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ORIGINAL STUDY

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Separation of congenital embryonic syndactylies in children: dorsal or volar flap? About a comparative retrospective series

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Congenital syndactylies account for 1 to 2 out of 2000 birth defects. Although several types of syndactylies exist, we only studied embryonic syndactylies. The goal of our study was to compare 2 types of coverage flap for the reconstruction of the finger web spaces: a volar flap described by Blauth and a dorsal flap described by Gilbert. Between 1993 and 2015, children affected by simple and complex syndactylies (partial or complete) were treated in 2 french pediatric hospitals and were selected for our analytic, comparative, retrospective review. The 2 hospitals used different surgical techniques: one used a volar flap described by Blauth and the other a dorsal flap described by Gilbert. The children were followed up to look for signs according to the stages of the Classification of Withey and to evaluate a global result according to the score of Withey. Our secondary criteria of judgement were the aspect of the surgical scar according to the VSS (Vancouver Scar Scale) and the satisfaction of the parents and children. The age of the children, need for a surgical revision and time of last follow-up were also studied. We found statistically significant differences between group I (volar flap) and group II (dorsal flap) in favor of the volar flap: higher scores of Withey (even when the number of commissures was increasing) and better VSS (regardless of the number of web spaces treated). There was no statistically significant difference between the 2 groups in terms of age, follow-up, or rate of surgical revision. All in all, the volar flap presented less sequelae in terms of scar retraction. Regardless of the flap used, the cosmetic results of the full-thickness skin graft used impacted the result both on the receiving site (dyschromia, hairiness) and the donor site.

Keywords : Congenital defects of the hand; congenital syndactyly; flap coverage; full-thickness skin graft; scar contracture.

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INTRODUCTION

Congenital defects represent 1% of all birth, and congenital syndactylies 1 to 2 births out of 2000 *(1,2)*. They belong to type II according to Swanson and IFSSH (International Federation for Societies for Surgery of the Hand) classifications *(3,4,5,6)*. Type I syndactylies according to Temtamy-McKusick classifications are the most common *(7,8,9)*. Embryologic syndactylies happen during the 7th week of pregnancy due to a lack of apoptosis between the digits *(8,9)*. Different types of syndactylies can be encountered: incomplete or complete, simple or complex (fusion of the distal phalanx sometimes associated to vascular or nervous defects) *(10,11)*. Last types of syndactylies are those associated to other birth defects or syndroms *(7)*.

Syndactylies are usually operated between 12 to 18 months of age in order to improve the function of the hand. If several web spaces are affected, they should be operated before the age of 1 and even earlier if the 1st web space is affected *(10,11,12)* or when there is a significant difference of length between the 2 affected digits like between the 4th and 5th digits *(10,13)*. Indeed, a difference of length will lead to an axial deviation and then a flexion deformity *(10)*.

The goal of the surgical procedure is to recreate a web space as functional and cosmetic as possible without creep or retraction *(14)*. The original surgical technique combines coverage flaps and Z-shaped incisions to separate the digits as well as skin grafts to fill the lack of skin created by the separation of the digits *(4,15,16)*. However, some post-operative complications are sometimes reported such as: scar retractions, web creep, hairiness, dyschromia of the grafted zone *(17)*, nail dystrophies and a psychological impact due to the imperfect cosmetic result of the operated hand *(14)*.

To reduce complications, several flaps have been described with or without an associated skin graft: dorsal *(18,19)*, volar *(20,21)*, omega-shaped *(22,23)*, diamond-shaped losangique *(24)*, rectangular *(25)* and V-Y flaps *(26,27)*.

The hypothesis is that the dorsal flap is equivalent to the volar flap for the web space.

This study compares 2 types of coverage flaps for the treatment of syndactylies: volar flap described by Blauth *(21)* versus dorsal flap described by Gilbert *(28)*, in terms of scar retraction, global aspect of the scar, complications, rate of surgical revision, and satisfaction of the children and their parents.

