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CASE REPORT

Dermatosis Neglecta and Chilblain Lupus Erythematosus Occurred Post Surgery of Basal Cell Carcinoma: A Case Report

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Abstract: Dermatosis neglecta (DN) is a rare condition, often linked to psychosomatic disorders, with an unclear etiology. Chilblain lupus erythematosus (CHLE) constitutes a rare variant of cutaneous lupus erythematosus, frequently mistaken for idiopathic chilblain due to its chilblain-like lesions. To date, no instances of concurrent DN and CHLE occurring at the site of a skin flap or graft have been documented. Herein, we present a case that the occurrence of DN and CHLE developed on a skin flap at once. The simultaneous occurrence of DN and CHLE on a skin flap is exceptionally rare. Clinicians must exercise heightened vigilance due to the common misdiagnosis of CHLE. Various skin disorders can manifest on skin flaps and grafts post-transplantation. Thus, clinicians must remain vigilant for both recurrence and metastasis during post-operative monitoring, while also attending to patients' mental well-being and the condition of skin flaps.

Keywords: dermatosis neglecta, skin flap, basal cell carcinoma, chilblain lupus erythematosus

Introduction

Basal cell carcinoma (BCC) ranks among the most prevalent skin cancers, predominantly manifesting on sun-exposed areas like the external nose, periorbital region, and cheek.[1] Surgical excision stands as a primary treatment modality for BCC. In instances of sizable lesions that cannot be directly closed post-excision, procedures such as skin flap and graft transplantation become imperative for repairing the surgical defect.[2]

DN represents a rare dermatosis with an elusive etiology, commonly categorized among psychogenic dermatoses. DN typically arises from neglect stemming from various factors, including surgical history, skin hypersensitivity, pain, and cognitive impairment. This neglect leads to the accumulation of sebum, sweat, keratinocytes, and dirt, culminating in the formation of thick, dirt-like crusts adhering to the skin surface.[3] While proper cleaning alone can effectively eliminate these crusts, addressing the underlying causes of neglect remains paramount.

CHLE constitutes a rare subtype of chronic cutaneous lupus erythematosus, predominantly afflicting middle-aged women and potentially associated with microcirculatory impairment.[4] CHLE lesions typically manifest as mauve patches and papules on the face, hands, and feet, closely resembling idiopathic chilblains. Given its rarity and the striking similarity of its lesions to chilblains, CHLE often faces underdiagnosis and misdiagnosis in clinical settings.[5]

Herein, we present a case of DN coexisting with CHLE, occurring on a flap subsequent to BCC excision and skin flap transplantation.

Case Presentation

A 53-year-old female presented with a black maculopapule on the tip of her nose. Approximately 1 year prior, she noticed a rash, initially the size of a rice grain, without accompanying pain or itching. Over time, the rash progressively

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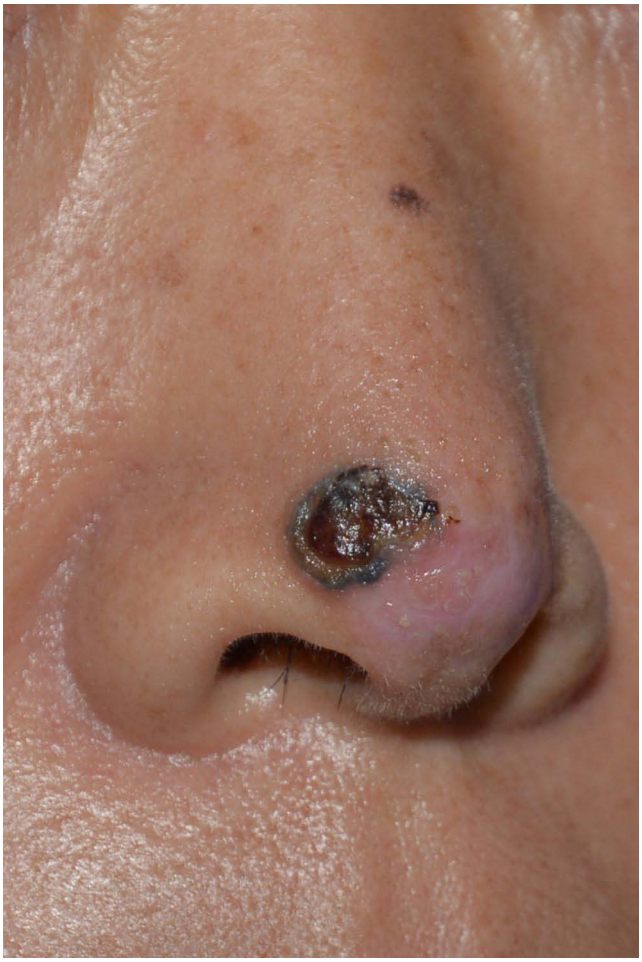

