

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

Le Wang, MD, PhD^a, Yousheng Fang, MD, PhD^{b,c,d,*} 

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.^[1] ABS is a

Editor: Mohamed Fahmy.

The authors have no conflicts of interests to disclose.

The datasets generated during and/or analyzed during the current study are available from the corresponding author on reasonable request.

^a Department of Pediatric Surgery, Shanghai Jiao Tong University Medical School Affiliated Ruijin Hospital, Shanghai, PR China, ^b Department of Hand Surgery, Huashan Hospital, Fudan University, Shanghai, PR China, ^c Key Laboratory of Hand Reconstruction, Ministry of Health, Shanghai, PR China, ^d Shanghai Key Laboratory of Peripheral Nerve and Microsurgery, Shanghai, PR China.

* Correspondence: Yousheng Fang, Department of Hand Surgery, Huashan Hospital, Fudan University, Shanghai, PR China, Key Laboratory of Hand Reconstruction, Ministry of Health, Shanghai, PR China, Shanghai Key Laboratory of Peripheral Nerve and Microsurgery, Shanghai, PR China (e-mail: 1721129185@qq.com).

Copyright © 2021 the Author(s). Published by Wolters Kluwer Health, Inc. This is an open access article distributed under the terms of the Creative Commons Attribution-Non Commercial License 4.0 (CCBY-NC), where it is permissible to download, share, remix, transform, and buildup the work provided it is properly cited. The work cannot be used commercially without permission from the journal.

How to cite this article: Wang L, Fang Y. Clinical assessment of the results of one-stage circular incision techniques for limb ring constriction due to amniotic band syndrome. *Medicine* 2021;100:30(e26764).

Received: 28 January 2021 / Received in final form: 17 June 2021 / Accepted: 9 July 2021

<http://dx.doi.org/10.1097/MD.00000000000026764>

rare congenital disorder caused by entrapment of fetal parts (usually a limb or digits) in fibrous amniotic bands.^[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.^[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.^[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.^[5] The ratio between males and females affected is almost the same, and all ethnic groups are equally affected.^[6] This disease is not hereditary. Despite multiple hypotheses, this disease is not well understood.^[7] There are 3 theories to explain the mechanism: intrinsic theory, extrinsic theory, and intrauterine trauma theory.^[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.^[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.^[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 (2 years–10 years). The mean ages at surgery were 11.7 months (range, 0–72 months) (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,

Table 2

Esthetics evaluation visual analog scale with scar ranking scores on a scale of 0 (minimum satisfaction) to 10 (maximum satisfaction) were used to evaluate esthetic outcomes.

VAS scores	N (%)
Poor (0–2)	1 (3.7%)
Fair (3–5)	2 (7.4%)
Good (6–8)	14 (51.8%)
Excellent (9–10)	10 (37%)

VAS scores (Mean ± SD) for patients' satisfaction with esthetics were 7.55 ± 1.89. N = number of people.

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	Femal	L thumb, middle, little finger	Superficial	None/5
14	10 mo	Female	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	Female	L forearm; R forearm	Deep	None/9
24	6 mo	Female	L middle ring finger (Syndactyly); R index middle ring little finger (Syndactyly)	Superficial	None/7
25	3 mo	Female	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).



Figure 1. (A–B) Two months old boy with circumferential amniotic band syndrome affecting the right forearm.

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,



Figure 2. (A–C) Intra-operative pictures, resection of the amniotic membrane band along the entire circle under the dermis to loosen the deep ABS.

