INTRODUCTION

Basaloid squamous cell carcinoma (BSCC) is an uncommon variant of squamous cell carcinoma. It was first described by Wain et al in 1986.[1] BSCC is included as a distinct entity in the revised classification of tumors of head and neck by WHO in 1991.[2] Generally, it has a predilection for head and neck region, particularly the upper aerodigestive tract, i.e. larynx-hypo pharynx.[3] In the oral cavity, BSCC has a predilection for the tongue, though it has been described in other locations such as floor of the mouth, palate, retromolar trigone and gingival mucosa.[4]

The aggressive biological behavior of BSCC has been commonly associated with early recurrence, cervical lymph node involvement and distant metastasis with spread to the lungs and liver.[5] Most BSCC’s are diagnosed at advanced clinical stages and have an unfavorable prognosis because of poor overall patient survival rate.

Clinically, patients with BSCC present features similar to those of the patients with squamous cell carcinoma and have the same etiological risk factors, e.g. tobacco and alcohol consumption.[6] The recommended treatment for BSCC is surgery followed by radiotherapy and chemotherapy.[7]

CASE REPORT

A 65 year - old male patient reported with a chief complaint of pain in right lower back tooth region of 6 months duration. The patient gave a history of difficulty in mouth opening and swallowing. He had a history of beedi smoking for a period of 50 years, with a frequency of 12/day.

Extra oral clinical examination revealed palpable, mobile submandibular and upper jugular lymph nodes on the right side, measuring approximately 1×1 cm and they were firm and non-tender to palpation. On intra-oral examination, an ulceroproliferative lesion involving the right side of the retromolar trigone region, measuring about 2×2 cm. was seen [Figure 1]. The ulcer was tender on palpation, exhibiting irregular margins with ill-defined borders and white slough surrounded by erythematous area. Oral hygiene was poor with generalized stains and calculus. Generalized attrition was present with right posterior teeth tender on percussion. No limitation of mouth opening was seen. Tongue movements were not affected.

A provisional clinical diagnosis of malignant ulcer was given. After obtaining written consent from the patient, an incisional biopsy was performed under local anesthesia and sent for histopathologic examination. An orthopantomogram [Figure 2] was taken which showed irregular radiolucency distal to 3rd molar with erosion of the ascending border of the ramus of the mandible, measuring 2 × 2 cm. An ultrasonogram of neck was done. The report showed metastatic changes in submandibular and level II group of lymph nodes. The histopathologic report [Figure 3] was poorly differentiated squamous cell carcinoma. The lesion was T4 N1 Mx.
The treatment planned was hemimandibulectomy with supraomohyoid neck dissection under general anesthesia, followed by radiotherapy. An apron incision extending from the midline of the chin along the second crease of the neck extending to the mastoid process was made. Anteriorly, the incision was continued around the chin to split the lower lip in the midline. Dissection was done in the subplatysmal layer and supraomohyoid neck dissection was done. The nodes in the level I and level II regions of the neck were surgically removed. The submandibular salivary gland was excised [Figure 4]. Hemimandibulectomy was done. Hemostasis was achieved. Wound closure was done. Postoperative recovery of the patient was uneventful. The histopathologic picture of the excised specimen was reported as BSCC. The histopathologic report was given by three oral pathologists from different centers.

The patient has undergone radiotherapy with 5000 cGy fractionated over 6 weeks. Barium meal test, chest X-ray and endoscopic evaluation of the upper aerodigestive tract were done, which showed no evidence of lesion or metastasis. The patient has been reviewed regularly for the last 3 months and is disease free.

BSCC has been defined in the 2005 WHO blue book as an aggressive high grade variant of squamous cell carcinoma of both basaloid and squamous components. [8] Wain et al.[13] first introduced this histopathological entity and called it as BSCC in 1986. Till date approximately 45 cases of BSCC involving the oral cavity have been reported in literature, with a strong predilection for base of the tongue (61%) and floor of the mouth (30%). [9]

The histogenesis of this neoplasm is controversial. It is suggested that the tumor originates from the totipotential cells in the basal layer of squamous epithelium. When we review the etiology, it is seen that the possible relationship of BSCC and viruses is a matter of debate and has been reported in some locations like nasopharynx and penis. [10,11]. The obtained data are controversial, while Kleist et al. [12] and El mofty et al. [13] have very recently detected a high frequency of HPV and HSV in basaloid tumors than in conventional squamous cell carcinoma in the head and neck, others have found no difference.

