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A Review on the Safety of One-Stage Circumferential Ring Constriction Release

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The study was undertaken to investigate the use of one-stage circumferential ring-constriction release with Z-plasties regarding the safety, aesthetic appearance, and limb function. A thorough review was conducted on all English publications in PubMed during the period of 2001 through 2011. Titles and abstracts were identified using online search engine from National Library of Medicine's PubMed database under the keywords "limb constriction ring," "limb constriction band," "amniotic band," "annular constriction," and "circumferential constriction." We used Boolean operator and field of title. Evaluation was done to search indications, timing of the first surgical intervention, time interval between surgeries, patients' gender, anatomic location of the ring, wound healing problems, and scar quality. Fourteen publications met the criteria. There were 17 patients with 25 ring constrictions in total. Sixteen ring constrictions (64%) were circumferential; nine (36%) were semi-circumferential. Mean age of 14 patients treated with one-stage release was 4.8 years. Six articles mentioned about normal development of postoperative limb function. Mean age of three patients treated with staged release was 10.5 months. Two articles mentioned regained distal muscle function postoperatively. It is confirmed that surgeons may continue the practice to release circumferential CRS in one stage.

Key words: Amniotic band syndrome – Constriction ring – Surgical procedures – Pediatrics

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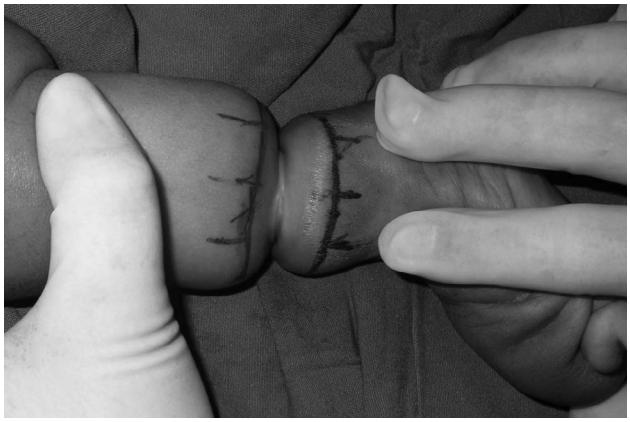

Fig. 1 Complete circular CRS (constriction ring syndrome). The picture shows a complete circular CRS.

Fig. 2 Complete dissection of the fibrous band. Constriction release is accomplished by completely excising the fibrous band. The first stage of a 2-stage release technique is shown.

Fig. 3 Postoperative result of half-circle approach. A half-circle release of CRS results in a good outcome.

Constriction ring syndrome (CRS) is a type of congenital anomaly of which the incidence ranges from 1 in 1200 to 15,000 live births.[2–5] The ratio between male and female affected is approximately 1:1 and all ethnic groups are equally affected.[3,5] Constriction ring syndrome (CRS) is characterized by a fibrous band that tightly encircles the limb, either partially or completely. Interestingly, this abnormality can appear on sites other than its usual place, such as the abdomen and the chest.[6–7] Various theories have attempted to explain the etiology of CRS, but they still cannot be inferred.[2] However, there are 3 theories that are likely to be accepted among hand and plastic surgeons; they are intrinsic theory,[3,8] extrinsic theory,[3] and intrauterine trauma theory.[1,9]

The recommended management for the correction of CRS is done in 2 or 3 stages of releasing the constricting ring (Figs. 1 and 2).[10] This technique is considered as a safe way to eluding vascular and lymphatic embarrassment and also usually gives a good outcome (Fig. 3) However, several modifica-

