



Aesthetic Reconstruction of Congenital Cerebriform Intradermal Nevus by Sequential Tissue Expansion at an Early Age: Ten-year Follow-up

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Cerebriform intradermal nevus (CIN) is a congenital tumoral abnormality of the scalp. The incidence of development of malignant melanoma in CIN can rise up to 10%. Surgical excision of the lesion is the sole treatment modality for CIN. In this case, we present the 10-year follow-up of a 45-day-old infant with congenital CIN covering 75% of his scalp who had scalp reconstruction by sequential tissue expansion. Sequential tissue expansion processes with rectangular-shaped expanders of 450, 400, and 250 ml in size, respectively, were applied over an eight-month period. The interval between the two procedures was two weeks. The tissue expansion technique is a useful option for hair-bearing scalp reconstruction. This technique can be used sequentially or gradually. In order to obtain homogenous follicle distribution, the rest of the scalp should be expanded as much as possible. Careful planning in large defects helps prevent complications. In this case, we applied a sequential tissue expansion procedure when the patient was 45 days old, and the expansion period was continued for eight months. This very early age is a unique case for tissue expansion applications when compared with examples in the literature. On the other hand, we want to underline the aesthetic result of this case rather than the reconstructive method. Reconstruction of huge scalp defects by re-expansion (sequential expansion) and leaving the expander beneath the expanded flap until the sequential procedure decrease flap contracture reduces the required number of operations, and produces better aesthetic outcomes.

Key Words: Cerebriform Intradermal Nevus; Tissue Expansion; Sequential Tissue Expansion.

Ardışık Doku Genişletme İşlemi ile Konjenital Serebriform İntradermal Nevüsün Erken Yaşta Estetik Rekonstrüksiyonu: 0n Yıllık Takip

Serebriform intradermal nevus skalpte görülen konjenital tümoral bir anomalidir. Malign melanoma gelişme insidansı %10'a kadar ulaşabilmektedir. Bu lezyon için cerrahi eksizyon tek tedavi seçeneğidir. Bu vakada, 45 günlük bir bebeğin kafa derisinin %75'ini kaplayan konjenital serebriform intradermal nevüsün ardışık doku genişletme metodu ile tedavisinin 10 yıllık takip sonuçlarını sunmaktayız. Ardışık doku genişletme işlemi sırasıyla 450, 400, 250 ml'lik dikdörtgen şekilli doku genişleticilerle sekiz aylık bir sürede uygulandı. Sözkonusu bu üç doku genişletme işlemi arasındaki bekleme süresi 2 hafta idi. Doku genişletme tekniği saçlı deri rekonstrüksiyonunda oldukça etkin bir seçenektir. Bu teknik ardışık veya aşamalı olarak uygulanabilir. Homojen saç folikülü dağılımı elde etmek amacıyla, kalan saçlı deri elverdiği ölçüde genişletilebilir. Geniş defektlerin rekonstrüksiyonunda dikkatli planlama, komplikasyonların önlenmesinde yardımcıdır. Bu vakada, 45 günlük çocuk hastaya ardışık doku genişletme işlemi sekiz ay boyunca uyguladık. Bu vaka, literatürdeki diğer vakalarla kıyaslandığında bu kadar erken yaşta doku genişletici uygulanan tek vakadır. Diğer taraftan, rekonstrüksiyon metodundan ziyade on yıllık estetik sonucu vurgulamak istemekteyiz. Geniş defektlerin rekonstrüksiyonunda ardışık doku genişletme işlemi ve genişleticinin ikinci doku genişletme işlemi başlayıncaya kadar flebin altında tutulması, flep kontraktürünü azaltarak ameliyat sayısını düşürmekte ve daha iyi estetik sonuçlar sağlamaktadır.

Anahtar Kelimeler: Serebriform İntradermal Nevüs; Doku Genişletme; Ardışık Doku Genişletme.

Introduction

Cerebriform intradermal nevus (CIN) is a congenital abnormality of the scalp that is characterized by the folding, thickening, and overgrowth of skin in a brain

or gyrata shape.^{1,2} CIN is a rare form of cutis verticis gyrata, and is also known as pseudo cutis verticis gyrata.² CIN is not associated with systemic diseases, whereas cutis verticis gyrata is usually accompanied by endocrinological or neurological diseases.^{1,3} CIN is a

potential risk factor for the development of malignant melanoma, and the incidence can rise up to over 10%.^{1,4} Due to psychological complications, aesthetic considerations, and the probability of developing malignancies, surgical excision of the lesion is required. Primary closure is possible after surgical excision of small or localized lesions, while larger lesions require more complicated reconstructive methods, such as tissue expansion applications, prior to excision.⁵⁻⁷

In order to replace the hair-bearing scalp defect, the use of local tissue of similar quality and structure is optimal. For reconstruction of large lesions or defects, tissue expansion presents a useful option.^{7,8} The tissue expansion technique can be used sequentially or gradually.

