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## Original Research

# Hemiepiphysiodesis Corrects Lower Extremity Coronal Plane Deformity in Children with Skeletal Dysplasia Irrespective of Intra-Articular Malalignment



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#### ARTICLE INFO

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#### ABSTRACT

Background: Skeletal dysplasias (SkD) are a rare group of disorders characterized by abnormal growth and development of bone and cartilage, often causing limb deformity. Many patients also have ligamentous instability that can accentuate the malalignment. This ligamentous instability can present either a generalized ligamentous laxity or focal coronal plane intra-articular malalignment. Temporary hemiepiphysiodesis (HE) is a commonly employed minimally invasive surgical technique for correcting coronal plane limb deformities. This study evaluated the effectiveness of HE in the correction of knee coronal plane deformity in children with SkD and the correlation of concurrent joint laxity.

*Methods*: A retrospective cohort study was conducted to evaluate radiological outcomes of HE for coronal plane knee deformities in patients with SkD (aged < 18 years) who had surgery between January 1, 2008 and December 31, 2020. Changes in distal femoral and proximal tibial mechanical angles, and knee joint line congruence angles (JLCA) prior to HE and at the final follow-up were analyzed. An increased JLCA of  $\geq \pm 2^{\circ}$  was considered a knee with ligamentous instability. Preoperative and postoperative patient-reported pain status was recorded.

Results: Fifty-six tibial and 42 femoral HE procedures in 32 children (mean age at HE: 9.8  $\pm$  2.8 years) were included. The deformity was fully corrected in 23/32 (72%) children, at an average annual rate of 6.3° at the distal femur and 3.3° at the proximal tibia, over 21.9  $\pm$  12.6 months. The overall pre-HE JLCA improved by an average of 5.3° (P < .05). The improvement was more pronounced in varus knees (n = 45; P < .05) than valgus knees (n = 12; P = .11) but regardless of the severity of joint instability, pre-HE JLCA did not impede the rate of femoral (r = -0.22) or tibial (r = -0.21) corrections. Preoperative pain was reported by 78% of patients whereas only 25% of patients reported pain postoperatively at the final follow-up (mean follow-up: 26.4  $\pm$  13.5 months).

Conclusions: The presence of coronal plane intra-articular malalignment did not affect the rate and the magnitude of correction with HE in this cohort. Coronal plane deformities and JLCA improve with deformity correction by HE in children with SkD.

## Key Concepts:

- (1) Intra-articular malalignment at the knee does not impede the success of hemiepiphysiodesis in children with skeletal dysplasia.
- (2) While all coronal plane knee deformities improved, a more significant improvement was noted in children with varus than valgus.
- (3) Joint line congruency angles improved with guided growth for coronal plane deformities. Level of Evidence: III

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#### Introduction

There are over 425 heterogenous skeletal dysplasias (SkD) with known pathogenic genetic variants [1–3]. These are rare disorders with a combined incidence of 1 in 5,000 live cases and over 450 distinct genotypes [2,4]. SkD result from errors in endochondral ossification, leading to generalized abnormal growth and development of bone and cartilage that can result in limb deformity [1–3]. The diseased ossification process and the unpredictable development of growth plates combined with varied ages of manifestation of different dysplasias result in a wide spectrum of deformities [2]. As such, many SkD patients are moderately to severely short-statured. Coronal and sagittal plane deformities are more pronounced at the knees and ankles, which are accentuated with weight gain, older patient age, and after attaining walking age.

Studies have shown that the correction of the lower-limb deformity can improve function and relieve pain [1-3,5]. Since the inception of hemiepiphysiodesis (HE), growth modulation of the physis using Hueter-Volkmann's principle has rapidly evolved and is widely accepted as a safe, effective, and minimally invasive option for limb deformity correction in skeletally immature patients [5,6]. To date, the use of HE in children with SkDs is sparsely reported. While many children with SkD have limb deformities, there is concern that HE is less effective as their condition can result in a slower growth velocity compared to average-stature children. Some studies have reported that HE for coronal plane deformities in children with SkD is inconsistent and less predictable [5,7,8]. In addition to deformities within the bone, children with SkD frequently have concurrent intra-articular deformity and ligamentous laxity, which may further impede the effectiveness of HE [9]. In this article, intra-articular malalignment refers to the resulting deformity from intra-articular bony irregularities and/or ligamentous joint laxity.

To the best of our knowledge, no correlation between joint intraarticular malalignment and the effectiveness of HE has been reported in children with SkD. Therefore, this study aimed to assess if coronal plane intra-articular malalignment might contribute to delayed or incomplete correction of coronal plane knee deformities in SkD. We anticipated that HE would improve limb deformity and lessen joint malalignment due to intra-articular deformities and joint instability.

### Materials and methods

A retrospective observational study was conducted with ethics committee approval. Children aged less than 18 years with a diagnosis of a SkD who underwent HE for coronal plane deformity of the knee over a 12-year period (January 2008 to December 2020) were included. Patient characteristics, diagnosis, and radiological features on weightbearing full-length lower-limb alignment radiographs obtained preoperatively until the final follow-up were reviewed for study inclusion.

The parameters measured on the radiographs included: (1) mechanical axis deviation, (2) lateral distal femoral angle (mLDFA) (normal range: 85-89°), (3) medial proximal tibial angle (MPTA) (normal range: 86-90°), and (4) joint line congruence angles (JLCA) (normal range:  $-2^{\circ}$  to  $+2^{\circ}$ ). The radiograph immediately preceding the procedure (which was used as the baseline for planning HE) was defined as the "pre-HE" radiograph. Similarly, the last radiograph taken prior to the removal of the HE implants was defined as the "final" follow-up radiograph. For those patients who did not or have yet to undergo implant removal, the most recent radiograph at the time of the study was considered as their final follow-up. All images were retrieved from the picture archiving and communication system. The clinical notes in the medical records were reviewed for documentation of patient-reported pain in the preoperative and postoperative periods.

