Long-Term Results of Callus Distraction-Lengthening in the Hand and Upper Extremity for Traumatic and Congenital Skeletal Deficiencies

William H. Seitz, Jr., Patty Shimko and Ryan W. Patterson


This information is current as of December 1, 2010

**Reprints and Permissions**

Click here to order reprints or request permission to use material from this article, or locate the article citation on jbjs.org and click on the [Reprints and Permissions] link.

**Publisher Information**

The Journal of Bone and Joint Surgery
20 Pickering Street, Needham, MA 02492-3157

[www jbjs org](http://www.jbjs.org)
Long-Term Results of Callus Distraction-Lengthening in the Hand and Upper Extremity for Traumatic and Congenital Skeletal Deficiencies

By William H. Seitz Jr., MD, Patty Shimko, OTR/L, and Ryan W. Patterson, MD, MPH

Investigation performed at the Cleveland Clinic, Cleveland, Ohio

Introduction

Children born with severe congenital upper-extremity limb deficiencies have been treated with many surgical techniques and use of prosthetic limbs. One surgical technique has involved the elongation of skeletal segments and their surrounding soft-tissue envelope through the process of distraction-lengthening.

Indications for Application of Distraction-Lengthening Technique

Indications for the application of distraction-lengthening techniques include traumatic or congenital bone loss of the humerus and congenital deficiencies such as phocomelia (Fig. 1).

In the forearm, traumatic amputation or segmental bone loss can be managed by segmental bone transport to fill a defect gap or to lengthen a short below-the-elbow segment after amputation.

The technique may be useful in the treatment of a very short forearm, as can occur in patients with radial or ulnar clubhand (Figs. 2, 3, and 4), or in the treatment of major discrepancies of radial or ulnar length, as can occur in patients with multiple hereditary exostoses.

Fig. 1

Radiographs showing distraction-lengthening process in a patient with right-sided phocomelia (A). A half-frame lengthening apparatus was applied laterally (B), with three sets of fixation pins in the scapula and humerus, to allow distraction-lengthening. A stable shoulder and a longer, more functional arm unit are now present (C).

Disclosure: The authors did not receive any outside funding or grants in support of their research for or preparation of this work. One or more of the authors, or a member of his or her immediate family, received, in any one year, payments or other benefits of less than $10,000 or a commitment or agreement to provide such benefits from a commercial entity (Stryker Orthopaedics).
Distraction techniques have been used to stretch soft tissues for correction of radial clubhand deformity in early infancy. In older children, this technique has been used concomitantly with forearm lengthening and centralization or radialization of the wrist\(^5\)\(^\text{10}\) (Fig. 2).

In the hand, distraction-lengthening can be useful in the treatment of congenital or posttraumatic partial absence of the digital rays involving either a thumb or multiple digital rays\(^4\)\(^\text{11-21}\) (Figs. 5-A through 8).

The primary treatment goal is to increase bone length in the hand to enhance prehension and grip, which provides functional improvement as well as an improved cosmetic appearance. This technique should not be used solely for cosmetic purposes. Its use in the treatment of an isolated short digit (e.g., isolated brachymetacarpia) must be carefully considered, and a lengthening procedure should only be recommended after a thorough evaluation has revealed that a meaningful functional impairment exists. When a functional deficit of the thumb or multiple digits involving the metacarpals and/or phalanges is present, the technique of digital lengthening can restore a very functional prehensile unit by distraction of the existing skeletal architecture or the addition of transplanted bone\(^4\)\(^\text{12,22-24}\).

