Case Report: 35-Year Follow-up for Nonvascularized Toe Phalangeal Transfer for Multiple Digit Symbrachydactyly

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Abstract

Background: A case is discussed in which a young girl was born with symbrachydactyly of multiple digits in whom nonvascularized proximal toe phalanges were transferred to the aphalangic digits at the age of four. At 39 years of age, she presented incidentally to our clinic and was observed to have a very functional hand with mobile metacarpophalangeal joints in all reconstructed digits. **Methods:** We present a case report which is discussed in the context of long-term follow-up, and phalangeal growth in the absence of distraction, and a review of the current literature in regards to outcomes for this modality of treatment. **Results:** We document growth of the transplanted phalanges, despite surgery occurring after the reported optimum age of before 18 months, and the patient not undergoing distraction. The patient reported no donor site morbidity in regards to function or psychosocial impact. Furthermore, we observed active function at the metacarpophalangeal joints of all operated digits. **Conclusions:** We report the longest follow-up (35 years) following nonvascularized proximal toe phalangeal transfer for short finger type symbrachydactyly. We highlight the long-term functional outcome of nonvascularized toe phalangeal transfers, and present an overview of the current outcome literature for this type of procedure, advocating that nonvascularized toe phalangeal transfers remain a viable treatment option for select cases of symbrachydactyly.

Keywords: pediatric, symbrachydactyly, toe phalangeal transfer

Introduction

Short digits occur in approximately 1 in every 30 000 births, the most common etiology of which is symbrachydactyly, where there is a proximal to distal foreshortening and a significant reduction in the bone content of the soft tissue envelope.⁶

We present a patient with short finger type symbrachydactyly of the right hand, treated with nonvascularized toe phalangeal transfers. The purpose of this case study is to highlight the long-term functional benefit of nonvascularized toe phalangeal transfers, and to synthesize the current outcome literature for this procedure.

Case Report

A 39-year-old female, left-hand dominant, hospital intensive care unit nurse presented to our senior author's clinic after sustaining a superficial laceration from a broken Christmas tree light bulb. She presented to the same hand surgeon 35 years prior at the age of 4 with a diagnosis of short finger type symbrachydactyly of the right hand. At that time, she had no other significant past medical history, an unremarkable birth history, and no family history of aphalangia or any other congenital anomaly. On examination, she was observed to have symbrachydactyly of the

right middle, ring, and small fingers with complete absence of proximal, middle, and distal phalanges, as well as shortening of the index finger proximal phalanx and absence of index finger middle and distal phalanges.

At the age of 4, she underwent nonvascularized toe phalanx transfer. In brief, the surgical technique employed utilized a dorsal foot incision from the proximal interphalangeal joint to the proximal aspect of the metatarsophalangeal joints of the small and ring toe of the right foot, and small toe of the left foot. Dissection was carried down to the extensor tendon, which was split longitudinally. The dorsal proximal interphalangeal and metatarsophalangeal joint capsules were incised and the collateral ligaments to the proximal phalanx were released. The proximal phalanx was removed extraperiosteally using a tenaculum and sharp release of the flexor tendon sheath, preserving the periosteum. The extensor tendon was repaired, and incision closed. Attention moved to the recipient extremity, where dorsal longitudinal incisions were made distal to the remnant metacarpal on the middle, ring, and small

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Figure 1. Left: appearance of hand 35 years after nonvascularized bone grafts to the middle, ring, and small fingers. Right: radiographic appearance at age 39.

Figure 2. Appearance of the feet showing 3 donor sites at age 39.

fingers. A soft tissue bed was created distal to the metacarpal for the transplanted toe phalanx. Blunt dissection was taken down to the extensor tendons, which were retracted. The extensor/flexor tendon hood was split and the toe phalanx inserted within the split. The transferred toe phalanx was wrapped within the soft tissue bed distal to the metacarpal, and secured with a K-wire, which was removed at 3 to 4 weeks, after which active mobilization was started. Local flaps were created to accommodate the increase in soft tissue envelope volume.

Despite the effort, we were unable to locate the patient's original radiograph at first presentation. Radiographs taken at the time of her most recent clinic visit (35 years following surgery) do indicate that the transplanted phalanges have grown, despite her surgery occurring after the reported optimum age of before 18 months, and her not undergoing distraction (Figure 1). The patient reported no donor site morbidity in regard to function or psychosocial impact (Figure 2). On follow-up evaluation, she was observed to have active function at the metacarpophalangeal joints of all operated digits (Supplemental Video S1).

Discussion

The primary goal of intervention for symbrachydactyly is improvement of digital length to enhance mechanical advantage and prehension.⁴ Surgery is usually performed at 6 months, so that surgical interventions are completed by school age.

