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Surgical Considerations in the Management of Constriction Ring Syndrome

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Surgical management of constriction ring syndrome (CRS) is individualised due to the heterogenic presentation of the condition. CRS includes constriction rings, acrosyndactyly, nubbins and short digits. Involvement of more than one limb is common and children often need multiple surgeries. Each limb may need staged surgeries. If the child has vascular or lymphatic compromise secondary to a constriction ring, the ring needs to be excised and released in the first few days of life. The rings are released using multiple big Z-plasties in one or two stages. Nerve palsy associated with the rings need early intervention. Tendon transfers may be advised when nerve procedures like neurolysis and nerve reconstruction fail. Acrosyndactyly can be corrected with separation of the fused fingertips. At times, the web is more distal than usual and requires web deepening using partial syndactyly separation techniques and may need full thickness skin grafting. Complex type of acrosyndactyly is difficult to treat as it needs proper planning and staged surgical correction to achieve a five-digit hand. Separation of the syndactyly with web deepening gives more functional length and independence to the digits. The short fingers, especially the thumb, need reconstruction. Non-vascularised toe phalangeal transfer or a microvascular toe transfer can reconstruct a missing digit.

Level of Evidence: Level V (Therapeutic)

Keywords: *Constriction ring syndrome, Amniotic band, Toe transfer, Syndactyly, Acrosyndactyly*

INTRODUCTION

Constriction ring syndrome (CRS) is a common congenital limb deformity that is frequently observed in Asian countries.[1] The clinical manifestations of CRS are varied and can affect multiple limbs. The clinical presentation in CRS includes constriction rings, acrosyndactyly, nubbins and short digits. The treatment strategy is individualised due to the heterogenic presentation of CRS. There is a significant amount of uncertainty regarding the aetiology.[2] According to the extrinsic theory, rings are formed when the amniotic membranes rupture and wrap around the limbs or parts of them, causing external compression, resulting in the distinct constriction rings. However, the extrinsic theory does not explain the anomalies like craniofacial anomalies, renal defects, cardiac defects and abdominal wall defects that are not band explicable.[1] This is supported by the intrinsic theory, which speculates that the deficiency originates from abnormalities in the germplasm.[4] Prenatal ultrasound imaging has the capability to occasionally identify constriction rings in developing foetuses. Upon delivery, the infant can undergo a more comprehensive evaluation with appropriate radiographs. This can help differentiate it from symbrachydactyly (Table 1).[5,6]

The aim of this article is to discuss surgical considerations in patients with CRS. These are based on our experience with 146 children with CRS who were treated

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Table 1. Symbrachydactyly versus Constriction Ring Syndrome

Symbrachydactyly	Constriction ring syndrome
• Unilateral	• Can involve multiple limbs.
• Generalised limb hypoplasia	• Hypoplasia is not a feature.
• The soft tissue nubbins can be associated with hypoplastic or absent metacarpals and phalanges.	• The short fingers have a conical bony amputation stump. The amputation occurs through the bony phalanx.
• The fingers are short and exhibit developmental syndactyly.	• Acrosyndactyly or fusion of the finger tips is characteristic. A proximal epithelium lined sinus will be present.
• Nail complex like nail plates, nail folds are present.	• Nail complex is usually absent in the short digits.

surgically at our centre between July 2013 and May 2024. We briefly discuss considerations with regard to anaesthesia followed by specific surgical considerations related to constriction rings, acrosyndactyly and short digits and conclude with a short segment on the late sequelae of CRS.

ANAESTHETIC CONSIDERATIONS

The surgery is performed under general anaesthesia and regional block to the limbs are provided for postoperative pain relief. Local anaesthetic infiltration is used if skin grafts are needed. Children with CRS require multiple surgeries due to multi-limb involvement. If both hands are involved, a single stage operation can be considered in patients who only require separation of the acrosyndactyly. If patients need web deepening and full thickness skin grafts, it is preferable to stage the procedure in view of the prolonged duration of anaesthesia. In addition to the surgeries, anaesthesia is usually needed for the first postoperative dressing that is typically done at 2 weeks from skin grafting. This allows inspection of the graft recipient and donor sites, facilitates fabrication of a new splint and allows skin grafting again in patients who have graft loss. If a toe transfer is planned in future, it is important to remind the anaesthesiologist to avoid the feet for venous access while inducing the child for the hand procedures, to avoid encountering thrombosed veins while harvesting the toe.

