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# Keystone Flap in Amniotic Band Syndrome—Innovative Approach of an Established Operative Technique for an Unusual Entity

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**Abstract:** Amniotic Band Syndrome (ABS) is a complex condition characterized by constricting rings and tissue synechiae, resulting in tissue necrosis and congenital anomalies. In newborns and infants with ABS, tissue necrosis can be profound, requiring a tissue defect reconstruction, realized by a Keystone Perforator Island Flap (KF). Primarily used for reconstruction after skin cancer excisions, KF's applications expanded to defects of various etiologies and disorders throughout the body. Subsequently, additional KF types adapted to the particular tissue defects were developed. The KF's preparation is relatively simple to perform leading to shorter operative times, and the postoperative monitoring is less laborious. Individualized surgical approaches and timing are essential for addressing the varied manifestations of ABS, with immediate treatment recommended for vascular compression, all-layered tissue necrosis, and nerve compression cases. To our knowledge, there is no published case in which a KF was used for the reconstruction of tissue defects and release of constriction rings in the context of an amniotic band syndrome. Therefore, the purpose of this article is to introduce the established surgical technique of KFs as an innovative surgical approach with satisfying reconstructive results for tissue defects and constriction ring release in ABS.

**Keywords:** keystone flap; amniotic band syndrome; constriction band syndrome; perforator flap; reconstructive surgery; plastic surgery

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## 1. Introduction

The Amniotic Band Syndrome (ABS), first described by Montgomery in 1832, is a very rare condition of a clinical constellation of various congenital anomalies [1,2]. A diverse spectrum of clinical deformities is associated with ABS and varies from simple ring constrictions to tissue synechiae, tissue necrosis, body wall defects, and major craniofacial and visceral defects [3–5]. The incidence for ABS ranges from 1/1200 to 1/15,000 live births. The fact that over 30 different names have been used to describe this entity shows the variability of clinical presentation between patients [6]. Although the etiology of the ABS remains controversial. The extrinsic theory states that amniotic tissue entangles the fetal parts, thereby causing constrictions, whereas the intrinsic defect theory claims that inherent abnormalities, in the subcutaneous germplasm, cause mesenchymal hypoplasia and scarring [7–9].

Amniotic band syndrome is commonly identified postnatally through clinical observations that align with the presence of constricting bands and related congenital anomalies distal to these bands. During prenatal stages, ABS can be recognized via routine ultrasound imaging, revealing delicate echogenic bands attaching to both the uterine wall and impaired fetal structures. Yet, detecting these bands directly can be intricate, underscoring the importance for clinicians to scrutinize any instances of extremity asymmetry, amputation, or syndactyly, as these signs may signify underlying band-related issues. Furthermore, it is noteworthy that amniotic bands can sometimes resemble amniotic sheets, a harmless discovery without clinical implications. Consequently, the visibility of bands on prenatal ultrasound alone does not substantiate an ABS diagnosis, unless accompanied by evident fetal anatomical irregularities [10].

In some manifestations with severe tissue necrosis and extended constriction, amputation of the affected extremity might become necessary distal to the constriction bands, which leads to a loss of function and devastating deformities [11,12]. Therefore, the early recognition of this entity and the subsequent surgical treatment of the presenting necrosis and body wall defects are important for the prognosis with respect to wound closure and scaring as well as contractures. In the case of remaining tissue defects after necrosectomy, primary wound closure is often impossible.

The Keystone Perforator Island Flap (KF) presents sufficient and satisfactory tissue coverage [13,14]. Its inception can be traced to the evolution of perforator-based techniques, which aimed to minimize donor site morbidity and improve flap viability. Over time, the keystone concept evolved, emphasizing the central perforator vessel as the pivotal element sustaining the flap, leading to the development of various keystone flap variations tailored to different anatomical regions and clinical demands. Furthermore, it minimizes the requirements for skin grafting, and it is less technically demanding than other locoregional perforator flaps or microvascular-free tissue transfer options. The KF has earned the moniker of a "workhorse" within the domain of locoregional and fasciocutaneous flaps [13]. Its exceptional versatility across diverse body regions, coupled with its proficiency in providing ample soft tissue coverage, has solidified its status. Furthermore, the KF consistently yields remarkable aesthetic outcomes, underlining its significance as a reliable and effective reconstructive technique.

