

A rare case of basaloid squamous cell carcinoma in the labial commissure region with literature review



ARTICLE INFO

Keywords

Squamous cell carcinoma
Oral cavity
Head and neck neoplasms

ABSTRACT

Basaloid squamous cell carcinoma (BSCC) is a rare variant of oral squamous cell carcinoma (SCC), representing less than 1% of all SCC in the mouth. Despite being similar to the conventional SCC regarding etiological factors and clinical presentation, the clinical course and prognosis of BSCC remain controversial in the literature. Although the microscopic aspects of the BSCC are well defined, its accurate diagnosis might be a challenge for the pathologist, especially regarding the quality and quantity of the analyzed fragment, being extremely relevant to considering possible differential diagnoses. We describe a rare case of an 83-year-old male who presented a rapidly growing lesion in the labial commissure, asymptomatic and extending to the buccal mucosa, resulting in a peculiar clinical presentation. Histopathological examination of incisional biopsy revealed neoplastic epithelial cells with moderate pleomorphism, figures of mitosis, foci of comedonecrosis, invading the submucosa and skeletal striated muscle tissue, confirming BSCC diagnosis. We also present a discussion on the clinical aspects, differential diagnosis, staging, and evolution, and in addition a relevant summary of reported BSCC clinical cases in recent literature.

1. Introduction

Basaloid squamous cell carcinoma (BSCC) was first described by Wain et al. [1] in 1986 and only eleven years later it was included by the WHO as a variant of squamous cell carcinoma [2]. The BSCC represents few cases of squamous cell carcinoma and is considered rare. Despite that, this malignancy has been considered for many years as aggressive, with high metastatic potential and poor prognosis, its clinical evolution remains controversial [2–4].

BSCC presents distinct histopathological features with a biphasic morphological pattern, affecting mainly older men with a history of tobacco and alcohol consumption, in the upper aerodigestive tract. The cases of BSCC in the oral cavity are limited to case reports and/or retrospective studies with a small sample [1–6].

This report presents an unusual location of an asymptomatic BSCC occurring in the labial commissure of the elderly patient highlighting the importance of precise diagnosis to better understand how this variant progresses.

2. Case report

An 83-year-old man with a chief complaint of “a lump in the mouth”, reported the presence of a rapidly growing lesion in the right buccal mucosa, asymptomatic, with 4 months of duration, making it difficult to eat, associated with cutaneous lesion involving the commissure labial with approximately one year. Medical history revealed difficulty in walking, heart failure, systemic arterial hypertension, Alzheimer's disease, history of stroke, and smoking (2 packs/day) for 30 years, discontinued for 20 years.

Intraoral physical examination showed a hardened nodule in the right buccal mucosa continuous with an ulcer in the adjacent labial commissure (Fig. 1A–D).

The patient underwent an intraoral incisional biopsy and the histopathological examination revealed numerous islets of neoplastic epithelial cells with moderate pleomorphism, palisade, and hyperchromatic peripheral cells, several figures of mitosis, foci of comedonecrosis, invading the submucosa and skeletal striated muscle tissue. In addition, a clear separation between the neoplastic epithelial islets and the adjacent connective tissue was observed (Fig. 1E–H). The diagnosis of BSCC was established. The patient underwent surgical removal of the tumor without lymph node dissection, and the histopathological analysis of the surgical specimen confirmed the previous diagnosis. After three years of follow-up, the patient remains without signs of local recurrence or metastasis.

3. Discussion

In 1986, Wain et al. [2] described a rare type of carcinoma affecting the base of the tongue, hypopharynx, and larynx. In the histopathological evaluation of these tumors, these researchers highlighted the basaloid pattern associated with areas of squamous cell carcinoma, carcinoma in situ, or focal squamous differentiation, calling it BSCC. These same researchers proposed that a primitive totipotent cell with divergent differentiation could be the precursor of BSCC, which could be located at the base of the pseudostratified columnar epithelium or in the proximal ducts of salivary glands of the larynx, hypopharynx, and tongue [2]. Subsequently, Raslan et al. [7] explained that this tumor could arise from stem cells located in the basal layer of the affected mucosa.

BSCC represents less than 1% of all squamous cell carcinomas of the mouth [8,9]. On the other hand, this tumor subtype was found in 13% of all squamous cell carcinomas of the tongue [10].