MATERIAL AND METHODS

Between 1993 and 2015, children presenting an embryonic syndactyly were operated in 2 French pediatric hospitals and were selected for our analytical, comparative, retrospective study. After separation of the digits, the newly formed web space was reconstructed using a different coverage flap in the 2 hospitals. In the group I, a volar flap described by Blauth was performed *(21)*, in group II a dorsal omega-shaped described by Gilbert was used *(28)*.

Children presenting syndactylies due to burns or from amniotic band constrictions were excluded, as well as syndactylies resulting from polymalformative syndroms.

A total of 25 children were included among which 20 males (10 in each hospital) and 5 girls (3 in group I and 2 in group II). A family history of syndactylies existed in 6 cases. Twenty right hands (8 for the volar flap and 11 for the dorsal flap) were operated. 20% of the syndactylies were bilateral (5 out of 25 children in both hospitals). A total of 35 syndactylies were separated (17 in group I and 18 in group II).

In group I, the 3rd web space was the most frequently affected (76.5%), followed by the 2nd web space (11.75%), the 4th web space (11.75%) and the 1st web space (0%). In group II, the 3rd web space was also the most frequently affected (66.7%), followed by the 2nd web space (22.2%), the 4th web space (5.55%) and the 1st web space (5.55%) (Figure 1). In group I, there were 47% of type A syndactylies, 29.4% of type B syndactylies, 5.9% of type C syndactylies and 17.6% undetermined according to Malek's Classification. In group II, there were 33.3% of type A syndactylies, 33.3% of type B syndactylies, 27.8% of type C syndactylies and 5.5% undetermined according to Malek's Classification (Figure 2).

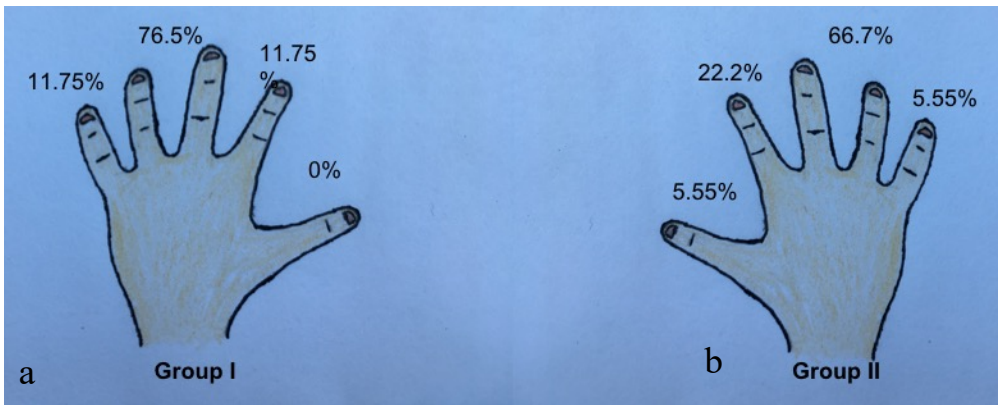

Fig. 1. – Global percentage of web spaces affected of embryonic congenital syndactylies: a) In group I, b) In group II.

