

Case Report

Basaloid squamous cell carcinoma of gingiva: A rare case report

Sunitha Jagaluru Doddanna¹, Meghanand T. Nayak², Aparna K. Sanath², Mohammad Zanul Abedeen²

¹Department of Oral Pathology and Microbiology, MNR Dental College and Hospital, Sangareddy, Telangana, ²Department of Oral and Maxillofacial Pathology, Teerthanker Mahaveer Dental College and Research Centre, TMU, Moradabad, Uttar Pradesh, India

Received: 03-Dec-2019

Revised: 17-Feb-2021

Accepted: 19-Apr-2021

Published: 25-Sep-2021

Address for correspondence:
Dr. Sunitha Jagaluru
Doddanna,
Professor & HOD
Department of Oral
Pathology and Microbiology,
MNR Dental College
and Hospital, Fasalwadi,
Sangareddy - 502 294,
Telangana, India.
E-mail: drsunithajd@
gmail.com

ABSTRACT

Basaloid squamous cell carcinoma (BSCC) is an exceptional, aggressive variant of squamous cell carcinoma (SCC) because of its unique histological feature and an ominous clinical behavior. Recently, it has been recognized as a high-grade SCC. The most preferential site of occurrence is the upper aerodigestive tract. Because of its aggressive behavior and tendency to metastasize, BSCCs are considered to have poor prognosis. We present a BSCC case in a 60-year-old male, which was clinically diagnosed as an aggressive inflammatory lesion with a differential diagnosis of granulomatous lesion, involving the mandibular anterior gingiva. Till now, only 17 cases of BSCC with gingival involvement have been reported in the literature. Here, we present one additional case of BSCC involving gingiva.

Key Words: Aggression, basaloid, carcinoma, gingiva, squamous cell

INTRODUCTION

Basaloid squamous cell carcinoma (BSCC) is considered a rare variant of squamous cell carcinoma (SCC).^[1] It was first described by Wain *et al.* in 1986.^[2] In 1991, it was included in the revised edition of the World Health Organization (WHO) classification^[3,4] and was considered as a distinct variant of SCC due to its more aggressive nature and morphological features.^[5] BSCC arises preferentially in the upper aerodigestive tract. Dimorphic pattern is the histological hallmark BSCC of with a characteristic feature of basal cells showing nuclear pleomorphism, hyperchromatic, and indistinct cytoplasm associated with squamous portion. Overlying epithelium showing carcinomatous changes and areas of comedo-necrosis are other common histological features.^[1,6,7] In the oral cavity, the tongue is the most common site of

occurrence. However, it is also found in other sites such as floor of the mouth, palate, retromolar area, buccal mucosa, and the gingiva.^[4-6] BSCC on gingiva is very rare, and only a few reported cases are seen in the literature. This study reports and reviews a rare BSCC case involving the gingiva.

CASE REPORT

A 60-year-old male patient reported to the outpatient department of the authors' institution with the complaint of pain and swelling in the lower front tooth region for 15 days. The patient gave a history of extraction 1 month back due to loosened teeth following which he noticed a soft-tissue overgrowth in his mandibular anterior tooth region. The patient had the habit of bidi smoking for 30 years. Extraoral

This is an open access journal, and articles are distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 4.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as appropriate credit is given and the new creations are licensed under the identical terms.

For reprints contact: WKHLRPMedknow_reprints@wolterskluwer.com

How to cite this article: Doddanna SJ, Nayak MT, Sanath AK, Abedeen MZ. Basaloid squamous cell carcinoma of gingiva: A rare case report. Dent Res J 2021;18:80.

Access this article online



Website: www.drj.ir
www.drjjournal.net
www.ncbi.nlm.nih.gov/pmc/journals/1480

examination did not reveal any significant findings. He also experienced dull pain associated with this overgrowth. The submandibular and submental lymph nodes on the ipsilateral side were palpable.

