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Nonvascular phalangeal transfer from toes to hand in congenital aphalangia

Doğuştan afalangialı olgularda ayaktan ele nonvasküler falanks transferi

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Amaç: Doğuştan afalangianın rekonstrüksiyonunda ayaktan ele serbest nonvasküler falanks transferi, tercih edilen cerrahi seçeneklerden biridir. Bu çalışmada transfer sonrası falanksların gelişimi değerlendirildi.

Çalışma planı: Klinik ve radyografik olarak afalangia tanısı konan altı hastaya (3 erkek, 3 kız) ayaktan ele 18 serbest nonvasküler falanks transferi yapıldı. Bir olguda iki taraflı anomali vardı. İleri yaştaki (11.5 yaş) bir hasta dışında, hastaların ameliyat dönemindeki yaş ortalaması 27 ay (dağılım 14-48 ay) idi. Hastalar fonksiyonel sonuçlar, falanksların yıllık uzama miktarı, epifiz açıklığı, kemik rezorbsiyonu ya da nekrozu, parmak ucu nekrozu, kırık, açılanma, subluksasyon, enfeksiyon ve verici saha sorunları açısından izlendi. Takip süresi ortalama 2.5 yıl (dağılım 2-4 yıl) idi.

Sonuçlar: Transfer edilen 18 falanksın yıllık ortalama uzama miktarı 2.4 mm idi. Son grafilerde epifiz açıklığı devam eden falanks sayısı 14 (%78) idi. Fonksiyon değerlendirmesinde üç hastanın transfer edilen parmakla sadece stabilizasyon sağlayabildiği, üç hastanın ise daha ileri hareketler yapabildiği görüldü. Bir olgu subluksasyon nedeniyle tekrar ameliyat edildi. Diğer olgularda 15 dereceyi geçen açılanma veya kırık görülmedi. Üç hastada (%16) kısmi, bir hastada (%5) total kemik rezorbsiyonu gelişti. Yumuşak doku kaybı olarak bir hastada (%5) parmak ucunda kısmi nekroz görüldü. Hiçbir hastada enfeksiyon gelişmedi. Tüm olgularda verici parmakta hafif kısalma gözlendi.

Çıkarımlar: Vasküler parmak transferine uygun olmayan afalangia gibi el anomalilerinde prostetik cihazların kullanımını kolaylaştırmak ve fonksiyon kazanımı açısından nonvasküler falanks transferi alternatif bir rehabilitasyon yöntemidir.

Anahtar sözcükler: Parmak/anormallik/cerrahi; el deformitesi, doğuştan; ayak parmağı falanksı/transplantasyon.

Objectives: Free transfer of the toe phalanges to the hand without vascular anastomosis is a proposed option for reconstruction of congenital aphalangia. We evaluated the growth behavior of the phalanges in patients after transfer.

Methods: Six patients with congenital aphalangia underwent 18 nonvascular phalangeal transfers from toes to the hand. The anomaly was bilateral in one patient. Except for one patient who was 11.5 years old, the mean age at the time of surgery was 27 months (range 14 to 48 months). The patients were followed-up for a mean of 2.5 years (range 2 to 4 years) with respect to functional results, yearly longitudinal growth, epiphysial closure, bone resorption or necrosis, digital tip necrosis, fracture, angulation, subluxation, infections, and donor site morbidity.

Results: The mean yearly longitudinal growth rate of transferred phalanges was 2.4 mm. On final radiographic examinations, epiphysial closure was not completed in 14 phalanges (78%). Functional improvements included stabilization using the affected digit in three patients, and performing further movements in three patients. One patient underwent reoperation for postoperative subluxation. No fractures or angulation above 15 degrees were seen. Bone resorptions were partial in three patients (16%), and total in one patient (5%). Concerning soft tissue complications, digital tip necrosis developed in one patient (5%). No infections were encountered during the follow-up. All the patients exhibited minimal donor digit shortening.

Conclusion: Nonvascular phalangeal transfer is an alternative rehabilitation method by which use of prosthetics may be facilitated and functional capacity may be increased in congenital hand anomalies such as congenital aphalangia, in which vascular phalanx transfer is not suitable.

Key words: Fingers/abnormalities/surgery; hand deformities, congenital; toe phalanges/transplantation.