Figure 1 Preoperative dermatologic examination revealed a black maculopapule measuring approximately 0.7 centimeters in diameter at the tip of the nose. The lesion exhibited ulceration and crusts on the surface, with rolled edges.

enlarged to the size of a peanut grain before abruptly rupturing and bleeding, prompting her visit to the dermatology department 5 months ago. Dermatological examination revealed a black maculopapule, approximately 0.7 centimeters in diameter, with a crusted surface and rolled edge (Figure 1). Clinically diagnosed as BCC, the patient underwent skin tumor excision, with the surgical defect repaired using a bilobed flap. Postoperative pathology confirmed the diagnosis of large nodular BCC (Figure 2). Three months post-surgery, the patient presented again, with the nasal skin flap exhibiting an edematous mauve plaque with poorly defined borders and thick fissured tawny crusts, which proved challenging to remove (Figure 3) and led to concerns about flap necrosis. Notably, the patient had a medical history of diabetes mellitus, managed with subcutaneous insulin aspart and insulin glargine, maintaining fasting blood glucose below 6.1 mmol/L and 2-hour postprandial blood glucose below 7.5 mmol/L.

Direct immunofluorescence staining of nasal crust scrapings revealed mycelia, indicative of Malassezia furfur infection (Figure 4), subsequently confirmed by fungal culture (Figure 5). Histopathological examination of the nasal plaque demonstrated hyperkeratosis with hyperplasia, follicular plugging, mild hyperplastic hypertrophy of the epidermis, extensive vacuolar liquefaction of basal cells, and lymphocytic infiltration around superficial and deep dermal blood vessels and appendages (Figure 6). Hematological examinations indicated no abnormalities in the antinuclear antibody spectrum (ANAs), markedly accelerated erythrocyte sedimentation rate (ESR), mildly elevated leucocyte count, lymphocyte count, and neutrophil count. No abnormalities were observed in infectious diseases. Additionally, the patient scored 62 on the Self-rating Anxiety Scale (SAS),[6] indicative of moderate anxiety.

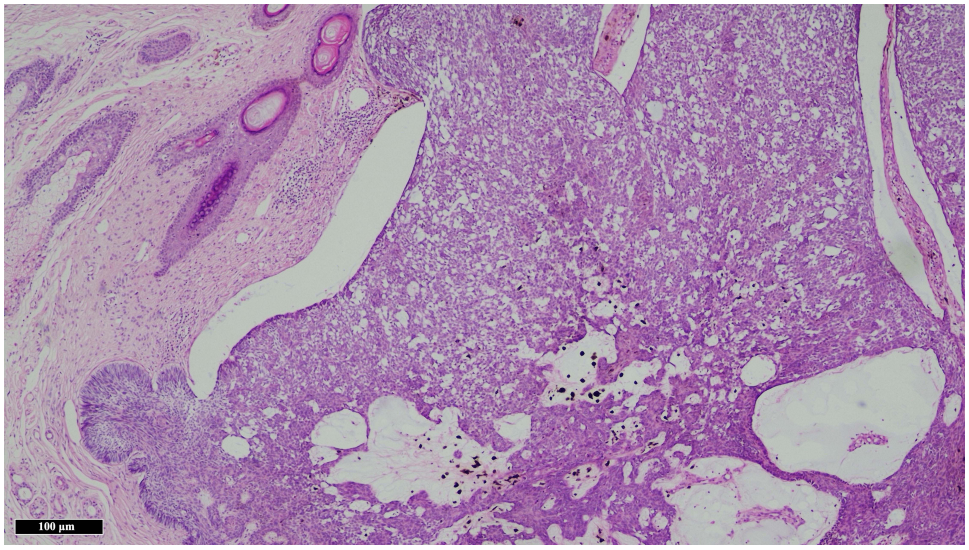

Figure 2 Postoperative pathology depicted a mostly normal epidermis, with the dermis exhibiting a cluster of tumor cells comprising basaloid cells (H&E staining).