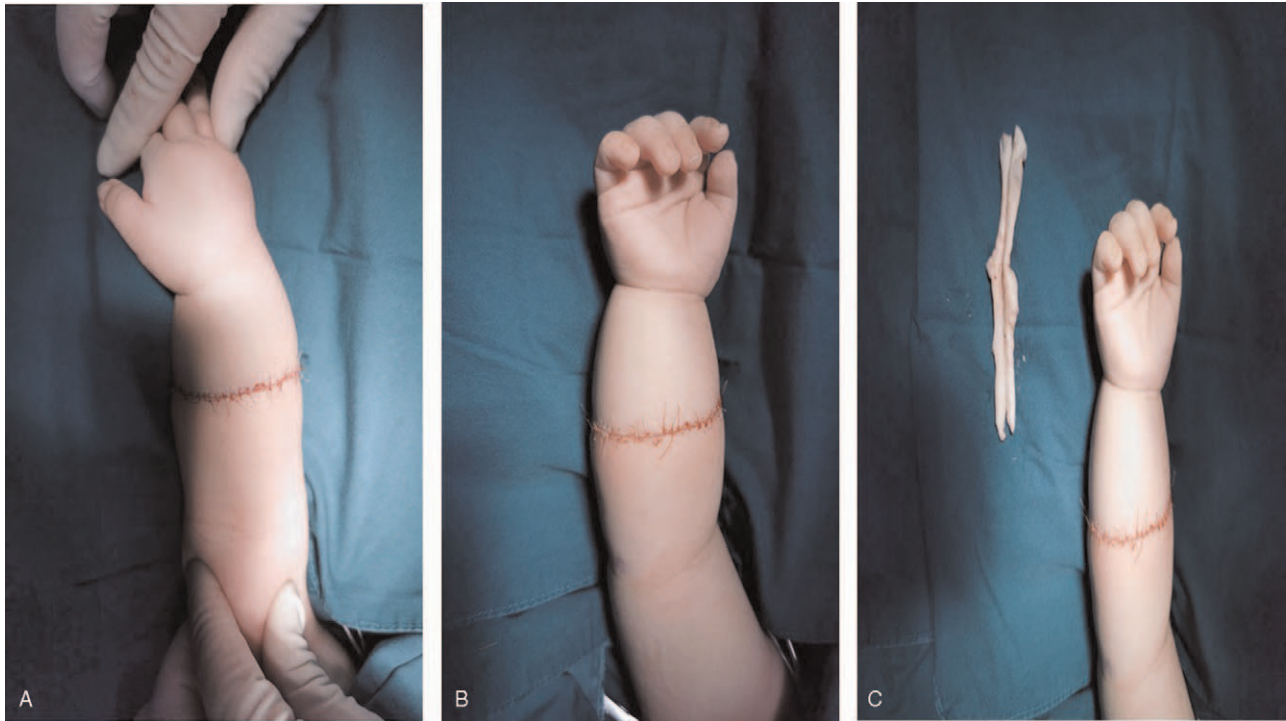


Figure 3. Picture A and B: One-stage excision of the right forearm’s amniotic band, complete circle suture. Picture C shows the excess skin removed.

