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A Case of a Giant Congenital Melanocytic Nevus Treated by Curettage with the Application of Cultured Epidermal Autografts before 6 Months of Age

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Summary: Curettage is common in the treatment of a giant congenital melanocytic nevus (GCMN) in infants and should generally be performed before 6 months of age. Post-curettage retarded epithelialization often interferes with the ability to perform multiple operations within a short interval, and thus, it is difficult to treat large lesions in the neonatal period. We herein report a case of a GCMN comprising 20% of the total body surface area, which required multi-stage curettage, in which a cultured epithelial autograft was used to promote epithelialization of the post-curettage wound. The patient was a 1-month-old boy with a GCMN in his head, neck, chest, back, buttock, left upper arm, and a few satellite lesions. A four-stage operation was performed between 3 and 6 months of age; the cultured epithelial autograft took well after each operation, and complete epithelialization was observed at postoperative days 20, 23, 27, and 12, respectively. Seven months after the last surgery, hypertrophic scar formation was only observed in a small area of the left upper arm without axillary contracture. The color of the treated area improved, except for slight partial re-pigmentation. A skin biopsy was obtained from the re-pigmented area. The results demonstrated that nevus cells remained in the basal layer of the epidermis, hair follicles, and deep layer of the remaining dermis, suggesting that the recurrent nevus cells in the regenerated epidermis migrated from hair follicles. We conclude that the combination of curettage and the application of a cultured epithelial autograft is a promising option for GCMN treatment. (*Plast Reconstr Surg Glob Open* 2021;9:e3600; doi: [10.1097/GOX.0000000000003600](https://doi.org/10.1097/GOX.0000000000003600); Published online 25 May 2021.)

A giant congenital melanocytic nevus (GCMN) is a type of large melanocytic nevus that is present at birth. The most common definition is a nevus that is >20 cm in diameter in adults, and ≥6 cm on the body or ≥9 cm on the head of neonates.¹ Surgical excision is recommended to prevent the incidence of malignant melanoma as a result of a GCMN.¹ However, melanoma can occur from the remaining nevus cells in the subcutaneous or deeper tissue, even after the excision of skin lesions,² and complete removal of nevus cells is difficult. Furthermore, complete removal of a large skin lesion often causes functional and cosmetic disorders.

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Curettage is another option for treating GCMN, described by Moss³ as a resection technique to remove the superficial layer of the GCMN with a sharp curette. When the nevus is too large to excise, curettage offers a good alternative to surgical excision. Curettage should be performed before 6 months of age, as nevus cells lie mainly in the upper dermis in newborns before migrating to deeper tissues with growth.³ Multiple operations are needed in the neonatal period to complete the curettage of large GCMN; however, this is made particularly difficult by delayed epithelialization.¹

In Japan, cultured epidermal autografts (CEA; JACE, Japan Tissue Engineering Co., Ltd., Gamagori, Japan), prepared using Green's technique,⁴ have been approved and covered by public health insurance for the treatment of GCMN patients since 2016. The application of CEA after curettage is expected to promote epithelialization and reduce hypertrophic scarring.⁵⁻⁸ Here, we present a case of a GCMN covering 20% of the total body surface area (TBSA) of an infant in whom the GCMN was completely curetted with application of CEA before the age of 6 months using a 4-stage surgery with an interval of approximately 1 month.

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Fig. 1. Black and dark brown melanocytic nevi are located in the neck, chest, and back regions, with a few satellite lesions.

CASE PRESENTATION

A 1-month-old boy had a GCMN covering 20% of his TBSA, including on his neck, chest, back, buttock, left upper arm, and a few satellite lesions (Fig. 1). We planned a staged curettage in combination with CEA application.

At the age of 2 months, full-thickness skin (size: ~1 cm²) was harvested from his left groin to prepare the CEA under local anesthesia. After preparing the CEA for 3 weeks, the first curettage of the anterior chest and neck was performed at the age of 3 months. The superficial part of the nevi above the cleavage plane was removed with a curette, and the remaining nevus was removed at the same layer using the hydro-surgery system (Versajet II; Smith & Nephew, Tokyo, Japan) and a CO₂ laser (AcuPulse; Lumenis, Tokyo, Japan). The CEA was then applied to the resected wound surface and covered with a silicone-faced wound dressing (SI-Mesh and SI-AID; ALCARE Co., Ltd., Tokyo, Japan) and tie-over dressing. The tie-over dressing was removed 7 days postoperatively. The remaining nevus of the left shoulder and arm was removed at 4 months, that of the occipital region and back was removed at 5 months (Fig. 2), and that of the buttock was removed at 6 months in the same way. The CEA took well, and the healing time of the abovementioned areas,⁸ which was the time to complete epithelialization without any dressing, was 20, 23, 27, and 12 days, respectively. Finally, the whole lesion of the nevus was completely treated before 6 months of age.