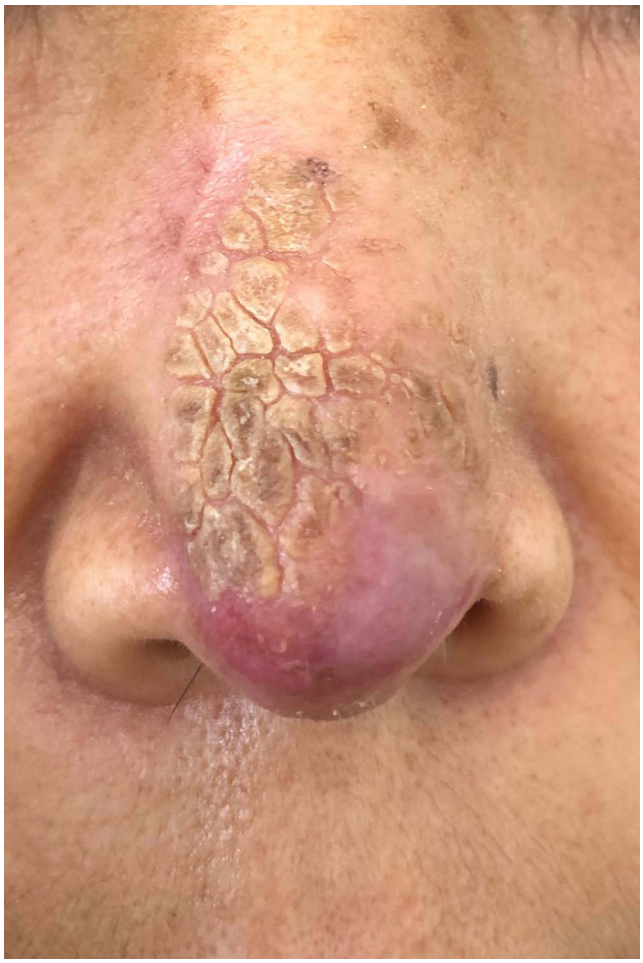

Figure 3 Three months post-surgery, the nasal tip flap exhibited a poorly defined edematous plaque. The nasal dorsal flap was covered with thick, tawny cracked crusts that were challenging to remove.

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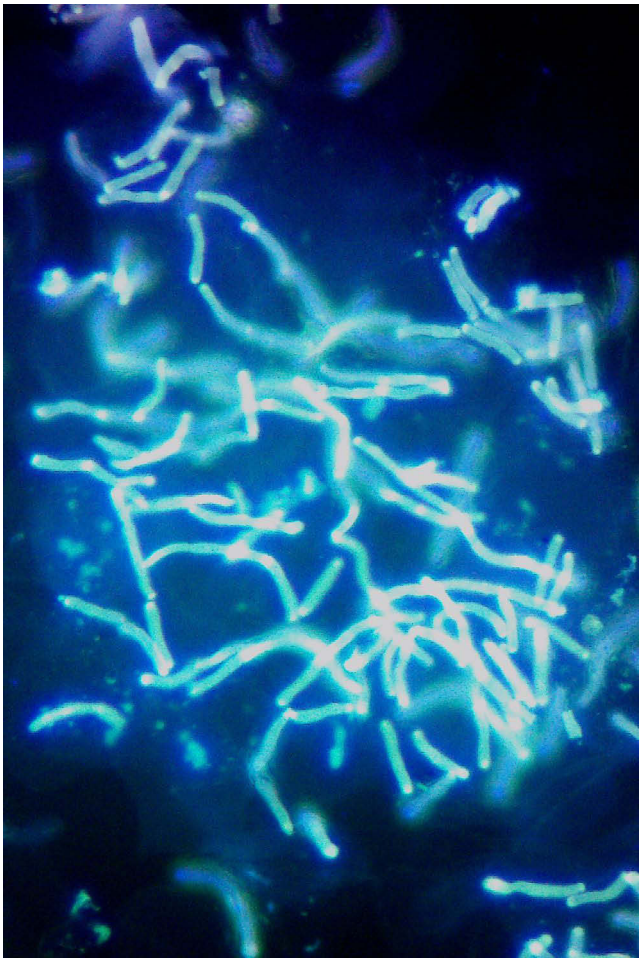