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Aphalangia, one of the congenital short finger anomalies, results either from a developmental defect or prenatal injury to the foetal appendages. Aphalangia can be classified in two categories according to Swanson.[1] These are, transverse defects (category I), and developmental deficiencies (category V). Another similarly presenting condition; constrictive band syndrome, on the other hand is classified differently (category VI) because of its different etiological basis. The incidence of congenital aphalangia is 1,5 in 10,000 live births.[1-3] Soft tissue remnants may be found in aphalangia fingers as tubular finger stumps, nail remnants, and rudimentary tendons (flexor or extensor). Although there are reported cases with an intact base of a proximal phalanx, generally the phalanx is totally absent. [4] Surgical treatment options found in the literature includes web space deepening, distraction, lengthening with a bone graft, vascular, and nonvascular phalanx transfer from the foot. Among these, nonvascular phalanx transfer is the most used technique, especially in patients with intact sensory functions and tendon rudiments who may use prosthetic devices. Nonvascular phalanx transfer is not an option for patients lacking metacarpal bones or pollices.[3-9] In this study, the results of the cases undergone free nonvascular phalanx transfer from toe to hand were evaluated.

Patients and methods

Six patients (3 male, 3 female) with congenital aphalangia, who had undergone non-vascular toe transfer procedure between 2001-2005 were included in this study. Except for one case who was 11.5 years old, mean operative age of the patients was 27 months (range 14 to 48 months). The total number of the aphalangia digits was 27, while 18 of them were transferred (Figure 1). Involvement was in the left hand in two patients and in the right hand in three patients. One of the cases was affected bilaterally while no patient had any additional congenital anomaly. All of the patients were operated by same surgeon. Mean follow up time was 2,5 years (range 2 to 4 years). No phalanx remnant was detected in any of the hypoplasic fingers, hence all of the transferred phalanges were fixated to metacarpal stumpends. The patients were followed up for their functional capacity, yearly mean longitudinal growth, epiphysial viability, bone resorption or necrosis, digital tip necrosis, fracture, angulation, subluxation, infection and donor site deformities. The functional results were evaluated in two categories; fulfilling simple stabilization functions and achieving associative functions. Growth of the phalanges was evaluated by anteroposterior direct roentgenograms.

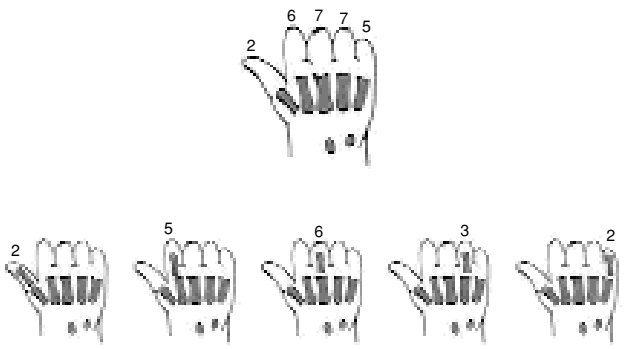

Figure 1. Number of the fingers with aphalangia and number of transfers.

Surgical Technique

Concerning the donor site selection of the phalanx to be transferred, 4th toe of the non-dominant foot was preferred in the first place, while 4th and 3rd toes of the same foot were preferred in double transfers. In multiple transfers, 4th and 3rd toe of the dominant side, followed by 4th, 3rd, and 5th toes of the contralateral foot were selected consecutively.

Under general anesthesia and pneumotic tourniquet; the dorsal skin of the proximal phalanx of the toe to be transferred was incised in a zig-zag fashion to expose the extensor tendon which was longitudinally incised afterwards. Fine dissection of the phalanx was performed extraperiosteally, in an effort not to harm the epiphysial plate. The phalanx was evacuated while delicately preserving the collateral ligament, dorsal and plantar joint capsules, and flexor tendons. Collateral ligaments were sutured to the extensor and flexor tendon stumps with 5–0 polypropylene sutures.