Here, we describe an innovative surgical approach for the simultaneous release of constriction rings and tissue defect reconstruction using Keystone Perforator Island Flaps on the upper extremities in newborns with all-layered tissue necrosis and constriction rings due to an amniotic band syndrome. This approach brings assessable complexity, low perioperative risks, and short surgical time making it an ideal reconstructive option.

## 2. Surgical Strategy

Generally, presented newborns can show all-layered tissue necrosis and constriction rings on either upper or lower extremities. Given the rarity of amnion band syndrome as an entity, our cohort for this manuscript comprises a limited number of three Keystone Flaps. Affected localities were particularly the upper extremities. The localizations exhibited a defect size of approximately 1–3 cm. However, you should keep the corresponding body size in mind, with newborns sometimes not even 1 day old. In the prenatal screening, there was no indication of amniotic band syndrome.

Due to often partially demarked necrosis and an incipient reddening of the wound surroundings, prompt surgery should be arranged. Intraoperatively, the tissue necrosis partly presented, leaving behind a profound defect in which primarily wound closure was not possible anymore (see Figure 1). Therefore, we opted for the release of the constriction rings and subsequent reconstruction utilizing Keystone Flaps at various locations, a novel approach to our understanding, hitherto unexplored in newborns diagnosed with amniotic band syndrome.

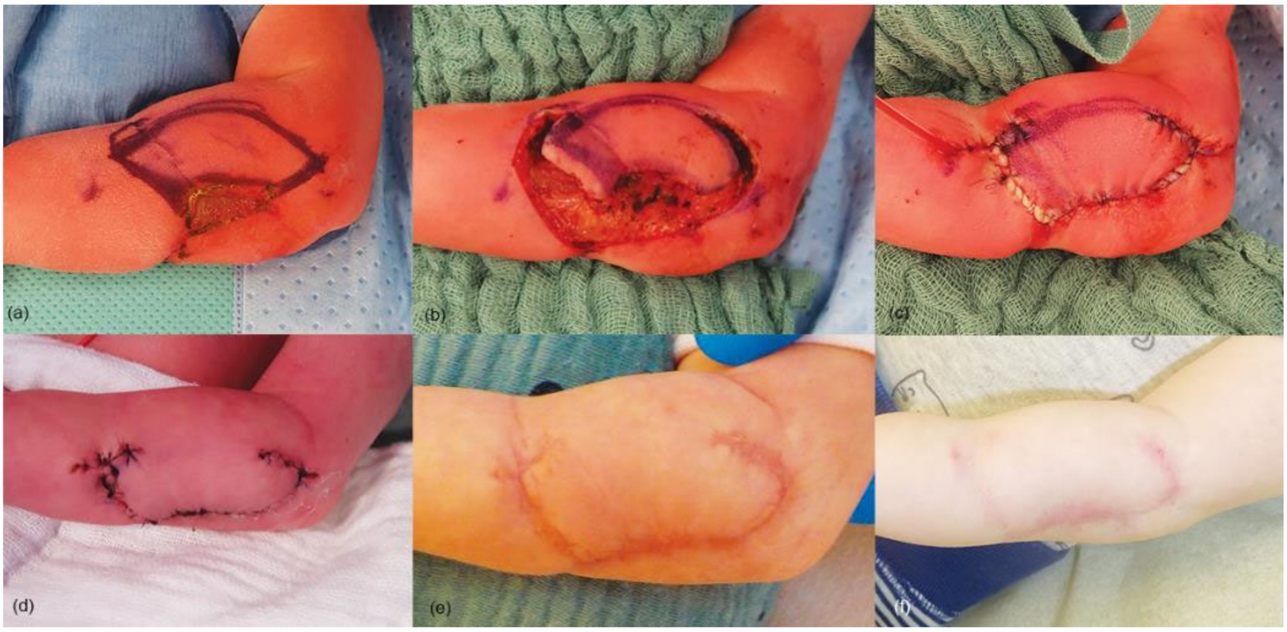


**Figure 1.** (**a**) Preoperative Keystone Perforator Island Flap (KF) marking; (**b**) KF's blunt dissection mobilization of a SMU modification of a Type IV KF; (**c**) defect reconstruction with KF on the left forearm; follow-up after (**d**) 7 days, (**e**) 1 months, and (**f**) 6 months postoperative.