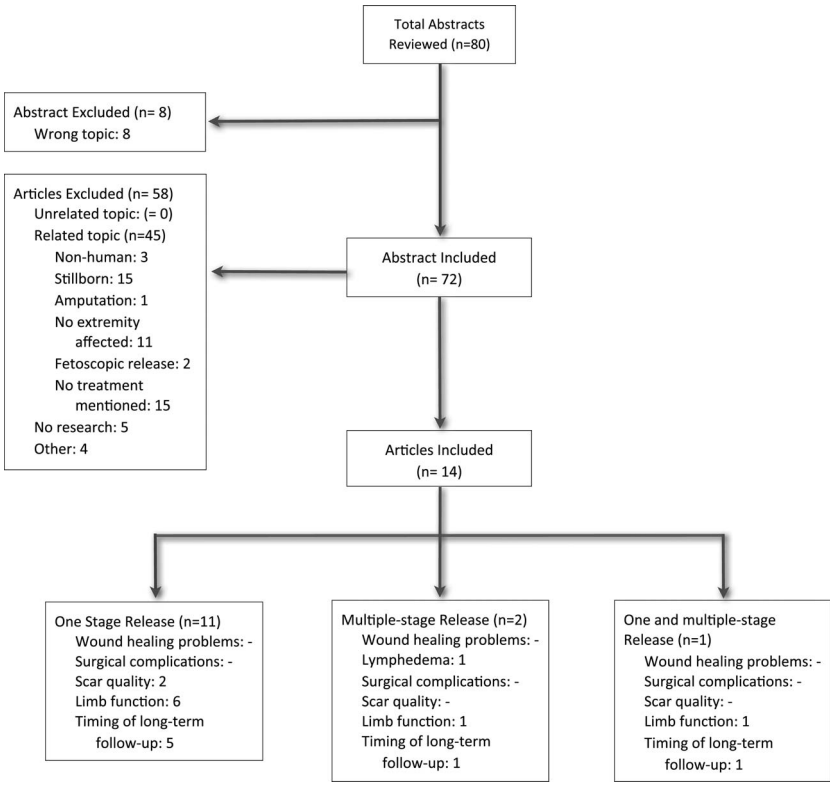

Fig. 4 Study flow diagram.

tions on the management of CRS have been attempted; one of them is the 1-stage circumferential ring constriction release with Z-plasty.[11–15] The safety of this method is still deemed to be debatable regarding the viability of the limb distal from the ring.[5]

The aim of this study is to investigate the outcomes from application of 1-stage circumferential constriction-ring release with multiple Z-plasty techniques for the correction of CRS Patterson type 1, 2, and 3.

Methods

This study is a thorough review to answer the question of how effective the 1-stage circumferential ring-constriction release procedure with skin recontouring using multiple Z-plasty techniques to correct the ring-constriction in patients with CRS Patterson type 1, 2, and 3 is compared to multiple-stage ring-constriction release with multiple Z-plasties.

Titles and abstracts were identified using online search engine from the National Library of Medicine's PubMed database under the keywords (limb* AND constriction* AND ring*) OR (limb* AND constriction* AND band*) OR (amnio* AND band*) OR (annular AND constriction*) OR (circumferential AND constriction*). The search used Boolean operator, field of title. The search was limited to articles published between 2001 and 2011, written in English, and human subjects. Studies involved were prospective and retrospective cohort studies on CRS type 1, 2, and 3 cases according to Patterson classification along with the treatment. Exclusion criteria included patients diagnosed with constriction ring syndrome (CRS) with intrauterine auto amputation (type 4) and/or necrotic distal limb, ring constriction that appeared on areas other than limb(s) and finger(s), nonhuman patients, and stillborn fetus.

Evaluations of interest included in this study are indications; age at the time of the first surgical intervention; time interval between the first and the next stages; gender of the patients; anatomic location of the ring; wound healing problems such as infections, wound dehiscence, delayed wound healing; other surgical complications such as lymphedema and vascular compromise; the scar quality [based on *e.g.* the visual analog scale (VAS)]; the limb function; and the timing of long-term follow-up.

Result

Eighty citations (Fig. 4) were successfully extracted from PubMed. The types of citations were letter to

Table 1 Patient demographic, number of patients and ring constriction, age at the first surgical intervention for ring constriction release, sex, number and anatomic location of ring constriction, indication of the surgical intervention (lymphedema, vascular disturbance, viability disturbance), stage of constriction release (single-stage and multiple-stage)

No.	Authors and year of publication	Number of patients	Sex		Age	Number and site of the ring constriction	Indication	Constriction release stage	
			Male	Female				One	Multiple
1	Eren *et al*[15] 2010	1		1	10 month	Complete constriction in the left cruris and thigh. Semiconstriction in right lower extremity, between femur and tibia. Ring constriction around the right distal cruris	Complete constriction at the left lower extremity Semiconstriction at the right lower extremity	One	
2	Blyth *et al*[16] 2010	1		1	10 month	Circumferential bands at the left wrist, forearm, and upper arm	Circumferential bands cause restriction of the circulation resulting in lymphedema at the affected limb	One	
3	Fawzy *et al*[17] 2008	1		1	11 years	Noncircumferential constriction band along abdominal wall Tight constriction bands on both calves Constriction band on left 3rd toe	Both the abdominal and right calf constriction bands become symptomatic with pain	One	
4	Morovic *et al*[18] 2004	2	1	1	2yr10mo 3yr10mo	Ring constriction of the little finger of the left hand Asymmetric digital ring constriction with distal atrophy, amputations, and severe left clubfoot	NA	One	
5	Beidas *et al*[19] 201	2	2		1. 12 mo 2. 15 mo	Deep circumferential constriction band extending into the soft tissues of the distal forearm		One (case 1)	Multiple (case 2)
6	Ronderos-Dumit *et al*[20] 2006	1	1		1week	Constriction ring in left and right leg Fibrous annular ring in 3rd and 4th right toes.	NA	One	
7	Sentilhes *et al*[21] 2004	1	1		2 days	Constriction ring at the lower 3rd part of the leg	Severe edema of the distal limb	One	

