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# GUEST EDITORIAL What's New in Limb Lengthening and Deformity Correction

Stewart G. Morrison, MBBS, FRACS, Andrew G. Georgiadis, MD, and Mark T. Dahl, MD

This review summarizes selected articles regarding limb lengthening and deformity correction that were published in 2019 and early 2020. Our goal was to select articles that encapsulate a broad spectrum of pathologies and interventions, investigated at a variety of institutions. These articles evaluate current practice techniques as well as describe innovations and emerging frontiers within our subspecialty.

#### **Congenital Limb Deficiencies**

Congenital limb deficiencies in children are an ongoing subject of intense study because of the psychosocial effects of the diagnosis, difficult early treatment decisions (amputation versus reconstruction), and the technical execution of challenging lengthenings and multiple procedures throughout childhood. Mano et al. performed a novel investigation in which children with congenital limb deficiencies were asked to draw whole body portraits, and their knowledge of basic anatomy was evaluated<sup>1</sup>. The children with congenital limb deficiencies were equivalent to controls with respect to visuospatial body knowledge, but they demonstrated less lexicalsemantic knowledge (i.e., affected children were less familiar with general terms for body parts). Gettys et al. reported that children with a congenital limb deficiency and scoliosis had a high rate of clinically important intraspinal pathology on magnetic resonance imaging (MRI) and high rates of neurosurgical intervention, and therefore recommended that screening spine MRI be performed whenever the 2 entities coexist<sup>2</sup>.

In a retrospective investigation involving children with congenital femoral deficiency, monolateral external fixation lengthening was compared with motorized intramedullary nail limb lengthening (MILL). Despite similar lengthening parameters, the MILL group had lower complication rates and better range of motion at the end of distraction and at consolidation<sup>3</sup>.

In another recent study, staged reconstruction and amputation were compared in the treatment of Achterman-Kalamchi Type-II (severe) fibular hemimelia. Despite clear socioeconomic differences between those undergoing reconstruction versus amputation, psychosocial adjustment and health-related quality of life were comparable between the groups and achieved near-normal values for age<sup>4</sup>. A report on a staged approach to fibular hemimelia (systematic utilitarian procedure for extremity reconstruction [SUPER]ankle then lengthening) suggested durable results in most patients at an average of 9 years of follow-up<sup>5</sup>.

A novel staged-reconstruction strategy for Jones Type-IV tibial deficiency (distal tibial-fibular diastasis, absence of ankle mortise, shortening) was described by Ernat and colleagues<sup>6</sup>. The technique included soft-tissue distraction, tibiotalar fusion, and later tibial lengthening, and resulted in functional independence at skeletal maturity in 2 patients.

Litrenta et al. reviewed the treatment of congenital tibial deficiency, describing an incidence of 1 per million, noting a broad spectrum of involvement and unknown etiology, and reporting that associated pathologies are common<sup>7</sup>. The authors described that the overriding goal is to create a stable limb capable of independent ambulation, usually through amputation and prosthetic fitting. Fundamental to surgical decision-making is knee stability and extensor-mechanism function. In those with a functional knee joint, proximal cross-union of the tibia and fibula and distal amputation are most successful. Limb lengthening and foot centralization, in select cases, can be performed, but multiple surgeries, the high risk of complications, and unknown functional results must be addressed carefully with the family.

#### **Guided Growth**

A retrospective investigation of the screw divergence angle in plate + screw tension-band constructs was performed, in order to investigate whether greater or less screw divergence resulted in a superior correction rate for hemiepiphysiodesis about the knee<sup>8</sup>. Although there are biomechanical reasons that either screw configuration could result in superior correction, both appeared equivalent, and the authors suggested finite-element

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analysis as a means of further investigation of all potential variables.

Several groups have reported on the use of guided growth for the proximal part of the femur with cannulated screws (medial proximal femoral hemiepiphysiodesis) to correct coxa valga in patients with neuromuscular hip subluxation<sup>9</sup>. Although largely successful in the slow correction of neck-shaft deformity and migration percentage, physeal growth "off the screw" was common after approximately 2 years and resulted in difficulty with screw exchange.