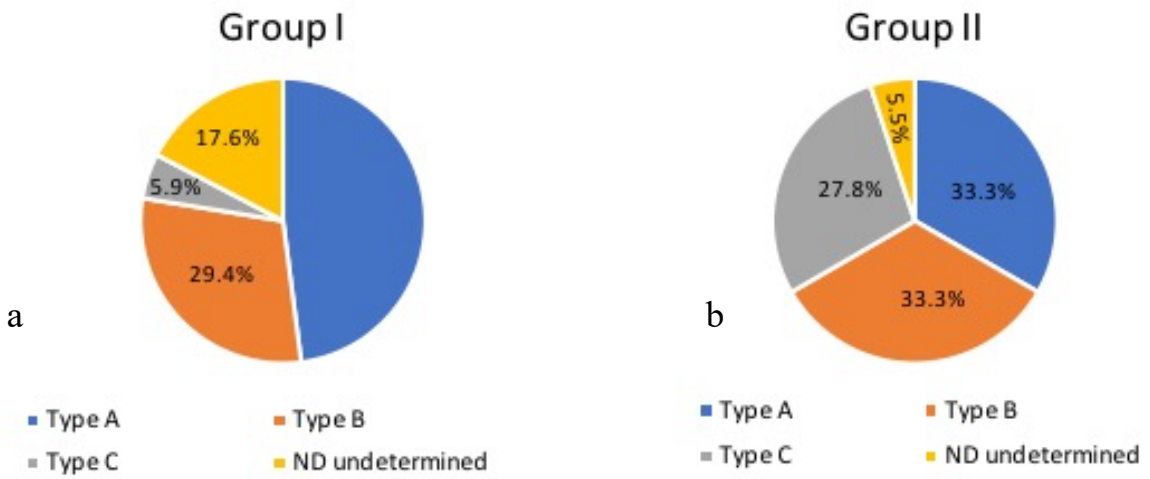

Fig. 2. – Distribution of the syndactylies according to Malek's classification: a) In group I, b) In group II. Type A: Syndactylies where the digit is joined all the way to the fingertip. Types B: Syndactylies where the fingers are joined to the middle phalanx. Type C: Syndactylies where the fingers are joined to the proximal phalanx. ND: Undetermined.

Both surgical techniques aimed at separating the two digits and reconstructing a new web space without scar contracture, creep or surgical revision. The goal of the coverage flaps was to create new web spaces as close to normal as possible. A native web space presents a dorsal slope at a 45° angle reaching from the head of the metacarpal to the half the length of the proximal phalanx. On the volar aspect, the web space stops halfway between the Proximal Inter-Phalangeal joint (PIP) and the distal volar crease. Syndactylies affecting 2 adjacent web spaces were separated in 2 consecutive operations to avoid vascular complications of the central digit.

In group I, the reconstruction of the web space was performed using a volar flap described by Blauth *(21)* and in group II using a dorsal flap described by Gilbert *(28)*.

In both groups, the procedure was performed under general anesthesia. The child was in dorsal decubitus, arm at 90° of abduction on an arm table. The incisions were drawn at the beginning of the procedure using a surgical skin marker to limit the dorsal and volar flaps. A pneumatic tourniquet was inflated at a pressure depending on the systolic pressure of the child. After performing Z-shaped incisions, the dissection was performed

progressively using magnifying glasses, and the different flaps created with care to preserve the adjacent vascular elements *(10)*

When a bony fusion of the distal phalanx existed, it was separated using a number 11 surgical blade and a Buck-Gramcko skin plasty was performed *(29)*. Once the 2 digits were separated, the lateral flaps obtained from the Z-shaped incisions were used to cover most of the exposed tissues exposed on the lateral aspects of the fingers. For the parts that could not be covered, usually at the proximal part of the digits a full thickness skin graft was used. The skin sutures were performed using absorbable thread (Ethicon® 5/0, Vicryl Rapide®), then the tourniquet was deflated to assess the vascularization the separated digits before putting in place the dressings.

In group I, the volar flap was drawn with a tip reaching to the crease of the PIP joint. The base of the flap was located at 2/3 of the distance between the distal volar crease and the proximal digital crease. On the dorsal aspect the hand, the incision was extended to the most proximal point of the created web space (Figures 3 and 4). The full-thickness skin graft was always taken at the inguinal crease. A paraffin gauze dressing was left in place for 15 days and changed between the 5th and 7th day. In a few cases, a silicon piece was used for pressotherapy purposes in the web space to limit the appearance of hypertrophic scar tissue (Figure 5) *(30)*. The average follow-up for children operated by the volar flap technique was 107.2 months (8.9 years, with a difference between 11 and 270 months).