### *2.1. Operative Technique*

In general, the timing of surgical intervention in constriction rings depends on the extent of the disease and skeletal growth influence as well as the extension of tissue necrosis. When a constriction ring results in limb contour deformity, with or without lymphedema, surgical intervention is recommended. In essence, the overarching goal in rectifying ABS constriction rings surgically revolves around the liberation of tissue constriction, the adept closure of tissue defects, and, when necessary, the alleviation of compression on functional structures like nerves, vessels, and muscles. Circulatory compromise with tissue necrosis or peripheral nerve palsy secondary to compression demands rapid attention.

At large, the Keystone Perforator Island Flap was established to close elliptical defects entailing a highly efficient geometry that resembles the trapezoidal, apical, and curvilinear stone of a Roman arch [13]. This keystone plays a pivotal role in stabilizing the arch by efficiently distributing gravitational forces. Much like this architectural principle, the keystone flap is designed to secure itself within the defect through its crafted shape, thereby providing not only coverage but also essential structural reinforcement to the reconstructed area. For a sufficient wound closure, the KF should be designed on the defect's side of greater cutaneous laxity. In this regard, a pinch test, a simple yet crucial assessment, should be conducted preoperatively. This test allows for the evaluation of tissue elasticity and laxity, providing valuable insights into the optimal planning of flap design and ensuring that the chosen reconstructive approach aligns harmoniously with the patient's individual anatomical characteristics. For the classical marking (see Figure 2), lines are drawn at the ends of the primary defect with average angles of 90 degrees, reaching a 1:1 ratio with the amplitude of the initial defect and ending with a curvilinear line that joins these two lines at the outer edge of the KF [15]. The available tissue is optimized by this design and equates to two or even three V-Y island flaps [16]. Placing the main axis parallel to the defect favors tissue recruitment in the flap center. This preparation allows the distribution of the tension necessary for the wound closure throughout the periphery [17]. Furthermore, these principles facilitate primary closure of both the defect and donor area, often obviating the need for split-thickness skin grafting to cover the donor site defect.

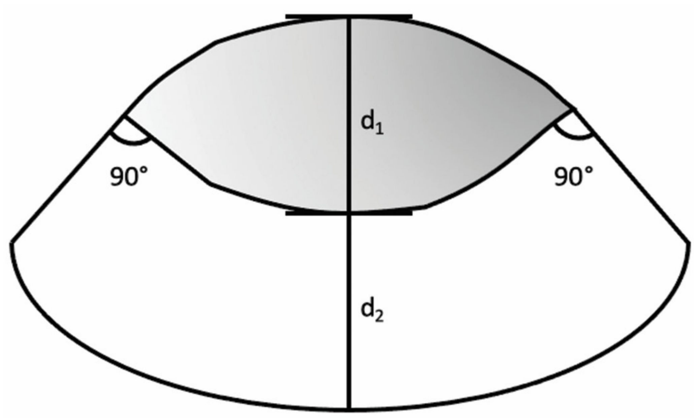


**Figure 2.** Keystone Flap (KF) design considering the 1:1 ratio ($d_1$ = $d_2$) of defect width to flap width and regarding the trapezoidal shape with 90° angle at the limits of the KF.

Certainly, the greatest surgical concern is the vascular safety of the KF, since the perfusion is based on the underlying vascular bed that remains attached to the area [13]. A various network of blood vessels with anastomotic linkages in the intradermal, subdermal, and subcutaneous layer is supplied from the profound tissues via perforators of varied size and course securing their perfusion [18]. Therefore, the KF's major axis should be designed parallel to the cutaneous nerves and/or known perforators to include the vascular perforator structure.

For KF it is not required to identify and skeletonize a source vessel or a specific perforator, unlike for typical perforator flaps. Therefore, the perioperative risk, complexity, morbidity, and surgical time is reduced compared to classic perforator flaps [19]. Furthermore, it is recommended to preserve the cutaneous nerves to exploit neurocutaneous circulation and preserve the sensitivity of the Keystone Flaps [19,20].

Beyond the aforementioned foundational blueprint of the Keystone Perforator Island Flap, a tapestry of innovative variations has been woven. This rich spectrum of designs has been thoughtfully classified into four distinct types, with their respective modifications.

In the case of a Type I KF, a solely cutaneous incision is executed, preserving the profound fascia. This specific KF iteration is particularly advisable for defects with a diameter not exceeding 2 cm, finding its frequent application in the head and neck region.