Our case shows some typical features of this neoplasm, such as involvement in men and advanced age. However, oral BSCC affecting buccal mucosa, as described in the present patient is uncommon. Table 1

<https://doi.org/10.1016/j.oor.2023.100027>

Received 21 March 2023; Accepted 21 March 2023

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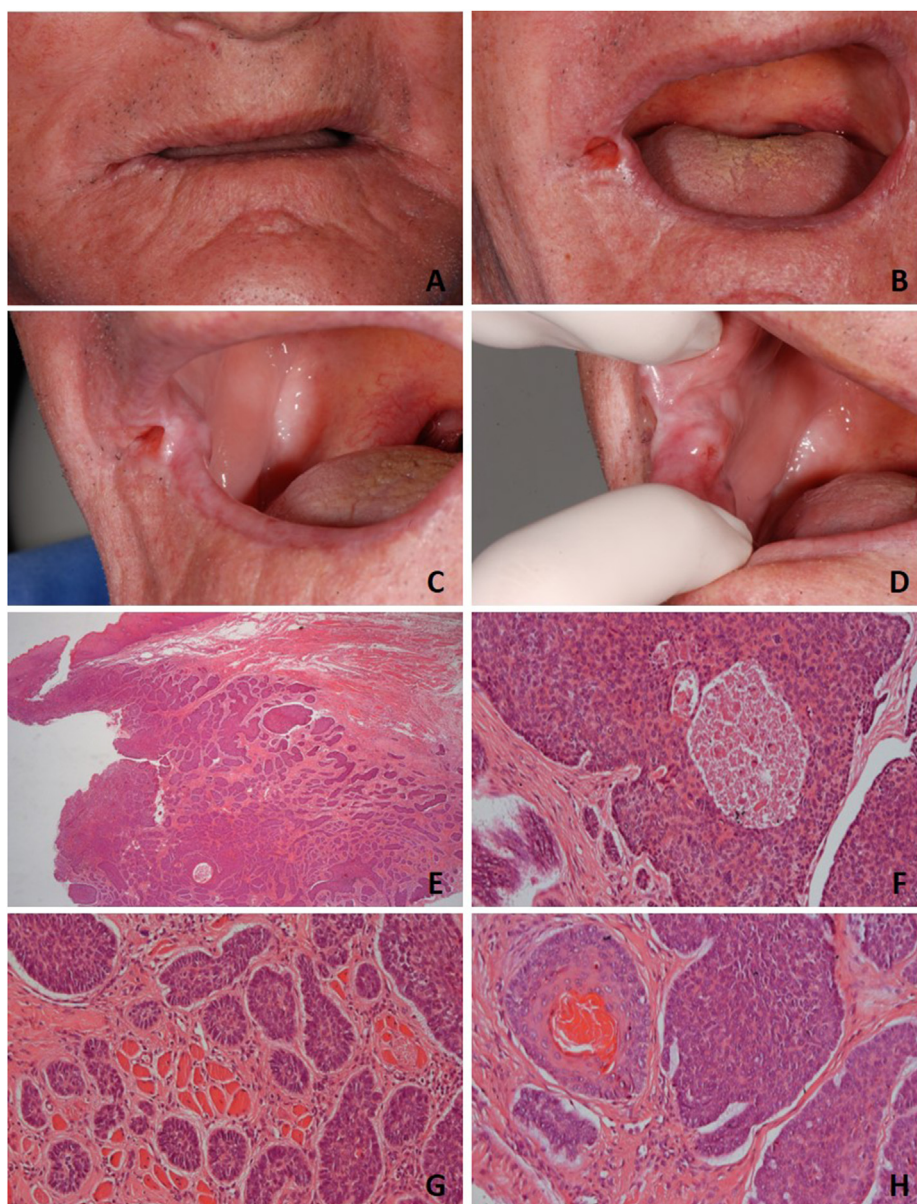


Fig. 1. Clinical aspects of the intraoral lesion presenting as a nodule continuous with an ulcer in the adjacent commissure labial (A–D). Microscopic sections revealed mucosa BSCC presenting the oral hyperplastic squamous stratified epithelium in continuity with tumor (E). Islets of neoplastic epithelial cells composed of hyperchromatic and palisade peripheral basaloid cells infiltrating muscle tissue (F), presence of comedonecrosis (G) and keratin pearls (H); (Hematoxylin-eosin stain, E= 25x, F–H= 200x).

illustrates cases of BSCCs in the head and neck published in the last fifteen years and reveals that the most common anatomical sites are the gingiva, retromolar trigone, and tonsils region. However, an important study concluded that most BSCCs of the oral cavity were located on the floor of the mouth [4].

