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Original Research

Parallel Hatching: A New Method of Hand Deformity Surgery in Epidermolysis Bullosa

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Purpose: In dystrophic epidermolysis bullosa (EB), progressive blistering, scarring, and fusing of digits lead to mitten-hand and pseudosyndactyly in early childhood, inflicting psychosocial stress and low self-esteem in EB patients. Treatment is a challenge in the hand surgery field because of the recurring nature of this inherited disease. We aim to evaluate the results of our surgery method for the hand deformities in EB patients.

Methods: We retrospectively reviewed the chart of the EB patients from 2013 to 2023 and included the patients that had the surgery by the same surgeon with the technique of parallel transverse hatching incisions in their hands with using amnion as a dressing and no grafts. The results of the surgery were assessed by using the pseudosyndactyly grading. To compare the data, t test and χ^2 test were used for variables as appropriate.

Results: We presented the results of our surgery method for hand deformities in EB patients. Twenty hands (11 right and 9 left hands) from 11 patients were included. The mean ± SD of grades before and after surgery in all of the patients were 2.2 ± 0.9 and 1.2 ± 1.2, respectively. The differences in grades from baseline in both left and right hands were significant (*P*-values of .009 and .001, respectively).

Conclusions: In this method, we use parallel transverse incisions in epidermis and dermis of the palmar interphalanges without grafting. This will limit the trauma imposed on EB patients who are susceptible to reblistering and poor wound healing. Our patients demonstrated improvement in the function and appearance of their hands with up to 3 years of follow-up.

Type of study/level of evidence: Therapeutic, III.

Epidermolysis bullosa (EB) is a congenital, blistering disorder triggered by minor mechanical trauma which has a prevalence of approximately 8–10 cases per million births. It has four major types including simplex, junctional, dystrophic, and Kindler syndrome.[1,2]

One of the most disabling complications of EB is hand deformities caused by repetitive trauma to the hands because of daily usage and consequential blistering and scarring in these patients. Hand deformities range from pseudosyndactyly to flexion contractures and mitten and cocoon hands in their severe forms. Given the high psychosocial impact of function and appearance of the hands on children's self-esteem, management of this condition is important.[3] Hand deformities management has a high rate of recurrence and failed results because of the nature of the disease. Different surgical methods are used to release the contractures and achieve successful epithelialization. Various methods are used to cover the degloved skin such as allogenic skin graft, dermal graft, and bioengineered skin equivalents.[3,4] In our center, EB patients with hand contractures undergo a specific method of surgery with transverse parallel hatching incisions and further use of the human amniotic membrane dressing.

An amniotic membrane is a thin, complex structure composed of different layers of epithelium and fibroblasts.[5] Human amniotic membrane is a natural scaffold with low immunogenicity and wide availability and has been used as a wound dressing in different wounds. It has anti-inflammatory effects and produces various

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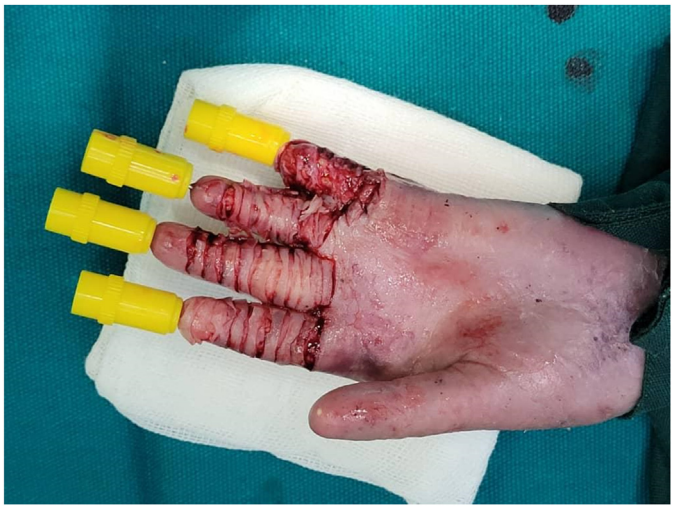

Figure 1. The parallel hatching method. Small parallel transverse incisions are made in the palmar surface of the digits of the hand.

cytokines and growth factors that promote wound healing.[6] In this study, we aimed to evaluate the healing process of our patients by a visual grading introduced by Colville.[7]

Materials and Methods

Patient selection

We retrospectively reviewed the chart of the EB patients admitted in our hospital from 2013 to 2023. We included the EB patients that were operated on their hands for the first time by the same surgeon with the technique of parallel transverse hatching incisions with amnion as the dressing and no grafts. Moreover, the patients should have had two photographs of their operated hand in their medical record with complete data and history between these two photographs. The patients that did not meet our inclusion criteria were excluded.

