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Case Report

Surgery Section

An Unusual Synchrony of Bowen's Disease and Merkel Cell Carcinoma: A Case Report

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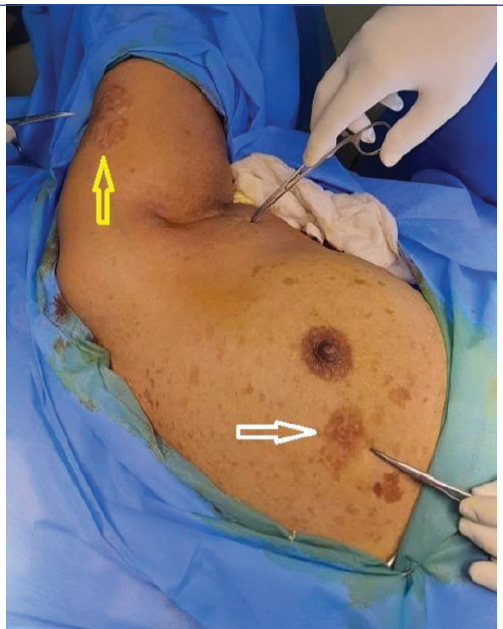
ABSTRACT

Bowen's Disease (BD), recognised as Squamous Cell Carcinoma (SCC) in situ, typically affects older individuals and postmenopausal women, whereas Merkel Cell Carcinoma (MCC) is an aggressive neuroendocrine skin malignancy with a predilection for elderly white males. This case report delves into the unique convergence of BD and MCC in a 45-year-old woman, presenting as BD in the left breast and arm, coupled with MCC in the left wrist with metastasis to the left axilla. The patient was treated with wide local excision of the breast and arm lesions, coupled with en masse clearance of the left axilla due to metastatic MCC. This unique case underscores the intricate interplay between different cutaneous malignancies, emphasising the need for continued research to unravel the underlying mechanisms and optimise therapeutic strategies in rare and challenging dermatological presentations.

Keywords: Dermis, Epidermis, Malignancy

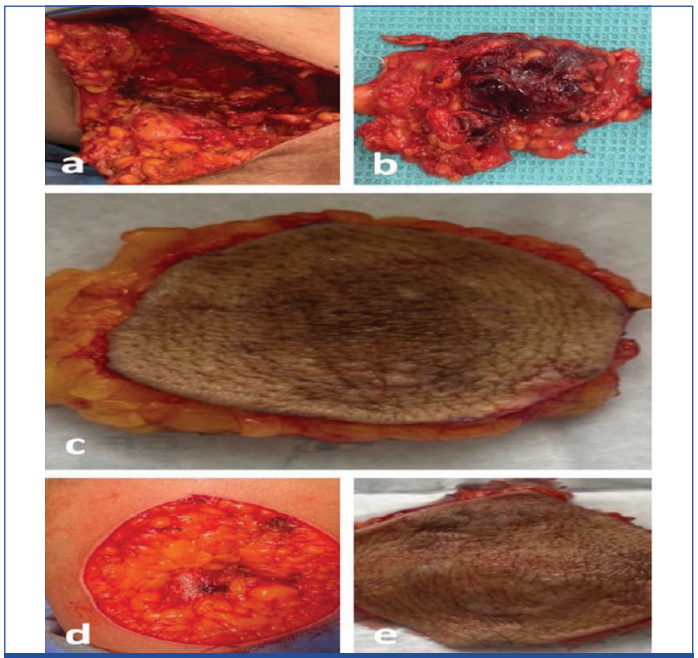
CASE REPORT

A 45-year-old woman presented with a reddish patch on her left breast and left upper arm that had been present for the past five years. Initially, the patch exhibited slow, progressive growth without associated symptoms, leading her to delay seeking medical consultation. The patient sought medical attention after noticing an increase in the size of the lesions along with the appearance of a nodular swelling in the left axilla. The patient gave a history of excision of a lesion on her left wrist two years prior, identified as MCC upon Histopathological Examination (HPE). Upon examination, an erythematous patch of approximately 5×2 cm was noted on the left arm, and a 5×3 cm patch on the left breast [Table/Fig-1]. These lesions were slightly elevated from the skin surface, displaying a scaly appearance, and were tender to touch with no signs of inflammation.