SURGERY FOR CONSTRICTION RINGS

CRS is a complex spectrum of constrictive rings, acrosyndactyly and terminal amputation of digits. Constriction rings around the digits are more common than in the proximal part of the limbs. The fingers are more affected than the thumb. The rings can be shallow or deep and circumferential or partial. The rings not associated with distal lymphoedema are usually considered shallow. Upton and Tan[7] classified rings into shallow and deep depending on the presence of subcutaneous fat and/or dorsal veins between the dermis and tendon. Deep rings do not have soft tissue beneath them. Hall et al.[8] classified the rings based on the depth into three types: mild, moderate and severe, depending on the presence of lymphoedema and amputation. The rings can be so shallow that only a skin dimpling may be seen. The rings can involve the underlying blood vessels, lymphatics, nerves and tendons leading to vascular compromise, venous congestion, lymphoedema, nerve palsy and joint contractures. Deep rings can involve the bone and result in amputation; usually happens in utero.

Management ranges from observation to urgent limb salvage. If detected early during gestation, foetoscopic release of the bands can be done if there is vascular compromise. However, foetal surgeries are associated with premature labour and injury to the mother and/or foetus.[9] After birth, if vascular compromise, gross distal lymphoedema or peripheral nerve palsy (PNP) is noted, band release should be performed within the first few days. Superficial rings that do not cause distal swelling can be observed. They may be observed for contour deformity as the child grows and may be released electively for cosmesis. Deep rings need surgical attention. Persistent lymphoedema will eventually result in fibrosis and need early attention.

The rings can be released using various techniques and this can be done in single or multiple stages. Initially, these rings were released using simple Z- or W-plasties.[10] Later, Upton and Tan[7] reported their experience in correcting 116 rings. They recommended the excision of the circular groove with mobilisation of the adipose tissue and transposition of the flaps for attaining better contour. They incorporated Z-plasties on the lateral aspect of the fingers with a dorsal straight-line closure. Various techniques like linear circumferential skin closure, Mutaf and Sunay technique and Sin plasty have been reported to avoid the Z-plasty scars.[11–16] The ring release can be single or multi-staged. Single staged linear circumferential skin closure has been reported to give optimum results with no vascular complications.[11,14,15] DiMeo

and Mercer had no circulatory disturbances in the four patients who underwent ring excision and Z-plasties in a single stage.[17]

The authors prefer a single stage multiple Z-plasty release for circumferential deep rings around the proximal part of the limbs. We follow the Upton and Tan[7] technique of excising the ring and fibrous tissue with mobilisation of the adipose tissue and transposition of the skin flaps (Fig. 1). But we perform big Z-plasties all around the site. In fingertips with deep circumferential ring with no soft tissue beneath, we prefer a partial ring release to avoid vascular embarrassment and is done in stages. Usually, parents are satisfied with this and do not opt for another stage. On the palmar aspect, loupe magnification is used to avoid injury to the neurovascular bundle. The bulbous look of the fingertips caused by lymphoedema is corrected with ring release and a better contour is achieved (Figs. 2 and 3).

Large 60° Z-plasties used by the authors provide sizable skin flaps that alleviate stress, increase length and maintain the skin viability. Multiple small Z-plasties often result in unfavourable scars. Compared to linear closure, hourglass deformity is prevented with Z-plasties. We prefer a single stage release to staged ring release as we can avoid an additional surgery and anaesthesia and facilitate early nerve regeneration post nerve decompression. When we plan a single stage release, the tourniquet is deflated after releasing half of the ring, to assess the distal vascularity. We then complete the ring release. The skin flaps are found to be supplied by the musculocutaneous arteries that originate from the muscles and penetrate the subcutaneous tissue to reach the skin. Hence, wound healing and venous compromise are rare. It is also reported that the non-operated part of the ring pulls the operated tissue leading to recurrence.[15] Buck-Gramcko[18] has recommended doing a thorough removal of the constricting bands using magnification, which is expected to improve the blood flow. Amongst the 29 cases of ring release done by the senior author, for the extremities (digits excluded), 26 were done as a single stage procedure.