Fig. 2
Radiographs showing the upper limb of a young boy with ulnar agenesis and divergent forearm bones with a dislocated radial head (A). Distraction-lengthening was performed to elongate the ulnar remnant and realign the radiocapitellar joint through distraction (B and C). Photograph of the patient after the performance of muscle transfers to provide pincer grasp between the two forearm bones, similar to the result obtained with a Krukenberg procedure (D). (Fig. 2, A through D, reprinted from: Seitz WH Jr. Distraction lengthening in the hand and upper extremity. In: Green DP, Hotchkiss RN, Pederson WC, Wolfe SW, editors. Green’s operative hand surgery. 5th ed. Philadelphia: Elsevier Churchill Livingstone; 2005. p 1913-36; with permission from Elsevier.)
Contraindications for Application of Distraction-Lengthening Technique

Aside from lengthening solely for cosmesis, other contraindications to this technique do exist. The technique should not be used in noncompliant patients or families. The patient and family must be fully aware of the duration of the procedure, the potential for complications, the necessity of daily monitoring and pin-site care, and the need for avoidance of certain activities. They must also be aware of the importance of adhering to the schedule of regular follow-up appointments to the surgeon and therapist. Trying to accomplish successful follow-up in patients who live a long distance from the treatment facility can be difficult; therefore, family counseling is extremely important. Problems and complications are not uncommon, so families must be very aware of these issues and be ready to deal with them when they occur so that minor problems can be prevented from evolving into major complications.\textsuperscript{19,25}

For treatment of a shortened forearm segment or when centralization is being considered for the treatment of radial agenesis, distraction-lengthening is contraindicated in the presence of a stiff or ankylosed elbow joint, as the wrist may be functioning as the elbow joint and reconstructive techniques could result in loss of function.

Neo-Osteogenesis Through Callotasis

The concept of distraction osteogenesis uses the principle of slow distraction through an area of healing fracture callus. This technique in the long bones has been pioneered by Ilizarov\textsuperscript{16}, De Bastiani et al.\textsuperscript{26}, Monticelli and Spinelli\textsuperscript{27}, and Paley et al.\textsuperscript{28-31}, and the biology behind this technique has been elucidated by Aronson, Green, and others\textsuperscript{1,10-12,15,19,23,28-30,32-63}.

Although the principles of callotasis have only recently begun to be applied to the upper extremity, the relevant cases and series that have been published in the last few decades have demonstrated that the process results in the successful elongation of skeletal segments with complete consolidation to normal healthy bone and that a single-surgery procedure results in fewer complications than those resulting from a staged surgical procedure\textsuperscript{1,17,21,31,64-83}.

Fig. 3

Radiographs of a child with a severe case of ulnar agenesis and an unstable elbow (A). The radius remnant has been brought underneath the humerus more centrally and stabilized with a neutral outrigger, while osteotomy and distraction was done through the forearm (B). (Fig. 3, A and B, reprinted from: Seitz WH Jr. Distraction lengthening in the hand and upper extremity. In: Green DP, Hotchkiss RN, Pederson WC, Wolfe SW, editors. Green’s operative hand surgery. 5th ed. Philadelphia: Elsevier Churchill Livingstone; 2005. p 1913-36; with permission from Elsevier.)
Multiple daily lengthenings of very small distances are associated with two distinct advantages: (1) less discomfort, and (2) adaptation of the surrounding soft-tissue structures.

The technique includes the preinsertion of fixation pins or wires, local periosteal incision at the site of planned bone division, careful elevation and preservation of the periosteum, controlled osteotomy, periosteal repair, wound closure, and device assembly. After the operation, there is a delay period to allow early healing and callus formation, followed by a period of slow distraction with multiple small lengthenings on a daily basis. Circular frame and hybrid devices have been recommended for correction of the upper extremity, but they are complex to apply, cumbersome to wear, and require so-called “through-and-through” placement of pins, which can injure soft-tissue structures as the lengthening proceeds. In the hand, circular frames are extremely cumbersome, whereas half-frame lengtheners with threaded half-pins provide adequate fixation and minimize the bulk of the fixator while allowing angular correction acutely.31,78,84

Predistracttion Nonvascularized Toe Phalangeal Transfer for Symbrachydactyly
In congenital hypoplasia of the phalanges and/or metacarpals, such as in symbrachydactyly, the technique of callotasis can be used to create fingers of a functional length. However, when the soft-tissue envelope is not supported by underlying skeletal architecture, performance of bone transfer before the lengthening procedure can be beneficial. Once revascularization has taken place, the bone transfer can ultimately be used either as a site for insertion and anchorage of the fixator pin or as a skeletal segment to be lengthened. This requires a period of at least six months for revascularization to occur and for a healthy fibrous capsule to form between the transplanted bone and the host bone.24