On inspection, a soft nodular growth measuring about approximately 5 cm × 3 cm × 2 cm was seen on the mandibular anterior region. The lesion was pinkish white in color and ulceroproliferative in nature. The lesion was extending labiolingually from labial attached gingiva to lingual attached gingiva and mesiodistally from mandibular left canine to mandibular right first premolar region [Figure 1]. The growth was tender and firm in consistency. Based on clinical findings, a provisional diagnosis of aggressive inflammatory lesion with a differential diagnosis of granulomatous lesion and carcinoma of gingiva was made. No significant findings were noticed in an orthopantomogram. After routine hematological investigations, an incisional biopsy was performed. The tissue specimen was routinely fixed and processed. The H- and E-stained sections showed dysplastic epithelium invading into the connective tissue stroma. Invaded epithelial cells were in the form of nests, islands, and sheets composed of basaloid cells showing hyperchromatic large nuclei and indistinct cytoplasm [Figures 2 and 3]. Areas of comedonecrosis were seen in between the sheets of cells [Figure 4]. Frequent mitotic figures were also evident. The stroma also showed numerous blood vessels with areas of hemorrhage. Based on the histopathological findings, a diagnosis of BSCC with differential diagnosis of adenoid cystic carcinoma (ACC) (solid variant) was made. To differentiate between the two tumors, periodic acid–Schiff (PAS) stain and immunohistochemical staining for pan-cytokeratin were done. The negative results of PAS and positive for pan-cytokeratin [Figure 5] lead to the final diagnosis of BSCC. The patient did not comply with any further treatment and succumbed to the disease within 1 month.

DISCUSSION

BSCC is a rare variant of SCC with a frequency of >1% of all SCCs.^[4] According to the WHO, it is defined as “an aggressive and distinct variant of SCC, which is composed of basaloid and squamous components.”^[7]

BSCC is a particularly rare tumor of the oral cavity having predilection to occur in the base of the



Figure 1: Photograph showing the ulceroproliferative lesion with indurated margins in the mandibular anterior gingiva.

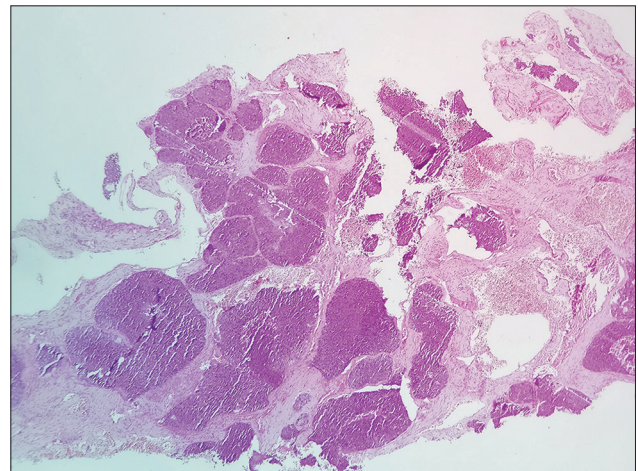


Figure 2: Photomicrograph showing connective tissue stroma showing along with island and sheets made of basaloid cells invaded into connective tissue (H and E stain, ×4).

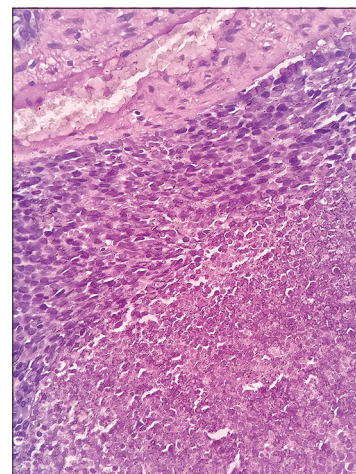


Figure 3: Photomicrograph showing hyperchromatic large nuclei and indistinct cytoplasm (H and E stain, ×40).

tongue (61%) and the floor of the mouth (30%).^[7,8] Its occurrence in the gingiva is very rare.^[7,9] BSCCs

are often diagnosed at advanced clinical stages and their prognosis is unfavorable due to the overall poor patient survival rates.^[4]

The etiopathogenesis of BSCC is similar to conventional SCC. Most of the patients will have a long history of tobacco smoking and consumption of alcohol. In the pathogenesis of BSCC, risk factors such as smokeless tobacco and other exogenous carcinogens such as occupational, environmental, and nutritional factors also play a role.^[7]

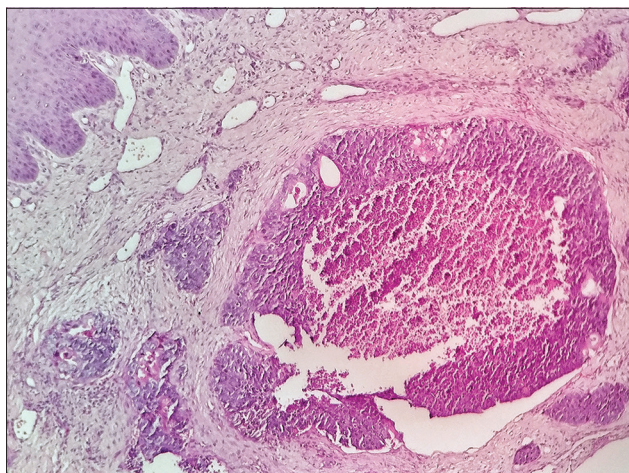


Figure 4: Photomicrograph showing areas of comedonecrosis in between the sheets of cells (H and E stain, x40).