In the context of a Type II KF, the procedure extends to encompass the incision of the profound fascia, giving rise to the trilaminar unit composed of skin, fat, and profound fascia. This type of KF, in turn, features subdivisions based on the potential for primary wound closure (Type IIA) or the requisition of a split skin graft to close the donor site (Type IIB). These types are most frequently used for defects located at the upper and lower extremities.

A Type III KF embodies the concept of a double keystone flap, manifesting in either a symmetrical placement on both convex sides of the soft tissue defect, known as the double-opposing design, or a nuanced adaptation akin to the "Yin–Yang" variant. In the latter, two slightly offset flap configurations intertwine along the longitudinal axis of the soft tissue defect. These innovative types are particularly effective for addressing more extensive defects ranging from 5 to 10 cm. Their versatile application finds a notable presence in regions such as the trunk, calf, and sacrum.

A Type IV KF is defined by a partial de-epithelialization and a subfacial placement of the according part of the flap. The wound closure is achieved by the comprehensive mobilization of the flap. Therefore, a split skin graft is necessary for the closure of the donor side. This flap type is often used for defects at the lower extremities or cross-joints defects. In the so-called "omega" variant of Type IV, KF further undermining of the flap's lateral parts while preserving the central hot spot of perforators is necessary. This facilitates an additional rotational movement. The defect is then closed in a fish-mouth fashion. A further modification of the Type IV KF is the SMU modification. In this modification, a skin bridge is maintained along the greater arc of the KF.

The selection of the appropriate flap type depends on several factors, including the anatomical region, its specific location, and the size of the defect. See Table 1 for the flap types we used. For Types I–III, the design ideally follows the course of nerves and muscles along their longitudinal axes, facilitating seamless advancement into the corresponding defect along this axis. For a Type IV flap, the inclusion of a well-defined vascular base within the flap's structure takes on paramount significance, ensuring optimal functionality. When dealing with closures over convex structures like the mandible or tibial edge, exercising caution is essential. Such areas usually exhibit limited tissue mobility, thereby predisposing the flap to unnecessary tension.

**Table 1.** Details of defect location, size, and utilized Keystone Flap type.

| Localization | Defect Size [cm] | Keystone Flap Type |
|---|---|---|
| Right Forearm | $1 \times 2$ | III |
| Right Forearm | $1 \times 1$ | IIA |
| Left Forearm | $3 \times 1$ | IV—SMU modification |

The procedures were performed under general anesthesia due to the patient's age with the patient in the supine position. In general, keystone flap surgery can be conducted under local anesthesia in an ambulatory setting in many cases, depending upon the dimensions and location of the soft tissue defect. Furthermore, if the tissue defect occurs in different locations, the patient can also be in a prone position or positioned sideways, enabling the best surgical access to the defect side.

In the pursuit of minimizing potential complications related to flap perfusion and enhancing confidence in vascular viability, the integration of advanced techniques becomes indispensable. In this context, the employment of tools like hyperspectral imaging analysis and thermography examinations proves helpful, both during and after surgery. These sophisticated methodologies act as observers, offering real-time insights into the vascular status and contributing to a more assured surgical outcome (see Figure 3).

After fitting the KFs into the defects, closure by planes is to be performed depending on the flap thickness. A continuous suture is used with absorbable material in children compared to a polypropylene suture in adults.

In the final intraoperative hyperspectral and thermography analyses after wound closure, the perfusion of the KFs was shown to be satisfying (see Figure 3).

Flap monitoring and meticulous wound care are paramount elements in the early stages of postoperative management. In the initial days following the procedure, close clinical monitoring of the flap's perfusion, color, and overall health is imperative to swiftly identify any signs of compromise. Concurrently, daily dressing changes facilitate a hygienic environment conducive to optimal healing. As the healing process progresses, the frequency of dressing changes is gradually reduced in alignment with the wound's stabilization.

The affected extremities were dressed in a cotton bandage and covered in a self-adhesive gauze bandage.