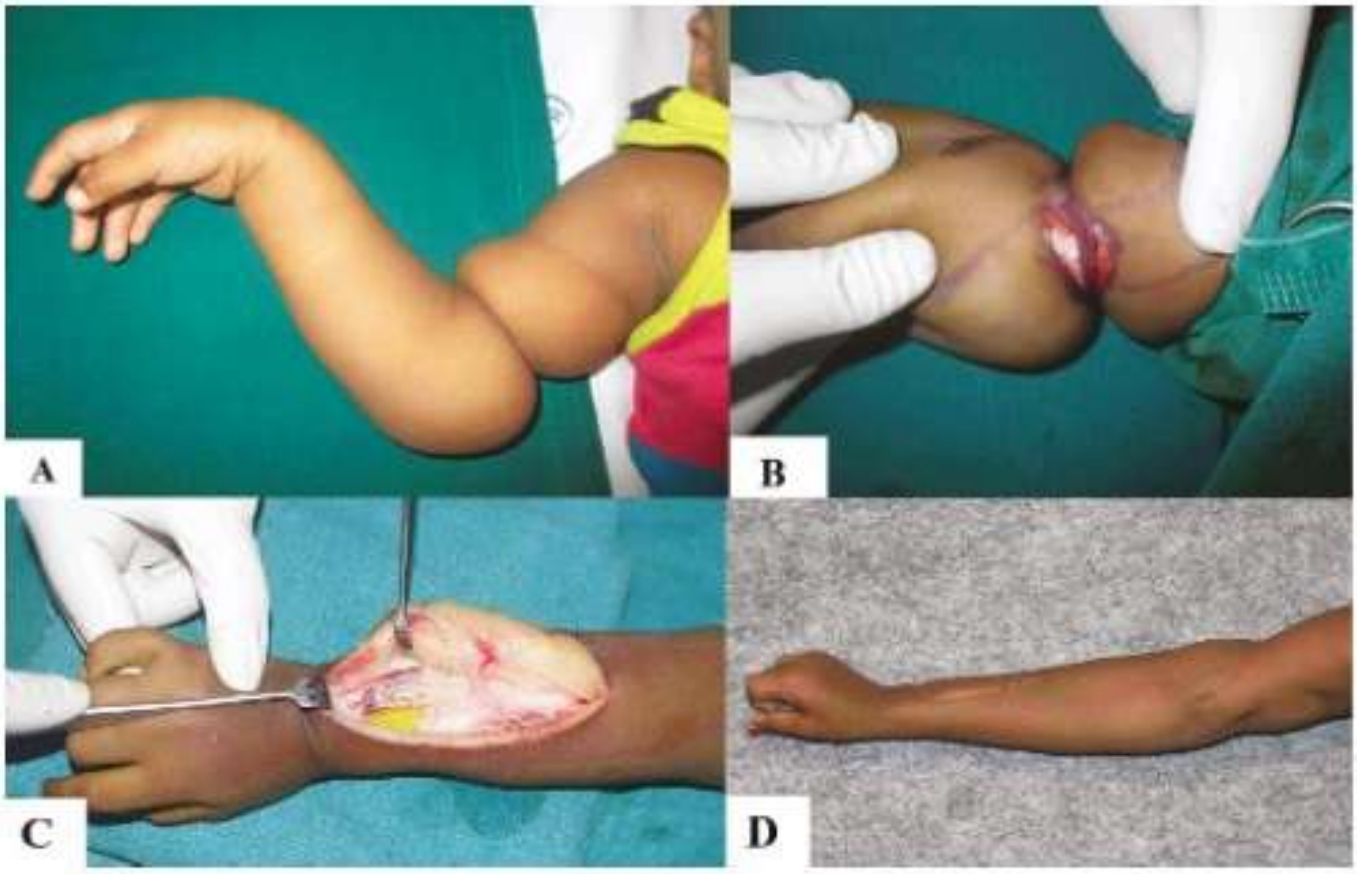

Fig. 1. Constriction ring associated with peripheral nerve palsy. (A) A 1-year-old girl with a deep circumferential ring in the distal third of her right arm. (B) Incision markings for big Z-plasties for ring release. Excision of the circular groove. (C) Pronator teres was transferred to extensor carpi radialis brevis and flexor carpi ulnaris was transferred to the finger and thumb extensors. (D) Final follow-up at 3 years after ring release and 15 months after tendon transfer showing good wrist and finger extension with good arm contour and well settled scars.