The toe phalanx is favored over other types of bone grafts (such as an iliac crest graft) because the cortical/periosteal envelope of the toe phalanx minimizes resorption of the graft.⁴ There is also potential for growth, and an articular surface with which to reconstruct the metacarpophalangeal joint. There is always significant resorption of iliac crest grafts when placed on the end of the bone as opposed to interposition bone grafting, and has essentially been deemed unreliable. There are conflicting results obtained from on-end toe phalangeal bone grafting. Some have had poor results, but others have noted that the epiphyseal plate can stay open in the transferred toe phalanx and provide further growth in 90% of cases.⁴ To withstand resorption, it seems that nonvascularized toe phalangeal grafts should come from proximal toe phalanges, be performed before 18 months of age, and be transferred with intact periosteum and collateral ligaments.³ If nonvascularized bone grafting does not provide sufficient length, further distraction lengthening of this transferred phalanx can be performed.⁵ We observed growth of the transplanted phalanx in the absence of distraction. Although we unfortunately do not have the original radiographs with which to compare, we still appreciate that transplanted phalanges have grown in size from the time of surgery.

Several studies have looked at outcomes following nonvascularized proximal toe phalangeal transfer. Cavallo et al described their technique and experience with 22 children who underwent a total of 64 transfers of the proximal (35) or middle (29) toe phalanges (average 3 per child).¹ The mean age at initial surgery was 15 months, and the mean follow-up was 5 years. When looking at recipient site outcomes, digital complications included skin necrosis of the recipient fingers, infection leading to the loss of the transplanted phalanges, scarring requiring secondary release with Z-plasty or local skin flaps, and phalangeal displacement and instability, which occurred most often at a rate of 17%. Proximal phalangeal transfers averaged 5.7 mm in length (range, 4-11 mm) and subsequent follow-up revealed an average of 0.7 mm resorption (range, 6-mm resorption to 1-mm growth). Middle phalangeal transfers were more robust, possibly because of the intact cortical barrier, and averaged 5.5 mm length on harvest (range, 3-8 mm) and subsequently had an average net growth of 0.8 mm over the follow-up period (range, 3-mm resorption to 2-mm growth). Another study observed that physeal patency was maintained in 22% of surviving transfers, and the length of the transferred phalanx averaged 75% of the contralateral comparable toe phalanx and 44% of the contralateral digit proximal phalanx.³

A major problem in the assessment of efficacy of toe phalangeal transfers (vascularized or nonvascularized) is that there is very little information regarding subsequent function. Between 1975 and 1995, 20 children with congenital hand deformities underwent reconstruction by 56 nonvascularized free toe phalanx transfers.⁷ The average age at initial surgery was 4.8 (range, 0.5-22) years. Retrospectively, the children were examined after an average of 3.5 (range, 1.5-17.6) years for function of the hand, transplanted phalanx growth, assessment for the epiphyseal plate, and assessment for psychologic performance with their parents. Seventy-five percent of the parents confirmed significant improvements of the functional performance, and 75% of the parents confirmed improvements in manual skillfulness of their child. The assessment of aesthetic improvement was not so clear with 50% of parents reporting only moderate improvements. Similarly, only 50% of parents confirmed improvements of self-confidence. Sixty-six percent assessed the benefit of the operation and deemed the procedure justifiable. In younger patients (<1.5 years), grafts were well tolerated and showed good growth and only few resorptions. In patients aged 1.5 to 4 years, grafts showed no growth. In older patients (>4 years), grafts were mostly resorbed. Our patient endorsed functional utility of her reconstructed hand and a high degree of self-confidence, displayed growth of the transplanted phalanx, and felt that the procedure was justified.

There are few reports on donor site outcomes following this reconstruction. One study utilized the Oxford Ankle Foot Questionnaire (OAFQ), which is a validated questionnaire for children aged 5 to 16 years, with separate versions for the children and parents.² Each version includes the same 15 questions subdivided into 4 categories: physical, school and play, emotional, and footwear-related problems. Responses are rated on a 5-point scale based on the frequency of the issue affecting the child. The authors converted these scores to percentages, where 100 means complete satisfaction and 0 complete dissatisfaction. They reviewed 40 patients (17 boys, 23 girls) operated on between 1991 and 2007 for digital hypoplasia, by a single surgeon. The most common diagnosis was symbrachydactyly (n = 33). Mean postoperative follow-up was 122 months (range, 36-228 months). The mean age at transfer was 32 months (range, 7-108 months). A total of 36 families and 30 patients gave valid and complete answers to the OAFQ. Thirty-two families (89%) and 28 patients (93%) expressed variable degrees of dissatisfaction. For footwear-related problems of the OAFQ, only 10 patients (33%) and 14 families (39%) were completely satisfied (ie, had no complaints). However, all patients interviewed admitted a tendency to hide their feet. Most patients avoided wearing open shoes. In 4 cases, other people had teased and bullied patients about the appearance of the patients' feet. For some patients, shoe fitting was a problem because of toe instability.

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finger type symbrachydactyly. We highlight the long-term functional outcome of nonvascularized toe phalangeal transfers, and present an overview of the current outcome literature for this type of procedure, advocating that nonvascularized toe phalangeal transfers remain a viable treatment option for select cases of symbrachydactyly.

Ethical Approval

This study was approved by our institutional review board.

Statement of Human and Animal Rights

All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2008. Informed consent was obtained from all patients for being included in the study.

Statement of Informed Consent

Informed consent was obtained from all individual participants included in the study.

Declaration of Conflicting Interests

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Supplemental Material

Supplemental video S1 is available on the *HAND* website at <http://hand.sagepub.com/supplemental>.

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