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

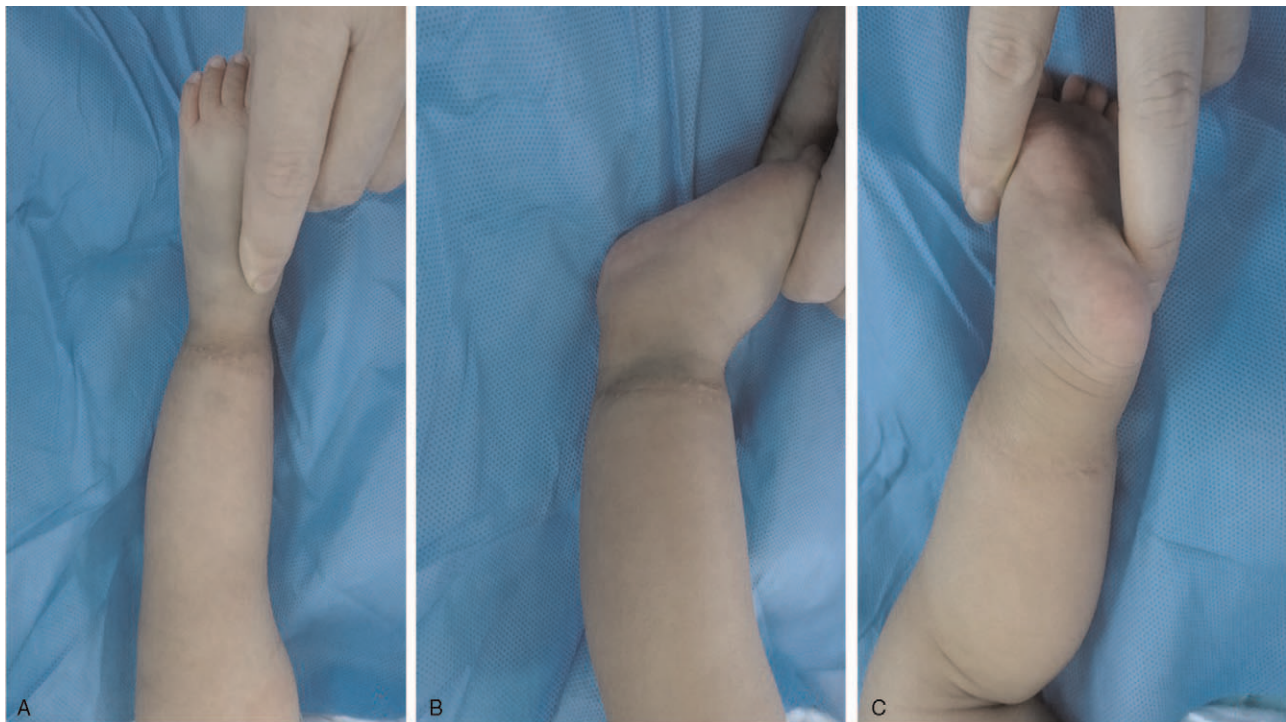


Figure 4. (A–C) Two-year follow-up photo after the releasing of the left calf amniotic band, no recurrence was found, and small scar.