Nonvascularized transplantation of a toe phalanx to the hand and the subsequent use of distraction-lengthening restores functional length to a skeletally deficient, poorly functioning hand while maintaining an overlying layer of vascular
and sensate tissue\(^8,9,28,74,85-90\). The primary goal is improvement of digital length to enhance hand function. This technique utilizes a surgical approach to the dorsum of the second and/or third toes for extraperiosteal dissection of the entire proximal phalanx. Following phalanx harvest, the extensor tendon, which was split longitudinally, is repaired and the skin is closed\(^{24}\). A subcutaneous pocket is created in the hypoplastic hand to create a pseudocapsule for placement of the transferred bone at the distal end of the existing host bone. Frequently, an extensor tendon and flexor tendon are present, and the created “pocket” preserves these tendons. The toe phalanx is then inserted into the pseudocapsule, which is sutured closed. The transplanted bone is held in place with longitudinally placed Kirschner wires, and the skin is closed.

To create a thumb and functional finger posts in a hypoplastic hand, the index ray can be resected through a z-plasty incision to create a functional first web space, while the extraperiosteally harvested index metacarpal remnant can be transferred to the thumb position to create enhanced thumb bone stock. In doing so, the first dorsal interosseous muscle is
preserved and transferred to the thumb to function as a thumb adductor.

**Materials and Methods**

Four hundred and twelve individual bone lengthenings were performed in a total of 141 patients ranging in age from six months to thirteen years (average age, 1.2 years at the time of the index operation). Lengthened segments included the humerus (eleven), the ulna (sixteen), the radius (nine), the metacarpals (140), and the phalanges (236). Fifty-three patients underwent preliminary nonvascularized toe phalanx transfers and/or rudimentary metacarpal free transfer prior to distraction-lengthening. As of this writing, the patients have been followed for a minimum of one year and a maximum of twenty-one years (mean duration of follow-up, 8.6 years).

**Surgical Technique**

The technique of distraction-lengthening was performed in the hand through dorsal incisions. When multiple fingers were lengthened, fixator pins were placed in a fanlike array to allow best access to individual fingers. A half-frame lengthening apparatus was used. The Mini-Hoffman digital lengthening device (Stryker, Mahwah, NJ) was initially used, but difficulty in adequately aligning this device for multiple fingers led to the development of the MicroFix digital lengthener (Stryker). A variety of half-frame designs that were designed by a number of manufacturers were used in the humerus, but most recently the Monotube Dynamic Lengthener (Stryker) has been utilized.

When prior bone transplantation has been performed and the subsequent six-month revascularization period has passed, the toe bone is sufficiently revascularized and is ready
for distraction-lengthening. A dorsal longitudinal skin incision is made over the area of toe-pin insertion. Dissection is carried down to bone, and the periostium is incised. A Kirschner wire (0.035 inches [0.889 mm] in diameter) may be used to create a superficial pilot hole for the drill bits. Under direct vision, the bone is drilled (with use of a 1.5-mm drill bit) bicortically, with the first bit left in place to serve as a drill guide so that the second pin could be placed parallel to the first. Next, 2-mm, self-tapping, threaded half-pins are placed obliquely in the transferred toe bone to avoid injury to the extensor mechanism (Fig. 5-A). When multiple devices are used in the hand, placing them along the natural arch of the hand limits overcrowding (Fig. 5-B). A third pin is inserted if adjacent joint stability is a concern. The periostium is incised between the two pins and elevated, and an osteotomy is performed in the metaphyseal region with use of an osteotome in adults and a number-69 Beaver blade (Becton Dickinson, Franklin Lakes, New Jersey) in younger children (Fig. 5-A).

**Closure**
The periostium is closed with number 6-0 absorbable suture. The flexor and extensor tendons are closed around the distal aspect of the transferred toe phalanx. Closure of subcutaneous tissue and skin is standard. The four lengthening devices are then secured in place, and image intensification is used to confirm alignment. A bulky dressing is applied for five days.