After reviewing, the English literature showed only 17 reported cases of BSCC involving the gingiva. Table 1 discusses and summarizes the clinicopathological features of BSCC cases of gingiva, including the present case. BSCCs of the gingiva have a strong predilection to males ($n = 13$) than females ($n = 4$) with 40–85 years of age range and a mean age of 62.5 years. The present case is consistent with the literature with male predilection. Most frequently, it was seen in the mandibular gingiva ($n = 13$) as compared to the maxillary gingiva ($n = 4$). Five patients were presented in Stage I, three in Stage II, four in Stage III, and two in Stage IV, according to the AJCC standard tumor-node-metastasis staging. Staging was not assessed for the other three cases. Surgery was the treatment of choice for all the cases. Six patients were treated with neck dissections and four were also given adjuvant radiotherapy. Eleven of 17 patients of BSCC had survived till 56 months of median follow-up time.

Wain *et al.* first described the BSCC in 1986,^[2,6] and BSCC was diagnosed on the basis of four principal histologic features: (a) solid groups of cells in a lobular configuration closely apposed to the surface mucosa; (b) small, closely packed cells with scant cytoplasm; (c) dark, hyperchromatic nuclei without

Table 1: Clinicopathological findings of 18 cases of basaloid squamous cell carcinoma that occurred on the gingival

First author/ reference	Year	Age/ gender	Location of lesion	Stage	Treatment	Final outcome	Follow-up period, months
Wedenberg <i>et al.</i> ^[10]	1997	55/male	Oral mucosa and maxillary tuberosity	I	Surgery	Alive	5
Ide <i>et al.</i> ^[11]	1997	63/male	Mandibular gingiva	-	-	Died	-
Abiko <i>et al.</i> ^[12]	1998	79/female	Mandibular gingiva	I	Surgery	Alive	24
Peddapelli ^[7]	2008	61/male	Mandibular gingiva	II	Surgery + FND	Alive	120
Peddapelli ^[7]	2008	56/male	Mandibular gingiva	IV	Surgery	Died	180
Peddapelli ^[7]	2008	65/male	Mandibular gingiva	III	Surgery	Died	2.5
Subramania <i>et al.</i> ^[13]	2009	72/female	Mandibular gingiva	III	Surgery + FND + RT	Alive	12
Hirai <i>et al.</i> ^[14]	2009	55/male	Mandibular gingiva	II	Surgery + FND	Alive	79
Hirai <i>et al.</i> ^[14]	2009	65/male	Mandibular gingiva	I	Surgery + RT	Alive	60
Xie <i>et al.</i> ^[9]	2010	40/male	Maxillary gingiva	III	Surgery + FND + RT	Alive	25
Sun <i>et al.</i> ^[15]	2013	85/male	Mandibular gingiva	IV	Surgery + FND	Alive	4
Patel <i>et al.</i> ^[16]	2013	59/male	Maxillary gingiva	I	Surgery + RT	Alive	36
Peddapelli <i>et al.</i> ^[7]	2013	60/male	Mandibular anterior gingiva	II	Surgery	Alive	24
Patil <i>et al.</i> ^[17]	2014	45/female	Mandibular gingiva	III	-	-	-
Iyoda <i>et al.</i> ^[18]	2019	67/male	Mandibular gingiva	I	Surgery + FND	Alive	44
Dai <i>et al.</i> ^[19]	2020	78/male	Maxillary gingiva	-	Maxillectomy	Alive	12
Protyusha and Sivapathasundharam ^[20]	2021	57/female	Maxillary gingiva	-	-	-	-
Present case	2019	60/male	Mandibular anterior gingiva	-	No treatment	Died	1