The various sites of origin of BSCC reported by authors were gingiva (Eiji Hirai et al.[14]), oral mucosa and maxillary tuberosity (C. Wedenberg et al.[15]), floor of the mouth (Kunal Sah et al.[16]), retromolar trigone (Marcia Sampaio Campose et al.[17]), nasal cavity (Joong Seob Lee et al.[18]), hypopharynx with extensive spindle cell component (Tokuhiro kimura et al.[19]), conjunctiva and paranasal sinus (Pooja Vasudev[20]), urinary bladder (Funda Vakar –Lopez et al.[21]) and uterine cervix (Yong Soon Kwon[22]) [Table 1].

Domenico Coppola et al. [23] studied the clinicopathological and immunohistochemical features of eight BSCC of floor of the mouth which reveals high recurrence, worse prognosis, metastasis, mortality and shorter survival than squamous cell carcinoma. Grossly, most of the previously reported BSCCs are flat or slightly elevated tumors, often with a central ulceration similar to our case. Very few cases show a polypoid pattern. Interestingly these cases are always associated with a spindle cell component. [18,19] Jung Yeon Kin et al.[24] reported that all BSCC showed positivity for high molecular weight cytokeratin (HMW CK) with heterogenous or diffuse staining pattern, but lacked activity for neuroendocrine markers and bcl-2 oncoprotein. Pinar Atasoy et al. [25] reported a case of BSCC of lungs whose cells showed a high mitotic rate and peripheral palisading. The immunohistological examination for neuroendocrine markers was negative. Tie–Jun Li et al. [26] studied BSCC of esophagus without adenoid cystic features. Grazia Salerno et al. [27] reported that low levels of p27Kip 1 expression significantly correlated with poor prognosis, biological aggressiveness and consequent shortened survival. The supposed higher clinical aggressiveness of BSCC compared with the conventional Squamous cell carcinoma remains a continuous matter of debate. Banks et al., Luna et al and De sampaio et al. [28] did not find significant differences in behaviour between these two neoplasms in different anatomical sites while others did. Cosmo E et al. [29] discussed the clinicopathological and follow-up study of 40 cases and review of literature and concluded that solid nest with typical cell population, basaloid at the periphery [Figure 5] and squamous at the centre, are the most common growth patterns of BSCC, which was the histopathologic picture seen in our case.

**DISCUSSION**

BSCC is a rare and aggressive variant of squamous cell carcinoma which is reported to occur predominantly in men between 60 to 70 years of age. [1] It is reported in individuals with history of tobacco and alcohol abuse. The most frequent site to be affected by BSCC is the upper aerodigestive tract with strong predilection for the base of the tongue, supraglottic larynx and hypopharynx, but it is also found in the anus, thymus and uterine cervix. Clinically, it is an aggressive tumor with high rates of nodal (64%) and distant metastases (44%). Results of a case control study by Soriano et al found a 6 times higher risk of distant metastases compared to the usual type of squamous cell carcinoma. Some authors recommend a chest CT and FDG- PET in all cases to rule out early distant metastases. Treatment of choice is complete surgical excision of the lesion with neck dissection supplemented by radiotherapy or adjuvant chemotherapy.

Our case report was a BSCC in the retromolar trigone which is an unusual site of occurrence. Marcia sampaio campos et al.[7] has also reported an atypical presentation of oral BSCC in the retromolar trigone in a 39 year old man, in which he described the immunohistochemical characteristics of the lesion. Due to its biological and morphologic features, it may be confused with adenoid cystic carcinoma of the solid subtype, small cell neuroendocrine carcinoma undifferentiated carcinoma,
basal cell adenocarcinoma and squamous or adenosquamous carcinoma. In the large majority of cases, the distinction between these tumors is readily made on the basis of standard H&E morphology. However, immunohistochemical markers have been reported to be useful in differentiating these tumors.

Our case mimics squamous cell carcinoma clinically and was reported on incisional biopsy as poorly differentiated squamous cell carcinoma. On examining the deeper sections of the excisional biopsy, the histologic picture showed nests and cords of closely packed pleomorphic basaloid cells with nuclear palisading along the periphery of the neoplastic nests surrounded by a fibrous stroma with prominent areas of comedo necrosis. Hence the histopathologic report of BSCC was given.

Wain et al.[1] and recently Barnes et al.[30] put down the following criteria to diagnose cases of BSCC.

The features included:
- Predilection for head and neck region in men in their 60s or 70s.
- An ulcerated or exophytic mass with submucosal soft tissue infiltration.
- Solid basaloid appearing dysplastic island with biphasic pattern showing comedo type necrosis [Figure 6] and pseudo-glandular pattern.
• Abrupt foci of squamous differentiation with or without keratin pearls, and surface mucosal epithelium showing dysplastic features.