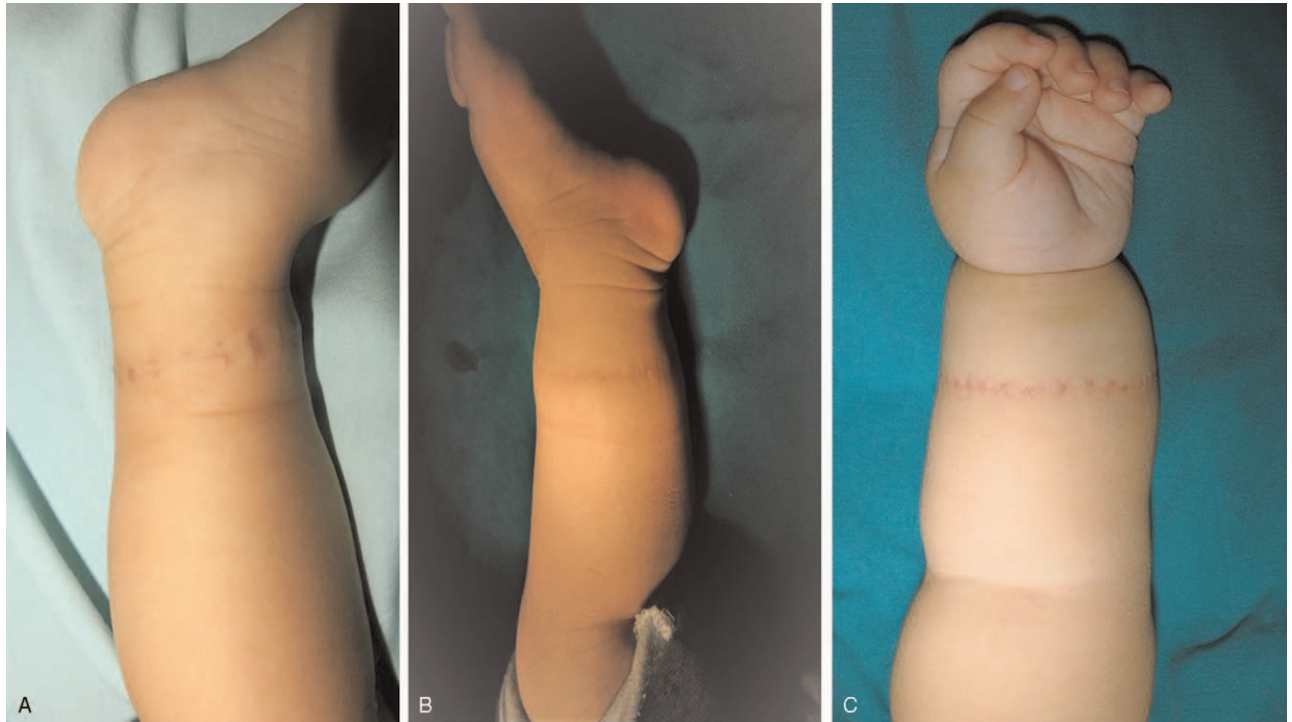


Figure 5. (A–C) Picture A: Three-year follow-up photos after the release of the amniotic membrane band of the right calf, no recurrence was found, and the cosmetic effect was good. Picture B: Two-year follow-up photos after the release of the amniotic membrane band of the left calf, no recurrence was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

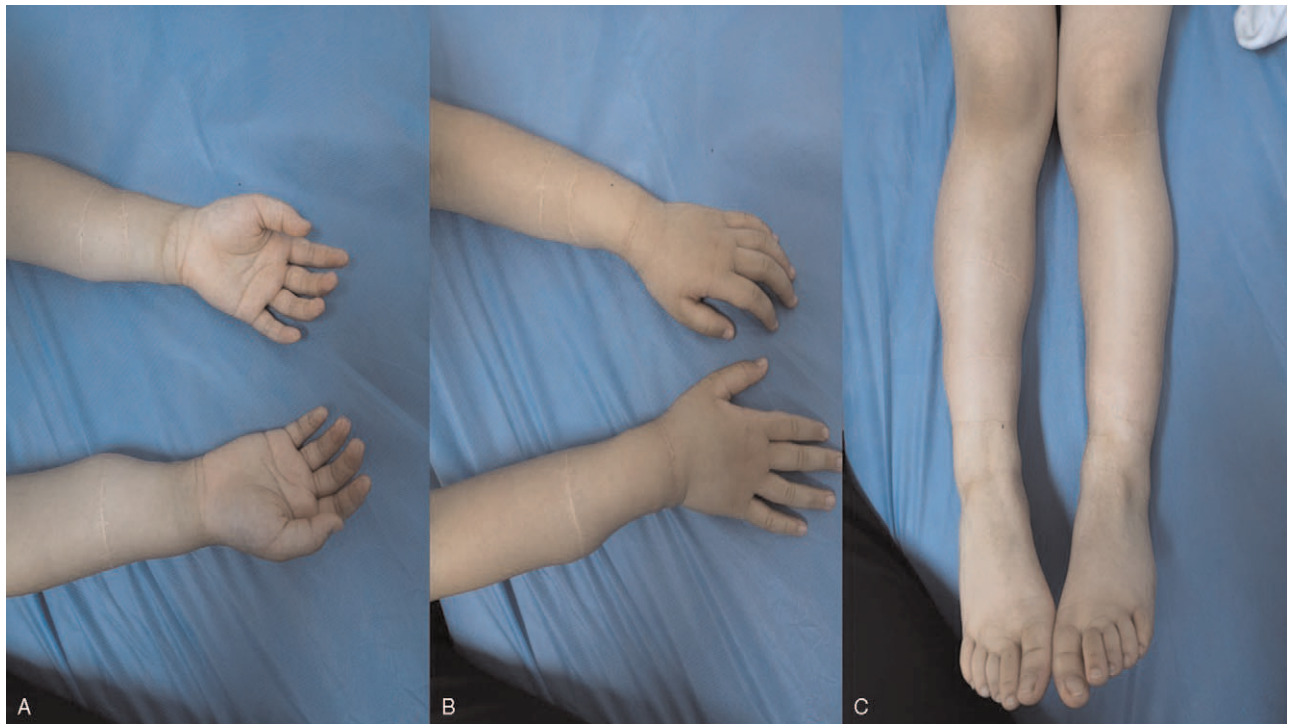


Figure 6. (A–C) Picture A–C: Ten-years follow-up photo after the release of the multi-amniotic band. No recurrence of the constrictions or distal lymphoedema could be observed.