FND: Functional neck dissection; RT: Radiotherapy

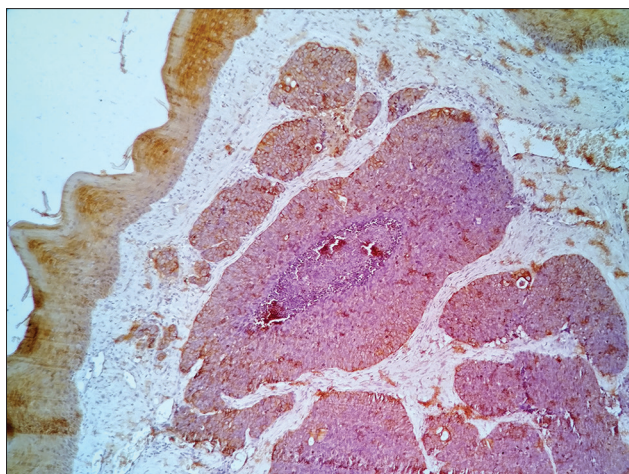


Figure 5: Photomicrograph showing positivity for pan-cytokeratin AE/AE3. (immunohistochemical stain, $\times 10$).

nucleoli; and (d) small, cystic spaces containing mucin-like material.^[6,8] All these features were histologically consistent with the present case.

Clinically, BSCCs are similar to the conventional SCC; therefore, it becomes very difficult to differentiate it from SCC. Therefore, the diagnosis of BSCC is largely dependent on histopathological and immunohistochemical characteristics.^[8] Histopathologically, the BSCC shows that basaloid cells are usually arranged in cords, nests, islands, and lobules. The cells display an increased nuclear/cytoplasmic ratio with scant cytoplasm and oval hyperchromatic nuclei with nucleoli. The cells also show nuclear pleomorphism, and mitotic figures are often observed. The tumor islands exhibit basaloid cells with areas of comedonecrosis and focal keratinization.^[4,9]

BSCC should be histologically differentiated from solid ACC, adenosquamous carcinoma, mucoepidermoid carcinoma, neuroendocrine carcinoma, basal cell and polymorphous low-grade adenocarcinoma, small-cell undifferentiated carcinoma, conventional SCC, basal cell carcinoma, spindle cell carcinoma, and adenoid SCC.^[7]

All these tumors show overlapping histopathological features, and distinction becomes difficult. ACC is one of the minor salivary gland tumors with a palate and retromolar region as their favorite sites. Usually, the solid variant of ACC is difficult to differentiate it from BSCC.^[2] In the present case, the basaloid pattern of ACC was ruled out because of the presence of squamous component and surface dysplastic epithelium which was close to the tumor nests. Certain

features such as mitosis, nuclear pleomorphism, and comedonecrosis are common in BSCC compared with ACC, as observed in our case. The mucin positivity and true ductal-acinar differentiation in adenosquamous carcinoma differentiate it from BSCC.^[21]

By immunohistochemistry, BSCC expresses cytokeratins, epithelial membrane antigen, pan-cytokeratin AE/AE3, and squamous epithelial marker 34 β E12 which are the most useful marker for this tumor.^[7] The present case was immunohistochemically positive for pan-cytokeratin AE/AE3. Immunohistochemical studies have shown that the indices of AgNOR and PCNA (Proliferating Cell Nuclear Antigen) were significantly higher in BSCC than in SCC cases. Immunostaining for p53 protein showed a higher percentage of positive cells and more intense staining in the BSC tissues than in the SCC tissues. In addition, MMP-1, MMP-2, and MMP-9 expression was higher in BSC cells than in SCC cells.^[11]

The clinical course and the prognosis of BSCC have been considered worse than conventional SCC with an increased tendency for local invasion and distance metastasis. It commonly metastasizes to lymph nodes, lung, bone, skin, and brain, through lymphatics and blood vessels. Hence, BSCCs are generally been treated with intensive multimodality therapy. It includes excision by radical surgery, dissection of the neck, and radiotherapy along with chemotherapy. BSCC shows a high (60% die of the disease) mortality rate.^[22] Immunotherapy as adjunct treatment has shown improved treatment with a reduction rate of mortality.^[7,9]

CONCLUSION

BSCC of mandibular gingiva is an unusual location. Here, we add an additional case of BSCC involving gingiva to the literature. BSCC is an uncommon malignant tumor and is a distinct clinicopathological entity of SCC. It has aggressive clinical course, high rate of metastasis, and poor prognosis than conventional SCC. Its diagnosis still depends on hematoxylin and eosin sections by recognizing the defined histological criteria and histopathological evaluation. Hence, awareness of BSCC's clinical and pathological characteristics is thus essential for its early diagnosis and treatment.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/

her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

The authors of this manuscript declare that they have no conflicts of interest, real or perceived, financial or nonfinancial in this article.