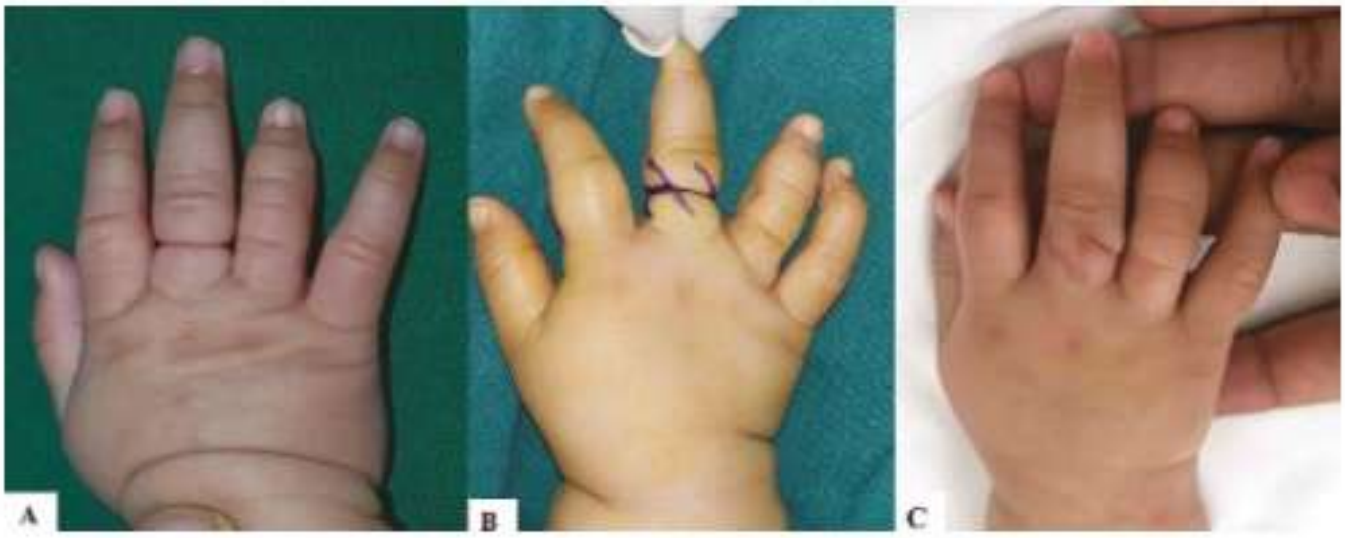

Fig. 2. Release of constriction ring around the finger. (A) Clinical photograph showing constrictions rings around the proximal phalanx of the middle finger. (B) Ring release was done using multiple Z-plasties all around the finger except the palmar aspect. (C) Final follow-up picture. The finger contour looks good.

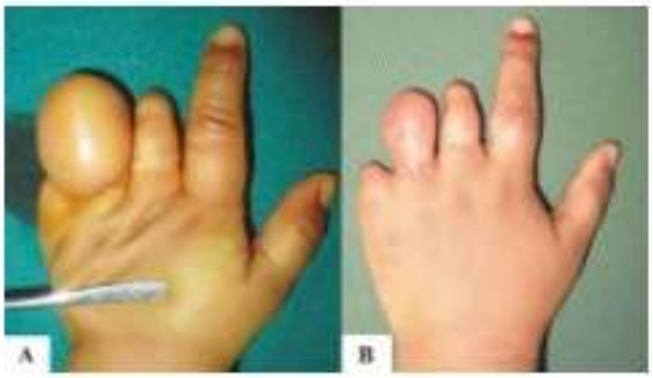

Fig. 3. Reduction of the distal lymphoedema after ring excision. (A) Pre-operative photograph showing the distal lymphoedema secondary to the constriction ring around the ring finger. Ring release was done with multiple Z-plasties at 5 months of age. (B) Final follow-up photograph taken 3 years after surgery, showing the reduction in oedema after surgery.

Except for a child with haemophilia B, we have not had any flap vascularity issues with one stage release.[19]

In deep rings, there might be tight fascial bands compressing the underlying structures like muscle or nerves. These tight fascial layers must be looked for and decompression should be done by dividing them. The subcutaneous tissue is sutured using absorbable suture material after the interdigitating triangular flaps are approximated correctly. The skin is closed using non-absorbable sutures in proximal areas like arm and forearm and absorbable materials in digits. The sutures are removed after 10 days in hand and 14 days in proximal parts. If the hand is involved, an above elbow plaster of Paris slab is applied. Ring release with Z-plasties and interdigitating flaps re-establishes the lymphatic connections resulting in reduction of oedema and a better contour. Scar massage and an elasticated tubular garment (proximal limb parts) are advised for a period of 6 months postoperatively.