A zig-zag incision was made to the dorsal skin overlying the junction of the tubular soft tissue remnant and distal metacarp of the recipient hypoplastic finger. At this point care was taken for not to harm the extensor tendon rudiment if found to be intact. A pouch for the graft was prepared with fine dissection (Figure 2a, 2b). The graft was inserted to the pouch and was fixed with a 0,7 mm Kirschner wire (K-wire) in a neutral position (0º) between the tip and the metacarpal head (Figure 2c). If any flexor or extensor tendon rudiment was found, it was inserted to the proximal of the graft with 5-0 polypropylene

sutures. Skin incisions were closed primarily with 5-0 rapidly absorbable polyglactine sutures. Antibiotic prophylaxis with sefazolin sodium was administered for seven days, postoperatively. K-wires were removed at sixth week, postoperatively. Cast splints were applied to the hands for eight weeks and to the feet for three weeks (Figure 3a-f).

Results

The mean longitudinal growth rate of the 18 transferred phalanx was 2,4 mm. Number of the phalanges with persistant epiphysial plates was 14 (78%) in the last roentgenogram. The epiphysial plate was closed in the patient who has advanced operating age (11.5 years old). Subluxation was observed in one patient due to loosening of the K-wire, consequently a second operation was performed and reduction is established. No angulation beyond 15 degrees, subluxation or fracture was seen in other patients. The neutral alignment (0º) was preserved in all patients. Partial bone resorption was seen in three phalanges (16%), while total loss of the graft was seen in one phalanx (5%). Epiphysial plate was intact in the totally resorbed phalanx. One digital tip necrosis was seen as a soft tissue complication (5%). This tissue loss was left for healing by secondary intention. In the functional evaluation, three patients were found to be able to perform simple stabilization functions, while more complex functions as grasping, feeding, dressing, and drawing were observed in three patients. No infections complicated the procedure but in the patient with digital tip necrosis, minimal discharge was observed at the time of healing with secondary intention. Direct microscopy of the discharge revealed rare polymorphonuclear leukocytes and 1-2 bacteria per field under Gram stain. Aerob cultures were negative. Blood analysis of the patient didn't reveal any elevation in the leukocyte counts and no fever was detected in the patient. Minimal shortening of the donor fingers was seen in all patients. The middle phalanges were found to be shorter in respect to the contralateral side in two of the patients whereby the amount of shortening was greater.

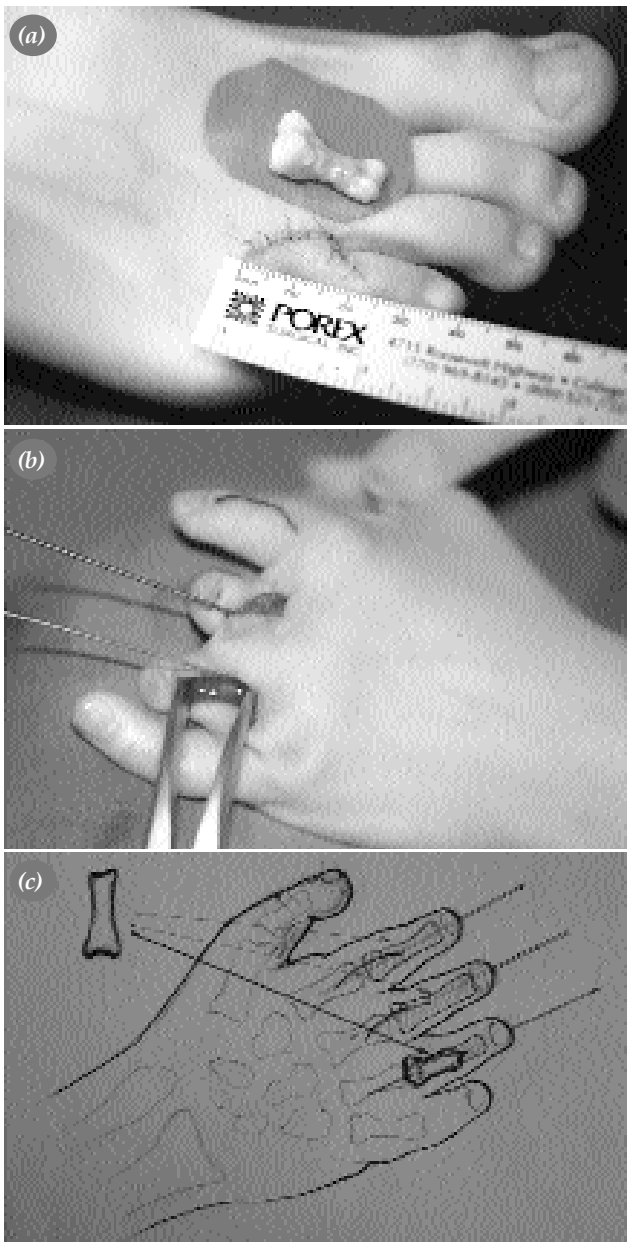

Figure 2. **(a)** Taking of the phalanx from the toe, **(b)** formation of the pouch in recipient site. **(c)** implantation of the phalanx.