Quite recently, however Coletta et al. [31] have demonstrated the importance of cytokeratins 1, 7 and 14 in the diagnosis of SCC and have shown significantly higher AgNOR and PCNA positivity in BSCC when compared with squamous cell carcinoma. Immuno staining for p53 also showed a higher percentage of positive cells in BSCC. The glandular carcinomas can be excluded as glandular lesion present immunoreactivity for CK7 unlike BSCC. Expression of MMP-1, MMP-2 and MMP-9 were reported higher in cells of BSCC than in cells of squamous cell carcinoma, suggesting of aggressive behaviour. Emanuel et al. [32] have stressed the value of p63 in making the distinction between BSCC and adenoid cystic carcinoma of head and neck.

Though in a comparative study of oral BSCC and squamous cell carcinoma done by Ferrada C Grizza et al. [4] they concluded that their study supported the opinion that the prognosis of BSSC does not differ from that of conventional squamous cell carcinoma of the oral cavity when matched for clinical classification. [4]

Nodal metastasis is quite common in laryngeal, hypopharyngeal and tracheal BSCC. Regional nodal metastasis was seen in 75% and distant nodal metastasis was seen in 35-50% of the cases in Wizenburg et al. series, lung is reported to be the main target for distant metastasis in BSCC. In our case, the patient showed no clinical or radiographic evidence of lung metastasis. Finding a second primary tumor is a common clinical situation in the head and neck. Cosmo E et al. have observed in 17.5% of their patients, a second primary tumor and therefore support Thompson’s advice for keeping in mind the possibility of finding a second primary tumor in any sites either synchronic or metachronic, when diagnosing a BSCC in the head and neck.

CONCLUSION

BSCC, a distinct clinicopathological entity with aggressive clinical behavior, which usually is reported to occur in the upper aerodigestive tract, has been reported by us occurring

Table 1: Review of the site, clinical and immunohistological features

| Journal / Author          | Study done                  | Pt. volume | Site of the lesion | Outcome                                                                 |
|---------------------------|-----------------------------|------------|--------------------|-------------------------------------------------------------------------|
| Eiji Hirai et al. [14]    | Case report                 | 2          | Gingiva            |                                                                         |
| C. Wedenberg et al. [15]  | Case report                 | 1          | Oral mucosa and maxillary tuberosity |
| Kunal et al. [16]         | Case report                 | 1          | Floor of the mouth |                                                                         |
| Marca Sampaio Campose et al. [17] | Case report | 1          | Retromolar trigone |                                                                         |
| Joong Seob Lee et al. [18] | Case report                 | 1          | Nasal cavity       |                                                                         |
| Tokuhiro kimura et al. [19] | Case report                 | 1          | Hypopharynx        |                                                                         |
| Pooja Vasudev [20]       | Case report                 | 1          | Conjunctiva and paranasal sinus |                                                                         |
| Funda Vakar – Lopez et al. [21] | Case report | 1          | Urinary bladder    |                                                                         |
| Domenico Coppola et al. [22] | Clinicopathological and immunohistological features | 8          | Floor of the mouth | Reveals high recurrence, worse prognosis, metastasis, mortality and shorter than sq. Cell carcinoma positivity for high molecular weight cytokeratin (HMW CK) but lacked activity for neuroendocrine markers and bel-2 oncoprotein high mitotic rate and peripheral palisading and neuroendocrine markers was negative without adenoid cystic features low levels of p27Kip1 expression significantly correlated with poor prognosis, biological aggressiveness and consequent shortened survival |
| Jung Yeon Kin et al. [24] | Immunohistochemical features |           |                    |                                                                         |
| Pinar Atasoy et al. [25] | Histopathological and immunohistological features | 1          | Lung               |                                                                         |
| Tie–Jun Li et al. [26]   | Clinical features           |            | Esophagus          |                                                                         |
| Grazia Salerno et al. [27] | Immunohistochemistry |           |                    |                                                                         |
| Cosmo Ereno et al. [29]  | Clinical and follow up study | 40         |                    |                                                                         |
in the retromolar trigone area which is an uncommon site for an uncommon lesion such as BSCC.

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How to cite this article: Rachel JR, Kumar NS, Jain NK. Basaloid squamous cell carcinoma of retromolar trigone: A case report with review of literature. J Oral Maxillofac Pathol 2011;15:192-6.

Source of Support: Nil. Conflict of Interest: None declared.

Journal of Oral and Maxillofacial Pathology: Vol. 15 Issue 2 May - Aug 2011