As shown in the literature, smoking, and alcohol consumption are closely related to the development of BSCC. The human papillomavirus (HPV) is another etiological but controversial factor for this malignant tumor. Begum et al. [11] investigated the presence of HPV types 16, 18, 31, 33, 35, 45, 51, and 52 in BSCCs and found that all oral cavity tumors were negative, but 16 out of 21 oropharyngeal tumors (76.2%) were HPV positive. Our patient was not investigated for the presence of HPV types in tumor cells.

The clinical features of BSCC are similar to that of conventional squamous cell carcinoma, i.e., an exophytic, firm, and often ulcerated mass (Table 1). Despite the unusual location, the possibility of malignant neoplasm was one of the diagnostic hypotheses in the present case. Furthermore, our patient's lesion was asymptomatic. Interestingly, a literature review showed that cases that maxillary BSCCs can cause some

symptoms such as cheek pain, nasal obstruction, sinusitis, headache, diplopia, epistaxis, and even tooth loss [12].

Although we initially suspected the origin of the tumor (skin or mucosa), the intraoral incisional biopsy confirmed that the neoplastic cells arose from oral squamous cell epithelium (Fig. 1 E). The diagnosis of BSCC appears to be challenging to the pathologist. A superficial or small biopsy can make it difficult to identify the basaloid pattern, and a detailed microscopic analysis of the surgical specimen is essential, observing the relationship with the surface epithelium and adjacent tissue structures [13,14]. Of the five cases cited by Altavilla et al. [5], four were initially misdiagnosed. Three of these cases were diagnosed with conventional squamous cell carcinoma and one was diagnosed with adenoid cystic carcinoma [5]. However, adenoid cystic carcinoma does not show areas of squamous differentiation, is not related to mucosal lining epithelium, and has a smaller amount of pleomorphism, mitosis, and areas of necrosis compared to BSCC [1,15].

Additionally, other tumors can be confused with BSCC such as ameloblastoma, small cell undifferentiated carcinoma, low-grade polymorphic carcinoma, adenosquamous carcinoma, small cell

undifferentiated carcinoma, neuroendocrine carcinoma, and salivary duct carcinoma [1,16]. In the present case, a biopsy with a representative sample and the experience of the pathologist allowed for an accurate diagnosis of BSCC, confirmed by histopathological analysis of the surgical specimen.

Our patient underwent only tumor resection, due to the absence of clinical lymph node involvement, age, and general health status.

Moreover, although our patient did not have clinical lymph node involvement, a recent study showed that over 70% of patients with BSCC present N+ [17]. A limitation of the case reports presented in Table 1 is that most studies did not specify the staging of patients with BSCC. Furthermore, there is no consensus on the treatment protocol for BSCC. Important studies have concluded that radical surgical treatment was performed in all patients with BSCC and most of them underwent lymph

Table 1

Summary of case reports of basaloid squamous cell carcinoma of oral cavity available in Pubmed database (2007–2021).

