Guest Editorial
What's New in Limb Lengthening and Deformity Correction

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This review summarizes selected articles regarding limb lengthening and deformity correction published from January 2020 to April 2021. Our intent was to encapsulate a diverse spectrum of clinical pathology, treatment paradigms, and geographies. These articles review current widespread practice techniques, recent innovations, and potential frontiers within our surgical subspecialty. Some studies may be mentioned within 2 sections, as their findings are important for multiple subdisciplines.

Bone Defects

Ocksrider et al. wrote a comprehensive review article for radiologists describing distraction osteogenesis and how this is used for bone transport, with a focus on the radiographic assessment of regenerate or bone grafts and graft substitutes[1]. In this large review of 78 studies, the authors concluded that successful union, refracture rates, and complication rates were similar between bone transport and the Masquelet technique for the treatment of large segmental bone defects. Meng et al. addressed the mixed results that have been reported with the Masquelet technique, suggesting that failures with the Masquelet technique resulting in recurrent infection could be attributed to an incomplete debridement of the infection and emphasized that 2 opportunities exist to eradicate infection: (1) debridement and antibiotic spacer placement, and (2) antibiotic spacer removal and second debridement[2].

A wide-ranging instructional course lecture comprehensively reviewed autologous graft harvest techniques and location and an overview of principles for each: the induced membrane technique, bone transport, and vascularized bone transfer[3].

Zeckey et al. described a case report of a previously infected 7-cm distal tibial defect treated successfully with a fully implanted PRECICE (NuVasive) bone transport nail after eradication of the infection had been achieved, indicating the efficacy of this newly developed device approved by the U.S. Food and Drug Administration (FDA)[4].

Liu et al. reported 282 Ilizarov bone transports performed at a single institution over 10 years, with 189 problems, 166 obstacles, and 406 complications for a total of 761 actual complications, underscoring the difficult nature of bone defect treatment with external fixation[5].

A systematic review and meta-analysis of 11 studies involving 210 patients with infected nonunion defects averaging >8 cm in size was performed, evaluating the combination of antibiotic spacer placement with Ilizarov methods for the treatment of an infected nonunion. Deng et al. reported excellent osseous results in 65%, and the rate of the recurrence of infection was 6.99%. They concluded that combining antibiotic cement spacers with the Ilizarov transport method was efficacious[6].

Xu et al. described treating 31 massive bone defects accompanied by large soft-tissue defects with 2-level Ilizarov bone transport without soft-tissue transfer. The soft-tissue distraction was performed by dragging soft tissue with the bone ends to allow gradual granulation of tissues. They considered this a reasonable alternative to flap coverage when that was not available, but described that the bone ends must be covered with soft tissue for it to be effective[7].

Limb Lengthening

Wright et al. described 17 consecutive patients undergoing tibial lengthening with a magnetic motorized intramedullary nail, noting valgus and procurvatum deformity without blocking screws. They recommended the routine use, during tibial lengthening, of medial proximal blocking screws to prevent valgus mechanical axis deviation and of posterior proximal blocking screws to prevent procurvatum[8]. In another study, 21 adult patients with posttraumatic deformities of the femur and tibia were treated with magnetic intramedullary lengthening. Complications were higher than in comparable procedures in children, with smoking and a prior operation associated with delayed consolidation[9].

A novel case was reported by Alder et al., describing an allograft reconstruction of a proximal tibial defect caused by Ewing sarcoma resection employing proximal and distal plate fixation. A concomitant intramedullary lengthening nail was inserted and used to lengthen 5 cm through the native tibial diaphysis after union and adjunctive therapy were completed[10].

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A retrospective review of 10 patients with preexisting ankle and hindfoot fusion demonstrated the efficacy of retrograde lengthening nail placement and tibial lengthening. All lengthening sites healed, 3 with persistent ankle or hindfoot deformity. The authors observed predictable regenerate bone formation at the distal tibial lengthening sites, but cautioned to watch for hypertrophic and hypotrophic bone formation[11]. Bafor et al. found that a radiographic pixel value ratio of ≥0.93 in 3 of 4 cortices could be used to initiate full weight-bearing after intramedullary limb lengthening, without adverse events[12].

Sheridan et al. reviewed the existing comparative literature on classic limb lengthening (namely, all-external fixation) compared with integrated limb lengthening (namely, external fixation with an adjunctive plate or nail), finding that all parameters of interest were improved if using an integrated technique, including patient time in a fixator and time to consolidation, but there were higher complication rates with integrated lengthening and specifically higher infection rates with lengthening over nails[13].

In young patients in whom intramedullary nails are not appropriate for femoral lengthening and whose families wish to avoid external fixation, Dahl et al. employed an internal extramedullary technique to perform 11 femoral lengthenings[14]. Complication rates and induced deformities through the regenerate were comparable with those for other techniques, suggesting that newer technology could be developed for all-internal lengthening in this age group.

