Case Reports and Reviews

Case Report

Case Report - Basal Cell Carcinoma of Skin with Myoepithelial Differentiation (BCC-MED) : A Rare Diagnostic Entity

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Abstract

Basal cell carcinoma (BCC) with myoepithelial differentiation is a rare histopathologic variant that may pose significant diagnostic challenges because of its unusual cytomorphology and broad immunophenotype.

We report the case of an 83-year-old man with multiple facial cutaneous malignancies, including the current unusual BCC of the left upper cheek showing prominent myoepithelial differentiation. The lesion was initially interpreted as a poorly differentiated malignant neoplasm with a broad differential diagnosis including melanoma and plasmacytoid squamous cell carcinoma. Subsequent histochemical and immunohistochemical studies supported the diagnosis of BCC with myoepithelial differentiation.

We discuss the clinicopathologic features, immunophenotype, differential diagnosis, and current understanding of this rare entity in the context of the existing literature.

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Introduction

Basal cell carcinoma is the most common cutaneous malignancy and is typically characterized by indolent behavior and recognizable histologic features such as basaloid nests, peripheral palisading, and stromal retraction. Numerous histologic variants have been described, including tumors demonstrating divergent differentiation along follicular, sebaceous, eccrine, neuroendocrine, and squamous lineages. Basal cell carcinoma with myoepithelial differentiation (BCC-MED) represents one of the rarest of these variants and is characterized by the presence of tumor cells with plasmacytoid or signet-ring–like morphology, abundant eosinophilic cytoplasm, and hyaline inclusions admixed with areas of conventional BCC [1–3].

Because of its rarity and atypical appearance, BCC-MED may be misdiagnosed as a primary adnexal neoplasm, melanoma, metastatic carcinoma, or plasmacytoid squamous cell carcinoma. Awareness of this entity and judicious use of immunohistochemistry are essential to avoid diagnostic error.

We present an additional case of BCC-MED occurring on the face of an elderly man, review the salient features that distinguish this tumor from its mimics and correlate the findings with prior published reports.

Case Presentation

An 83-year-old man was evaluated in the plastic surgery clinic for multiple facial skin lesions suspicious for cutaneous malignancy. His past medical history was significant for chronic ultraviolet exposure due to a long career in construction work, with no history of sunscreen use. He had a known history of multiple non-melanoma skin cancers, including a basal cell carcinoma of the right nasal ala diagnosed in 2022, which had been treated with radiation therapy followed by excision.

At the current presentation, the patient reported four facial lesions. Lesions on the right lateral brow and nasal dorsum were clinically and histologically diagnosed as basal cell carcinoma, and a lesion on the left inferior cheek was diagnosed as squamous cell carcinoma in situ. The lesion that is the subject of this report was located on the left upper cheek (nasolabial region) and measured approximately 2 mm, with a red central scab.

Excisional procedures were performed on the suspicious lesions.

Pathologic Findings

Microscopic examination of the excision from the left medial upper cheek revealed a dermal-based malignant neoplasm composed of sheets and clusters of atypical cells. The

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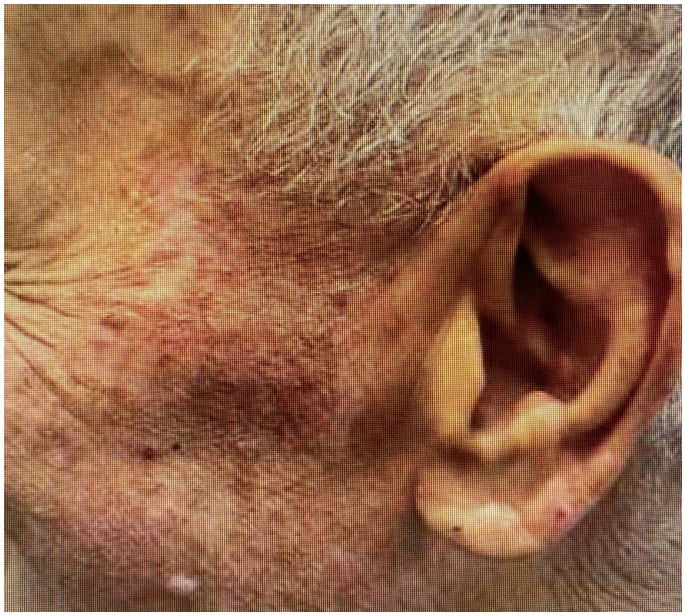