Table 1 Continued

No.	Authors and Year of publication	Number of patients	Sex		Age	Number and site of the ring constriction	Indication	Constriction release stage	
			Male	Female				One	Multiple
8	Soldado F *et al*[22] 2009*	2	NA	NA	Postnatal period	Constriction in the distal third left leg, limb constriction on the right thigh Distal left-leg constriction	Distal edema	One	
9	Piero *et al*[23] 2009*	1	NA	NA	Postnatal period	Constriction band at distal-third left leg	Mild distal edema	One	
10	Altamirano *et al*[24] 2009	1	NA	NA	NA	Circumferential deep-scarred grove in the right ankle		One	
11	Samra *et al*[25] 2006	1		1	12 hours	Deep complete circumferential constriction band at the distal right thigh	Severe edema, cyanoses of the right leg		Multiple
12	Tanabe *et al*[26] 2002	1		1	2 years	Constriction band encircle the finger at the distal interphalangeal joint of left ring	NA	One	
13	O'Driscoll *et al*[27] 2008	1		1	6 month	Constriction band around the fingers (excluding thumb) of the right hand Constriction band joining all the fingers (apart from the thumb) of the left hand	NA		Multiple

NA, not available.

*Highly possible for duplication in publication.

Table 2 Distribution of clinical response from surgical intervention of ring constriction release [wound problems, scar quality based on Visual Analog Scale (VAS)], limb function, and timing of long-term evaluation)

No.	Authors and year of publication	Number of patients	Location of the ring constriction	Wound healing problems		
				Infection	Wound dehiscence	Delayed healing
1	Eren *et al*[15] 2010	1	Left and right lower extremity	NA	NA	NA
2	Blyth *et al* [16] 2010	1	Left upper limb	NA	NA	NA
3	Fawzy *et al* [17] 2009	1	Abdominal wall Left and right calves Left 3rd toe	NA	NA	NA
4	Morovic *et al* [18] 2004	2	Left little finger	NA	NA	NA
5	Beidas *et al* [19] 2010	2	Left distal forearm	NA	NA	NA
6	Ronderos-Dumit *et al* [20] 2006	1	Left and right leg Right 3rd and 4th toes	NA	NA	NA
7	Sentilhes *et al* [21] 2004	1	Lower 3rd part of the right leg	NA	NA	NA
8	Soldado *et al* [22] 2009*	2	Distal 3rd left leg, right thigh	NA	NA	NA
9	Piero *et al* [23] 2009*	1	Distal-third left leg	NA	NA	NA
10	Altamirano *et al* [24] 2009	1	Right ankle	NA	NA	NA
11	Samra *et al* [25] 2006	1	Distal right thigh	NA	NA	NA
12	Tanabe *et al* [26] 2002	1	Distal from interphalangeal joint of the left hand	NA	NA	NA
13	O'Driscoll *et al* [27] 2008	1	Fingers of each right and left hand (except the thumbs)	NA	NA	NA
14	Findik *et al* [28] 2006	1	Left thigh	NA	NA	NA

NA, not available.

*Highly possible for duplication in publication.

editor, case report, and case series. Among the 80 citations, only 14 met the inclusion criteria.

There were 17 patients with ring constriction with a total of 25 ring constrictions. Eight ring constrictions (32%) appeared on the upper extremity, and 17 (78%) appeared on the lower extremity. Sixteen ring constrictions (64%) were circumferential and 9 (36%) were semicircumferential (Table 1).

Fourteen patients were treated with 1-stage circumferential ring-constriction release with skin recontouring using multiple Z-plasties (Table 1). The age range of patients spanned from less than 24 hours of life to 16 years old (mean 4.8 years).

According to data from the articles, none of the articles discussing 1-stage circumferential ring constriction release elaborated on wound healing problems and surgical complications following surgical intervention. Two articles mentioned scar quality. Fawzy *et al*[18] stated that the scar became hypertrophic, whereas Ronderos-Dumit *et al*[21] stated that the scar following the surgery had excellent cosmetic results. Six articles mentioned postoperative limb function result; they showed normal development, looked excellent, healed satisfactorily, had no functional impairment, recovered, and demonstrated full function (Table 2). Five articles mentioned the timing of long-term evaluation after 1-stage release. The follow-up timing ranged from 9 months to 26 months postoperative (mean 16 months).