In the absence of vascular or lymphatic or neurological problems, fat injection has been tried in correction of the deformity caused by the ring, as an alternative to surgery. Khouri et al.[20] introduced the technique of aponeurotomy and fat injection. The fat harvested from the abdomen or thighs is injected under the ring. Care should be taken not to increase the compartment pressure as it will cause extravasation of the fat into the normal subcutaneous area as well as affect the survival of the injected fat. Though isolated reports of successful fat grafting to treat shallow constriction rings have been published, there is no consensus regarding the indications, fat injection volume and timing.[20,21]

When the constriction rings are deep, they can compress the nerves and cause PNP (Fig. 1A).[22-26] This is usually seen in proximal areas like the arm where two or more nerves can be affected. Triple nerve palsy when encountered is devastating. Literature review as well as our clinical experience shows that nerve decompression should be performed as soon as it is detected. However, decompression or procedures like nerve grafting or neurolysis cannot guarantee nerve recovery.[22,25] One of the possible explanations for this is the fact that the nerve compression occurred in the foetal stage and once the baby is born, considerable time has passed and the motor end plates are no longer

excitable. Hence, the features mimic those of a late presenting traumatic nerve palsy. In such cases, tendon transfers are advised.[22] However, we could find only one such report in literature.[26]

We have treated seven children with PNP at our centre, in the past 16 years. Tendon transfers were done in four out of the seven children, who did not have any improvement after neurolysis or nerve reconstruction (Fig. 1D). Pedicled Latissimus dorsi muscle transfer and biceps reconstruction using fascia lata graft were done for biceps discontinuity. All of them had British Medical Research Council (BMRC) 3/5 power after tendon transfer for radial nerve palsy ($n = 4$) and 4/5 elbow flexion, after muscle transfer ($n = 1$). The grip/pinch strength ($n = 4$) after tendon transfer was found to be improved in three children. The children became independent after the tendon transfers. However, the challenge here was the availability of good donor tendons and the hypoplastic nature of the recipient tendons. In such cases, after the tendon transfer, we continue splinting for a longer period than is recommended normally for the particular transfer.

SURGERY FOR ACROSYNDACTYLY

Acrosyndactyly is a distinct feature of CRS characterised by distal fusion of the fingers with a proximal sinus tract between them. Patterson[27] classified CRS into four types and the presence of acrosyndactyly was termed type 3. The fusion may involve just the fingertips with adequate or inadequate webspace or it may be of greater degree with just a tiny sinus opening proximally with absent webs. The fingers are usually short with a smooth stump and absent nails as a result of intrauterine amputation secondary to deep rings. In complex types, the fingers may be fused together with one stacked upon the other and it is often difficult to delineate them. The central digit in the fused complex is often pushed palmarly.

The goal of surgery for acrosyndactyly is separate fingers of adequate length and spacing. Separation of the fused fingertips require only primary closure. But if the web is not fully formed and is more distal than normal, commissuroplasty is needed. Standard syndactyly separation techniques are used. If the sinus is small and narrow, it is advised to pass a vessel loop or artery forceps through it before marking the incisions. The epithelium lined sinus tract is not useful and is often found to be macerated, requiring excision. A dorsal square flap is used for commissuroplasty and fingers are covered with interdigitating flaps. If zig-zag incisions are not possible distally due to the tapered ends of the amputated stumps, a linear incision is made between the fused fingers as in Apert syndrome. The resultant raw areas are covered using full thickness skin grafts. We prefer to use the groin skin as the donor site morbidity is less and gives an aesthetically favourable scar. We close the donor wound in two layers using absorbable sutures with subcuticular suturing for the skin. The separated fingers look longer and have independent functions. Local flaps can be used to cover big raw areas resulting from syndactyly separation.

In case of a scrambled type of acrosyndactyly, where the fused cluster of fingertips resemble 'a bunch of grapes', digits cannot be distinguished and radiographic evaluation is necessary.[4] Separation of the syndactyly will lead to large raw areas. It is usually difficult to cover them with full thickness skin grafts for lack of availability. In such situations, split thickness skin grafts are used to separate the fingers for the time being so that the child starts using the functionally independent fingers as early as possible (Fig. 4). It is important to counsel the parents of the need for a later surgery as chances of contracture are higher. At a later stage, we can deepen the web spaces and use full thickness skin grafting for the raw areas. This will give more functional length to the fingers.