Figure 4 Direct fungal fluorescent staining revealed mycelia.

Based on clinical manifestations, pathological findings, hematological results, and microbiological analysis, the patient received a diagnosis of dermatosis neglecta combined with chilblain lupus erythematosus occurring on a skin flap following BCC tumor excision. Psychosocial support was provided, alongside olive oil for crust softening, soap for cleansing, and topical ketoconazole cream, resulting in DN resolution within one week (Figure 7). Treatment with hydroxychloroquine (HCQ) 0.2g twice daily and 0.01% tacrolimus ointment continued topically, leading to flattening of the plaque after one month of therapy. The patient remains under follow-up care.

Discussion

DN represents a rare dermatologic condition with multifactorial etiology, including psychosocial factors, Malassezia colonization, trauma, previous surgical history, and poor hygiene practices. And the concomitant pain could be an important reason for the lack of washing.[7] For our patient, who had a major surgery, the postoperative pain could contribute to poor hygiene initially. DN diagnosis primarily relies on its characteristic clinical presentation and patient history. Palaniappan et al underscored the nonspecific histopathological findings in DN, advocating against invasive tests such as biopsy.[3] In our case, post- BCC surgery, the patient's anxiety and neglect of hygiene, compounded by Malassezia infection, precipitated DN development. Terra firma-forme dermatosis (TFFD) is characterized by idiopathic retention hyperkeratosis, resulting in dirt-like brown, asymptomatic plaques.[8] Distinguishing DN from TFFD is pivotal, with key differentiation points outlined in Table 1. Additionally, an alcohol swabbing test aids in differentiation: DN crusts can be removed using various substances, including oil cleansers, emollients, 70% alcohol and isopropyl alcohol, while TFFD

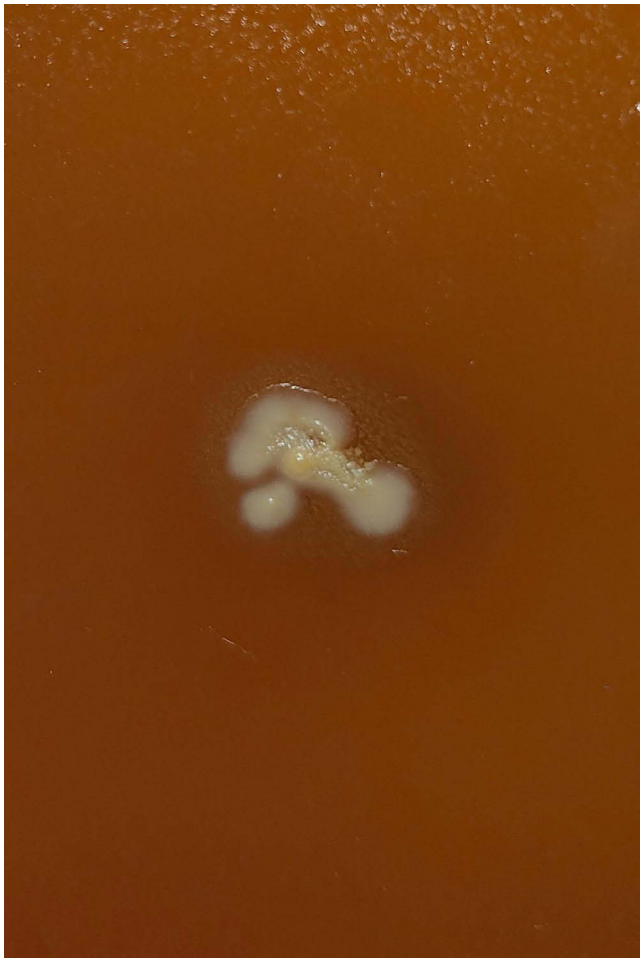

Figure 5 Fungal culture exhibited pale yellow colonies.