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.^[7,12] 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 constricting 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

Project administration: Yousheng Fang.

Supervision: Yousheng Fang.

Writing – original draft: Le Wang.

References

- [1] Seeds JW, Cefalo RC, Herbert WN. Amniotic band syndrome. *Am J Obstet Gynecol* 1982;144:243–8.
- [2] Esparza M, Tran E, Richards BS, et al. The ponseti method for the treatment of clubfeet associated with amniotic band syndrome: a single institution 20-year experience. *J Pediatr Orthop* 2021;41:301–5.
- [3] Minella C, Costantino B, Ruano R, et al. Fetoscopic release of amniotic band syndrome: an update. *J Ultrasound Med* 2021;40:1039–48.
- [4] Javadian P, Shamsirsaz AA, Haeri S, et al. Perinatal outcome after fetoscopic release of amniotic bands: a single-center experience and review of the literature. *Ultrasound Obstet Gynecol* 2013;42:449–55.
- [5] Graham E, Garlick JW, Johns D. Congenital amniotic constriction band of the pelvis: first reported case and proposed classification criterion. *Plast Reconstr Surg Glob Open* 2019;7:e2336.
- [6] Singh AP, Gorla SR. Amniotic Band Syndrome. In: *StatPearls. Treasure Island (FL)* 2021.
- [7] Goldfarb CA, Sathienkijkanchai A, Robin NH. Amniotic constriction band: a multidisciplinary assessment of etiology and clinical presentation. *J Bone Joint Surg Am* 2009;91(Suppl 4):68–75.
- [8] Prasetyono TO, Sitorus AS. A review on the safety of one-stage circumferential ring constriction release. *Int Surg* 2015;100:341–9.
- [9] da Silva AJF. Amniotic band syndrome with placenta-encephalocoele adhesion: an uncommon case. *J Pediatr Neurosci* 2020;15:160–1.
- [10] He T, Xu H, Sui P, Wang X, Sun Y. Amniotic constriction band syndrome resulting in amputation caused by septate uterus: a case report. *J Int Med Res* 2020;48:300060520949755.
- [11] Yang XD, Zhao SF, Wang YX, et al. Use of extended lateral upper arm free flap for tongue reconstruction after radical glossectomy for tongue cancer. *Aesthetic Plast Surg* 2015;39:562–9.
- [12] Light TR, Ogden JA. Congenital constriction band syndrome. Pathophysiology and treatment. *Yale J Biol Med* 1993;66:143–55.
- [13] Mutaf M, Sunay M. A new technique for correction of congenital constriction rings. *Ann Plast Surg* 2006;57:646–52.
- [14] Yengo-Kahn AM, Plackis AC, Bonfield CM, Reddy SK. Correction of a vertex encephalocele related to amniotic band syndrome. *BMJ Case Rep* 2020;13:3.
- [15] Daya M, Makakole M. Congenital vascular anomalies in amniotic band syndrome of the limbs. *J Pediatr Surg* 2011;46:507–13.
- [16] Dufournier B, Guero S, de Tienda M, et al. One-stage circumferential limb ring constriction release and direct circular skin closure in amniotic band syndrome: a 14-case series. *Orthop Traumatol Surg Res* 2020;106:1353–9.
- [17] Chiu DTW, Patel A, Sakamoto S, Chu A. The impact of microsurgery on congenital hand anomalies associated with amniotic band syndrome. *Plast Reconstr Surg Glob Open* 2018;6:e1657.
- [18] Gueneuc A, Chalouhi GE, Borali D, Mediouni I, Stirnemann J, Ville Y. Fetoscopic release of amniotic bands causing limb constriction: case series and review of the literature. *Fetal Diagn Ther* 2019;46:246–56.
- [19] Habenicht R, Hulsemann W, Lohmeyer JA, Mann M. Ten-year experience with one-step correction of constriction rings by complete circular resection and linear circumferential skin closure. *J Plast Reconstr Aesthet Surg* 2013;66:1117–22.