Metaizeau et al. described a new technique of guided growth in the transverse plane, involving percutaneous cannulated screws and a cable around the distal femoral physis to effect slow distal femoral derotation<sup>10</sup>. Transverse-plane correction occurred at about 1.2° per month, although small amounts of recurvatum were introduced in 8 of 20 knees, and stiffness requiring formal knee manipulation under anesthesia occurred in 6 of 20 knees.

Definitive percutaneous epiphysiodesis (PE) was compared with temporary epiphysiodesis (TE) with medial and lateral plates (distal part of the femur and/or proximal part of the tibia)<sup>11</sup>. Correction of limb-length discrepancy was greater in the PE group, although the differences did not meet traditional thresholds for statistical significance. Complication rates were higher in the TE group, because of unplanned reoperation for coronal malalignment, notwithstanding the need for routine implant removal.

Lawing et al. examined factors associated with late follow-up and overcorrection when guided growth techniques are employed<sup>12</sup>. In a retrospective series of 112 patients, they reported a late follow-up rate of 39%, which was associated with overcorrection, obesity, and a primary language other than English, highlighting the need for close and appropriate patient education and communication with patients undergoing guided growth. A review article was also published pertaining to guided growth<sup>13</sup>.

## Dysplasia and Tumors

Cirstoiu et al. described current treatment options for bone defects after resection of extremity bone sarcomas, including modular endoprosthetic reconstruction, bone-graft reconstruction, bone transport, resection arthrodesis, and rotation-plasty<sup>14</sup>. Specific concerns with bone transport included infection risk and immunosuppression, possible tumor cell activation, regenerate fracture, muscle contractures, implant failure, nonunion, malalignment, and high psychological burdens.

Jang et al. describe a small group of patients with "proximal tibial dysplasia" associated with congenital pseudarthrosis of the tibia, who underwent lengthening either with a transphyseal distraction or an osteotomy directly next to the physis<sup>15</sup>. Those with lengthening through the physis had a lower healing index (faster healing) than those with metaphyseal corticotomy. The authors acknowledge that this technically demanding surgery (transphyseal distraction) was developed for particularly severe cases of proximal tibial dysplasia associated with congenital pseudarthrosis, best done near maturity.

Ko et al. reported on a series of extensive lengthenings among 14 patients with achondroplasia and 1 patient with hypochondroplasia, comparing complications by bone segment and whether the patient had simultaneous bilateral femoral lengthenings and separate bilateral tibial lengthenings (transverse technique), or crossed lengthenings (simultaneous femoral and contralateral tibial, followed later by the opposite)<sup>16</sup>. Humeral lengthenings were associated with significantly fewer complications and quicker healing than lower-extremity lengthenings, and the authors also found that humeral lengthening aided in perineal self-care. The crossed lengthenings had a greater incidence of malalignment and leg-length discrepancy compared with the transverse technique.

Memeo et al. described the use of a protocol for sequentially injecting bone marrow aspirate concentrate in patients with achondroplasia during distraction osteogenesis for the treatment of delayed union<sup>17</sup>. The concentrate was injected in multiple areas of poor regenerate, and the site was manipulated to improve penetration. The authors did not describe the influence of frame stability, implying that the concentrate acted alone to improve healing.

Singer and Johnston reported the results of 34 patients treated with the Charnley-Williams procedure for congenital pseudarthrosis of the tibia, who were followed to skeletal maturity<sup>18</sup> (mean follow-up of 11.9 years after the initial surgery). The authors considered patients whose fibula was not treated to have had inferior results. Thirteen of the 34 patients experienced fracture after a period of union, 10 of whom healed on retreatment. All 6 patients who did not have fibular surgery had poor results and requested amputation.

#### **Foot and Ankle**

A "round table discussion" was conducted pertaining to tibiotalocalcaneal arthrodesis in patients with and without diabetes<sup>19</sup>. Patient outcome measures, including the Grimby scale, Lower Limb Activity Scale, and visual analog scale (VAS) for activity, were used in 2 papers from the same series of 47 patients by Morasiewicz et al.<sup>20,21</sup> that reported clinical results as well as physical activity levels after ankle arthrodesis. While ankle arthrodesis was found to be effective using either external or internal fixation, better outcome scores were reported in the external-fixation group.