In group II, the omega-shaped dorsal flap had a base drawn at the level of the metacarpal heads and a tip reaching to the PIP joints. On the volar aspect, an anchor-shaped incision with 2 lateral flaps was drawn (Figures 6 and 7). A full-thickness skin graft was performed either at the inguinal crease or the elbow crease. A dressing was left in place for 10 to 15 days and renewed under general anesthesia in most cases. The average follow-up for children operated by the dorsal flap technique was 66 months (5.5 years, with a difference between 14 and 108 months).

The classification of Withey was chosen to evaluate the retraction of the web space in 5 stages (Table I) *(31)* and the score of Withey was used for a more global evaluation of the hand, assessing the scar in flexion and extension, clinodactyly, and deformation in rotation. The score of Withey ranges from 1 to 11 (Table II). The Vancouver Scar Scale (Table III) was also used to assess the scar at last follow-up *(32)*. Finally, the cosmetic satisfaction of the parents and children (when possible) was assessed using a binary criterion (yes or no).

The statistics were performed using regression analysis (for the score of Withey, VSS, and satisfaction), estimated by bayesian techniques of Markov chains and Monte Carlo integrations (McMC).

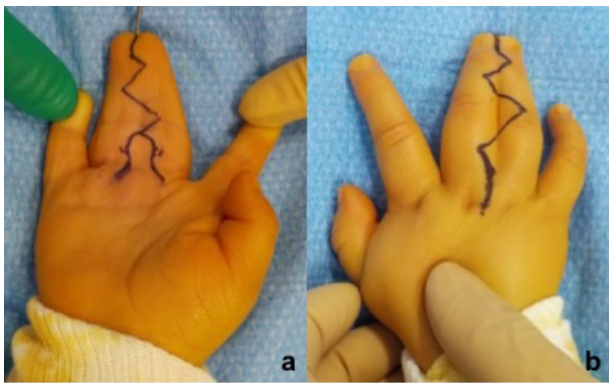

Fig. 3. – Drawing of the incisions in group I (volar flap described by Blauth): a) Volar view, b) Dorsal view.

Fig. 4. – Silicone piece used in pressotherapy to improve healing in group I.

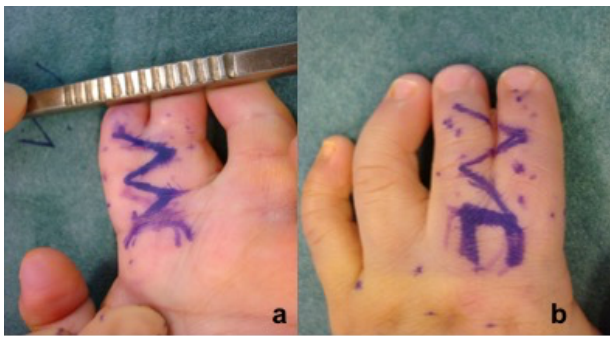

Fig. 5. – Drawing of the incisions in group II (dorsal flap described by Gilbert): a) Volar view, b) Dorsal view.

Fig. 6. – Result at last follow up of a syndactyly of 3rd web spaces of both hands using a volar flap described by Blauth (group I): 168 months after surgery: a) Volar view of the right hand, b) Volar view of the left hand, c) Dorsal view of the right hand, c) Dorsal view of the right hand, d) Dorsal view of the left hand.

RESULTS

The 2 groups studied both had a predominant male population (Sex Ratio: 4) with 3 females for 10 males in group I (23%) and 2 females for 10 males in group II (16%). The average age at the time of the procedure was 14 months in group I (standard