Congenital Limb Deficiencies

Tibial hemimelia continues to be a rare entity, with Laufer et al. contending that amputation provides superior results in tibial aplasia or Paley Type-V tibial hemimelia in their review of 10 patients[15]. They discussed limb salvage compared with amputation in a cultural context and employed a staged femoral-pedal distraction followed by reconstruction when salvage was undertaken.

The methods of internal augmentation of lengthening by external fixation continue to be presented, as mentioned above, primarily to reduce the duration of fixator application. A study under the aegis of the European Pediatric Orthopaedic Society (EPOS) presented the results, from 3 centers, of patients with congenital femoral deficiency or fibula hemimelia who underwent lengthening via external fixation augmented by flexible intramedullary nailing. There were no differences in results between the use of titanium nails and hydroxyapatite-coated flexible nails[16].

In a sobering narrative article, Hootnick et al. examined the treatment of congenital limb deficiencies from a different perspective: that of a patient and parent. The authors (1 of whom was a patient and 1 of whom was a parent) presented their recollections pertaining to a childhood "interrupted" by multiple limb-lengthening procedures[17]. The article highlighted the fact that limb lengthening is a major undertaking with social and familial implications well beyond the surgical considerations most often discussed.

Westberry et al. highlighted that even patients with fibular hemimelia managed with early amputation require ongoing clinical follow-up, reporting on a series of 17 patients who required guided growth of the proximal part of the tibia for valgus deformity[18]. Such attention to alignment is important in order to optimize prosthesis function. In another study, Westberry et al. reported that tibial corrective osteotomy can be safely performed at the time of foot ablation in patients with fibular hemimelia[19]. Morrison et al. reported on the concept of ideal leg length in patients who have undergone Syme amputation for any indication (including congenital limb deficiency in 37 of their 47 participants), contending that excessive length of an amputated limb impacts prosthesis fitting and patient function[20].

Huser et al. investigated the anatomy of the femoral neurovascular bundle in congenital femoral deficiency, finding that it lies significantly closer to the anterior inferior iliac spine and further from the lesser trochanter compared with the unaffected limb, suggesting that preoperative imaging may be warranted for a surgical procedure in the anterior aspect of the hip for these patients[21].

In a 3-dimensional anatomical study of the pelvis and acetabulum in congenital femoral deficiency, Musielak et al. quantified the anatomic distortion in many ways, concluding that there is not simply a posterolateral wall deficit, but frequently a complete malorientation of the acetabular axis, perhaps suggesting that reorienting osteotomies are preferable to acetabuloplasty[22].

Bone Dysplasias and Tumors

A heterogenous group of often rare conditions can result in limb-length discrepancy or short stature. Many of these conditions have subtle features that must be considered in the application of limb reconstruction techniques. Mindler et al. reviewed considerations regarding deformity correction and prevention, skeletal protection, and lengthening in a number of specific conditions, including achondroplasia, X-linked hypophosphatemia, and osteogenesis imperfecta[23]. They suggested a highly multidisciplinary approach to both the treatment and decision-making regarding these patients.

Progress continues to be made into the treatment of congenital pseudarthrosis of the tibia. A novel application of the well-accepted technique of hemiepiphysiodesis was demonstrated by Laine et al. to potentially prevent fracture of Crawford Type-II or III tibiae[24]. They reported a mean follow-up of 5.1 years with resolution of cystic change in several cases, and no fractures in any of the 10 patients. Meselhy et al. reported on the results of a hybrid technique for congenital pseudarthrosis of the tibia in 19 patients between the ages 3 and 20 years who had undergone failed previous treatment; the hybrid technique involved an induced membrane technique in addition to structural grafting[25]. Two series involving treatment

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of congenital pseudarthrosis of the tibia via vascularized fibular grafting have been reported: 1 with 6 patients in whom a massive allograft was also added (Capanna technique)[26], and 1 with 39 patients[27], which is the largest series of its type in the literature to date, to our knowledge. All of the above series involving congenital pseudarthrosis of the tibia reported the problem of progressive ankle valgus, highlighting the importance of this parameter when reporting long-term follow-up in this population.

The management of osseous deficits after tumor resection also brings its own challenges specific to the patient population. A systematic review by Portney et al. identified 292 skeletally immature patients treated with expandable prostheses for tumor resections[28]. The mean Musculoskeletal Tumor Society (MSTS) score was 80.3 points, the overall complication rate was 43%, and, at the end of treatment, patients had undergone a mean of 4.4 lengthening procedures, for a mean 46 mm of lengthening, with 36% still having a limb-lengthening discrepancy of >2 cm[28]. Zou et al. reported the use of their own prosthesis in a similar patient population, achieving a very similar mean lengthening magnitude (4.2 cm) and MSTS score (83.2%)[29]. In both studies, lengthening was achieved by prosthesis elongation, rather than distraction osteogenesis.