**Rehabilitation and Postoperative Care**
The lengthening process is started five days postoperatively. Four daily lengthenings of 0.25 mm each are performed at breakfast, lunch, dinner, and bedtime. Once the desired length is achieved, the consolidation process takes approximately two
to three times the duration of the lengthening itself. Pin-site care is performed twice daily with hydrogen peroxide on a sterile cotton-tipped swab after the dressing is removed. After two weeks, the patient may shower daily, towel dry, then clean pin sites with alcohol. The device may be removed when radiographic evidence of cortical continuity of three sides and confluent appearance of intramedullary bone are present. A clinical example of this technique is shown in Figures 5-A and 5-B.

Outcomes and Method of Assessment
A questionnaire was developed by our therapists as an evaluation tool to evaluate the functional capacity of those patients doing activities of daily living as well as their ability to interact in family, social, and personal relationships, participation in leisure and recreation and play activities, effect on sleep and rest, effect on cognition and thinking, ability to do housework and schoolwork, and ability for employment in the older patients. The questionnaire had sixty items, of which forty-five were graded zero to four on the basis of degree of difficulty, presence of pain, and impact on social interaction and satisfaction (with zero indicating the most difficulty and four indicating the least difficulty), fifteen were graded as zero or one (yes or no questions), twenty-two were age-dependent, and the maximum total number of points available, if all questions were answered in all three categories of testing, was 585. A scoring index was developed that essentially divided the total score by the number of items answered. An excellent outcome score was ≥9.0, a good outcome score was 8.0 to 8.9, a fair outcome score was 7.0 to 7.9, and a poor outcome score was <7.0. In addition, as part of a secondary research project, we developed a separate questionnaire to specifically look at the social and emotional impact on patients as well as families of children with congenital hand differences who were managed with use of this technique. The initial functional outcome was performed by our therapists in person; the latter was performed by a personal interview, phone interview, or e-mailed and faxed questionnaires.

Measurements were calculated to determine the degree of lengthening of each skeletal segment both in millimeters and as a percentage of the segment lengthened. All complications were noted and categorized.

Source of Funding
No funds were received for this study.

Results
The functional outcome evaluation, combining all of the activities of daily living and functional activities, demonstrated the following results.

With regard to difficulty performing functional activities: 88% of patients had no difficulty, 7% had some difficulty, 0.7% had much difficulty, and 5% were unable to perform functional activities.

With regard to pain: 97% of patients never had pain, 2% sometimes had pain, 0.5% had pain most of the time, and 0.5% always had pain.

The outcome score averaged 8.9 (range, 2.33 to 9.98) for the group. Sixty-seven percent of patients scored in the “excellent” range, 24% scored in the “good” range, 3% scored in the “fair” range, and 6% scored in the “poor” range.

Ninety-two percent of patients were very satisfied with their outcome, 2.8% of patients were somewhat satisfied with their outcome, 4.5% of patients were dissatisfied, and 0.7% of patients were very dissatisfied.

Static two-point discrimination with regard to sensation demonstrated virtually no difference between the involved hand and the contralateral hand. Object manipulation and writing was improved in all patients following surgery. Physeal growth was assessed and demonstrated either no change from anticipated growth or an increase in anticipated growth for that particular skeletal segment. For those patients whose limbs were lengthened for purposes of prosthetic use, prosthetic function was considered improved in all patients. Activities of daily living were enhanced in virtually every patient.