Authors, (Year)	Age (Sex)	Symptoms/complaints	Location	Clinical features	Stage (AJCC staging)	Treatment	Outcome
Present case	81 (M)	Asymptomatic	Labial commissure (R)	Ulcer + Nodule	pT2N0M0	Sx	Free of recurrence or metastasis after 3 years
Nikkhah M [18]	50 (M)	Sore throat, dysphagia, reflux, and pain in left ear	Tonsil (L)	Multiple ulcers	NR	Sx + CTX + RT	Good response to therapy
Aggarwal et al. [19]	65 (M)	Difficulty in mouth opening and swallowing	RM trigone (R)	Ulceroproliferative growth	NR	NR	NR
	56 (M)	Pain	Gingiva (Ant/Mand)	Ulcerated growth	NR	NR	NR
Gupta et al. [16]	60 (M)	Pain	Tongue, soft palate and faucial pillars (L)	Ulceroproliferative lesion	NR	Sx + ND + CTX + RT	NR
Peddapelli et al. [20]	60 (M)	NR	Gingiva (Ant/Mand)	Swelling	NR	Sx	Alive after 24 months
	46 (M)	Pain	RM trigone (R)	Ulcer	NR	Sx	Alive after 24 months
Raychaudhet al. [21]	62 (M)	Difficulty in swallowing and throat pain	Tonsillar fossa (R)	Ulceroproliferative growth	NR	NR	NR
Kumari et al. [22]	39 (M)	Pain	Tongue (L)	Ulcer	cT2N0	Sx	In follow up
Hara et al. [23]	71 (M)	NR	Uvula	Ulcers over swelling	cT1N1	Sx + ND	Disease free 1 and half year after ND
Heera et al. [1]	54 (M)	Burning sensation and diffuse pain	FOM	Ulceroproliferative growth	NR	NR	NR
	57 (M)	Limited tongue movements and mouth opening	Tongue (L)	Ulceroproliferative growth	NR	NR	NR
Cheng et al. [24]	74 (F)	NR	Gingiva (R/Mand)	Swelling mass	NR	Sx	NR
Kuan et al. [12]	32 (M)	Pain	Hard palate and alveolar ridge (L)	Mass	T4aN0M0	Sx + RT	Completed treatment without complications
Pathak et al. [25]	60 (M)	Pain and nasal discharge	Gingivobuccal sulcus (R) to palatine raphe	Ulceroproliferative growth	NR	Sx + RT	Lost to follow up
Shivakumar et al. [26]	72 (M)	Pain and burning sensation	Tonsillar region (R)	Ulcer	NR	NR	NR
Tan et al. [27]	54 (M)	Pain	RM trigone (L)	Tumor	cT4N2M0	Sx + RT	Good response
	61 (M)	Pain	Tonsillar region (L)	Ulcerative mass	cT1N2cM0	Bilateral ND + CTX + RT	Good outcome
Xie et al. [28]	40 (M)	Nasal obstruction	Gingiva (R/Upper)	Mass	pT4N0M0	1st Sx + CTX + RT + 2nd Sx	Free from tumor recurrence after 3 years
Patel et al. [29]	46 (F)	Limited mouth opening	Buccal Mucosa (L)	Ulceroproliferative growth	NR	Sx + ND + RT + CTX	Died within a year after surgery
	59 (M)	Cough and breathlessness	Gingiva (L/Upper)	Exophytic growth	NR	Sx + RT + CTX	Free of disease after 3 years
Chaidas et al. [30]	56 yo (M)	Mild dysphagia	Tonsil (R)	Mass	T3N1M1	CTX + RT	Alive 3 years after initial diagnosis
Shetty et al. [31]	53 (M)	Difficulty in swallowing and foreign body sensation	Tonsillar fossa (L)	Mass	NR	Sx + CTX + RT	No evidence of complications, local recurrence or distant Metastasis after 1 year
Rachel et al. [32]	65 (M)	Pain, difficulty in mouth opening and swallowing	RM trigone (R)	Ulceroproliferative lesion	pT4N1Mx	Sx + ND + RT	Disease free after 3 months
Campos et al. [33]	59 (M)	Symptomatic	RM trigone (R)	Nodular mass	NR	Sx + RT	Died due to mesenteric thrombosis after 8 months
Hirai et al. [34]	55 (M)	Pain	Gingiva (L/Mand)	Mass	T2N0M0	Sx + ND	No evidence of disease after 6 years
	65 (M)	Asymptomatic	Gingiva (R/Mand)	Mass	T1N0M0	Sx + RT	No evidence of disease after 4 years
Subramania et al. [35]	72 (F)	NR	RM trigone (L)	Nodular growth	NR	Sx + ND + RT	Free of recurrence or metastasis after 1 year
Karatzanis et al. [36]	68 (M)	Asymptomatic	Soft palate (R)	Tumor	T1N0M0	1st Sx + RT + 2nd Sx	Free of disease for 10 months after recurrence treatment
Joshi et al. [15]	51 (F)	Asymptomatic	FOM	Swelling	NR	Sx + RT	In follow up

AJCC staging (American Joint Committee on Cancer staging system); Ant (Anterior); CTX (Chemotherapy); F (Female); FOM (FOM (Floor of the mouth)); L (Left); M (Male); Mand (Mandibular); ND (Neck dissection); NR (Not reported); R (Right); RM (Retromolar); RT (Radiotherapy); Sx (Surgery).

node dissection and postoperative radiotherapy [4,17]. Only few patients undergo chemotherapy for the treatment of this tumor [4,13].

Several studies have shown that this tumor has high rates of regional and distant metastases, suggesting a more aggressive clinical profile when compared to conventional squamous cell carcinoma of the mouth [3,8]. However, Sampaio-Góes et al. [4] compared the clinical and prognostic characteristics of patients with BSCC with patients with conventional squamous cell carcinoma, concluding that the clinical course and prognosis of BSCC were similar to conventional squamous cell carcinomas of the mouth. Also, Linton et al. [9] found no statistically significant difference in the risk of death rate between patients with BSCC and patients with conventional squamous cell carcinoma. Our patient is under follow-up without clinical evidence of locoregional recurrence or distant metastases.

In summary, this clinical case reinforces the importance of performing an intraoral incisional biopsy in the face of suspicious lesions and allows the recognition of the peculiar histopathological aspects of this subtype of oral cancer, a malignancy that the dentist or physician cannot neglect.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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