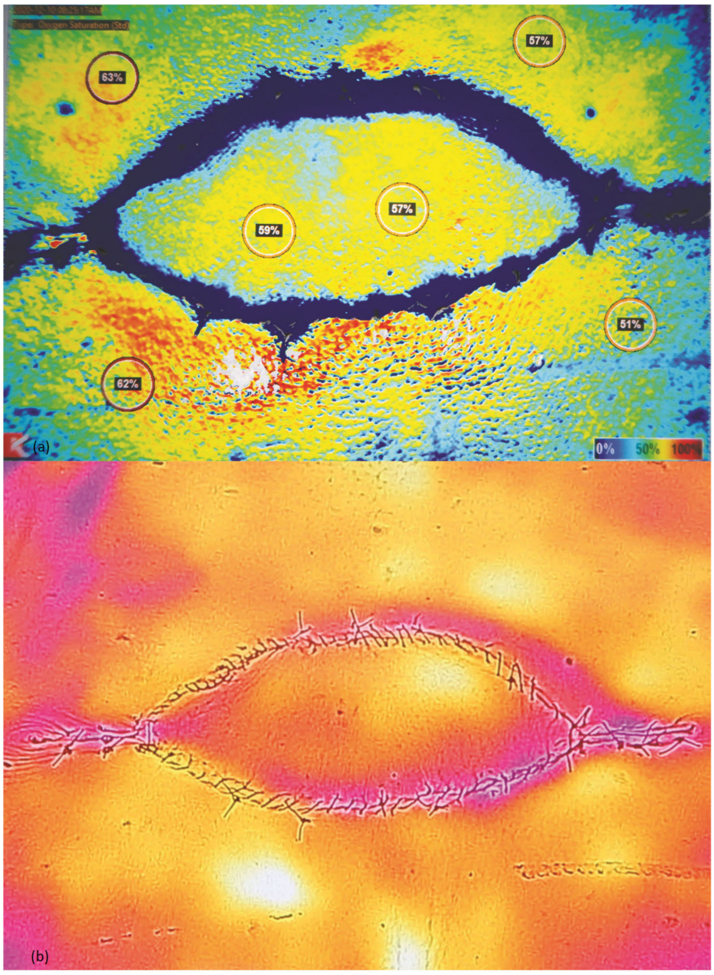


**Figure 3.** Exemplary perfusion analysis in (**a**) hyperspectral imaging and (**b**) thermography.

### 2.2. Operative Timing

Given the diverse range of potential manifestations of ABS, the surgical approach and timing should be individually customized to suit each patient's distinct needs and presentation. In cases where constrictive amniotic rings or associated defects are detected, surgical repair is recommended. Immediate treatment is necessary in the case of vascular compression or all-layered tissue necrosis and constriction rings [21]. Additionally, prompt intervention is warranted if signs of nerve compression emerge within the initial week of life, as early treatment has been shown to enhance the potential for functional recovery [22]. Monitoring the clinical response through techniques like electromyographic and nerve conduction studies can further guide the course of early nerve decompression [23]. Furthermore, if there is active bleeding in the area of the band abrasion at birth, the constriction might exacerbate as new scar tissue forms, potentially resulting in vascular compromise, emphasizing the need for prompt surgical treatment.

If there is no prompt intervention warranted, consensus among many researchers suggests delaying surgical intervention until the child reaches an age of at least 3 to 6 months. This precautionary approach aims to mitigate the risks associated with general anesthesia. However, experts also advocate for completing band resection prior to the child turning 2 years old, in order to minimize the potential for psychological trauma [24]. In terms of contraindications, aside from standard anesthesia-related considerations, there are no specific barriers to surgically correcting amniotic bands or associated defects.

## 3. Results

In the course of our conducted postoperative perfusion evaluations, the Keystone Flaps consistently demonstrated robust vascularization and vitality. Our observations yielded an absence of wound dehiscence, (partial) flap loss, healing complications, or any indication of lymphedema.

The patients were regularly re-examined postoperatively (see Figure 1). We documented the results of surgery based on function, appearance, and development of scarring and contractures.

In one case the right elbow showed a minor extension contraction. Besides that, there were no adhesions or extensive scarring, and the children's development showed to be regular.

## 4. Discussion

Since 1832 when Montgomery originally mentioned the term amniotic constriction band, the term has been used to represent a variety of associated congenital abnormalities, including constriction rings of the extremities, disfigurements, and infrequent manifestations such as the complete absence of a limb, craniofacial deformities, and trunk defects [1,9,12].

In 1961 Patterson classified the extent of constrictions of the amniotic band as (1) involvement of simple annular constriction of the extremities; (2) the extremity distal to the ring is normal despite the presence of subcutaneous tissue defects at the level of the ring; (3) there is acrosyndactyly or fenestrated syndactyly; and (4) amputation at any level of the extremity or a digit (see Table 2) [25].