Surgery technique

Anesthesia

As EB patients are prone to oral and airway blisters, we used both intravenous midazolam and ketamine as well as inhaled isoflurane gas as anesthesia with a gauze under face mask instead of intubation to reduce further damage.

Hand surgery

The contracture of the hand was first released by making transverse mid palmar incisions and syndactyly was enhanced by using the blunt side of scalpel or a blunt-tipped delicate dissector in the web spaces.

Flexion contractures of the fingers were released by parallel transverse hatching incisions at the level of the distal palmar crease and interphalangeal joints; the incisions were made in the epidermis and dermis by scalpel with the avoidance of the adipose tissue (hypodermis). Each parallel incision with 2–3 mm distance from each other. The neurovascular bundles were identified to avoid further damage but there were no exploration or manipulation of nerves and tendons. Finger extension was achieved by external manipulation which is performed by gently extending the fingers until a neutral position is achieved. Longitudinal Kirschner wires (Smith & Nephew Company, Memphis, TN) of 0.8–1 mm were inserted after contracture release to maintain interphalangeal joints extension. The representative picture of the parallel hatching is demonstrated in Figure 1. Then, hands were dressed with amnion dressing. Dressings were changed in operating room every week thereafter under sedation until 4 weeks post operation. Moreover, Kirschner wires were removed after 3 weeks post operation.

Assessment of results

The results of the surgery were assessed using the pseudosyndactyly grading proposed by Colville[7] in 1989: no fusion as grade 0, fusion to the proximal interphalangeal joint as grade 1, fusion to the distal interphalangeal joint as grade 2, and fusion to the tip of the digit as grade 3.

Limitation of thumb adduction was considered to be grade 1, limitation of thumb abduction without the thumb overlying the palm to be grade 2, and if the thumb was held overlying the palm to be grade 3.

Statistical analysis

Statistical evaluation was carried out using SPSS v26 (IBM Corp., USA). Kolmogorov–Smirnov statistics was used for checking the normal distribution assumption of continuous variables. Quantitative variables are presented as mean with SD and qualitative variables are presented as number and percentages. To compare the data, t test and χ^2 test were used for variables as appropriate. $P < .05$ was considered as statistically significant.

Ethical considerations

All of the authors ensured that the patients' anonymity was protected and verified that any experimental investigation with human subjects reported in the manuscript was performed with written informed consent. All the guidelines for experimental investigation with human subjects required by our institution and the ethical standards of the Helsinki Declaration of 1975 were respected.

Results

We retrospectively reviewed EB patients with hand deformities that were operated on with hatching method and amnion from 2010 to 2023. Twenty hands (11 right and 9 left hands) from 11 patients were included. The characteristics and studied variables of them are presented in Table 1, individually.

Most of our patients were women and the mean ± SD of age of the patients were 11.2 ± 6.3 years. The follow up time of our patients was different based on their available medical records. However, the mean ± SD of follow up duration was 13.25 ± 10.83 months.

The mean ± SD of the grades before and after surgery in all of the patients were 2.2 ± 0.9 and 1.2 ± 1.2, respectively. The mean difference between the grades was 1.0 ± 0.8. The grades before and after surgery and their differences were not significantly different between the right and left hands (*P* values of .923, .664, and .586, respectively) (Table 2). The changes in the grades of before surgery compared to after surgery are presented as a parallel graph in Figure 2. Moreover, one sample t test showed that the differences in grades from baseline in both left and right hands were significant (*P* values of .009 and .001, respectively) (Table 3). Figure 3 shows the consecutive photos of a patient that were operated on his right hand as representatives of our technique.

