

A Case of Surgical Excision of Nevus Comedonicus

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Abstract

Nevus comedonicus (NC) is a hamartoma of epithelial origin that is centered on a pilosebaceous unit. NC presents with grouped follicular openings and plugs that are similar in appearance to comedones. We introduce a case of congenital nevus comedonicus, involving a large area of the trunk with multiple abscesses. A 26-year-old man who was referred to our hospital presented with a nevus over half of his trunk. The nevus measured 15 cm×6 cm and was limited to the dermis. After 4 staged resection procedures, the overall appearance was greatly improved except for a hypertrophic scar. The histopathological results showed aggregation of dilated follicular infundibula with prominent keratotic plugging and absent or rudimentary sebaceous elements. The lesion was diagnosed as nevus comedonicus. This case shows that when repeated inflammation causes morphological changes to occur as complications of nevus comedonicus, the potential for other treatment options for recurrences should be considered. We suggest surgical excision as the treatment of choice.

Keywords: Nevus, Intra-dermal, Nevus, Surgical procedures, Operational

Introduction

Nevus comedonicus (NC) is a hamartoma of epithelial origin that is centered on a pilosebaceous unit. NC presents with grouped follicular openings and plugs that are similar in appearance to comedones. NC can appear on any part of the body, but has a predilection for the face and neck areas [1]. We introduce a rare case of NC, involving a large area of the trunk with multiple abscesses. By introducing this case, we suggest that surgical excision is the treatment of choice for NC.

Case

A 26-year-old man who was referred to our hospital presented with a nevus over half of his trunk (Fig. 1). The nevus was present from birth and progressively worsened with the onset of puberty. He had surgical incision and drainage for abscess formations in the nevus twice before presenting to our hospital. However, he still complained of pain and malodorous discharge from the lesion. On physical examination, the nevus had purulent drainage, evidence of residual scarring, and grouped, dilated, follicular openings with plugs resembling black comedones.

The patient's health status was otherwise normal. He had no congenital abnormalities or internal organ involvement. Signs of infection were observed, so we prescribed empiric antibiotic treatment.

Once the signs of infection subsided, we recommend flap surgery after total excision, but he refused based on potential scarring and donor site morbidity. He also wanted to minimize his length of stay in hospital. Finally, he decided to proceed with



Case Report

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Fig. 1. Initial appearance of the lesion including multiple abscesses with grouped follicular openings and plugs covering half of the trunk.

serial staged excision.

He was admitted to the Plastic Surgery unit and underwent a surgical excision under general anesthesia. The nevus measured 15 cm × 6 cm and was limited to the dermis (Fig. 2).

The histopathological results showed aggregation of dilated follicular infundibula with prominent keratotic plugging and absent or rudimentary sebaceous elements. Additionally, there were large-grouped, close-set follicles of various sizes and marked acanthotic and papillomatous epidermal hyperplasia with hyperkeratosis (Fig. 3).

The patient underwent two additional surgical procedures under general anesthesia during which most of the lesion was removed. Some scattered remnants of the lesion were additionally removed using local anesthesia. His overall hospitalization period was 11.4 days.

After serial staged resection procedures, the overall appearance was greatly improved except for a hypertrophic scar. The scar was subsequently treated with silicone gel (Fig. 4). At his 10-month follow-up visit, the lesion was well-healed. He had no complications and no other remnant or newly developed lesions were found.



Fig. 2. The NC specimen measuring 15 cm × 6 cm.

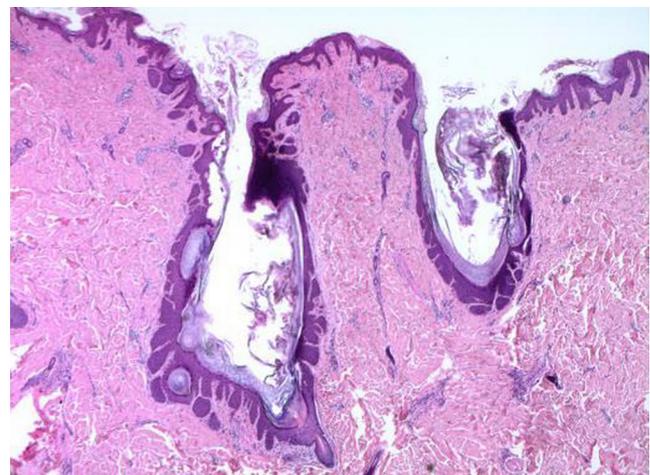


Fig. 3. Histopathologic findings of nevus comedonicus show marked acanthotic and papillomatous epidermal hyperplasia with hyperkeratosis, and invaginations with a laminated keratin plug (H&E, ×40).

Discussion

Kofmann initially described nevus comedonicus in 1895 [2]. Currently, it's estimated prevalence rate is 1 in 45,000 with no age or gender predisposition [3]. Nevus comedonicus clinically presents as expanded follicular ostia with a linear distribution along the lines of Blaschko. Ostia contain lamellar keratinocytic material similar to comedones [2]. Nevus comedonicus can appear in most of the body, including the trunk, face, scalp, neck, genitalia, soles, palms, or upper limbs. However, it rarely occurs bilaterally [4].

The differential diagnosis for nevus comedonicus should include dermatological disease presenting with lesions that resemble comedonal acne, neonatal acne, chloracne, prokera-



Fig. 4. Two-month postoperative clinical photo showing the hypertrophic scar and no evidence of recurrence.

otic ostial nevi, and familial dyskeratotic comedones [5,6]. Engbers et al. reported rare cases of NC that occurred simultaneously with other internal disorders [1]. He termed this condition nevus comedonicus syndrome. Nevus comedonicus syndrome (NCS) belongs to the larger category of epidermal nevus syndromes, wherein epidermal nevi appear concurrently with other organ disorders, such as skeletal abnormalities (scoliosis and spina bifida), neurological deficits (epilepsy and electrocardiogram abnormalities), and ophthalmic problems (congenital cataracts) [1]. The physical examination and laboratory test results in this case were not consistent with nevus comedonicus syndrome.

Nevus comedonicus is classified into two types according to its clinical features [7]. The first type is non-pyogenic NC with acne-like characteristics, and the second type is NC with cysts, papules, pustules, or abscesses that are associated with morphological changes. This case represents the latter type of NC,

with multiple cysts and abscesses.

Nevus comedonicus should be treated not only because of the potential cosmetic complications, but also to prevent infectious complications and residual scarring. However, as with other epidermal nevi, treatment is challenging. Retinoids can be useful, but they do not achieve complete resolution and relapses are common. Procedures such as shaving, extraction, and dermabrasion can be helpful temporarily, but these are not curative. Sometimes ambiguous borders of the nevus lead to difficult 'complete surgical excisions,' but surgery seems to be the most appropriate treatment. However, patients with large affected areas require staged surgical excision or skin grafting. The extent and location of NC can often limit the utility of surgical management [6].

Conclusion

There are several options for the treatment of NC, but considering the potential for recurrences, we suggest surgical excision as the treatment of choice.

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