Chappell et al. reported on circular fixator use for concurrent tibial and hindfoot deformity correction and distal tibial lengthening in a series of 19 patients<sup>22</sup>. For distal tibial deformity, a technique was described for dome osteotomy creation using a single circular ring as well as a 5-hole Rancho The Journal of Bone & Joint Surgery · JBJS.org Volume 00-A · Number 00 · June 15, 2020 WHAT'S NEW IN LIMB LENGTHENING AND DEFORMITY CORRECTION

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cube (Smith & Nephew) to "trace out" the osteotomy prior to fixation with cannulated screws<sup>23</sup>.

The infected neuropathic ankle presents a further management conundrum. Tomczak et al. presented a series of 7 such cases managed with a hybrid technique of circular external fixation and an intramedullary nail coated with antibiotic cement<sup>24</sup>. A silicone tube filled with antibiotic bone cement was placed over a standard hindfoot nail, and after cement setting, the tube was incised and removed.

#### **Hip and Knee**

Limb-reconstruction techniques continue to find applications in the management of complex articular pathology. Dumlao et al. described the management of a chronic, traumatic posterior hip dislocation in an 8-year-old boy by open reduction, interpositional grafting, femoral shortening, and stabilization with articulated iliofemoral external fixation<sup>25</sup>. Articulated hip distraction was also described by Papachristos et al. for the management of idiopathic femoralhead osteonecrosis, augmented by core decompression and grafting<sup>26</sup>.

The role of the posterior proximal tibial angle (PPTA), commonly referred to as "tibial slope" in non-reconstruction literature, is increasingly recognized for its role in knee stability. In a cadaveric study, the authors compared the effects of axial force across a newly implanted anterior cruciate ligament (ACL) graft in 10 knees, varying flexion angle and PPTA<sup>27</sup>. They demonstrated a strong linear relationship between graft force and PPTA, concluding that a low PPTA ("high slope") may contribute to ACL graft failure. The group also reported on the effect of PPTA on the posterior cruciate ligament (flatter slope was associated with graft failure)<sup>28</sup>.

Takahashi et al. performed a meta-analysis on the role of knee-joint distraction in the management of degenerative osteoarthritis, concluding that it may represent a treatment option<sup>29</sup>. The 3 studies included in the analysis all originated from the same research group, and it is unclear whether patient cross-over is acknowledged or accounted for.

Mayer et al. reported comparable radiographic outcomes for 41 patients treated for Blount disease with Ilizarov or Taylor Spatial Frame (Smith & Nephew) external fixation, with an average patient age of 9.6 years<sup>30</sup>.

#### **Upper Limb**

Circular-fixation techniques play a key role in the management of Monteggia-spectrum injuries. A case of infected ulnar nonunion and a 2.5-cm osseous defect was managed with the application of circular fixation, distal osteotomy, and retrograde ulnar transport to fill the defect, followed by a second ulnar osteotomy and lengthening in order to reduce the radial head<sup>31</sup>. Notably, no radial fixation was used in order to allow pronation and supination during the extended treatment course. Gallone et al. reported on 20 children managed with corrective ulnar osteotomy and lengthening with unilateral external fixation for chronic Monteggia injuries (3 to 38 months from injury to treatment), extensively reporting a number of radiographic and functional parameters<sup>32</sup>. The level of the osteotomy significantly influenced radial-head reduction success.

Ulnar lengthening is also central to the management of forearm deformity due to multiple hereditary exostoses, which disproportionally affects ulnar longitudinal growth, leading to forearm deformity, radial-head dislocation, and functional impairment. Thirty-three patients (average age, 10.4 years) were included in a study by Li et al.<sup>33</sup>, who used either unilateral or circular external fixation for lengthening. Patient age, ulnar diameter at the osteotomy site, and body mass index (BMI) were found to affect the bone-healing index, although only age achieved significance. Concurrent exostosis removal and lengthening percentage were nonsignificant.

In patients with distal radial deformity with shortening, Guan et al. described a technique for bifocal distal radial osteotomy, consisting of acute angular correction distally and external fixator lengthening more proximally. They reported results among 8 patients managed with the technique<sup>34</sup>.

## Trauma

The use of external fixation in pediatric fracture management has been employed by surgeons who are constrained by the physis for fixation options, seek to maintain motion during the initial healing period, and wish to minimize periosteal bloodsupply disruption. Korobeinikov and Popkov reviewed the use of external fixation for juxta-articular and periphyseal fractures, in both upper and lower extremities, in children in Kurgan, Russia<sup>35</sup>. Most frames were removed within 30 days of injury (elbow, wrist, knee, ankle), and full motion was routinely achieved within 30 days of frame removal.

