Clinical assessment of the results of one-stage circular incision techniques for limb ring constriction due to amniotic band syndrome

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Abstract

Background: Amniotic band syndrome (ABS) is a congenital malformation that results in abnormalities in many parts of the body. Most surgical treatments for ABS used multi-stage Z-plasties. The purpose of this study was to assess the clinical results of one-stage circular incision techniques for limb ring constriction due to amniotic band syndrome.

Methods: We reviewed 27 patients with limb ring constriction in ABS from 2010 to 2020. The mean ages of the patients were 11.7 months (range, 0–72 months). The complete circular incision release the ABS. All patients’ operations were used one-stage circular incision surgical techniques, including patients with multiple bands. All the patients were followed up with a period ranges from 2 years to 10 years. Patient-reported visual analog scale (VAS) scar ranking on a scale of 0 (minimum satisfaction) to 10 (maximum satisfaction) were used to evaluate esthetic outcomes.

Results: After our surgery, all the limbs, toes, and fingers were rescued, and the lymphedema reduced remarkably. The VAS scores (mean±SD) for patient satisfaction were 7.55±1.89. The surgical treatment of amniotic band syndrome in a one-stage circular incision is safe and effective.

Conclusion: The one-stage circular incision surgical techniques have many advantages, including reduced surgical invasiveness, scar formation, and the cost of treatment.

Level of Evidence: Level IV—retrospective case series.

Abbreviations: ABS = amniotic band syndrome, VAS = visual analog scale.

Keywords: amniotic band syndrome, circular incision, congenital constriction band syndrome, constriction ring, surgical treatment

1. Introduction

Amniotic band syndrome (ABS) is a congenital malformation that results in abnormalities in many parts of the body.\textsuperscript{[1]} ABS is a rare congenital disorder caused by entrapment of fetal parts (usually a limb or digits) in fibrous amniotic bands.\textsuperscript{[2]} The etiology is debated. Many scholars believed that amniotic band syndrome consists of a group of congenital abnormalities caused by strands of the amniotic sac that entangle some parts of the fetus.\textsuperscript{[3]} Those strands result from premature rupture of amnios. The incidence of amniotic band syndrome is 1:1200 to 1:15,000 live births, depending on case studies.\textsuperscript{[4]} While there are many diverse manifestations of ABS, constriction bands about the extremities are among the most common, with consequential lymphedema, vascular and neural deficits, and aesthetic loss.\textsuperscript{[5]} The ratio between males and females affected is almost the same, and all ethnic groups are equally affected.\textsuperscript{[6]} This disease is not hereditary. Despite multiple hypotheses, this disease is not well understood.\textsuperscript{[7]} There are 3 theories to explain the mechanism: intrinsic theory, extrinsic theory, and intrauterine trauma theory.\textsuperscript{[8]}

Mostly affected parts of the fetus are limbs (amputations, syndactyly, and clubbed foot), but facial and other parts of abnormalities have also been described.\textsuperscript{[9,10]} ABS is characterized by a fibrous band that tightly encircles the limb, either partially or entirely. Some studies released circumferential ABS in 1 stage with Z-plasties techniques.\textsuperscript{[8]} This method proved to be safe and effective.

To investigate the outcomes of a one-stage circumferential limb ring constriction in ABS release with a complete circular incision technique for the correction of ABS. We hypothesized that a one-stage circular incision for ABS is a safe and feasible method.
2. Materials and methods

This investigation was reviewed and approved by the Research Ethics Board at the Huashan Hospital (2015M-004–163). Parents and patients received detailed information. All patients presenting with ABS affecting the limbs were evaluated in the period between 2010 and 2020. The study comprised 27 patients with a total of 47 ring constrictions. Thirty ring constrictions (63%) appeared on the upper extremity, and 17 (37%) appeared on the lower extremity. Twenty ring constrictions (43%) were deep, and 27 (57%) were superficial. The patients’ ages ranged from 24 hours to 6 years. The male-to-female ratio was 10:17. All patients were treated with a one-stage circular incision and circular suture without Z-plasties. The mean follow-up was 3 years (2years–10years). The mean ages at surgery were 11.7 months (range, 0–72months) (Table 1).