Figure 1: *Clinical photograph of the left upper cheek lesion*

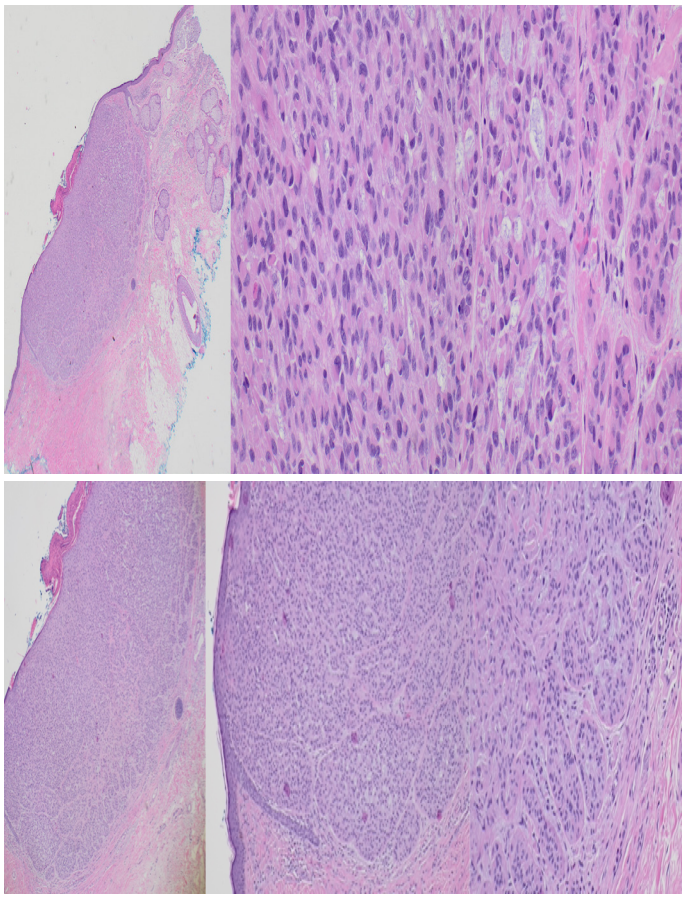

Figure 2 and 3*: Hematoxylin and eosin–stained section (10X, 20x and 40x) showing sheets of plasmacytoid tumor cells with abundant eosinophilic cytoplasm and eccentric nuclei.*

tumor cells exhibited abundant eosinophilic, cytoplasm with hyaline inclusions, vesicular and pleomorphic nuclei with conspicuous nucleoli, and numerous mitotic figures. In the initial diagnostic interpretation, the lesion was reported as a poorly differentiated malignant neoplasm, and the differential diagnosis included plasmacytoid squamous cell carcinoma and melanoma. Owing to the unusual cytomorphology, the case was referred for expert dermatopathology consultation.

Special histochemical stains demonstrated that periodic acid–Schiff (PAS) and PAS with diastase digestion (PAS-D) were negative within the tumor cells, arguing against true mucin-filled signet-ring cell differentiation, and consistent with prior reports of BCC-MED [1,2].

An extensive immunohistochemical panel was performed. The tumor cells showed positivity for AE1/AE3, BER-EP4, p63, p40, CK5, CK8/18, MA903, CD56, smooth muscle actin (SMA), smooth muscle myosin heavy chain (SMMHC), and calponin. The tumor cells were negative for EMA, vimentin, S100, SOX10, melanoma cocktail, HMB45, desmin, myoglobin, myogenin, CK7, and CK20. The Ki-67 proliferation index was estimated at approximately 10%.

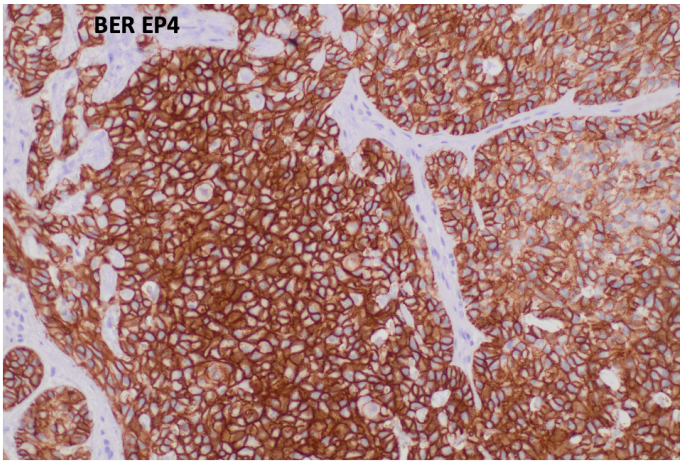

Figure 4: *Immunohistochemistry showing BER-EP4 positivity supporting basal cell carcinoma differentiation.*