Discussion

Keeping in mind that aphalangia cannot be detected easily in prenatal period, a tremendous emotional stress is seen in parents with children born with congenital abscence of the fingers. Severe aesthetic frustration of the parents is complicated with the stress of functional incapacity as the child grows. [3, 4] The reconstructive spectrum for aphalangia patients is broad. Vascular transfer of the toes to hand is the gold standart procedure among this spectrum but requires intact and normal flexor and extensor tendons in the recipient site in order to perform it properly. In the constrictive band syndrome where the number of anomalous fingers is fewer and the recipient site is more developed, thus suitable for transfer; vascular transfers of the toes may be a better option. But absence of the normal flexor and

extensor tendons, and generalized involvement of the fingers limit the use of the vascular transfers in congenital aphalangia cases.[3-9]

Nonvascular transfer of the toes from feet to hands is one of the most suitable treatment options for those patients with congenital aphalangia for restoration of their hands. The most suitable aphalangia cases for nonvascular transfer are those patients with available tubular soft tissue remnants among with enough tendon rudiments on fingers

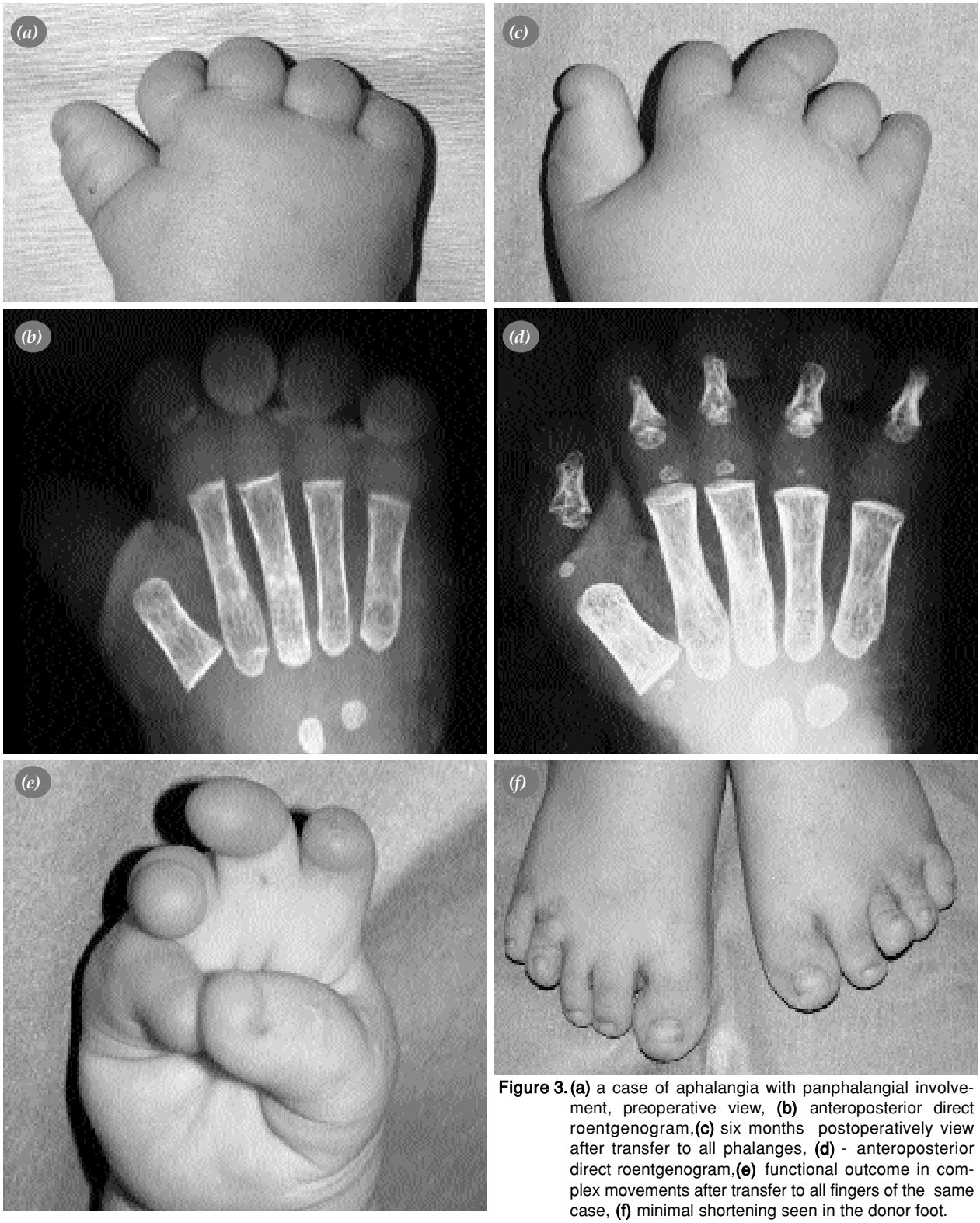

Figure 3. **(a)** a case of aphalangia with panphalangial involvement, preoperative view, **(b)** anteroposterior direct roentgenogram,**(c)** six months postoperatively view after transfer to all phalanges, **(d)** - anteroposterior direct roentgenogram,**(e)** functional outcome in complex movements after transfer to all fingers of the same case, **(f)** minimal shortening seen in the donor foot.

with intact sensory functions. This technique is not suitable for patients lacking metacarpal processes. The thumb is the least involved finger in this anomaly which is advantagous in these patients, as expected. In the cases where the thumb is also involved, the first thing would be to reconstruct the pollex.[4] The ideal age for this procedure is documented in all studies as ages where the epiphysial linings are still intact. But the advised age groups are the ages before the child integrates to his/her self image (before 3 years old) and the ages when the child is able to apprehend the reasons and the aims of the procedure (9 years and after). Between these ages (3–9 years), the child may be subject to severe psychogenic stress owing to the abrupt changes in the appearance of his/her body. Buck-Gramcko classified the patients according to their ages as; lesser than 18 months, 18–48 months, and older than 48 months in their study with 74 transfers. They admit that the best results were achieved in the 18–48 months group. The explanation they offer is that the earliest time in which smaller sized bones can be surgically manipulated harmlessly is the 18–48 months period. This period is also observed to be the earliest time where the bone is enough developed to be fixated by a K-wire.[5, 9] Unglaub et al.[10] in the other hand suggested that their best results were achieved in the so called before 18 months group. Goldberg and Watson [7] stressed on the importance of preserving the periost for growth and viability. They compared the growth of the phalanges to the contralateral toes in their fifteen patients study in which they found 80–90% growth rate in respect to the normal toes. Buck-Gramcko found their median longitudinal growth as 3,5 mm in their 18–48 months age group, while Radocha et al.