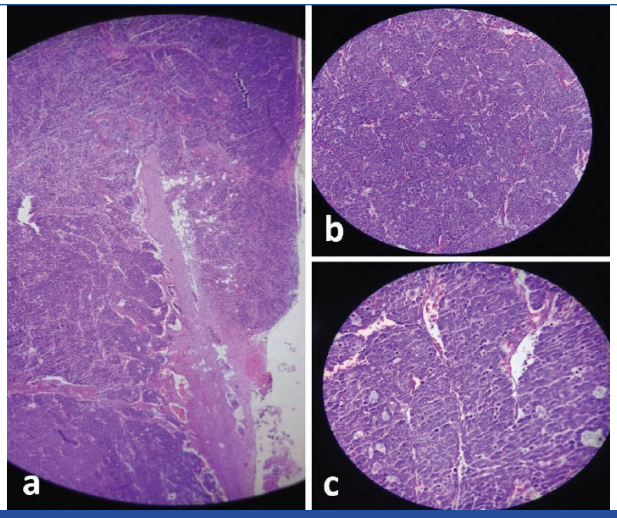
[Table/Fig-1]: Breast (white arrow) and arm lesion (yellow arrow) with axillary lymphadenopathy.

Examination of the ipsilateral axilla revealed enlargement of multiple lateral group of axillary lymph nodes, with the largest node measuring 2×1 cm. The lymph nodes were firm and exhibited restricted mobility. The rest of the ipsilateral and contralateral breasts appeared normal. To further investigate the lesions, a wedge biopsy was performed, and the patient sought the opinion of a dermatologist. The skin biopsy report for the lesions on the breast and arm confirmed the diagnosis of BD. As a definitive treatment, a wide local excision of the breast and arm lesions was performed, along with complete en masse clearance of the left axilla [Table/Fig-2].



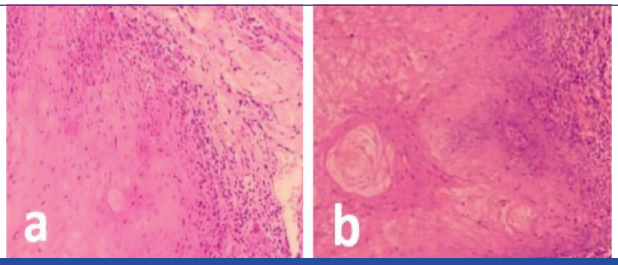
[Table/Fig-2]: Intraoperative and specimen photographs: a) Intraoperative view of axillary dissection; b) Excised axillary lymph nodes following dissection; c) Excised breast lesion (gross specimen); d) Intraoperative view of the arm after excision of the lesion; e) Gross appearance of the excised arm lesion.

The HPE results [Table/Fig-3,4] were consistent with BD (SCC in situ) for the breast and arm lesions, while the axillary lymph nodes indicated metastatic MCC.-Stage III (pT1–2, pN1, M0; AJCC 8th edition). The axillary lymph nodes demonstrated complete architectural effacement with extracapsular extension [Table/Fig-3a], tumour cells arranged in nests separated by vascular septa [Table/Fig-3b], and cells with coarse salt-and-pepper chromatin and frequent mitotic figures [Table/Fig-3c], consistent with metastatic



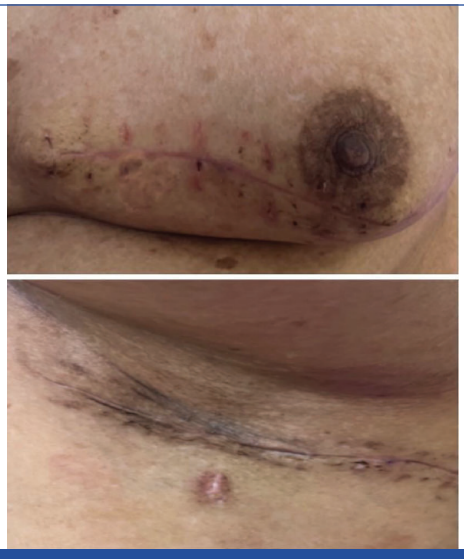
[Table/Fig-3]: Haematoxylin and Eosin stain at 10x magnification shows Lymph node architecture replaced by tumor cells with extracapsular extension (a); Tumour cells arranged in nested architecture separated by vascular septa (b); Tumour cells with coarse salt and pepper chromatin and multiple scattered mitotic figures (c).