**Table 2.** Classification system of congenital ring constriction by Patterson [25].

| Type | Definition |
|---|---|
| I | Simple ring constriction |
| II | Ring constriction with presence of subcutaneous tissue defect at the level of the ring |
| III | Ring constriction with any syndactyly or acrosyndactyly |
| IV | Any (intrauterine) amputation |

While the etiology of ABS is not yet fully understood, it is known that the anatomy proximal to a constriction ring remains preservable and recoverable. Hence, it is one of the guidelines for reconstructive surgeons to restore form and function [21]. Therefore, reconstructive strategies have been developed over time, which meet the following criteria: (1) providing similar tissue in terms of function, color, texture, and sensitivity; (2) causing no or minimal functional or aesthetic morbidity of donor areas; (3) being versatile for any reconstructive requirement; (4) requiring short operation durations; and (5) being replicable with quick learning curves.

Full-thickness defects in a newborn infant may pose a considerable infectious complication and deserve early coverage with adequate tissue. Tissue Engineering and Regenerative Medicine are emerging techniques that might prove valuable for infants but are not clinically available yet [26,27]. So, flap coverage remains the gold standard up to now [28]. The Keystone Flap satisfies all the aforementioned requirements and presents a valuable option within the armamentarium of a reconstructive surgeon.

In 2003, Behan et al. first introduced the KF for reconstruction after skin cancer excisions [13]. Since then, the KF applications have expanded to treat complex defects of over 150 various disorders and etiologies distributed over the body, highlighting the increasing applications of the keystone flaps [16,17,29–31]. The classification of Keystone Flaps encompasses four distinct types, each denoted by a progressive level of preparation involving the deep fascia and the meticulous undermining of subcutaneous tissue (see Section 2.1: Operative Technique). Implicit within these types is the prominent role of bilateral V-Y advancement of the flap tissue, exemplifying a pivotal facet of Keystone Flaps' foundational principles. Particularly, in the context of infant or neonatal patients, the significant potential for skin relaxation over time emerges as a notable asset, potentially conferring a distinct advantage over alternative reconstructive avenues. There is no doubt that in comparison to a defect reconstruction with a skin graft, KFs are more efficient and are superior concerning tissue retraction, pigmentation, texture, lack of volume, and donor site morbidity. Nevertheless, in some large defects, a skin graft may become necessary for the KF's donor site depending on the type of utilized KF. Employing a tissue expander to enlarge adjacent tissue primarily offers additional tissue for reconstruction. Nevertheless, the process of tissue expansion demands multiple procedures extended over a prolonged timeframe, rendering it unsuitable for newborns or infants. While microsurgery has become a clinical routine, it includes some pitfalls and possible resulting complications. The extended surgical duration, intricate demands for specialized microsurgical expertise, potential for prolonged anesthesia exposure, and the orchestration of postoperative care needed for microsurgical free flaps collectively present as shortcomings when compared with the perioperative management advantages offered by the KF technique. Moreover, executing microsurgery in a newborn or an infant bears a resemblance to super-microsurgery, demanding extensive training and a mastery of the most delicate microsurgical techniques due to the minute vessel diameters involved. In this light, the KF stands out for its capacity to streamline and simplify the overall management process, enhancing efficiency while still achieving sophisticated reconstructive outcomes.

Compared to typical perforator flaps, the distinct identification and preparation of the specific perforator or the source vessel are not necessary in the case of KF, reducing the complexity, morbidity, perioperative risks, and surgical time [13]. Therefore, while various flap options are available for ABS reconstruction, the keystone flap demonstrates a balance between simplicity, efficiency, and favorable outcomes for all-layered tissue necrosis.

