. Cross-section of lymph nodes showed possible involvement. Four bits of soft tissue, larger bit measuring 11.5 × 7.5 × 2.8 cm and three small bits measuring 0.8 × 0.6 cm, 1.1 × 1.3 cm, 2 × 1.6 cm, soft to firm in consistency, grayish-brown in color, irregular in shape and surface were taken for processing.

Histopathology features
The histopathology report reveals skin with subcutis and salivary gland tissue. An infiltrating tumor was seen, composed of cells arranged in nests, solid sheets and focally in a glandular pattern. The cells were round to polygonal with pleomorphic vesicular nuclei and eosinophilic cytoplasm. Foci of keratinization was seen [Figure 2]. Few mitotic figures and many giant cells were also seen [Figure 3]. A few questionable lymphovascular emboli were seen. No peri-neural invasion was seen in these sections. The overlying skin was not involved by tumor. A dense mixed inflammatory cell infiltrate and focal necrosis was noted. No mucous cells were seen in these sections. The special stains like periodic acid-Schiff (PAS), mucicarmine, alcian blue revealed absence of any type of mucin. Immunohistochemistry (IHC) was done and it showed positivity for cytokeratin (CK, 90%) (Figures 4,5)
and p63 (80%) (Figures 6, 7). Epithelial membrane antigen (EMA) and S-100 were negative. The immune profile favored a squamous cell carcinoma and was diagnosed as moderately differentiated squamous cell carcinoma arising from submandibular salivary gland. Neck nodes were free of...
metastasis. The patient reported for regular follow-up with no significant complaints.

DISCUSSION

The reported frequency of primary squamous cell carcinoma among all major salivary gland tumors varies from 0.9-4.7%.\[1\] They have been reported to represent 9% of the malignant parotid tumors and 2% of the malignant submandibular tumors. Past exposure of radiation therapy appears to increase the risk of developing this tumor. Affected patients often have been previously irradiated for acne, benign or malignant neoplasms, enlarged thymus, thyroid gland or tonsils. The median time between irradiation and diagnosis of salivary carcinoma is about 15.5 years, with a range of 7 to 32 years. The average age of patients with primary squamous cell carcinoma is between 61 to 68 years, with a strong male predilection.

When squamous cell carcinoma is identified in the submandibular salivary gland, an effort must be made to identify the etiology. When no other primary lesion exists, it seems logical to consider the cancer a primary tumor in the submandibular salivary gland. Primary squamous cell carcinoma of the submandibular salivary gland is unusual.\[2\] When the submandibular salivary gland is involved, other lymph nodes in the neck at risk should be removed. The incidence of distant metastasis is increased in patients with metastasis to the neck compared with those with submandibular salivary gland squamous cell carcinoma alone.

The clinical outcome was similar between patients with metastatic squamous cell carcinoma and primary squamous cell carcinoma of submandibular salivary gland, regardless of treatment plan.\[3\]

Differential diagnosis for the present case considered were mucoepidermoid carcinoma and lymphoepithelial carcinoma.

Mucoepidermoid carcinoma was excluded because PAS and alcian blue were negative. Immunohistochemical expression of Epstein Barr virus was negative in the present case. Thus lymphoepithelial carcinoma was ruled out.

The submandibular tumors are usually painful and duration of the symptoms is less than one year. Fixation to the skin and deeper tissues occur in some cases.\[4\] Treatment includes resection of the involved submandibular salivary gland with concurrent neck dissection, if cervical metastases are detected or suspected. Follow-up information in literature showed regional recurrence occurring in 66% of cases within one year. There appears to be some evidence that post-operative radiotherapy may help to control local recurrence.\[5\] Therefore, when squamous cell carcinoma is identified in the submandibular gland, one must consider all possibilities so that the proper treatment plan for a particular setting is selected. After therapy, all patients require lifelong follow-up.\[6\]

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