[8] found their growth as 1 +/- 0,6 mm in their 12–24 months age group, 0,5 +/- 1,5 mm in their "older than 24 months age group". Unglaub et al.[10] found the growth ratio as 73% in patients younger than 1,5 years, on the other hand they observed no growth in their 1,5 – 4 years age group. They recorded that bone resorption was prominent in their patients operated after 4 years of age. One of the longest follow-up time in the literature is the study of Tonkin et al. [11] with 7,4 years. They found the epiphysial viability ratio as 22% and the median growth ratio as 75% compared to the contralateral toes at the end of the follow-ups.[5, 11] In our study the dissection is extraperiosteal as in many other studies in the literature. Mean yearly longitudinal growth was found to be 2,4 mm in 14-48 months age group. In three patients multiple transfers were needed (4 or more) eliminating the possibility of comparison of the growth to the contralateral toes. As a result, repetitive anteroposterior direct graphies were used in order to evaluate the growth ratio of the transferred phalanges.[12] Different results among the documented studies obviate the fact that further studies must be undertaken in this subject. Any injury or air leakage to the chondroepiphysis in the time of surgery (intraperiosteal dissection of the phalanx), recipient site without enough vascularity or injury to the vasculature when dissecting the pouch, injury to the periosteal lining at the time of fixation with K-wire, or tendon rudiment insertion are the factors affecting the growth of the transferred phalanges. Retardation of the phalanx growth is a predictable outcome when the epiphysis is lost. But Unglaub et al. [10] observed no statistical correlation between the amount of epiphysial opening and growth rate. Like this we observed a near total resorption of a phalanx with completely intact epiphysial lining. Long term results are also important in evaluating the epiphysial continuity and growth. Tonkin et al.[11] found that some of the phalanges that tend to grow in early periods (at the end of 1st year) were found to be shortened in long term follow-up (7,7 years). The possibility of epiphysial fusion in the long term follow-up may lead to misinterpretation of the resultant amount of growth. Cavollo et al. [9] found the middle phalanges more resistent and robust than the proximal phalanges, possibly for their intact cortical barriers. They showed less graft shortening in their transfers with middle phalanges compared to the proximal ones. In our study we never used the middle phalanges. For minimalisation of the donor site morbidity we used 4th, 3rd, contralateral 4th, 3rd, and 5th toes consecutively. Minimal shortening of the donor toes is a well predicted outcome. The collateral ligaments, along with the flexor and extensor tendons must be sutured to each other in order not to end up with a morbid shortening or instabilty of the toes after the phalanx is evacuated. The possible reason for shortening of the middle phalanx in two of our cases may be an injury to its epiphysial vascular supply while harvesting the proximal phalanx, or direct injury to the epiphysial plate.[4, 9, 11] The 5th toe must not be selected for transfer at all times because this toe is the margin of the foot and lacks any lateral