Our questionnaire, completed by fifty-six patients and/or families, demonstrated that a congenital upper-limb difference initially provided a very stressful situation, both by placing stress on the parents’ marriage and by placing siblings in a secondary role. Most parents were eventually able to accept the congenital difference and treat the child normally through various mechanisms of coping, perseverance, and assistance from others in their family, community, and support groups. Parents...
noted that, with increasing age, the children did demonstrate some emotional changes as a result of their congenital hand difference. When asked about signs of isolation, depression, and sadness in the child at the point in life when the child understood the nature of his or her congenital hand difference, 47% saw none of these signs in their children, 17% noted some slight signs, 16% noted noticeable and/or prominent symptoms, and only 1% noted severe symptoms of depression, isolation, and sadness. Twelve percent of families indicated that, overall, their family life had not been negatively affected, while the remaining 88% noted at least some changes, if not overwhelming ones, in the way their lifestyle had been affected. When asked what their decisions were for electing to have surgery, everyone stated their primary interest was to provide maximum function for their child, to provide their child with a more normal life, to allow better mobility, and to improve upper-extremity appearance. The family’s greatest fear of surgery was the fear of anesthesia at a young age, but other fears included pain, potential complications, lack of function, infection, postoperative care, the need for multiple surgical procedures, what the ultimate appearance would be, the cost of the surgery, and even death.

Parents described the positive social and emotional effects that evolved from having a child with a congenital hand difference that required distraction-lengthening surgery. Over time, they stated that extended family relationships improved, that the process provided an opportunity for increased family time together and improved interactions within their community and circle of friends, and that they developed new forms of social support groups. Parents listed more than twenty positive traits that they believed that their child developed as a result of the treatment, including an improved sense of maturity, confidence, compassion, and a heightened ability to use their intelligence.

Families did note that, while going through the decision-making process, initially there was potential for creating disputes within the family regarding the correct choice of treatment options, which resulted in tensions and impact on interpersonal relationships at the outset but which improved with time.

Of the 141 patients and/or families who were evaluated, the ultimate question asked was: Were the results worth the surgery and aftercare? One hundred and thirty-nine (98.5%) of the 141 stated that they would go through the procedure again.

Complications included failure of adequate bone formation requiring supplemental bone-grafting in 6%, mainly in post-traumatic and transplanted bones that were lengthened. Other complications included premature consolidation (0.7%), soft-tissue injury (0.7%), tip necrosis due to inadequate soft-tissue coverage in a posttraumatic condition, angular deformity due to inadequate distal segment control and/or loosening around one or more of the fixation pins, and joint luxation or stiffness (0.7%) due to an unrecognized adjacent unstable joint, a regenerate bone fracture, or an infection. Major complications, as noted above, have occurred in 9% of lengthenings. Of these, approximately 5% required some form of secondary surgical correction. Minor complications, essentially confined to pin-track infections, had an occurrence rate of 46%. All of these have been completely managed by oral antibiotics without the need for additional surgical intervention.

**Objective Measurements**

Lengthening ranged between 15 and 135 mm between the phalanges and the humerus. The degree of lengthening ranged between 20% and 400% of the specific skeletal segment lengthened.

**Discussion**

Over twenty years of progressive experience in utilizing the technique of callotasis lengthening has demonstrated its efficacy in the management of functionally deficient skeletal architecture in some children with upper-limb deficiencies. Through careful evaluation of the complications that we have encountered, lessons have been learned. For example, poor bone regeneration occurs with either inadequate fixation or lengthening through dysvascular bone. To prevent this, we have ensured stable fixation by creating the osteotomy through healthy vascular bone and through the preservation and repair of the periosteum at the site of osteotomy.

Premature consolidation occurs because of delay of the onset of the lengthening process, after an early healing period of more than a week. Usually, this is due to noncompliance by the family, perhaps secondary to inadequate education on the part of the surgeon. We have recognized the importance of educating parents in the need for returning for regular follow-up visits. We have become stricter in patient selection and about providing families with educational material and contact information for families who have previously been through this treatment process. Soft-tissue injury can be avoided by ensuring adequate soft-tissue coverage, exposure and release of dense scar tissue, and avoiding the use of “through-and-through” pins, instead using only half-pins in a stable half-frame configuration.

Angular deformity can result from inadequate control of the distal segment, choice of an unstable device, and unrecognized soft-tissue tightness. The ability to control the distal segment has evolved through adequate pin fixation in healthy bone, use of a new generation of devices that can provide stable fixation in a variety of configurations, and, when needed, soft-tissue release and/or flap coverage distally with use of local rotation flaps.

