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SINGLE STAGE SURGICAL RELEASE OF LOWER LIMB CIRCUMFERENTIAL CONSTRICTION BAND IN A PATIENT WITH AMNIOTIC BAND SYNDROME: CASE REPORT

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SUMMARY

Amniotic band syndrome (ABS) also known as constriction band syndrome is a congenital condition caused by a fibrous ring from the amniotic sac, that encircle a developing foetus during pregnancy resulting in multiple deformities such as constriction bands, acrosyndycty and amputations. Circumferential constriction band pose a particular challenge due to the fear of vascular compromise during surgical release hence some literature advocates for staged release.

This report presents a case of a A 15-month-old female with amniotic band syndrome (ABS) affecting all limbs, including a circumferential left leg band, which was treated successfully with a single-stage release using serial Z-plasty.

Postoperatively, healing was excellent, with good palpable distal pulses and complete resolution of lymphedema. This case illustrates the feasibility and safety of single-stage circumferential release in ABS.

Keywords: Amniotic band syndrome, circumferential constriction band, case report

INTRODUCTION

ABS is a rare congenital disorder, reported in approximately 1 in 1,200–15,000 live births, The condition most often affects the extremities but may also involve craniofacial, spinal, abdominal, or visceral structures. Diagnosis typically relies on prenatal imaging using ultrasound visualisation of the resulting deformities, and clinical findings, as the fibrous bands themselves are rarely visible. (1–3)(4).

The exact cause of ABS is still unknown, but several theories have been suggested, including extrinsic theory which suggest that early rupture of amniotic sac result in fibrous bands which entangles foetal parts, The intrinsic theory proposed by Streeter claims that ABS results from underlying intrauterine anomalies that can lead to localised foetal malformations, and this supports the presence of major internal and craniofacial anomalies that are sometimes associated

with ABS a (2,5). Lastly, the vascular theory which suggests that interruption of blood supply to the foetal tissues results in tissue necrosis and limb defects (6–8).

ABS clinical features can be classified into four main categories: 1). Limb defects, 2). constriction bands, 3). neural tube defects and 4). craniofacial defects. Among the four clinical features, constriction bands is the most common feature of ABS especially constriction bands of the limbs which are usually accompanied by digital amputation and acrosyndycty, as seen in the above case (figure-1&2). There are many classifications used to grade limb involvement, with the Patterson classification being widely used. It categories the anomalies into four main types based on the severity of the deformity distal to the band; Type 1 - simple constriction ring without deformity (with the distal extremity being normal), Type II - constriction ring with distal deformity, such as lymphedema or muscle atrophy, Type III - constriction ring associated with

syndactyly or acrosyndactyly (distal fusion of digits , often with fenestration) and Type IV - intrauterine amputation of the extremity or any digit at any level (2,9).

No medical treatment exists for ABS; management is surgical and depends on neurovascular status, swelling, and severity. Historically, staged release of amniotic bands, especially for circumferential defects, was advocated, but emerging literature now supports single-staged complete release, which, when carefully done, offers faster functional recovery(10,11). Lastly, some surgeons advocate for excision of the constriction and direct closure, while some prefer Z or W-plasty (10,12). This report describes a leg circumferential amniotic band in a 15-month-old managed successfully with a single-stage circumferential release without adverse effects on distal perfusion.

CASE PRESENTATION

A 15-month-old female presented with deformities of both the upper and lower limbs. Patient was born at term via spontaneous vaginal delivery with no antenatal or perinatal complications. No family history of congenital anomalies, nor maternal history of drug or alcohol use, nor exposure to teratogens during pregnancy.

On examination, the distal left leg had a complete circumferential constriction band with stage-1 lymphedema and left hand acrosyndactyly of all the digits, auto-amputation of the bilateral great toes and left thumb, and brachydactyly of the right index and middle fingers, as seen in Figure1-2. Distal pulses were present bilaterally with no signs of ischemia.

Given the presence of a circumferential constriction band with lymphedema, surgical correction was planned. A single-stage circumferential release of the left leg constriction band with Z-plasty in series was done, Figure 3-4. The fibrous constriction tissue was released down to the healthy subcutaneous tissue, acrosyndactyly of the left hand managed with a single staged surgical separation of all fused digits and the defects closed primarily. Lastly reconstruction of the right great toe was performed by deepening the first web space with a local flap. Postoperatively, the distal left leg and foot oedema resolved completely; the distal leg and foot were well perfused, with no signs of ischemia and the right hand and left foot healed completely with satisfactory results as seen in figure 5-6. The patient was discharged home on postoperative day two in a stable condition, with a follow-up in clinic.

Figure 1: bilateral lower limb anomalies secondary to ABS



Figure 2: Bilateral hands anomalies



Figure 3: Z-plasty markings



Figure 5: 4-months post-operation, completely healed wound with resolved lymphedema.



Figure 4: Intraoperation after releasing the constriction band



Figure 6: 4-months post-acrosyndactyly release, showing a completely healed and functional left hand.



DISCUSSION

Management of ABS is individualised depending on the type, treatment ranges from conservative to surgical intervention (9,13). Shallow rings may require no treatment unless they impair lymphatic or vascular flow or cause cosmetic concern. Surgical treatment ranges from simple excision of the band and direct closure to z and w-plasty. Surgical intervention can be performed as an elective or emergency, vascular compromise diagnosed immediately postnatally may require urgent surgical intervention(2). Circumferential constriction rings, especially deeper ones, pose a challenge due to the risk of distal vascular compromise during surgical release. Therefore, multistage release of the band was traditionally advocated to avoid disruption of distal blood circulation (10,14). The two stage release of circumferential amniotic band was supported by some authors, the first release was usually performed while the child is 3-months old ,releasing 50% of the band and the remaining band was released after 6-12 weeks, some authors planned the second release at 1 month interval once the wound has healed, the reason for staged procedure was to prevent vascular compromise (14)(12)(10),

However, current literature supports single-stage circumferential release as safe and effective, preventing multiple surgical sessions and enabling immediate restoration of lymphatic and venous drainage. - as demonstrated by the positive outcome in this case (11,15)

In the present case, the decision to perform a circumferential single-stage band release was because the distal (distal to the constriction band) segment skin blood supply is mainly by musculocutaneous arteries hence protecting against secondary necrosis post excision (16). A release with serial z-plasties was performed (figure 3-4), providing immediate decompression and lengthening of the local tissues. The z-plasty technique reduces circumferential tension and the risk of postoperative ischemia. Postoperatively, the patient had immediate resolution of the lymphedema, complete wound healing, and no evidence of distal vascular compromise (figure 5). The above outcome further supports single-stage circumferential release as safe and effective. Moreover, it prevents repeated exposure to anaesthesia and surgery and provides a shorter recovery time, resulting in immediate functional improvement.

CONCLUSION

In this case, a single-stage circumferential release with serial z-plasty effectively relieved lymphatic obstruction and released the constriction band without compromising distal perfusion. This case illustrates the practicality and safety of single stage surgical release of lower limb circumferential constriction band in a patient with amniotic band syndrome and adds evidence to the existing literature.

Ethical approval: This study did not require ethical approval as it is a case report.

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