Three patients were treated with staged ring constriction release with skin recontouring using multiple Z-plasties (Table 1). Patients' age range spanned from 12 hours of life to 15 months old (mean 10.5 months old).

None of the articles on multiple-stage circumferential ring constriction release elaborated on the wound healing problems. One article mentioned lymphedema as a surgical complication. It stated that the lymphedema dissipated in 1 week following the first surgical management. None of the articles elaborated on the scar quality after the surgery. Two articles mentioned postoperative limb function result; they healed satisfactorily and regained distal muscle function (Table 2). The follow-up timing ranged from 6 months to 12 months (mean 9 months).

Table 2 Extended

Surgical complication				
Lymph-edema	Vascular disturbance	Scar quality	Limb function	Timing of long-term evaluation
NA	NA	NA	NA	NA
NA	NA	NA	Normal development	14 months postoperative
NA	NA	Hyper-trophic scarring	Excellent	NA
NA	NA	NA	NA	NA
NA	NA	NA	Healed satisfactory	12 months postoperative
NA	NA	Excellent cosmetic result	No functional impairment	26 months postoperative
NA	NA	NA	Recover	10 months postoperative
NA	NA	NA	Progressively full ankle function	1. 14 months 2. 10 months
NA	NA	NA	Full function	9 months postoperative
NA	NA	NA	NA	NA
Dissipated one week after 1st surgery	NA	NA	Regain distal muscle function.	6 months 18 months, and 4 yr 6mo
NA	NA	NA	NA	NA
NA	NA	NA	NA	NA
NA	NA	NA	NA	NA

Detail about the measurement criteria for wound healing problems, *e.g.*, infections, wound dehiscence, delayed wound healing, and vascular disturbance, cannot be acquired for both 1-stage and multiple-stage.

Discussion

One-stage circumferential ring constriction release is a common procedure to repair ring constriction of CRS, albeit it has not been a recommended procedure for CRS yet. Theoretically, it is safe to manage CRS in stages of ring constriction release in order to avoid vascularization embarrassment. Nevertheless, a careful management while doing a one-stage release of a circumferential CRS will yield a delightful outcome (Fig. 5).

There were 17 patients diagnosed with CRS with a total of 25 ring constrictions. Fourteen patients were treated with 1-stage circumferential ring-constriction release. However, steps of the technique were not clearly elaborated.

Age of the patients who undergo the surgical intervention, either with 1-stage or multiple-stage circumferential ring constriction, ranged from 12 hours of life to 16 years old. According to Moran *et al*[12], there is no specific age for the patients to undergo surgical intervention in order to release the band. The standard timing to release the ring constriction depends on the severity of the disease itself.

There are several criteria that we used in this study to evaluate the outcome of both 1-stage and multiple-stage circumferential ring-constriction release. The criteria are wound healing problems such as infection, wound dehiscence, delayed wound healing; surgical complications such as lymphedema and vascular compromise; the scar quality [based on *e.g.* the visual analog scale (VAS)]; and limb function.

Only 1 study stated about the surgical complications after the first surgery of multiple-stage ring constriction release; Samra *et al*[25] stated that lymphedema occurred following the first surgery, then dissipated in 1 week.

The scar quality evaluation is based on the visual analogue scale (VAS). Unfortunately, none of the articles used any standard parameter to evaluate the scar. Fawzy *et al*[18] stated that the scar became hypertrophic, whereas Ronderos-Dumit *et al*[20] stated that the scar following the surgery had excellent cosmetic results.

Fig. 5 Postoperative result of a single-stage approach. A single-stage circular complete release of CRS may result in a nice outcome as shown in the picture.

Limb function evaluation was based on the range of motion of the limb affected by the ring constriction. Although all the authors did not mention any parameter in evaluating the limb function progression, they mentioned that the limb after 1-stage circumferential ring constriction release showed normal development, looked excellent, healed satisfactorily, had no functional impairment, recovered, and demonstrated full function. For the multiple-stage circumferential ring constriction release, the authors stated the state of limb function as having healed satisfactorily and regained distal muscle function.

Conclusion

This study confirms that 1-stage circumferential ring-constriction release with skin recontouring using multiple Z-plasties is considered as a safe treatment for the correction of both circumferential and semi-circumferential congenital constriction ring.

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