Pediatrics

Each year, there are new and expanded uses for guided growth to treat lower-extremity pathology in lieu of larger open procedures. As mentioned above, Laine et al. reported on the use of anterolateral distal tibial hemiepiphysiodesis to treat congenital tibial dysplasia (anterolateral bowing), with successful deformity correction and fracture prevention in 10 consecutive patients[24]. The selected results of congenital pseudarthrosis of the tibia treatment are also mentioned above. Kocaoğlu et al. were able to achieve union in 15 of 17 cases of congenital pseudarthrosis of the tibia with a combination of intramedullary fixation, circular external fixation compression, and periosteal sleeve grafting from the iliac crest, albeit with 5 later-onset fractures[30]. Although congenital pseudarthrosis of the tibia has been the subject of much study, little has been explicitly written about fibular pseudarthrosis. Wang et al. reported on 15 cases of fibular pseudarthrosis, proposing a new classification based on the presence or absence of tibial deformity and dysplasia[31]. They proposed that inducing iatrogenic tibiofibular fusion with or without medial tibial hemiepiphysiodesis could be considered once tibial dysplasia is corrected to neutral alignment, or else there will be the risk of progressive ankle valgus, and only recommended attempting fibular osteosynthesis in cases without any tibial dysplasia.

Guided growth of the distal part of the femur for valgus deformity was used in a series of 6 patients with deformity secondary to chronic insensitivity to pain, with predictable but slow correction (approximately one-half the rate in typically developing patients with idiopathic deformities)[32]. Guided growth in patients with cerebral palsy with transphyseal screws at the anterior distal part of the femur (combined with hamstring lengthening and stretch casting) performed better in achieving knee extension than hamstring lengthening and casting alone[33]. At the ankle, transphyseal screws have also reportedly been successful in achieving deformity correction but are consistently difficult to extricate[34]. Aiming to improve on existing grading systems to quantify skeletal growth remaining, a serial radiographic study was performed on the morphology of the proximal part of the tibia in children (metaphyseal width, epiphyseal width, lateral epiphyseal height), with ratios of all 3 morphologic parameters demonstrating strong correlations with growth remaining and a high intraclass correlation coefficient between observers[35].

A multicenter group reported on the use of guided growth in the correction of Blount disease, finding that 63% to 88% of cases could be corrected at a mean of 24 months, with a mean rate of correction of 1°/month[36]. Another group reported a 41% failure rate in treating Blount disease with tension-band, plate-and-screw constructs, with data suggesting that cannulated screws and titanium implants were more prone to failure[37]. Transphyseal screws were also used successfully in a small series of patients with tibia vara, despite a theoretical disadvantage of this technique in having the fulcrum of deformity correction more medial (within the physis) than that of plate-and-screw constructs (exterior to the physis)[38].

A variety of osteotomy techniques have been employed in the treatment of Blount disease once guided growth fails or there is medial physeal arrest. Aly et al. reported on 19 modified Rab osteotomies as highly successful in achieving good radiographic correction of varus, procurvatum, and internal rotation deformity, with good functional results in most patients at 2 years[39]. For patients classified as Langenskiold stages V and VI, a group from Egypt reported durable results (5-year follow-up) with a combination of medial hemiplateau elevation and separate metadiaphyseal oblique osteotomy[40], and Maré et al. reported a similar technique for children at a mean age of 8 years[41], with 75% of children having good or excellent alignment at 3 years after the surgical procedure[42]. This latter group also suggested that children <7 years of age with a medial physeal slope of ≥60° were at particular risk for recurrence after osteotomy.

Trauma

A systematic review of open reduction and internal fixation compared with circular external fixation as definitive management for tibial plateau fractures was performed, suggesting that circular fixation was associated with shorter stays and earlier return to activity, but with overall higher infection and malunion rates[43]. A series of 24 high-energy proximal tibial fractures were managed by spanning circular fixation and delayed internal fixation (mean, 6 days) without any deep infections, recommending this paradigm[44]. In another study, the same group reviewed the literature on distraction osteogenesis compared with induced membrane techniques for segmental bone loss following Gustilo-Anderson grade-III open tibial fractures, finding a dearth of quality evidence but a suggestion of

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higher union and lower infection rates with distraction osteogenesis[45]. Abdou et al. reported high rates of fasciocutaneous flap success and limb salvage when paired with either distraction osteogenesis or an induced membrane for Gustilo-Anderson type-IIIB tibial defects[11]. In a pediatric setting, single-center results of management of open femoral and tibial fractures with bone loss was reported, including a variety of techniques (joint-spanning internal and external fixation), with an emphasis on identifying posttraumatic stress symptoms in these patients and providing early psychological support[46].