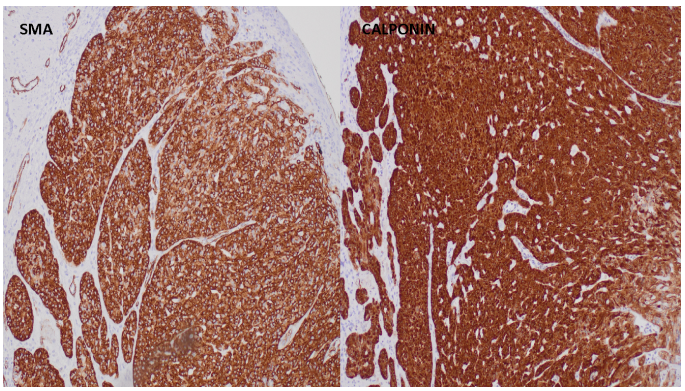

Figure 5: *Immunohistochemistry showing diffuse smooth muscle actin and/or calponin positivity supporting myoepithelial differentiation.*

The combined morphologic, histochemical, and immunohistochemical findings supported a diagnosis of basal cell carcinoma of skin with prominent myoepithelial differentiation (BCC-MED).

Discussion

Basal cell carcinoma with myoepithelial differentiation (BCC-MED) is an exceptionally rare variant of BCC. Since its initial description by Seo et al. in 1979, fewer than two dozen well-documented cases have been reported in the literature [1–5]. Cohen summarized 16 cases reported in the literature, highlighting frequent diagnostic difficulty due to unusual histologic features [1]. The majority of reported cases have occurred in older individuals, with a male predominance, and all have arisen on the face, most commonly involving the cheek, periocular region, nose, and forehead [2,5]. Our patient closely mirrors the clinicopathologic spectrum described in that review, including advanced age, facial location, and initial diagnostic uncertainty.

Histologically, BCC-MED is characterized by a dual population of tumor cells. One component exhibits features of conventional BCC, including basaloid nests with peripheral palisading and stromal retraction. The second component mimics a myoepithelioma and is composed of plasmacytoid cells with eccentric nuclei and abundant eosinophilic cytoplasm containing hyaline inclusions [2–4]. These inclusions represent aggregates of intermediate filaments rather than mucin-filled vacuoles, explaining the frequent negativity for PAS and mucin stains and distinguishing this entity from true signet-ring cell carcinomas [2,3].

Immunohistochemically, BCC-MED demonstrates a composite profile reflecting both basal cell and myoepithelial differentiation. Tumor cells typically express epithelial markers such as cytokeratins, p63/p40, and BER-EP4, confirming basal cell carcinoma differentiation [1–3]. Variable expression of myoepithelial markers, including SMA, calponin, smooth muscle myosin, S100 protein, GFAP, and vimentin, has been reported [2–5]. Importantly, SOX10 is consistently negative, which helps exclude primary cutaneous myoepithelioma or myoepithelial carcinoma [3-5].

The differential diagnosis of BCC-MED is broad and includes cutaneous mixed sweat gland tumor (pleomorphic adenoma), myoepithelioma, myoepithelial carcinoma, plasmacytoid squamous cell carcinoma, melanoma, and metastatic signet-ring cell carcinoma from gastrointestinal or breast primaries [2–6]. Identification of a conventional BCC component and confirmation with BER-EP4 immunostaining are critical features that support the correct diagnosis. Immunostaining are critical for accurate diagnosis and to avoid overinterpretation as a more aggressive malignancy [1].

Available data suggest that the biologic behavior of BCC-MED parallels that of conventional BCC. Complete surgical excision, preferably with margin control such as Mohs micrographic surgery, is the recommended treatment. To date, reported cases have not demonstrated aggressive behavior or metastatic potential when adequately excised [2,5,6].

Conclusion

We report a rare case of basal cell carcinoma with prominent myoepithelial differentiation (BCC-MED) arising on the face of an elderly man with extensive cumulative sun exposure and a history of multiple non-melanoma skin cancers. This case highlights the striking morphologic and immunophenotypic features of this rare BCC variant and underscores the importance of considering BCC-MED in the differential diagnosis of poorly differentiated plasmacytoid cutaneous tumors. Awareness of this entity and judicious use of immunohistochemistry can prevent misdiagnosis and ensure appropriate patient management..

References

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