Clinical, Cosmetic and Investigational Dermatology

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CASE REPORT

Extensive Nevus Comedonicus Treated by Comb Flap Reconstruction, Super Tension-Relieving Sutures, and Vacuum Sealing Drainage: A Case Report

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Purpose: There are few reports on the comprehensive surgical management of extensive nevus comedonicus, and the genetic mechanism of this rare disease is not yet fully understood.

Patients and Methods: We report a case of a 38-year-old woman with extensive nevus comedonicus, refractory to multiple medical treatments. She underwent comb flap reconstruction combined with super tension-relieving sutures and vacuum sealing drainage. In addition, whole-exome sequencing was performed on the lesional tissue samples.

Results: Nevus comedonicus was confirmed by physical and histopathological examination. Postoperative recovery was uneventful, and the patient was satisfied with the efficacy. Furthermore, no rare or deleterious mutations were detected in NEK9, FGFR2, ABCA12, or KRT10.

Conclusion: Comprehensive surgical intervention is a viable therapeutic option for extensive nevus comedonicus refractory to medical management. Further investigation into the underlying genetic mechanisms of this disease is warranted.

Keywords: nevus comedonicus, whole-exome sequencing, surgical management, super tension-relieving sutures, vacuum sealing drainage

Introduction

Nevus comedonicus (NC) is a rare hamartomatous malformation caused by a defective development of the folliculosebaceous unit. To date, more than 200 cases have been reported worldwide, with a global incidence of approximately 1 in 45,000 to 1 in 100,000.[1] NC is generally present at birth or develops before the age of 10, which tends to occur on the face, neck, trunk, and limbs.

Clinically, NC is typically categorized into two types: simple comedo-like rash and rash complicated with cysts, scars, and infections.[2] To our knowledge, spontaneous resolution of NC has not been reported. Conversely, recurrent cyst formation, secondary infection, and scarring with sinus tract formation are common in NC.[3,4] Surgical excision remains the radical solution for these lesions.

Herein, we report a case of extensive, refractory NC in an adult treated with surgical reconstruction combined with vacuum sealing drainage (VSD). Meanwhile, given the rarity of NC, whole-exome sequencing (WES) was conducted to explore the genetic basis.

Case Presentation

A 38-year-old Asian woman presented with a 30-year history of slowly progressive, unilateral, clustered comedone-like papules extending linearly from the right breast to the right scapular line, following Blaschko's lines (Figure 1a and b).

Received: 16 October 2025
Accepted: 16 December 2025
Published: 20 December 2025

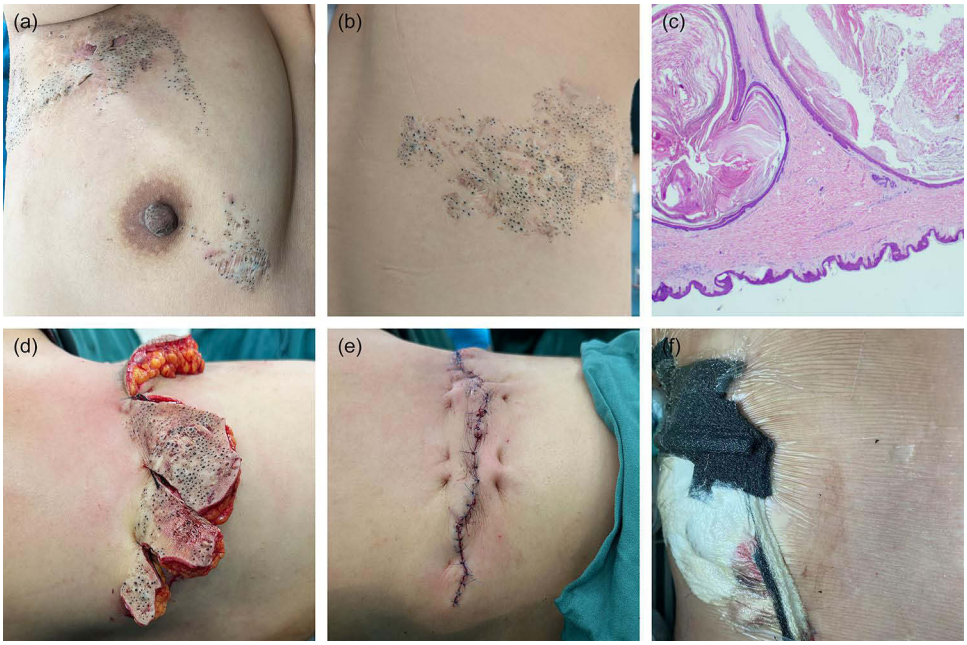

Figure 1 Clinical manifestations, histopathological findings, and treatment of nevus comedonicus. (**a**) Clustered comedo-like lesions are arranged in a unilateral, linear distribution along the right anterior chest, axilla, and across the lateral trunk. (**b**) The lesions consist of multiple dilated follicular openings filled with dark keratin plugs, following a Blaschkoid pattern. (**c**) Histopathological findings of the lesions. (**d**) Lesions were excised using the comb flap technique intraoperatively. (**e**) Super tension-relieving sutures were applied to close the wound and minimize tissue tension. (**f**) A vacuum sealing drainage device was applied to the wound postoperatively.