In an interesting report, Wang et al. described a skin-stretching technique wherein appendicular skin defects were treated with monolateral fixators with Kirschner wires purse-strung through the skin edges to slowly re-appose them for later closure (taking a mean of 4 days), and a variety of small and large wounds were all successfully treated[47].

A prototype plate-style external fixator underwent biomechanical testing compared with traditional external fixators in the management of tibial fractures, with superior load characteristics that were postulated to be secondary to adjustability and an interface with more angular stability[48].

A caprine fracture model developed by Glatt et al. suggested that reverse dynamization was superior for speed and strength of bone-healing (namely, beginning fracture fixation with a less rigid external fixator to encourage micromotion, then introducing rigidity to the construct to protect incipient neovascularization), bringing into question the concept of traditional frame dynamization in this context, which entails the opposite sequence[49]. Iobst et al. demonstrated a method to dynamize a hexapod fixator with modified shoulder bolts to allow for pure axial dynamization but preclude shear stress at the fracture site[50].

Upper Limb

A Polish group was able to achieve medium-term follow-up (7.8 years with functional outcomes via the QuickDASH-9 [a shortened 9-item version of the abbreviated Disabilities of the Arm, Shoulder and Hand questionnaire]) after pediatric forearm lengthenings via external fixation and distraction osteogenesis, reporting that only half of these now-adult patients reported functional improvement or cosmetic satisfaction[51].

A limited report from Tunisia of 58 humeral pseudarthroses showed that effective union was achieved with a no-touch compression technique via monolateral external fixation[52]. Zhao et al. reported on 9 cases of a suspension-bridge circular fixator construct for proximal humeral nonunions with skin defects, involving a single diaphyseal ring and multiple oblique olive wires for the fixation of 2, 3, or 4-part fractures[53]. Fifty-four humeral lengthenings were reported by Arenas-Miquelez et al., describing monolateral frame use, proximal-third corticotomies, and a mean lengthening of 9 cm with high satisfaction but 3 patients with resultant elbow flexion contractures[54].

Several groups reported the results of gradual ulnar lengthening by distraction osteogenesis with external fixation in the setting of multiple hereditary exostoses, with functional, cosmetic, and mental health improvement at a 3 to 4-year follow-up[55,56]. Another group suggested using the proportional ulnar length as a target for gradual ulnar lengthening in multiple hereditary exostoses (this metric is approximately 1.1 times the radial length in the general population) and was able to reduce the dislocated radial head in many cases while recommending no other operative treatment of radial deformity[57].

Practical Techniques

A team from Hospital Universitari Son Espases, Spain, raised the unsolved problem of the finite lengthening capacity of intramedullary motorized lengthening[58]. Retraction and redeployment of PRECICE nails have been described previously in the literature. Their case report provided an account of the successful use of the Fast Distractor to retract the nail while retaining it in vivo, after which new interlocking screws are placed. The procedure was performed on a traction table so that collapse or deformation of the regenerate did not occur, although many experts consider temporary spanning external fixation to provide ideal stabilization of the regenerate if such redeployment is to be performed. It must be acknowledged that both reuse of a distracted nail and the in vivo use of the Fast Distractor are not approved by NuVasive or the FDA. In a similar investigation, Eltayeby et al. reported on late reactivation of intramedullary nails, by using the Fast Distractor to test retraction and redeployment of nails ex vivo, after explantation, as part of the standard practice[59]. Of the tested nails, 84.3% successfully relengthened, but nails that been fully deployed at the end of the initial lengthening failed more frequently.

Hidden et al. (in conjunction with the senior author of this review) provided a case report, filled with technical details, of the removal of a broken intramedullary lengthening nail[60]. Such removal cases must be meticulously planned and the surgeon must be armed with multiple equipment options for extrication.

Therapeutics

There are continual advances being made in the understanding of distraction osteogenesis and osteotomy healing, with hopes that future applications of this knowledge will improve the deformity surgeon's ability to promote healing after lengthening or corrective procedures.

Zhang et al. demonstrated that inflammatory macrophages were increased in a mouse model of distraction osteogenesis and that bone regeneration was diminished by depleting inflammatory macrophages, suggesting that these cells have an important role in bone regeneration[61].

A retrospective cohort study by Fragomen et al. reviewed 155 limbs that underwent osteotomy and internal fixation with a pain regimen that either included nonsteroidal anti-inflammatory drugs (NSAIDs) or did not[62]. The group that received NSAIDs had no difference in time to union but had fewer milligram equivalents of morphine prescribed.

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