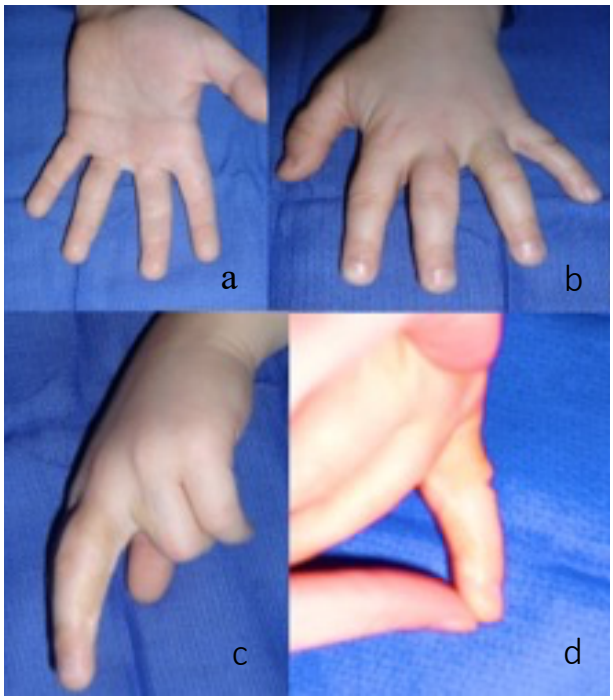

Fig. 7. – Result at last follow up of a syndactyly of the 3rd web space of both hands using a dorsal flap described by Gilbert (group II) 60 months after surgery: a) Volar view, b) Dorsal view c) Lateral view of the ulnar aspect of the 3rd digit, d) Lateral view of the radial aspect of the 4th finger.

The age or gender did not influence the VSS or the score of Withey in the comparison between the two groups. The parameter median age associated is 0.048 (with an interval of credibility at 95% [-0.384; 0.374]).

There was one complex form in group II and none in group I. There were 3 bilateral forms in group II (16.7%) and 2 bilateral forms in group I (11.8%), which was not statistically significant. According to Malek's classification, there were 47% of complete syndactylies in group I and 33.3% in group II which was not statistically significant (p=0.30). But the complete syndactylies were also complex which was not the case in group I.

The forms of operated syndactylies were mostly complete (40%) or incomplete (31.4%), there were 17.1% of simple syndactylies according to Malek's classification and 11.4% undetermined (Figure 7) *(33)*.

According to the classification of Withey, there was a low rate of web space creep with 70.6%

Table I. – Classification of Withey, global post-operative clinical evaluation of the separation of a syndactyly, ranging from 0 to 4. (PIP: proximal interphalangeal joint). (Withey S.J., Kangesu T, Carver N, Sommerlad B C. The open finger technique for the release of syndactyly, Journal of Hand Surgery (British and European Volume). 2001; p. 26-47 [31]).

0	Soft web
1	Thickening of the web with reduced span
2	Creep of web to 1/3 of the distance between base of the web and PIP crease
3	Creep of web to 2/3 of the distance between base of the web and PIP crease
4	Creep of web to the PIP crease

Table II. – Global score of Withey ranging from 1 to 11. (Withey S.J., Kangesu T, Carver N, Sommerlad B C. The open finger technique for the release of syndactyly, Journal of Hand Surgery (British and European Volume). 2001; p. 26-47 [31]).

	Grade	Description
Scare quality	1 2 3	Thin and narrow Wide and flat Raised and thick
Flexion–extension deformity	0 1 2	Normal digit Finger cannot be hyperextended Fixed flexion deformity
Web creep	0 1 2 3 4	Soft web Thickening of the web with reduced span Creep of web to 1/3 of the distance between base of the web and PIP crease Creep of web to 2/3 of the distance between base of the web and PIPJ crease Creep of web to the PIP crease
Lateral flexion deformity	0 1	Absent Present
Rotation deformity	0 1	Absent Present
Total	1 to 11	

patients with a score at 0, and 29.4% at 1 against 22.2% at 0 and 33.3% at 1 and 44.4% at 2.

The global score of Withey was statistically higher in the dorsal flap group (probability of superiority under 1.10-6). The score decreased as the number of operated web spaces increased: in the volar flap group it had a median of 2.6 [2.0; 3.4] (Table IV).