When only the tips of the fingers are involved in acrosyndactyly, or if the border digits (thumb and little finger) are involved, it is suggested that the separation be done at 6 months of age, to prevent angular deformities, disturbance in the growth and for earlier usage of the hand. Further surgeries can be done at 6 months interval. For the central digits, web deepening surgeries with full thickness skin grafting can be done in stages and can be commenced at 12 months of age. When both sides of a finger are involved in syndactyly, separation is done in stages to avoid vascular compromise. The position of the neurovascular bundle is often unpredictable in CRS.

Nubbins or rudimentary digits are globular soft tissue masses found attached to the fingertips. A circular bobble found on the dorsum occurs due to two adjacent constriction rings with intervening soft tissue. Floppy nubbins can be removed. After removal, the defect can be closed primarily or may require skin grafting. The bobble can be raised via a distal curved incision, thinned and sutured back to give a better appearance to the dorsum of the finger. When multiple nubbins are present in a grape like cluster, it is wise to plan how these can be utilised to cover the raw areas after syndactyly separation. The incisions are planned beforehand so that we know which fingertip gets the nubbin (Fig. 4).

Older children and adults often present with cellulitis and abscess associated with dirt entrapped inside the

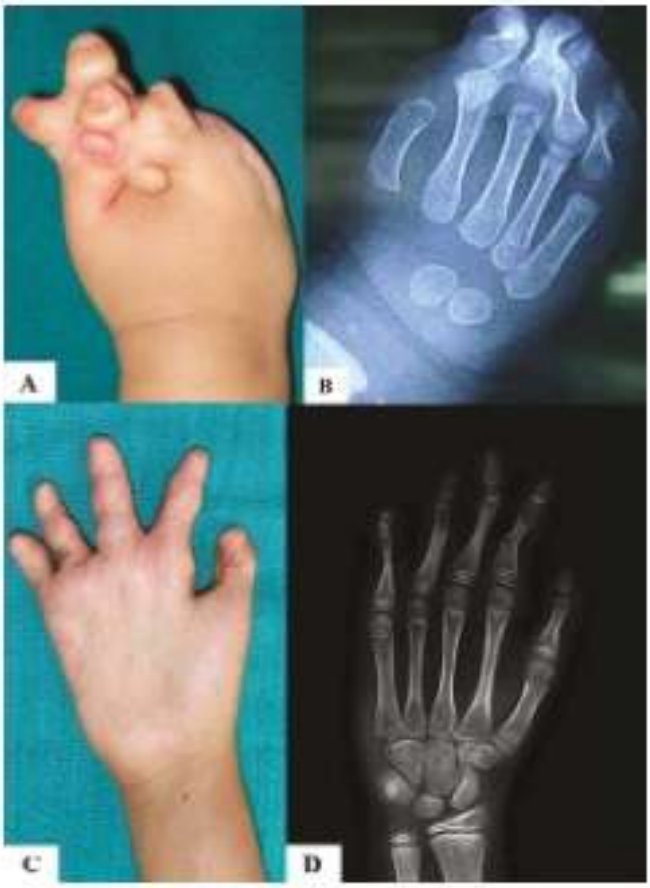

Fig. 4. Scrambled type of acrosyndactyly. (A and B) Pre-operative photograph of the complex acrosyndactyly with radiograph. The child has undergone four operations to achieve a five-digit hand as shown in C and D. Final follow-up photograph and radiograph were taken 10 years after the first surgery.

sinuses. Epidermoid inclusion cysts can occur when the fused fingers grow, and the epidermis trapped between the fingers is pushed into the subcutaneous tissue. Similarly, if the epithelium lined sinus is not excised entirely, inclusion cysts may occur. Toe acrosyndactyly separation is often done to facilitate wearing sandals and toe rings in girls of marriageable age.