Table 1
The Characteristics of Our Epidermolysis Bullosa Patients Presented Individually

Patient No.	Age (y)	Sex	Operated Hand	Grade Before Surgery	Grade After Surgery	Difference in the Grades	Follow-up Time (mo)
1	12	M	R	2	0	2	2
2	20	F	R	1	0	1	3
3	20	F	L	1	0	1	3
4	5	F	R	3	1	2	5
5	5	F	L	3	1	2	5
6	10	M	R	3	3	0	9
7	10	M	L	3	3	0	9
8	5	F	R	1	1	0	10
9	5	F	L	1	1	0	10
10	10	F	R	2	0	2	11
11	9	F	R	2	0	2	11
12	9	F	L	2	0	2	11
13	5	F	R	3	2	1	11
14	5	F	L	3	2	1	11
15	20	M	R	3	3	0	13
16	20	M	L	3	3	0	13
17	8	F	R	3	2	1	15
18	6	M	L	3	2	1	37
19	20	M	R	1	0	1	38
20	20	M	L	1	0	1	38

Table 2
The Summary of Data of the Patients Categorized by Right or Left Hand

Variables	Total (n = 20)	Right (n = 11)	Left (n = 9)	*P* Value
Age, years, mean (SD)	11.2 (6.3)	11.3 (6.0)	11.1 (6.9)	.956
Gender, n (%)				.714
M	8 (40%)	4	4	
F	12 (60%)	7	5	
Before surgery grade, mean (SD)	2.2 (0.9)	2.2 (0.9)	2.2 (1.0)	.923
After surgery grade, mean (SD)	1.2 (1.2)	1.1 (1.2)	1.3 (1.2)	.664
Difference in the grades, mean (SD)	1.0 (0.8)	1.1 (0.8)	0.9 (0.8)	.586
Follow-up duration, mean (SD)	13.25 (10.8)	11.6 (9.6)	15.2 (13.0)	.488

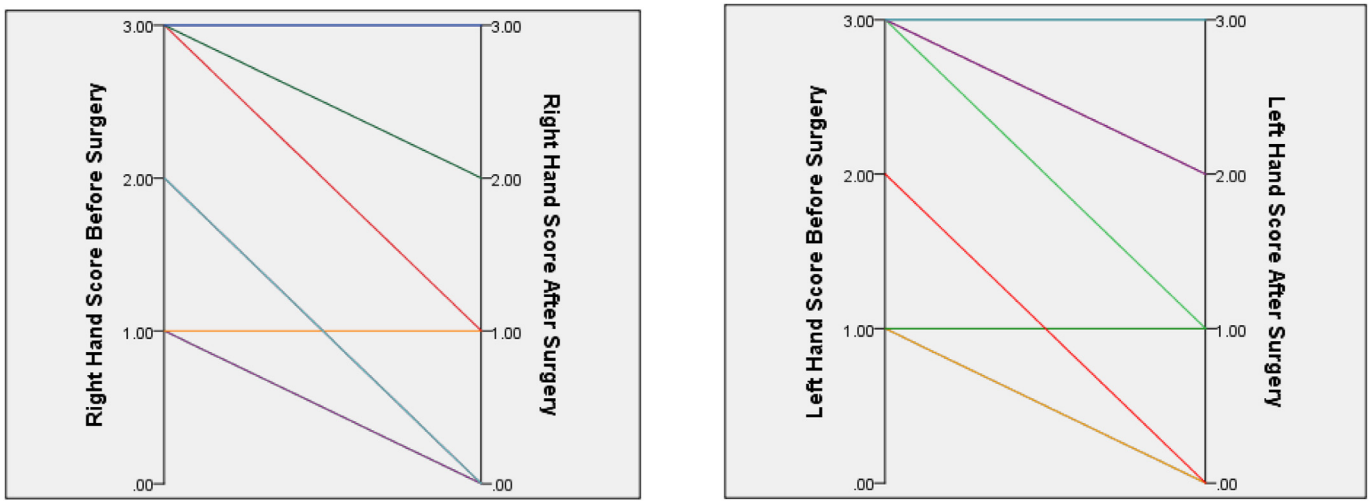

Figure 2. The parallel graph of grade changes in patients.