The lesions were previously diagnosed as NC based on clinical and histopathological features. Over the past six years, the lesions experienced recurrent infections, necessitating multiple treatments. However, the condition proved refractory to all therapies. Previous treatments included topical retinoic acid cream, oral isotretinoin, and CO_2 fractional laser; unfortunately, all yielded no significant improvement. One month prior to admission, the lesions on the anterior chest developed ulceration with secondary infection and purulent discharge. She therefore underwent surgical treatment to achieve a definitive cure. Her family history was unremarkable. The patient's parents, siblings, and two sons were all unaffected, with no similar cutaneous lesions or symptoms observed across three generations.

Cutaneous examination revealed a large, irregular plaque (19.3 cm×5.6 cm) featuring densely distributed comedone-like openings on its surface spanning from the anterior chest to the posterior thoracic region (around T2-T5 intercostal spaces). Localized scars were observed over the plaque, but active ulceration or infection was not present upon examination. A separate 3.9 cm×1.8 cm comedone-like plaque with localized scarring was also identified in the inferomedial quadrant of the right breast. No lymphadenopathy, ocular, neurological, or skeletal involvement was detected. Routine hematological examination results were unremarkable.

Histopathological examination (Figure 1c) revealed mild hyperplastic squamous epithelium, as well as multiple dilated dermal cystic cavities filled with keratinous material in the epidermis.

In accordance with the patient's preference for definitive resolution of the extensive lesions, comprehensive management was performed, including comb flap reconstruction (Figure 1d), super tension-relieving sutures (Figure 1e), and postoperative VSD (Figure 1f). Intraoperatively, isolated lesions were directly excised and sutured primarily. For high-tension areas spanning the anterior chest to the scapular line, comb flap reconstruction was employed to preserve tissue integrity, followed by super tension-relieving sutures to minimize scar widening. Given the history of recurrent infection, VSD therapy was initiated with continuous suction post-surgery, and the VSD device was removed on postoperative day 6. Sutures were removed on postoperative day 16, revealing well-approximated wound edges. At the 6-month follow-up, the patient reported high satisfaction with the efficacy.

WES performed on the lesional tissue sample (Supplementary material) did not detect any rare or deleterious mutations in previously reported genes (NEK9, KRT10, ABCA12, and FGFR2).

Discussion

Nevus comedonicus (NC) may occur as part of nevus comedonicus syndrome (NCS), a neurocutaneous disorder characterized by abnormalities of skin, skeletal, ocular, and central nervous systems.[5] The present case exhibited no systemic involvement, consistent with isolated NC. Therefore, during diagnosis, dermatologists must perform comprehensive history and physical examination to prevent missed and misdiagnosed cases. The differential diagnosis for nevus comedonicus includes epidermal nevi, histoid leprosy, Favre-Racouchot disease, lichen sclerosus and familial dyskeratotic comedones, among others. Histopathological examination remains the gold standard for definitive diagnosis.

Despite advancements in clinical recognition, the etiopathogenesis of NC remains incompletely elucidated. Current hypotheses primarily implicate postzygotic mosaicism involving somatic mutations in NEK9 and KRT10, FGFR2 genes, or potential germline defects.[6–8] Previous genetic molecular insights of NC have inspired exploration of novel biologic therapies, such as adalimumab, an inhibitor of TNF-alpha.[9] While the rarity of NC limits research into its pathogenic genes, further molecular investigation remains essential for developing novel therapeutic strategies.

The therapeutic management of NC poses clinical challenges. Current treatment options include excision, dermabrasion, cryotherapy, coagulation, extraction of comedones, topical agents, and ultrapulse CO_2 laser.[10] Conventional treatments, such as topical retinoids and chemical peels, often demonstrate limited efficacy, necessitating procedures like laser ablation, dermabrasion, or surgical excision for recalcitrant lesions. Emerging strategies encompass photodynamic therapy and biologic agents. However, watchful waiting remains appropriate for asymptomatic cases. The presence of inflammation frequently dictates the need for intervention, and surgical approaches typically demonstrate superior efficacy. While subcutaneous tissue expanders enable extensive lesion management, their use is constrained by prolonged treatment duration and demands on patient compliance.

This combined technique offers more options for dermatologists managing extensive NC and other large cutaneous lesions. Comb flap reconstruction allows intraoperative dynamic assessment of skin tension by sequentially anchoring the skin. Following wide excision of lesions in the high-tension chest wall region, the application of super tension-relieving sutures promotes wound healing, minimizes tension, and reduces the risk of scar widening. Furthermore, the use of vacuum sealing drainage was indicated by the history of recurrent infections and substantial local tension, effectively providing postoperative drainage and reducing the burden of wound care.

Conclusion

This case illustrates successful single-stage excision of extensive, refractory nevus comedonicus (NC), achieving reduction of tissue tension and infection risks. Although non-surgical treatments are necessary for many NC cases, we propose that this technique merits consideration as an effective treatment of extensive NC following evaluation by dermatologic surgeons.

Data Sharing Statement

Data sharing does not apply to this article.

Ethical Approval

Informed consent was obtained from the patient for publication. Institutional approval for publishing the case details was obtained from the Ethics Committee of The First Affiliated Hospital of Anhui Medical University.

Consent to Publish

Consent was obtained from the participant for publication in this report and any accompanying images.

Acknowledgments

The author(s) thank the patient in this work for her permission.

Funding

There is no funding associated with this work.

Disclosure

All authors have no conflicts of interest.

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