The current case presents the 10-year follow-up of a 45-day-old male who underwent scalp reconstruction by sequential tissue expansion to remove congenital intradermal cerebriform nevus covering 75% of his scalp.

Case

A 45-day-old male patient presented at our clinic with an extensive tumoral scalp mass present since birth. Physical examination revealed a dark-pigmented, brain (gyrus)-shaped tumoral mass covering 75% of the scalp (Fig. 1). Pathologic investigation of the incisional biopsy resulted in the diagnosis of congenital cerebriform intradermal nevus and the patient was hospitalized.



Figure 1. Preoperative view of 45-day-old infant.

Sequential tissue expansion processes with rectangular-shaped expanders 450, 400, and 250 ml in size, respectively, were applied over an 8-month period. The patient was 45 days of age when the first tissue

expansion process was applied. Incisions were made along the border of the lesion. Healthy scalp tissue was left in place. Inflation of the tissue expander commenced on postoperative day fourteen. Expanders were inflated twice weekly, and 20% of defective tissue was excised on each occasion. At the end of each process, reconstruction was performed by obtaining healthy skin, and replacing an average of 20% of the defective region (Figs. 2a and 2b). While expanded flap transferring to defective region, at the same time a new tissue expander was inserted under the flap for the second expansion process. The waiting time for the new expansion period was two weeks.



Figure 2a. View of after second expansion.



Figure 2b. At the end of the last expansion period.

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Although prosthesis infection occurred in the final session, there was no need to remove the prosthesis. After the third application of tissue expander, the entire scalp was reconstructed with autogenous scalp tissue. Permanent frontal nerve palsy and minimal asymmetry due to dog-ear deformity of expanded flap pedicle developed (Fig. 3a).

The patient was again referred to our clinic for scarring of the scalp at the age of 10. Physical examination revealed a linear scar in fronto-temporo-occipital direction (Figs. 3a and 3b). The scar tissue presented as widened and atrophic, so scar revision was planned.



Figure 3a. At the age of 10, anterior view.



Figure 3b Right side view when he was 10 years old.

Discussion

In this case, we applied a sequential tissue expansion procedure when the patient was 45 days old, and the expansion period was continued for eight months. The very early age of the patient presents a unique case for

tissue expansion application in comparison with examples in the literature. There are no good screening techniques for detection of the lesions and the only accepted treatment modality is surgical excision without delay.^{1,5} In the literature, the probability of malignant transformation in giant melanocytic lesions, such as cerebriform intradermal nevus is, in contrast, most likely to occur in the first decade. The formidable appearance of the scalp was very stressful condition for the parents. The insistence of the parents was the other indication that can be a relative indication for planning the surgery. We have not met any complication; despite this surgery series were very heavy for an infant.

Although extended scalp defects may be closed by traditional methods, such as local flaps and skin grafts, these methods cannot provide adequate tissue and optimal aesthetic results.⁷⁻⁹ The donor site containing the most similar and aesthetically appropriate tissue for scalp defect reconstruction is the rest of the scalp itself. The use of tissue expanders enables maximum use of the remainder of the scalp in the closure of scalp deformities.^{5,7,8} Careful planning prior to treatment of large defects helps prevent complication. Aggressive expansion in high-risk anatomic regions such as scar tissue may cause ischemic ulcerations and increase product failure.⁹

Whatever its basic pathology, reconstruction of large scalp defects using early re-expansion (sequential expansion) decreases flap contracture and the number of required surgical procedures, resulting in a better cosmetic outcome.

In conclusion, a sequential tissue expansion method would be preferable when compared to aggressive expansion methods that may cause complications, non-aesthetic reconstruction, and psychological disorders. According to our clinical experience, in the reconstruction of scalp defects, tissue expansion is easier in earlier ages. The sequential procedure decreases flap contracture, reduces the required number of operations, and produces better aesthetic outcomes.

In addition, we presented this unique case to share our experience about sequential tissue expansion method in earliest age with long term follow-up. Although early tissue expansion is better aesthetic results, we do not advocate performing early surgery unless malignant diagnosis.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Conflict of interest

There is no conflict of interest to declare.

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