Discussion

In dystrophic EB, progressive blistering, scarring, and fusing of digits lead to mitten-hand and pseudosyndactyly in early childhood. These deformities of hand and the resultant disability lead to psychosocial stress and low self-esteem in EB patients.[3] Treatment is a challenge in hand surgery field because of the nature and recurrence of this inherited disease. In a study by Zhou et al[4] in 2020, they described their surgery method with removing the cocoon epidermis and separating the web spaces and palm adhesions. They used parallel vertical incisions and blunt dissection between the digits to the metacarpophalangeal joints and also, Z-plasty incisions at the flexion creases. It is noteworthy that large incisions, tendon readjustments, and flexor digitorum superficialis muscle cut were also performed.[4] Tuncer et al[3] operated on moderate cases with surgical release, autologous dermal grafts, and a

Table 3
Evaluation of the Improvement of Hand Deformities Using Differences of Grades of Right or Left Hand Using t Test

	t Test Value	Degrees of Freedom	Mean (SD)	*P* Value (Two-Tailed)	95% CI of the Difference	
					Lower	Upper
Grade difference of right hand	4.353	10	1.1 (0.8)	.001	0.53	1.65
Grade difference of left hand	3.411	8	0.9 (0.8)	.009	0.29	1.49

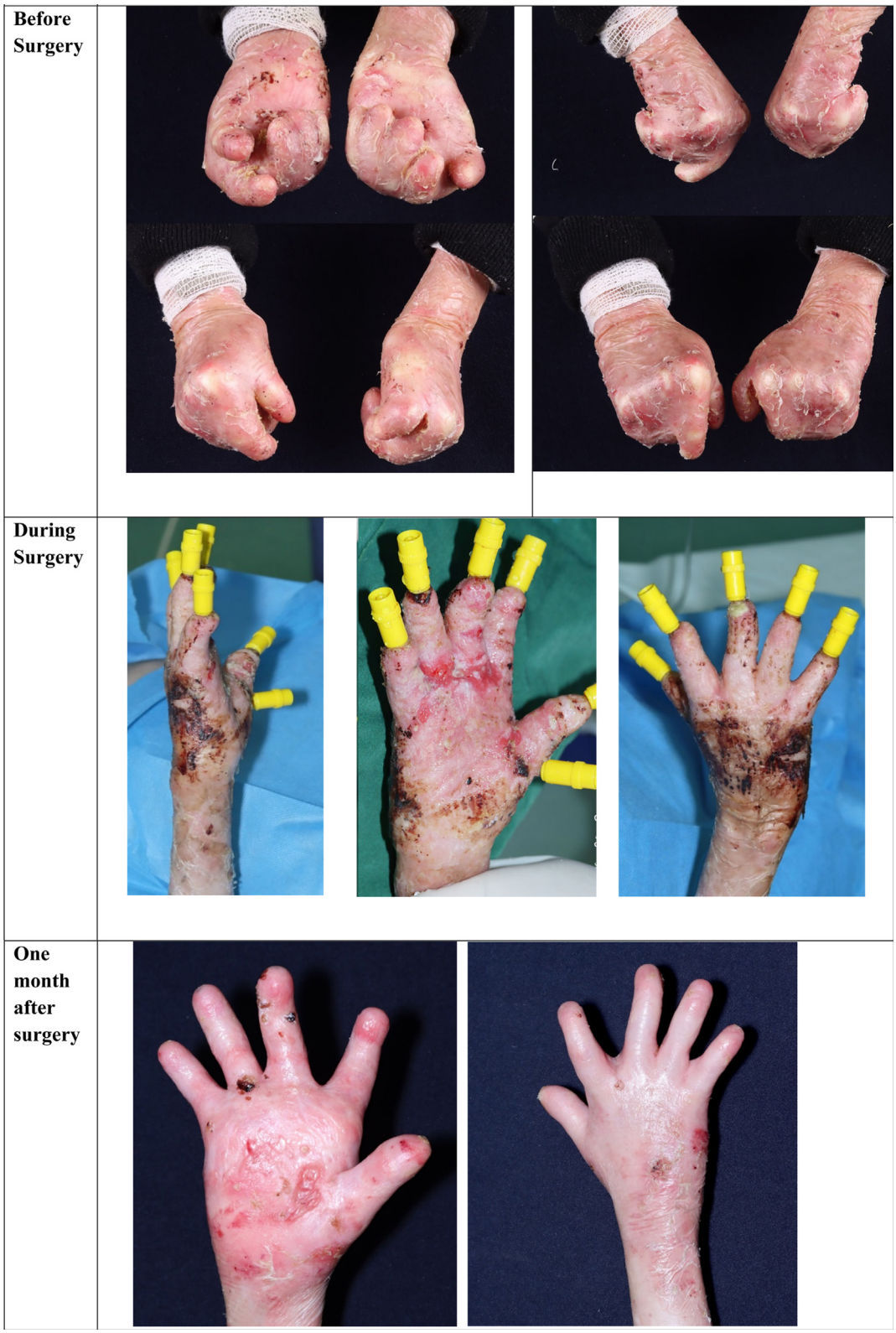

Figure 3. Consecutive photos of a patient who underwent an operation on his right hand.