Patient-reported visual analog scale (VAS) with scar ranking on a scale of 0 (minimum satisfaction) to 10 (maximum satisfaction) were used to evaluate esthetic outcomes.\[11\]

### 2.1. Surgical technique

The skin cut at the healthy skin level proximal and distal to the constriction ring. The incision depth does not exceed the dermis. Carefully remove the surface skin with the dermis using tweezers and scissors. The skin excised under cautious protection of underlying vessels, nerves, and tendons. At the bottom of ABS, a circular ring can be located, which can involve the muscle fascia and even reach the bone. This ring must wholly and carefully remove, conserving the underlying structures such as muscles, tendons, veins, arteries, and nerves. The muscle fascia is cut proximally and distally of the rings several times in a longitudinal direction. A complete reveal of the constriction ring and releasing the amniotic band. The skin and subcutaneous soft tissue proximal and distal to the ring organized in a circular pattern. The soft tissue of both sides, a multilayer linear, circular skin closure performed.

Patients with deep ring ABS generally have lymphedema, blood supply disorders, and severe nerve compression symptoms, so early surgical treatment is essential.

### 3. Results

All the patients were followed up with a period ranges from 2 years to 10 years. After our surgery, all the limbs, toes, and fingers were rescued, and the lymphedema was reduced remarkably. The

| Table 1 | Summary of the clinical series of amniotic band syndrome. |
|---------|----------------------------------------------------------|
| Case    | Age   | Sex      | Position                          | Depth | Complications/Scars (VAS scores) |
|---------|-------|----------|-----------------------------------|-------|---------------------------------|
| 1       | 1 mo  | male     | L hand; L index finger; L ring finger | Deep  | None/6                           |
| 2       | 3 mo  | Female   | L lower leg (Double bands)         | Deep  | None/7                           |
| 3       | 4 mo  | Female   | L lower leg; R lower leg           | Superficial | None/8                       |
| 4       | 2 mo  | Female   | L forearm                         | Deep  | None/8                           |
| 5       | 2 mo  | Male     | R forearm                         | Deep  | None/7                           |
| 6       | 4 mo  | Male     | L middle, ring, little finger (Syndactyly); First toe of L foot | Superficial | None/6                       |
| 7       | 3 mo  | Male     | L lower leg                       | Deep  | None/7                           |
| 8       | 3 mo  | Female   | L middle, ring finger (Syndactyly); L forearm | Superficial | None/8                       |
| 9       | 2 mo  | Male     | L middle, ring, little finger (Syndactyly); R upper arm | Superficial | None/7                       |
| 10      | 7 mo  | Female   | L index finger amputation         | Deep  | None/7                           |
| 11      | 3 mo  | Female   | L hand; L middle finger           | Superficial | None/2                     |
| 12      | 10 mo | Male     | L lower leg                       | Deep  | None/4                           |
| 13      | 2 yr  | Female   | L thumb, middle, little finger     | Superficial | None/5                     |
| 14      | 10 mo | Male     | R middle ring finger (Syndactyly); L index middle finger (Syndactyly); First toe of L foot | Superficial | None/9                     |
| 15      | 6 yr  | Male     | L middle finger                   | Deep  | None/10                          |
| 16      | 6 mo  | Female   | L middle finger; L ring finger     | Superficial | None/9                     |
| 17      | 3 yr  | Male     | L thigh                           | Deep  | None/10                          |
| 18      | 6 mo  | Female   | R middle ring finger (Syndactyly); L middle ring finger (Syndactyly); R foot 1–4 toes (Syndactyly) | Superficial | None/9                     |
| 19      | 5 mo  | Female   | R middle finger; L lower leg       | Deep  | None/9                           |
| 20      | 4 yr  | Male     | R thumb index middle (Syndactyly)  | Superficial | None/10                     |
| 21      | 10 mo | Male     | L forearm; L lower leg             | Deep  | None/9                           |
| 22      | 7 mo  | Female   | R index finger; R foot 3–5 (Syndactyly) | Superficial | None/10                     |
| 23      | 1 yr  | Male     | L forearm; R forearm               | Deep  | None/9                           |
| 24      | 6 mo  | Female   | R middle ring finger (Syndactyly); L index middle ring little finger (Syndactyly) | Superficial | None/7                     |
| 25      | 3 mo  | Male     | L forearm                         | Deep  | None/6                           |
| 26      | 24 h  | Female   | R lower leg                       | Superficial | None/8                     |
| 27      | 2 yr  | Male     | R forearm                         | Deep  | None/7                           |

L = left, R = right. VAS, Visual Analog Scale with scar ranking scale of 0 (minimum satisfaction) to 10 (maximum satisfaction).
VAS scores with scar ranking (mean ± SD) for patient satisfaction were 7.55 ± 1.89 (Table 2).