SURGERY FOR SHORT FINGERS (INTRA-UTERINE AMPUTATION)

Fingers may become foreshortened as a result of intra-uterine amputation from constriction rings. Hence, the level of amputation is through the bone (tapered type) in contrast to symbrachydactyly where it is through the joint (disarticulation type).[5] The fingers are more commonly affected than the thumb due to their outstretched position. The other proposed reasons are foetal posture that conceals the adducted thumb, its short length and an independent vascular supply and is believed that these protect it from the effects of the ruptured amniotic bands.[6]

The minimum requirement for a functional hand is the presence of two digits with some opposable power. Digital reconstruction is considered in the absence of a thumb of functional length or in the absence of an ulnar post for a functional thumb. A short thumb can be made to look longer and opposable with phalangisation or deepening of the first web space. Digital lengthening can be done by stump refashioning, distraction lengthening, non-vascularised toe phalangeal transfer (NVTT) or microvascular free toe transfer. The demerits of distraction lengthening in children are implant associated problems like infection and long treatment period. NVTT can be done if soft tissue pockets are available in the short fingers. One of the main challenges for free toe transfer and phalangeal transfer is the availability of a complete toe in the feet because, in CRS, the feet are often affected.

NVTT: NVTT is a good option to lengthen and provide stability to the hypoplastic digits with no skeleton. Though the most common indication is symbrachydactyly, NVTT can be used in CRS, where the short fingers have an adequate soft tissue envelope for accommodating a bone graft. The major determinants of survival of the grafted bone are extraperiosteal bone graft harvest, a tension-free skin closure and early age at transfer.[28-32] NVTT is rarely useful in CRS as the amputation is through the bone with skin shortage due to intrauterine necrosis and secondary healing.[30] There is often no soft tissue space within the finger for accommodating the toe phalangeal graft.

The two studies published by Carroll and Green[33] and Rank,[34] prior to 1980, showed no growth of the transferred phalanges, probably due to subperiosteal harvest of the toe phalanx. Extra-periosteal harvest of the phalangeal bone graft yielded favourable results.[28-32] The procedure gives the best results when done between 6 and 18 months of age.[28-32]

A dorsal longitudinal incision is made over the digital stump in the hand where the bone graft needs to be placed (Fig. 5). The soft tissue is dissected to create sufficient space as insufficient skin closure can lead to skin necrosis, premature closure of the growth plate of the toe phalanx and resorption of the bone graft. We select the proximal phalanx of either the fourth or third toe. In young infants, the middle phalanx of the fourth toe may be used in view of the tiny pocket sizes. A longitudinal incision is made over the toe phalanx and an extraperiosteal dissection is performed. The toe phalanx is harvested by dividing

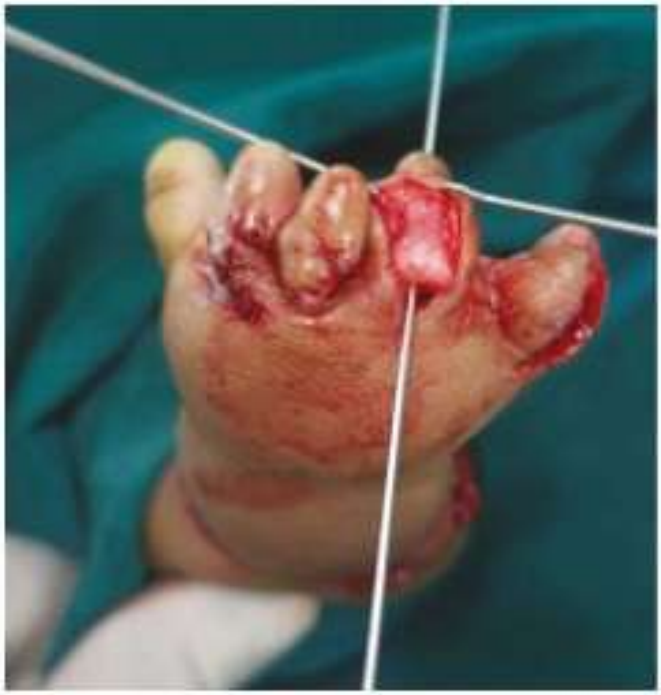

Fig. 5. Non-vascularised toe phalangeal transfer for a 7-month-old child with CRS. Toe phalanges were transferred to the ring and index finger to provide length and optimise prehensile function.

the collateral ligaments, volar plate and the flexor tendon sheath. The flexor and extensor tendons are sutured together to serve as an 'interposition spacer', as outlined by Buck-Gramcko and Pereira.[32] The bone graft is secured using a K-wire or sutures. The hand is immobilised in an above elbow plaster cast for 4 weeks. The K-wires are removed at 4 weeks without the use of anaesthesia. The foot is immobilised in a plaster for 2 weeks.