Joint subluxation and stiffness can be the result of an adjacent unstable joint, unrecognized ligamentous injury, inadequate rehabilitation, or poor placement of fixator pins in the device itself. The prevention for this has been to include a careful preoperative assessment of the adjacent joints, an initiation of early rehabilitation, careful pin placement, and avoidance of injury to any of the ligamentous structures at the adjacent joints.

Infection around the pin sites can be expected to occur in almost 50% of patients. However, aggressive pin-site care on the part of the parents and/or patient and meticulous surgical technique can avoid bone-burning and local necrosis, which can be a nidus for infection. Careful regular follow-up and awareness of the early signs of an infection can allow the use of oral antibiotics at an early stage to minimize pin-track infections.
Regenerate fracture can occur from premature removal of the device or inadequate support following device removal with high impact loading. Cortical bone formation throughout the area of regenerate bone must demonstrate a mature pattern of new bone formation. In older, active patients, protective splinting for a period up to four weeks following device removal is used. Gradual muscle-loading and grip-loading to the lengthened segments increases progressively over a four to six-week period following removal of the lengthening apparatus.

The evolution of technique; the understanding of pitfalls, problems, and complications; the development of new surgical devices; and the judicious use of preoperative bone transplantation have allowed us to use the technique of callotasis lengthening to provide a very functional tool to improve pahension and function throughout all aspects of activities in children and older patients who have substantial congenital and posttraumatic upper-extremity deficiencies. We have employed this technique in patients with phocomelia, radial agenesis, ulnar agenesis, complex symbrachydactyly, complex cleft hand and transverse terminal arrest, traumatic amputations, multiple hereditary exostoses, Apert syndrome, and constriction band syndrome in one or both upper limbs. A measurable increase in functional length has been achieved in the vast majority of patients. Nearly all patients and families at long-term follow-up have demonstrated a high sense of satisfaction following this reconstruction, with 98.5% willing to go through the procedure again to achieve the same degree of functionality.

From an emotional and psychological standpoint, we have developed a resource based on the experience of children and families who have gone through this procedure. We are in the process of formulating a parental handbook and have developed a family-support network. Children who had this surgery and have reached adolescence and adulthood have demonstrated excellent social skills and have been successful academically and in the workplace. A number have demonstrated exceptional athletic skills in soccer, volleyball, football, baseball, and hockey. Some have demonstrated surprising skills on musical instruments.

In a child with congenital symbrachydactyly and who requires preoperative toe phalangeal transfer, surgery can begin at age six months, with the initial lengthening at one year of age. After a period of approximately three months of lengthening and consolidation, the devices are removed and the syndactyly between some of the fingers is released. Three months later, a second syndactyly release is performed between the remaining fingers, and completion of the initial set of procedures is usually at an end point by approximately two years of age. In patients who have short limbs with transverse arrest and whose limbs are being lengthened for purposes of prosthetic fitting, the lengthening process can begin as early as six months, with early prosthetic fitting at the time of device removal, after consolidation. A passive prosthetic limb is worn until the child is approximately two years of age, at which point a cable device is applied and the child can begin training with a voluntary closing device prosthesis. Children with radial clubhand who have substantial soft-tissue contractures undergo an initial two-stage procedure, beginning at the age of six months, to gradually use the external lengthener to stretch the soft tissues and gradually realign the hand over the ulna. Once adequate soft-tissue stretching has been performed, a capsular reefing and/or centralization can be performed and the ulna itself can be secondarily lengthened through the technique of callotasis.

Children with ulnar agenesis have been treated with release of elbow contracture, transference of the radius to a short ulna component, when present, and subsequent lengthening. A pseudo-articulation between the radius and the humerus is created for patients in whom the ulna is absent and the radius is short, and a lengthening procedure can provide functional forearm length.

This paper demonstrates the wide array of applicability of distraction-lengthening throughout the upper extremity in children with congenital and posttraumatic differences. It is a technically demanding procedure with a relatively high complication rate; however, complications can be minimized and dealt with effectively through careful attention to detail and patient and family education.

References

<table>
<thead>
<tr>
<th>Page</th>
<th>Reference</th>
</tr>
</thead>
</table>