nonadhesive nylon dressing. In a study by Luria et al[8] on four EB patients, they also cut the epidermis of the web spaces until reaching the subcutaneous fat and used transverse incisions in both mid palm and distal metacarpals with split-thickness skin graft. They reported that significant rehabilitation goals were met; however, recurrence was occurred to some extent after average follow-up of 2 years.[8] However, in our method, we used parallel transverse incisions with no large or deep incisions to minimize the trauma to the tissues. In addition, muscles and tendons were intact.

In 2022, Abboud et al[9] reviewed their method of surgery on 30 hands of EB patients and revealed function improvement in 57% of patients after more than 3 years. Their method was using palmar, metacarpophalangeal, and interphalangeal transverse incisions with avoiding the dermal layer to release the flexion contractures which is similar to our method. Contrarily, they used full-thickness skin grafts compared to our method that we used no grafts and only amnion dressing.[9]

In 1993, Vozdvizhensky et al[10] introduced a simple surgery technique with limited trauma by using transverse palmar incisions on the fingers of 19 EB patients. However, they reported the recurrence rate of 53% in 15 patients with at least 1 year of follow-up. Our patients did not have the same follow-up duration given its retrospective nature and we could not report the recurrence rate. However, three of our patients who had a follow-up duration of more than 3 years demonstrated improvement in their contractures and syndactyly, despite the progressive nature of this disease. Ciccarelli et al[11] suggested that skin grafting is not necessary to avoid more trauma in donor-site as well. They reported the same duration of approximately 2 years for requiring repeated surgery in seven procedures with skin grafts and 58 procedures without skin graft.[11]

Using skin grafts in EB patients has remained a profound challenge because of inadequate donor sites, poor wound healing, and reblistering of donor sites, autologous skin grafts inflict another burden on these patients. As a result, alloplastic and bioengineered materials were introduced. Tissue-engineered skin grafts in EB patients were not as beneficial as in their other applications. In a study using Apligraf (Organogenesis, Inc.) in nine chronic wounds of EB patients, seven wounds were still open after 4 months.[12] In another study using OrCel (Forticell Bioscience, Inc., New York, NY) in EB patients, there was no significant reduction in the healing time compared to its collagen component and to standard care.[13] Adding these results to their cost and difficulty in obtaining them has led to the limitation of their application in EB patients.[14]

Studies have shown favorable results of amnion in the ulcers of EB patients. It competes with various biological dressings, such as autologous or allogenic grafts, as a result of being affordable and less time-consuming.[15] Amnion has an extracellular matrix capable of providing moisture to the wound bed and proliferation of different cell lineages and releases different growth factors that help with angiogenesis and wound healing.[16–18] Using amnion as a biological dressing promotes fibroblast and keratinocyte migration and differentiation, which contributes to epithelium regeneration.[18–22]

In a study by Lo et al,[6] eight applications of amnion in EB patients with significant improvement based on visual analog scale score in half of them with no adverse effects and reblisterings in the grafted areas. Hasegawa et al[15] used amnion for intractable ulcers in three EB patients, and they repeated the procedure weekly for up to 10 weeks. As a result, wound conditions improved remarkably after treatment with amnion for 2–10 weeks in all the patients, resulting in total re-epithelization of the ulcers.[15]

Our study used amnion for a great healing properties and small parallel transverse incisions instead of deep incisions and no skin graft to limit trauma to the tissue. However, our study had limitations of retrospective studies. Second, the number of and time interval between follow-up visits differed between patients, as these were scheduled to coincide with clinic visits and patients' compliances. Moreover, given the rare nature of EB disease, we could only collect the data of a limited number of patients. These limitations made us unable to report any recurrence rate. Lastly, the lack of any control group in our study to compare the results is another limitation that warrants further controlled research to support our results.

Conflicts of Interest

No benefits in any form have been received or will be received related directly to this article.

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