Early release of the bands in 1 stage with a transverse incision is a safe and effective technique for severe ABS, especially for multiple bands in unilateral limb involvement. The scar after the surgery had excellent cosmetic results.

The limb after 1 stage ABS release showed healthy development, healed satisfactorily, had no functional deficiency,
recovered, and demonstrated full function (Figs. 1–6). Complete circular resection never leads to a noticeable decline of blood circulation distal to the constrictions, even if blood flow already lower before surgery.

There were no postoperative complications that required additional treatment. No constriction could observe during the follow-up. All limbs (fingers, toes) were rescued, improved blood

Figure 3. Picture A and B: One-stage excision of the right forearm’s amniotic band, complete circle suture. Picture C shows the excess skin removed.

Figure 4. (A–C) Two-year follow-up photo after the releasing of the left calf amniotic band, no recurrence was found, and small scar.
Figure 5. (A–C) Picture A: Three-year follow-up photos after the release of the amniotic membrane band of the right calf, no recurrence was found, and the cosmetic effect was good. Picture B: Two-year follow-up photos after the release of the amniotic membrane band of the left calf, no recurrence was found, and good cosmetic results. Picture C: Two-year follow-up photo after the release of the left forearm amniotic band, no recurrence was found, the good cosmetic results.

Figure 6. (A–C) Picture A–C: Ten-years follow-up photo after the release of the multi-amniotic band. No recurrence of the constrictions or distal lymphoedema could be observed.
circulation, disappeared neurological symptoms, and reduced lymphedema.

4. Discussion

Resection and soft-tissue modification by multiple Z-plasties has been the treatment of choice for ABS. However, this study confirms that a one-stage circumferential ABS release with the circular incision is considered a safe operation for the treatment of both circumferential and semi-circumferential ABS. Some authors reported the pelvic constriction band in a newborn female with ABS.[11]

In the treatment of constriction rings, the key to success is the complete resection of all constric ting rings and the longitudinal incisions of the deep fascia. Scars should not cross joints longitudinally in order to prevent movement restrictions lead to scar contracture. By switching of 2 or more flaps, Z-plasties allowed additional relief of skin tension by rearrangement of the relative redundancy of adjacent skin.[13] Circular scars at the extremities generally refrained from scar contractures and circular constriction.[14]

The risk of vascular variation in deep bands was 100%.[15] The regular timing to release the ring constriction depends on the severity of the disease itself.[8] When amniotic bands affect nerves and blood vessels, surgery should perform without hesitation. Careful protection of the structures under the constriction rings prevented the circulation problem caused by the circular resection. In our experience, superficial ABS in young children do not deepen with growth. The one-stage release technique has the advantage of avoiding repeat anesthesia, painful postoperative care, and a lower hospital admission rate.[16]

Z-plasties would increase scars, and the linear, circular incision will reduce it.[16] There is no contracture with growth since complete release, and circular incision converted the deep constriction rings into superficial scars. Superficial scars and ABS in children do not deepen with growth. Since 2010, we released deep ABS at the extremities by complete circular incision and linear circumferential skin closure. Careful protection of the nerve and vessel under the fibrous bands can prevent related complications. Microvascular toe-to-hand transfer for severe hand deformity in Amniotic Band Syndrome is a valid surgical approach.[17] Some authors reported that fetoscopic release of the amniotic bands in the case of amniotic band syndrome is feasible in order to prevent amputation and dysfunction of the extremities.[18]

In all series, we released the ABS in 1 stage. All limbs (fingers, toes) were rescued, improved blood circulation, disappeared neurological symptoms, and reduced lymphedema. Our study showed the same result with this literature: our technique had better aesthetic results with circular incision than Z-plasty.[19] A single-stage correction approach provided satisfactory both functional and aesthetic results. Given many morphological variations of the syndrome, a decision on the strategy of treatment should be made individually for each patient. One-stage surgery with direct closure is a simple technique that provides satisfactory functional and esthetic results.

This surgery technique is not novel, but this technique is not very widely known. This technique offers several advantages over, including reduced surgical invasiveness, scar formation, and the cost of treatment. We believed that our technique is safe and reliable, but this study is limited by small sample size, retrospective design, and lack of a control group. Further studies with long-term follow-up could address these issues.

In the treatment of ABS, the key to success:

1. Deep amniotic bands must fully release by longitudinal incision.
2. Adequate skin coverage required after surgery.
3. Full consideration should give to the appearance of surgical incisions and surgical scars.

Author contributions

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