In group I, the median estimation of the score VSS was 3.591 [2.718; 4.533] and 4.878 [3.845; 5.954] in group II (Table V). There was no significant influence of the number of operated web spaces on the VSS.

The VSS of the donor site ranged between 0 and 5 out of 13 in the volar flap group with dominant values at 0 and 1 out 13. For the dorsal flap group, the values ranged between 0 and 3 out of 13 with predominant values of 0 and 1 out of 13 (Table VI). Hairiness was more frequently encountered in the volar flap group with a risk of 0.387 [0.094; 0.738] compared to the dorsal flap group with a risk of 0.064 [0.0044; 0.229].

The mean age of follow-up in the two groups was 107.24 months (8.9 years) ranging from 11months to 270months (22.5 years).

Parents were mostly satisfied in terms of global care with 92.3% of satisfaction in the volar flap group and 100% of satisfaction in the dorsal flap group. The children were satisfied with 76.5% of

Table III. – Vancouver Scar Scale ranging from 1 to 13 assessing the local aspect of the scar. (Thompsom CM, Sood RF, Honari S, Carrougher GJ Gibran NS. What score on the Vancouver Scar Scale constitutes. Results from a survey of North American burn-care providers. Burns. Nov. ; 2015 ; 41(7) :1442-1448 [32]).

Vancouver Scar Scale (VSS)		
Pigmentation	Normal Hypopigmentation Hyperpigmentation	0 1 2
Vascularity	Normal Pink Red Purple	0 1 2 3
Pliability	Normal Supple Yielding Firm Banding Contracture	0 1 2 3 4 5
Height	Normal 0-2 mm 2-5 mm >5 mm	0 1 2 3

satisfaction in the volar flap group and 66.7% of satisfaction in the dorsal flap group. The cosmetic satisfaction was of 86.7% in group I and 83.7% in group II for the parents and 92.3% in group I and 81.8% in group II for the children. There is a significant difference in terms of overall satisfaction for children (p=0.52) and parents (p=0.45), or regarding the cosmetic result for both (parents p=1.00, children p=0.58).

DISCUSSION

The 2 groups were comparable in terms of age and gender. Although more simple, complete, or familial syndactylies were encountered in group I, it was not statistically significant. In group II there were more complex forms (1 vs 0) and bilateral forms (3 vs 2), but it was not statistically significant.

Familial forms of syndactylies were encountered in 5 children in group I (3 males and 2 females), and in 3 children in group II (2 females, 1 male) making a total of 8 familial forms with a greater proportion of female patients (80%). There was no significant difference between the number of

Table IV. – Results of the Global score of Withey depending on the number of webs operated and, on the flap, used.

	Number of webs operated	Estimation	Average	Headcount
Group I	1	2.577 [1.957; 3.367]	2.80	10
	2	2.046 [1.623; 2.591]	1.25	4
	3	1.675 [1.309; 2.205]	1.00	3
	4	1.423 [1.095; 1.963]	-	0
Group II	1	5.313 [4.317; 6.345]	5.50	8
	2	4.280 [3.503; 5.081]	3.67	6
	3	3.399 [2.512; 4.399]	-	0
	4	2.709 [1.773; 3.946]	4.00	4

Table V. – Statistical results comparing the VSS on the receiving site of both groups.

	Estimation	Average	Headcount
Group I	3.591 [2.718; 4.533]	3.82	17
Group II	4.878 [3.845 ; 5.954]	4.39	18

Table VI. – Statistical results comparing the VSS on the harvesting site of both groups.

	VSS of the harvesting site of full-thickness skin graft				
	5	3	2	1	0
Group I	17.6%		11.8%	35.3%	35.3%
Group II		5.5%	16.7%	55.5%	22.2%

affected web spaces and the localization of web space for the different familial forms between the 2 groups. Indeed, in group I, there were 20% of familial forms affecting the 4th web space and 80% affecting the 3rd web space. In group II, all familial forms affected the 3rd web space.

Our study compared 2 surgical techniques of coverage flaps used to separate congenital embryonic syndactylies (excluding syndromic forms).