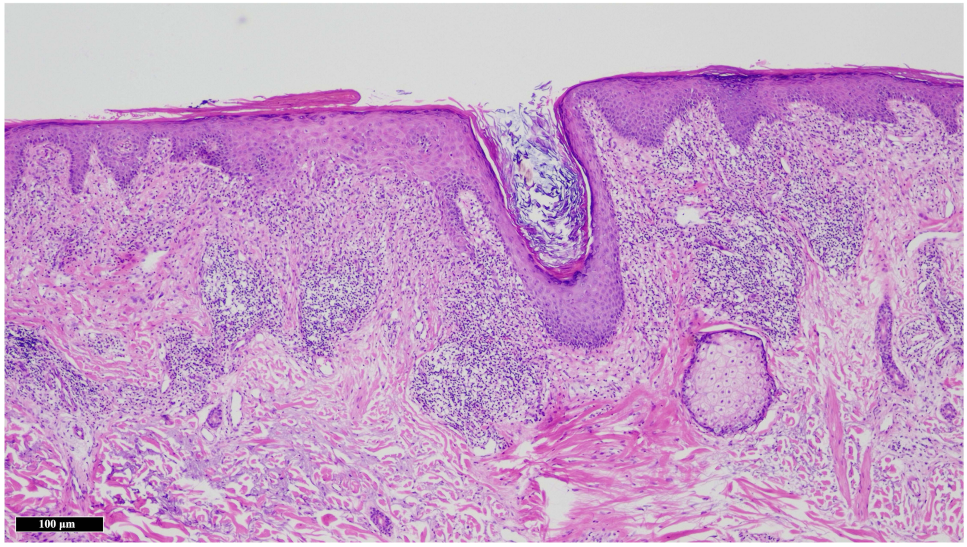

Figure 6 Pathological examination of the mauve plaque on the tip of the nose revealed a follicular plug, extensive vacuolar liquefaction of the basal cells, lymphocytic infiltration around dermal vessels and appendages (H&E staining).

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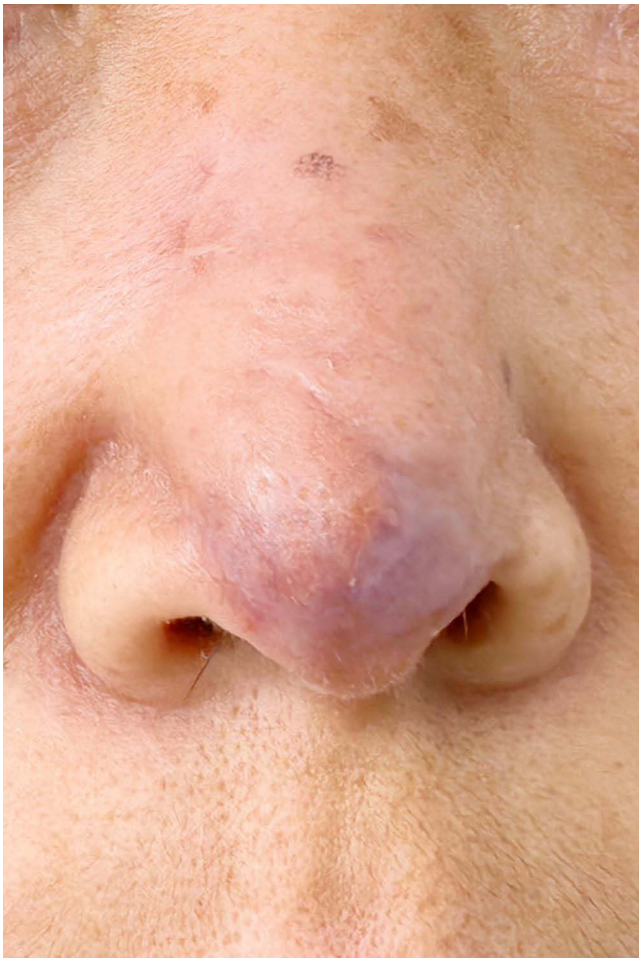