When Garagnani et al.[35] reported dissatisfaction related to toe shortening and instability, Raizman et al.[36] reported 'no measurable lower extremity morbidity or dysfunction'. Toe morbidity, especially shortening, can be prevented by placing a cylindrical iliac crest bone transplant as a spacer in the donor site. Bourke and Kay[37] have clearly shown that this step improves toe stability and length, leading to a better aesthetic result with minimal toe shortening. Despite these findings, we did not perform it more often because, after presenting the results of outcomes with and without bone graft preoperatively, parents chose not to have a bone graft. The primary influencing factor in this decision was likely the concern over operating on yet another site.

The study conducted at our centre, on 40 reconstructed digits in 19 children with symbrachydactyly, over a 14-year period, reported that the transferred phalangeal grafts had a growth of around 72% when compared to the contralateral phalanges.[28] The mean growth rate per year and comparative length were more in those who underwent the procedure before 18 months of age.

Microvascular Toe Transfer: The gold standard surgery for thumb reconstruction is microvascular free toe-to-hand transfer. Pollicisation is considered a better option than free toe transfer in conditions like thumb hypoplasia where the thenar musculature is underdeveloped.[38] In CRS, the thenar muscles are normal and the proximal recipient structures in the hand like the bone, tendons, nerves and vessels are intact. Hence, the hands with CRS resemble adult post-traumatic defects and toe transfers when done, are usually straightforward. Among all the indications for toe transfer in congenital hand conditions, the best results are obtained in CRS patients.[38,39] In symbrachydactyly, the proximal structures are hypoplastic and at times the metacarpal and phalanges are absent. In such cases, the toe has to be fixed to the carpal bone or distal radius. The level of amputation of the thumb in CRS is rarely at or proximal to the metacarpophalangeal joint, making fixation of the transferred toe easier. If a double toe transfer is needed in an adactylous hand to restore opposing pinch, two second toe transfers are preferred and is better performed sequentially.[39,40] The thumb is reconstructed first and in the next stage, the other toe is transferred to the finger and this can be adjusted to the position and mobility of the first transfer (thumb).[39] Staged transfer reduces anaesthesia risk.[40]

Amongst the 14 children who underwent free toe transfer for congenital hand conditions at our centre, two had CRS. The second toe was transferred to reconstruct the short thumb (Fig. 6). The second toe is chosen over the great toe in view of the better function and aesthetics attained.[39] The foot dissection is commenced only after dissection of the hand. The hand structures are identified and the positions of the bone, tendons, arteries, veins and nerves are noted down. Our goal is to reconstruct a thumb using a toe that is positioned in such a way that its pulp can meet the pulp of the fingers present in the hand. The toe is placed with adequate abduction and pronation depending on the presence of the fingers in the hand. At our centre, we perform the transfer at 3 years of age, when the vessels are of adequate size for microsurgical anastomosis and the child is capable of adjusting to the new thumb early.

The authors observed that after the second toe transfer for congenital hand conditions, the children attained a pinch strength of 73% of the opposite side and a grip strength of 53% of the opposite side. The transferred toe had a growth of 83% compared to the contralateral second toe.

Fig. 5. Clinical photographs of a child with an oligodactylous hand and a short thumb (A) Pre-operative photograph showing complete fusion of the right ring finger with the thumb, index and middle fingers (B) Radiograph showing the level of amputation of the thumb (C) Microvascular second toe transfer to reconstruct the thumb (5 years after separating the radial three digits. (D) Reconstruction leads to a functional pseudo-thumb. (E) Final operative radiograph showing the transferred toe in good position.

A well-planned operation combined with regular scar care and physiotherapy will result in excellent results. We advise twice daily scar massaging for 6 months. Each session involves circular scar massaging that lasts for 2 minutes. We utilise silicone gel sheets for the reconstructed comminutes as well as for the grafted skin. Once the grafts have healed well, we encourage the child to use the hand. Since each postoperative care period lasts for 6 months, the next stage of surgery is planned only after completion of the scar care regimen for the already operated fingers.

In conclusion, CRS is a congenital limb condition that involves multiple limbs in a heterogenous manner. Acrosyndactyly can be complicated by the presence of partial syndactyly or complex type syndactyly. Constriction rings can be associated with distal nerve palsy, lymphoedema and/or vascular compromise. Pseudoshortened digits need reconstruction in an adactylous hand or in a hand without either or both of the border digits. Children with CRS require multiple procedures and parental counselling regarding the same is essential.

DECLARATIONS

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