A group in Hanover, Germany who treated an adult with a floating knee injury utilized the time between initial stabilization (damage control) and definitive treatment to construct custom, 3D-printed external fixators through which an anatomic reduction could be maintained through the existing half-pins<sup>36</sup>. The authors discuss that such a technique could lessen the increased operative time and complication rates at definitive osteosynthesis, as damage-control frames with poor alignment can increase such complications.

Rohilla et al. evaluated the quality of regenerate with bone transport in cases of infected tibial nonunion, comparing monolateral rail fixators and circular fixators<sup>37</sup>. At 3 and 6 months, regenerate was similar, but at 12 months, the railfixator group appeared to have a significantly higher grade of mineralization. The authors postulate that a monolateral frame may be inherently less stable, and the resultant small increase in strain could favorably accelerate maturation.

A series of 14 patients with aseptic long-bone nonunion (femur or tibia) were treated with open debridement and compression with a magnetic expandable lengthening nail (in reverse)<sup>38</sup>. Interestingly, the nails were programmed to shorten

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an average of 9.5 mm and actually shortened 6.7 mm, the bone was actually shortened by an average of 3.1 mm, and the locking bolts at the telescopic end were observed to bend an average of 2.5°. Union was achieved in 13 of the 14 cases.

## **Bone Defects**

The management of bone defects continues to be a subject of interest in the orthopaedic literature. A common theme among 5 retrospective clinical investigations was the need for complete eradication of infection at the beginning of treatment through radical debridement<sup>39-43</sup>.

Borzunov and Shastov<sup>39</sup> reviewed posttraumatic defects of the tibia and forearm; they encountered poor bone regenerate during bone transport, solved by altering the mechanical environment. The authors employed 4 methods: secondary corticotomy, temporary compression through the poor regenerate, and compression performed acutely or gradually at the docking site, all of which were reported to be successful. The small and diverse sample makes it difficult to know which method to use.

In a review of 86 patients treated with bone transport for defects of >8 cm, Catagni et al. showed that trifocal treatment (with 2 lengthening sites) shortened fixator duration and was associated with better results compared with bifocal treatment (with 1 lengthening site), despite a longer operative time in the trifocal group<sup>40</sup>. The authors reported that the reduction in fixator duration was responsible for fewer complications.

Fifteen cases of deep femoral infection resulting from intramedullary fixation of closed femoral fractures were resolved with staged treatment, including radical defect excision, marrow debridement and continuous canal irrigation for 1 to 3 weeks, parenteral antibiotic administration, and subsequent monolateral bifocal bone transport<sup>41</sup>. Infection-free union was reported in all cases.

An L-shaped partial corticotomy was used for 8 cases of focal tibial osteomyelitis<sup>42</sup>. Hemicorticotomy with preservation of intact and uninvolved posterior tibial bone was described. The deficits averaged 8 cm after debridement, with the duration of circular external fixation averaging 169 days. The authors proposed that, in instances in which the infected defect is incomplete, posterior cortical and periosteal preservation may reduce fixator duration.

Conversely, Hosny and Ahmed queried the need for extensive debridement when managing chronic osteomyelitis in the pediatric population<sup>43</sup>. A range of techniques were employed in their series of 36 patients.

### **Limb Lengthening**

In a retrospective review of 107 femoral lengthenings using the PRECICE (NuVasive) motorized intramedullary nail, the authors compared antegrade and retrograde femoral lengthening techniques, finding "excellent" results with both. Knee motion was preserved in more patients in the antegrade than in the retrograde group<sup>44</sup>. The indications listed for retrograde lengthening were a narrow proximal femoral isthmus, large proximal soft-tissue envelope, or planned simultaneous distal femoral deformity correction.

Our group evaluated the use of motorized internal lengthening nails compared with external fixation in a series of humeral lengthenings<sup>45</sup>. While the lengthening parameters were comparable, motorized lengthening mitigated pin-site complications and allowed for repeat lengthenings via reuse of the implant. The ability for greater proportional lengthenings of the humerus was again demonstrated in a case report that involved a 15-year-old male patient after proximal humeral physeal arrest, in whom just over 10 cm of length was achieved in a single lengthening, 100% of the prior humeral length<sup>46</sup>.