Further operative techniques have been used to address the release of constriction rings and subsequent resection defects in amniotic band syndrome. Conventionally, the release of constriction rings of the extremities used to follow a two-stage approach, separated by a span of 6 to 12 weeks. This operative approach is intended to preserve vascular supply, especially to the distal extremity. However, the trend has shifted towards a single-stage circumferential band release, which has gained popularity and has not been associated with a heightened risk of ischemic complications or venous congestion [24,32]. The resection process should encompass not just the constriction ring itself, but also the surrounding skin where subcutaneous thickness starts to diminish. This ensures there is enough soft tissue available for closure without inducing tension. As well as in all-layered tissue necrosis, simply excising by the constriction ring affected tissue might expose the deep fascia, underlying musculotendinous units, ligaments, or bones. Therefore, comprehensive mobilization of the soft tissue is necessary, to ensure optimal closure of the resection defect. However, this frequently results in an hourglass-shaped defect due to the loss of subcutaneous tissue along with the resected bands. This soft tissue deficit can be mitigated using turnover dermal-fat flaps, a technique originally outlined by Mutaf and Sunay [33]. In this method, the proximal subcutaneous fat and a small portion of the deepithelialized dermis are released at a superficial level. The flap is then unfolded either from the upper aspect or the lower aspect of the defect, effectively serving as a turnover flap to fill the dead space. Modifications of the original technique by Mutaf and Suny have been described by Inglesby et al. executing the release and turnover of the subcutaneous fat without the inclusion of the overlying dermis [21]. Retaining a segment of the subcutaneous tissue along with the undermined dermis is believed to safeguard extra volume and blood supply for the incised skin edges. This becomes crucial when these turnover flaps are brought together with the distal skin edges for closure and might encounter tension. Subsequently, the turnover subcutaneous flaps can be secured in position by suturing them to the fascia underlying the soft-tissue defect resulting from the excision of the constriction ring. Additional methods employed to alleviate tension during the closure, avert contraction, and streamline subsequent linear excision-based reconstruction involve the use of Z-plasties, and triangular or rectangular-shaped flaps [32].

An alternative approach to filling the soft-tissue space, distinct from using local flaps, involves employing autologous fat grafting subsequent to creating perpendicular subcision releases into the band for circumferential release [34].

Recurrence rates concerning extremity constriction ring subsequent to excision and reconstruction occur in up to one third of the cases [35]. These rates are believed to correlate with cases, in which the complete depth of constriction ring tissue was not entirely resected during the surgical intervention. Postoperative deformities following keystone flap procedures are typically rare. However, in some cases, minor deformities or suboptimal outcomes can occur. These may include issues such as subtle contour irregularities, minor asymmetry, or scar contractures. Furthermore, complication rates are described to be low. The most common complications seen in the KF include dehiscence, cellulitis, maceration, sensory loss, contractures, and (partial) flap necrosis due to inadequate mobilization leading to flap loss because of circulatory compromise [19,29,30].

It is important to note that meticulous surgical planning, technique, and postoperative care can significantly minimize the risk of such deformities and complications. Different than expected, Doppler localization techniques or further circulatory diagnostic have often not been used perioperatively because the flap is aligned along the cutaneous nerve supply incorporating the perforators that accompany the peripheral nerves [13,36].

Since flap perfusion can be difficult to judge and detect any microcirculatory problems, technical aids such as indocyanine green laser angiography, hyperspectral imaging analysis, or thermography examinations can be easily used intra- and postoperatively [37–41]. These technical devices objectify and visualize perfusion differences securing a higher flap survival rate and can help to trim critically perfused edges of the flap eventually (see Figure 3).

Furthermore, a KF with its short surgery duration and simpler surgical technique is advantageous both for the patient and for the surgeon.

The following limitations should be acknowledged in this study. Firstly, due to the rarity of amniotic band syndrome, the included number of Keystone Flaps is relatively small. While our findings provide valuable insights into the application of Keystone Flaps in this specific context, we are aware that the limited number of Keystone Flaps might restrict the generalizability of our results. Additionally, the absence of long-term follow-up data is another notable limitation. Although we present our observations based on the immediate postoperative period and early outcomes, the lack of extended follow-up information prevents us from making conclusive statements regarding the durability and sustainability of the surgical outcomes over time. Consequently, while our study contributes valuable information, future research with larger cohorts and longer-term assessments is essential to establish a more comprehensive understanding of the efficacy and longevity of Keystone Flap reconstructions in the context of amniotic band syndrome.

## 5. Conclusions

In summary, the Keystone Flaps demonstrate impressive versatility, functionality, and aesthetic outcomes, supported by favorable results both in the literature and in our own experience. This easy-to-learn and efficient technique is suitable for novice and experienced reconstructive surgeons alike, offering a reliable option for reconstructing full-thickness defects. By utilizing adjacent tissue, KF yields superior aesthetic results compared to alternative approaches such as simple skin grafts or other flap types. As a result, this technique holds great promise for its application in newborn patients.

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