Seven months after the last surgery, a skin biopsy was obtained from the lesion with re-pigmentation in the back, and hematoxylin & eosin (HE) stained sections and immuno-stained sections with anti-SOX10 antibody were prepared. The epidermis regenerated well, with stratified



Fig. 2. The GCMN on the back was removed, and cultured epidermal autografts were applied on the curetted wound during the third operation.

keratinocytes and rete ridges. SOX10-positive cells were observed in the basal layer of the epidermis and hair follicles. In the dermis, dense SOX10-positive cell nests were located in the deeper layer, which were considered to be the remaining nevus tissue after curettage, while no SOX10-positive cells were observed in the superficial dermal layer (Fig. 3).

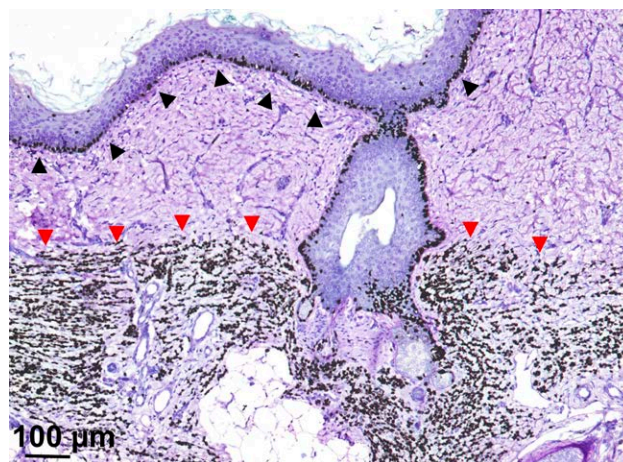


Fig. 3. SOX10-immunostaining section of the skin biopsy harvested 7 months after the last surgery. SOX10-positive cells were distributed in the basal layer of the epidermis (black arrow heads), hair follicles, and deep layer of the remaining dermis (red arrow heads). No nevus cells were observed in the superficial dermis layer above the red arrow heads.

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Fig. 4. Gross appearance at postoperative 1 year and 4 months. Although the spotty re-pigmentation and hair growth was partially observed, the color of treated area was improved and hypertrophic scar formation was trivial. No axillary contracture and no limitation of shoulder motion were present.

One year and 4 months after the last surgery, the color of the treated area was improved, with the exceptions of the spotty re-pigmentation and secondary hair growth. Hypertrophic scar formation was only observed in the left upper arm without axillary contracture (Fig. 4).

DISCUSSION

Curettage has been recommended to be started as early as possible. Nevus cells migrate to deeper tissues with age, making it difficult to remove them efficiently. Moreover, neonates and infants have lower risk of postoperative hypertrophic scar formation than children. Whang reported a case series of CEA grafting after curettage and/or erbium:yttrium-aluminum-garnet (Er:YAG) ablation and its effectiveness in terms of healing time.⁸ The mean wound healing time was 37.0 ± 21.7 days in the CEA group, which was significantly shorter than the 76.3 ± 48.4 days in the non-CEA group. In our case, the mean wound healing time was 20.5 days, which allowed the 4-stage operation with a 1-month interval to complete before the patient turned 6 months old. The color tone was improved, and hypertrophic scar formation was only observed in a limited area, which may indicate the effectiveness of both the early-start treatment strategy and the application of CEA.⁵

Biopsy of the lesion from the re-pigmented area at postoperative 7 months revealed that SOX10-positive cells remained in the basal layer of the epidermis, hair follicles, and deep layer of the remaining dermis. The localization of the nevus cells suggests that the recurrent nevus cells in the regenerated epidermis migrated via hair follicles.⁹ Kishi also reported rapid re-pigmentation after curettage

and dermabrasion around the hair follicles.^{5,10} Destruction of the remaining nevus cells in hair follicles with additional laser hair-removal treatment after curettage could be useful to prevent re-pigmentation. The histological studies also suggest the continued risk of malignancy. However, the risk could be lessened by a reduction in the total number of superficial nevus cells, although it has not been clarified.

In conclusion, the combination of curettage and CEA is a promising option for GCMN treatment. We will follow this patient carefully to observe the remaining nevus cells over a prolonged period.

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PATIENT CONSENT

The parents of the patient provided written consent for the use of the images.

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