According to the classification of Withey and the global score of Withey, assessing the neo web space: there was a significant difference in favor of group I. The VSS was also better in group I (Figure 8-not included). Many children operated using a volar flap described by Blauth presented a better global score of Withey. We found more scar-related complications (scar contracture, web space retraction…) in group II (Figure 9-not included), indeed it is the most frequently described complication of this type of dorsal flaps *(18)*. On another hand, some authors have shown that certain types of flaps decrease the rate of scar contractures. It is the case of the Gao study *(34)*, which studied 17 web spaces in 10 children treated using a dorsal pentagonal flap and did not present any case of scar contracture, scar hypertrophy or surgical revision. The silicone piece used for pressotherapy may have influenced the results in group I, by helping the healing process (Figure 3). Other factors influence the apparition of scar contractures: the experiences of the operator, the drawing of the coverage flap and of the skin graft.

If comparative studies between the dorsal flap described by Gilbert and other dorsal flaps have been conducted, no comparative study has yet been published between the volar flap described by Blauth *(30,3,36)* and a dorsal flap.

Some studies report a technique using a double flap: dorsal and volar. According to Gao in his series no surgical revision was needed *(34,37)*, and Lumenta only reports a rate of 12% of surgical revisions *(37)*. The combination of both flaps could therefore be interesting.

Regarding the aspect of the scar, there was a significant difference between the 2 groups. There was a lower VSS in group I than in group II. A frequent dyschromia could be frequently observed due to the skin graft, regardless of the coverage flap used. A hairiness was more frequently observed in group I. It was related to the zone of harvesting of the full thickness skin graft. In group I it was always taken from the inguinal crease whereas in group II it was taken from the elbow crease in 27.8% of cases, explaining a less frequent hairiness. A study conducted by Mishra et al. suggests good results of laser therapy for the treatment of post-operative hyper pigmented zones of syndactylies, although the population of the study was low with only 8 cases *(38)*.

Some studies reported similar rates of scar tissue *(39,40)* without grafting the loss of substance *(19,34,41)*. Some authors even use techniques of skin expansion using tissue expenders to enable a direct suture of the skin *(42)*. Sullivan and al. in his review finds a higher rate of revision of the scar when a skin graft is used compared to techniques using a coverage flap *(15)*. Some authors cover the lateral aspect of the digits using different flaps rather than a skin graft. Greuse and al. used a dorsal rectangular metacarpal flap *(26)*, and report 2 cases of complications, one case of infection and one case of scar hypertrophy. They described the resection of the subcutaneous fat tissue in the interdigital space to enable a direct suture of the skin without tension *(26,41)*. This technique is also described by Dong and al. who report good results on 35 web spaces on simple, incomplete syndactylies *(39)*. Other authors have described flaps that did not need skin grafts and report good global satisfaction of the parents due to the absence of dyschromia or hairiness. They do not report issues of skin coverage for simple syndactylies *(19,43)*, but it depends on the surface of loss of substance to cover. Despite all these techniques enabling to avoid skin grafting, Watson in his review concludes that the use of a skin graft should be used as often as necessary *(14)*.

The weaknesses of our study may be the difference technique employed in two centers by different surgeons and the treatment post-operative.

Finally, recent studies have also compared coverage of the lateral aspect of the digits using dermal substitutes such as Matriderm® [44], Integra® *(45)* and Hyalomatrix® *(22)* associated to ultra-thin skin grafting. This could be a solution to

avoid scar-related complications such as hairiness and dyschromia. Although only one retrospective study has evaluated the rate of scar contractures *(44)*. A multicentric comparative randomized study comparing dermal substitute associated to ultra-thin skin graft to full thickness skin graft seems necessary.

CONCLUSION

In the surgical treatment of embryonic congenital syndactylies in children, the reconstruction of the web space after separation of the digits seems to give better long-term results and less complications when using a volar flap compared to a dorsal flap.

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