REFERENCES

- Sah K, Kale A, Hallikerimath S. Basaloid squamous cell carcinoma involving floor of the mouth. *J Oral Maxillofac Pathol* 2008;12:61-3.
- Jain K, Harshaminder K, Madhushankari GS. Basaloid squamous cell carcinoma in retromolar ridge area: A rare case report. *Int J Oral Maxillofac Pathol.* 2011;23:27–31.
- Satish B, Kumar P. Basaloid squamous cell carcinoma – A case report. *Int J Dent Clinics* 2010;2:31-3.
- Gupta B, Bhattacharyya A, Singh A, Sah K, Gupta V. Basaloid squamous cell carcinoma-A rare and aggressive variant of squamous cell carcinoma: A case report and review of literature. *Natl J Maxillofac Surg* 2018;9:64-8.
- Campos MS, Modolo F, de Oliveira JS, Pinto-Júnior DS, de Sousa SC. Atypical presentation of oral basaloid squamous cell carcinoma. *J Contemp Dent Pract* 2009;10:98-104.
- Koshti S, Bartake A, Palaskar S, Prabhu P, Nagpal D. Basaloid squamous cell carcinoma involving the alveolar ridge, buccal & lingual vestibule-A case report. *J Dent Allied Sci* 2013;2:74-6.
- Peddapelli K, Rao GV, Sravya T, Ravipati S. Basaloid squamous cell carcinoma: Report of two rare cases and review of literature. *J Oral Maxillofac Pathol* 2018;22:285.
- Shivakumar B, Dash B, Sahu A, Nayak B. Basaloid squamous cell carcinoma: A rare case report with review of literature. *J Oral Maxillofac Pathol* 2014;18:291-4.
- Xie S, Bredell M, Yang H, Shen S, Yang H. Basaloid squamous cell carcinoma of the maxillary gingiva: A case report and review of the literature. *Oncol Lett* 2014;8:1287-90.
- Wedenberg C, Jesslén P, Lundqvist G, Lundgren J, Hellquist HB. Basaloid squamous cell carcinoma of the maxilla. *Oral Oncol* 1997;33:141-4.
- Ide F, Shimoyama T, Haga H, Horie N. Basaloid squamous cell carcinoma of the esophagus metastatic to the gingiva: A case report. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1997;83:584-7.
- Abiko Y, Muramatsu T, Tanaka Y, Ohuchi T, Satoh M, Okumura K, *et al.* Basaloid-squamous cell carcinoma of the oral mucosa: report of two cases and study of the proliferative activity. *Pathol Int* 1998;48:460-6.
- Subramanian B, Agrawal K, Panda K. Basaloid squamous carcinoma of mandible. *J Craniofac Surg* 2009;20:151-3.
- Hirai E, Yamamoto K, Yamamoto N, Yamashita Y, Kounoe T, Kondo Y, *et al.* Basaloid squamous cell carcinoma of the mandible: Report of two cases. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2009;108:e54-8.
- Sun Y, Kuyama K, Suemitsu M, Nemoto N, Yamamoto H. Basaloid squamous cell carcinoma of gingiva: A new case report with immunohistochemical study. *Int J Oral Med Sci* 2013;11:236-41.
- Patel PN, Mutalik VS, Rehani S, Radhakrishnan R. Basaloid squamous cell carcinoma of oral cavity with incongruent clinical course. *BMJ Case Rep* 2013;2013:???
- Patil AV, Tupsakhare SD, Shah K, Gabhane M, Ali FM. Basaloid squamous cell carcinoma of gingiva: A new case and review of literature. *Am J Cancer Case Rep* 2014;2:99-107.
- Iyoda, M, Koike, K, Saito, T, *et al.* Basaloid squamous cell carcinoma in the mandibular gingiva. *Oral Sci Int.* 2020; 17: 47– 49.
- Dai LL, Yang XX, Zhao HW, Han Q, Chen Y. Basaloid squamous cell carcinoma in gingiva: A case report. *Hua Xi Kou Qiang Yi Xue Za Zhi* 2020;38:343-6.
- Protyusha GB, Sivapathasundharam B. Basaloid squamous cell carcinoma in the maxillary gingiva. *J Can Res Ther.* [Doi: 10.4103/jert.JCRT_893_19].
- Heera R, Ayswarya T, Padmakumar SK, Ismayil P. Basaloid squamous cell carcinoma of oral cavity: Report of two cases. *J Oral Maxillofac Pathol* 2016;20:545.
- Thompson LD. Squamous cell carcinoma variants of head and neck. *Curr Diagn Pathol* 2003;9:384-96.