Figure 7 Following the removal of the thick crusts, the nasal flap displayed a mauve plaque.

lesions respond solely to 70% alcohol and isopropyl alcohol.[7] Our patient, a middle-aged woman with a two-month disease duration, anxiety, and reluctance to clean the surgical site, exhibited nasal crusts readily removed post-soaking in olive oil and washing with soap, aligning more closely with DN diagnosis.

Table 1 Differentiation Points Between Dermatosis Neglecta and Terra Firma-Forme Dermatosis

Points	**Dermatosis Neglecta**	**Terra Firme-Forme Dermatosis**
Age group	All Age group	Children and young people
Average duration	2–4 months	8 months
Hygiene	Poor	Usually maintained
Psychosomatic state	Often combined with anxiety, depression, cognitive impairment etc.	Usually normal
Lesions	Yellow to tawny fissured thick crusts	Brown hyperpigmented macule and patch, or velvety plaque
Malassezia	Often detected	Detected in a few cases, but now considered non-specific
Combined atopic diseases	None	Common

DN management centers on proper cleanliness, coupled with oral itraconazole and topical ketoconazole for fungal infections, alongside crucial psychotherapy. Studies have linked DN occurrence with psychosomatic disorders, underscoring the significance of psychological intervention.[9] In our case, comprehensive psychological counseling and topical antifungal treatment resolved DN within a week. While proper cleaning alone can effectively eliminate these crusts, addressing the underlying causes of neglect remains paramount. This underscores the importance of postoperative psychological support, emphasizing effective skin cleaning practices to prevent DN onset.

CHLE represents a rare subset of chronic cutaneous lupus erythematosus, with an approximate incidence of 15% among lupus erythematosus cases.[4] CHLE pathogenesis remains unclear, potentially associated with cold exposure and drug-induced triggers.[10] Postoperative CHLE cases are exceedingly rare, with only isolated reports documented.[11] Although our patient had no postoperative medication history, diabetes mellitus and surgical microcirculatory disturbances, combined with cold exposure, likely contributed to CHLE onset.

CHLE primarily affects middle-aged women, presenting as purplish or reddish infiltrating plaques on the nose, ears, hands, and feet.[12] Anti-Sjögren's syndrome A (SSA) antibodies serve as valuable diagnostic markers, with elevated positivity rates aiding CHLE diagnosis.[4] Differential diagnosis from idiopathic chilblains hinges on lesion persistence and absence of blisters or vesicles, distinguishing features of CHLE.[5] Although our patient's clinical presentation was typical, negative anti-nuclear antibodies did not preclude CHLE diagnosis, confirmed via clinical and histopathological assessment.

CHLE management entails warmth maintenance, topical glucocorticoids, and calcium-modulating phosphatase inhibitors for mild cases, escalating to oral glucocorticoids or hydroxychloroquine for severe presentations.[13] Given the risk of progression to systemic lupus erythematosus (SLE), regular follow-up is imperative to monitor disease stability and prevent SLE progression.[4]

Conclusion

Notably, postoperative DN and CHLE combination cases are exceedingly rare, lacking documented instances in literature. This case underscores the necessity for intensified postoperative follow-up following cutaneous tumor excision, emphasizing vigilance not only for tumor recurrence and metastasis but also for potential dermatologic comorbidities. Perioperative health education and patient-centered care are paramount, with long-term postoperative monitoring pivotal in ensuring optimal patient outcomes.

Ethics Approval and Consent to Participate

This study was performed in line with the principles of the Declaration of Helsinki. Approval was granted by the Ethics Committee of Affiliated Hospital of Shandong Second Medical University. The patient had given written informed consent for the publication of her clinical details and accompanying images.

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Disclosure

The authors declare no competing interests in this work.

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