support, thus absence of which may induce instability. In our study for only one patient we used the 5th toe who needed multiple grafts and no extra problems were observed. Second toe must also be excluded keeping in mind that the patient might need a vascular transfer thereafter.[10] Partial tip necrosis seen in one of the patients was left to heal by secondary intention because the soft tissues overlying the bone were spared. Preperation of the pouch is important by means of width and depth in order to overlie the transferred phalanx in the recipient site to avoid soft tissue problems that would occur if closed too tight. The quality of the pouch is important also for phalanx growth and resorption. Tightness of the soft tissues wrapping the graft compromises its growth.[11] The discharge seen at the secondarily healing stump was not interpreted as a sign of infection in the patient with digital tip necrosis. The Gram stain and direct microscopical examination of the discharge yielded rare polymorphonuclear leukocytes and 1-2 bacteria on every field, however aerob culture revealed no specific proliferation which receded us from a diagnosis of infection. Negative culture results may be attributed to antibiotic repression which we used as prophylaxis, but no other sign of infection was detected as fever or an increase in the leukocyte counts. Determining the range of motion, thus range of overall activity is rather challenging in children. Thus, detailed functional analysis could not have been accomplished. Simple stabilization purposes were fulfilled by three of our patients, whilst more refined actions as feeding, grasp, and clothing were achieved by the remaining three. Due to the difficulties in assessment of the functional outcome in children, Unglaub et al.[10] consulted the subject with parents at this point, recording an amount of functional improvement in most of the cases. Tonkin et. al [11] attained stabilization activities in 3 patients and more improved functions in 15 patients while Cavollo et. al[9] reported a mean 60º (0º to 110º) gain in range of motion. Current knowledge on advanced rehabilitation of the transfer cases remains controversial. Some authorities advise a second transfer procedure employing another phalanx or iliac crest bone graft at least six months after the first transfer. Nevertheless, our rehabilitative program includes a distraction osteogenesis procedure by age of nine; the approximate age we assume that the child is compliant with both the illness and the treatment. Compliance with distraction osteogenesis is low in younger children. While distraction of long bones such as radius and ulna is possible in younger patients, the procedure is rather difficult with small bones like phalanges.[11, 13, 14]

Conclusions

The reconstructive spectrum for aphalangia patients is broad. Vascular transfer of the toes to hand is the gold standart procedure among this spectrum but requires intact and normal flexor and extensor tendons in the recipient site in order to perform it properly. In the constrictive band syndrome where the number of anomalous fingers is fewer and the recipient site is more developed, thus suitable for transfer; vascular transfers of the toes may be a better option. But absence of the normal flexor and extensor tendons, and generalized involvement of the fingers limit the use of the vascular transfers in congenital aphalangia cases.[3-9]

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