MCC. Histopathology of the breast and arm lesions showed full-thickness squamous dysplasia with loss of maturation, nuclear pleomorphism, hyperchromasia, increased mitoses and intact basement membrane, consistent with BD [Table/Fig-4]. The primary lesion of MCC was in the left wrist, which was excised two years prior and confirmed on HPE.



[Table/Fig-4]: Histopathology of arm: a) and breast lesion; b). Haematoxylin and Eosin stain at 10x magnification shows full-thickness squamous dysplasia with loss of maturation, nuclear pleomorphism, hyperchromasia, increased mitoses and intact basement membrane, consistent with BD.

The patient experienced an uneventful postoperative recovery and was discharged on postoperative day 2 in a stable condition, with the operative sites showing healthy healing. No adjuvant chemotherapy or radiotherapy was given as the patient underwent complete surgical excision of the primary lesions with axillary clearance and showed no residual disease. Over the course of regular follow-ups spanning the last five months, the operative sites have healed nicely, leaving behind normal scars [Table/Fig-5].



[Table/Fig-5]: Healed scar site of breast and axilla region on post operative day 10.

DISCUSSION

BD, also recognised as SCC in-situ, manifests as a malignant lesion confined to the epidermis, lacking evidence of dermal invasion [1]. It predominantly afflicts older individuals and postmenopausal women and foreshadows the development of overt SCC [2]. Various factors contribute to its onset, including exposure to ultraviolet radiation, human papillomavirus, carcinogens such as arsenic, genetic predisposition and the presence of birthmarks [3]. Conversely, MCC is an infrequent neuroendocrine skin malignancy known for its aggressive nature. Primarily affecting elderly white males above 65 years old and immunosuppressed patients, MCC exhibits a predilection for the head and neck region [4]. The pathogenesis of MCC is linked to prolonged ultraviolet exposure and Merkel Cell Polyomavirus (MCV) [5]. The predominant locations of affliction of BD were the lower extremities in women and the scalp and ears in men, and the occurrence of BD in the breast is an unusual phenomenon [6].

MCC is believed to originate from Merkel cells, identifiable by the presence of electron-dense core granules- a characteristic feature of these cells [7]. Associations between MCC and other skin conditions like SCC, basal cell carcinoma, actinic keratosis, and rarely BD are documented. The exact cause of the rare association with BD remains unknown. Arsenic exposure is considered an inducer for MCC, but this patient had no history of arsenic exposure, viral infection, or immunosuppression [8]. MCC typically involves the dermis and subcutaneous tissue, with rare epidermal involvement.

BD characterised as an intraepidermal SCC in-situ was first documented by Bowen JT in 1912 [9]. Patients with BD often present with a gradually enlarging, well-defined erythematous plaque displaying hyperkeratosis, scaling, and itching. Although most of the cases involve postmenopausal women aged 69-84 years [10], few cases involved women below 60 as well, aligning with our case. Pathologically, BD exhibits full-thickness epidermal involvement with distorted architecture, abnormal mitoses, atypia, and dyskeratosis, without penetration into the dermis [2].

Treatment options for BD include topical chemotherapy, cryotherapy, curettage, photodynamic therapy, and surgery. Localised MCC is treated with surgical excision and adjuvant radiotherapy, with sentinel lymph node biopsy in clinically node-negative patients. Nodal disease requires lymph node dissection ± radiotherapy. Advanced MCC is managed with first-line anti–PD-1/PD-L1 immunotherapy [11]. In this case, a wide local excision was opted for, considering the planned left axilla clearance due to metastatic MCC in the left axilla.

Swain M et al., reported MCC concurrent with BD in a 32-year-old lady involving the dorsum of the hand with lymph node metastases [12]. Park HC et al., in 2012, reported that MCC is concurrent with BD in a 75-year-old female on the left mandibular angle [13]. A case of concurrent SCC on the forehead and MCC on the left cheek in a patient with generalised actinic keratosis was also reported by Yue ZH et al., [14]. Rare synchronous occurrence of BD and metastatic MCC in a young, immunocompetent woman highlights the need for thorough skin examination, vigilant surveillance, early staging, and multidisciplinary management in patients with unusual cutaneous malignancies.

CONCLUSION(S)

This rare case emphasises the unusual coexistence of BD and MCC. It underscores the significance of a multidisciplinary approach and vigilant follow-up in managing complex cases involving the convergence of distinct malignancies. Continued research and documentation of such cases contribute to a deeper understanding of the underlying mechanisms and optimal therapeutic interventions for these uncommon dermatological presentations.

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