It is evident now that multiple groups are working on motorized internal lengthening plates, to provide an allinternal solution for situations in which intramedullary nailing is contraindicated. Gaudreau et al. provided an ex vivo description of a prototype plate consisting of a cylindrical major segment 18 mm in diameter, capable of generating up to 735 N of distraction force<sup>47</sup>.

Cyclic, axial dynamization to promote osteotomy healing, popularly known as the "accordion technique," was evaluated in an ovine model by Reich et al.<sup>48</sup>. Their model used unilateral rather than circular external fixation, with 1,200 cycles per day of 0.2, 0.4, and 0.8-mm amplitude. Another animal-based model examined osteogenesis as well as the adaptive morphology of muscle tissue<sup>49</sup>. Canine tibiae were lengthened by 3 mm per day for 10 days, with histological examination being performed both at distraction completion and after 3 months. Interestingly, histological differences were observed in muscle tissue when fixation was augmented with intramedullary stabilization compared with no intramedullary stabilization.

Two studies looked at improving regenerate formation via pharmacological augmentation. Teriparatide, the bioactive component of parathyroid hormone, was delivered to patients after bone-transport docking via 8 weeks of a daily subcutaneous injection<sup>50</sup>. Sixteen patients were randomized, with 8 receiving treatment. Bone mineral density (BMD) was measured via dual x-ray absorptiometry (DXA) scanning at 8 weeks post-treatment; the treated group demonstrated significantly higher BMD. Functional outcome scores were also measured, but results comparing the groups were not reported, and the duration of frame treatment was not reported. Akçay et al. used vitamin E during mandibular distraction osteogenesis in a rabbit model<sup>51</sup>. Vitamin E was delivered intraperitoneally in the treatment group daily for 7 days. Following animal sacrifice, DXA measurement of regenerate demonstrated significant increases in BMD in the experimental group compared with the control group.

#### **Practical Management and Technique**

A practical and simple approach to leg positioning for circular frame application, utilizing a radiolucent triangle and

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U-bolster, is detailed in the Annals of the Royal College of Surgeons of England<sup>52</sup>. In a randomized, single-blinded study, Subramanyam et al. concluded that there is no role for antiseptic preparations in routine frame pin care<sup>53</sup>. One hundred and fourteen patients (2,363 pin sites) were randomized to either no preparation, chlorhexidine, povidone-iodine, or silver sulfadiazine. Participants were predominantly being treated for open or closed fracture (61%), and most frames were applied to the leg (62.3%). Neither the antiseptic preparation type nor performance of daily dressing changes appeared to affect the pin-site infection rate. Additionally, a review article published in *Injury*<sup>54</sup> details evidence pertaining to the diagnosis and management of pin-track infection, and highlights the absence of high-quality (Level I to III) evidence guiding care.

Finally, an "Ethics in Practice" article in *JBJS*, by Randall et al., presents the case of an 11-year-old girl with mild knee valgus resulting in mild mechanical-axis deviation (zone 2)<sup>55</sup>. Guided growth was performed. The authors use the case as the basis for a discussion about the merits of deformity-correction surgery with mainly "aesthetic" indications. This article high-

lights the importance of a holistic approach, shared decisionmaking, and the importance of medical ethical principles in the application of our incredibly specialized skill set.

Stewart G. Morrison, MBBS, FRACS<sup>1,2</sup> Andrew G. Georgiadis, MD<sup>2,3</sup> Mark T. Dahl, MD<sup>2,3</sup>

<sup>1</sup>The Royal Children's Hospital, Parkville, Victoria, Australia

<sup>2</sup>Department of Orthopaedic Surgery, Gillette Children's Specialty Healthcare, St. Paul, Minnesota

<sup>3</sup>Department of Orthopaedic Surgery, University of Minnesota, Minneapolis, Minnesota

Email address for A.G. Georgiadis: Andrew.g.georgiadis@gmail.com

ORCID iD for S.G. Morrison: <u>0000-0001-7979-7515</u> ORCID iD for A.G. Georgiadis: <u>0000-0002-3624-8273</u> ORCID iD for M.T. Dahl: <u>0000-0003-0530-8322</u>

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