In some patients, the joint line was not well-defined or delineated, especially in patients with either epiphyseal dysplasia or delayed ossification of secondary ossification centers. In such cases, the line joining

the mediolateral metaphyseal edge of the distal femur and proximal tibia was used to estimate the respective joint congruence lines [10,11]. The JLCA was used as a surrogate for objective assessment of intra-articular malalignment and joint laxity in these children. A JLCA of  $\geq \pm 2^{\circ}$  was considered a knee with ligamentous instability [12]. Full correction of the coronal plane deformity was defined as mild over-correction of mechanical axis deviation at which point implant removal was recommended. The failure to achieve this endpoint at the final follow-up was defined as "partial correction"/"persisting deformity." "Recurrence" was defined as a deformity that recurred after achieving full correction [5].

Descriptive statistics were derived, and unpaired T-test were utilized to compare the mean change before and after HE in the MPTA, LDFA, and JLCA. The rate of deformity correction at the distal femur and proximal tibia was calculated. Correlation coefficient models were used to analyze the degree of association between the rate of correction of MPTA and LDFA with preoperative coronal plane joint intra-articular malalignment (ie, JLCA) using SPSS software (version 21).

#### Results

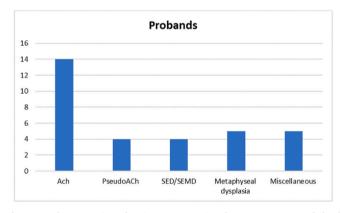
Thirty-two children with a SkD confirmed by genetic testing who underwent HE for knee coronal plane deformity met inclusion criteria. The mean age at the time of HE was 9.8  $\pm$  2.8 years, and 59% (19/32) were males. The most common SkD diagnoses in the cohort were achondroplasia (48%, 14/32), metaphyseal dysplasia (15.5%, 5/32), pseudoachondroplasia (12.5%, 4/32), spondyloepiphyseal dysplasia, and spondyloepimetaphyseal dysplasia (12.5%, 4/32). The remaining 5 children had oro-digito-facial dysostosis, Mucopolysaccharidosis IVA, Ellis-van Creveld syndrome, osteogenesis imperfecta, and metaphyseal anadysplasia (Fig. 1).

HE was done bilaterally in 81% (26/32). Of the children with bilateral deformity, 70% (18/26) had genu varus, 15% (4/26) had genu valgus, and 15% (4/26) presented with a windswept deformity. Among those with unilateral deformity, two-thirds (4/6) had valgus deformity (Table 1).

The coronal plane knee deformity fully corrected in 72% (23/32) of children, at an average rate of 6.3°/y at the distal femur (LDFA) and 3.3°/y at the proximal tibia (MPTA), over a mean period of  $21.9 \pm 12.6$  months (Figs. 2A and B and 3A and B).

Of these patients, 30% (7/23) had a recurrence of deformity. Deformity remained under-corrected in 9 children (including 3 patients who underwent repeat HE) and the mean age at the time of initial HE in these children was  $11 \pm 3.6$  years (P < .05).

In the varus knees, the mean preoperative MPTA (n=45) and mean preoperative LDFA (n=32) improved from 79.4° to 85.2° (P<.01)



**Figure 1.** The proportion of patients representing the 5 most common skeletal dysplasia diagnoses, including achondroplasia (Ach) (48%, 14/32), metaphyseal dysplasia (15.5%, 5/32), pseudoachondroplasia (12.5%, 4/32), and spondyloepiphyseal dysplasia (SED)/spondyloepimetaphyseal dysplasia (SEMD) (12.5%, 4/32).

**Table 1**Baseline patient characteristics.

Parameter	Frequency
Total patients	32
Female	13
Total hemiepiphysiodesis events	106
Proximal tibial	57
Distal femoral	49
Mean age at HE (year) (SD; range)	9.8 (2.8; 5.1-15.4)
Mean age at HE for those with undercorrection of	11 (3.6)
deformity (year) (SD)	
Mean follow-up (in months) (SD; range)	22 (12.6; 5-59)
Laterality	
Bilateral	26
Deformity	
Full correction*	16
Partial persistent deformity	9
Recurrence	7
Knee coronal plane deformity profile $(N = 56)$	
Genu varum (n = 39)	
B/L	19
Genu valgum (n = 17)	
B/L	4
Windswept deformity $(n = 8)$	4
Tibial hemiepiphysiodesis ( $N = 57$ )	
Genu varum (n = 45)	
B/L	16
U/L	4
Recurrence B/L	4
Recurrence U/L	1
Genu valgum (n = 12)	
B/L	4
U/L	4
Recurrence B/L	0
Recurrence U/L	0
Femoral hemiepiphysiodesis $(N = 31)$	
Genu varum (n = 16)	
B/L	5
U/L	6
Genu valgum (n = 15)	
B/L	5
U/L	5

HE, hemiepiphysiodesis; SD, standard deviation; B/L, bilateral; U/L, unilateral; MAD, mechanical axis deviation.

\* Full correction of the coronal plane deformity was defined as mild over-correction of MAD at which the point of implant removal was recommended. The failure to achieve this endpoint at the final follow-up was defined as "partial correction"/"persisting deformity." "Recurrence" was defined as a deformity that recurred after achieving full correction. Laterality of recurrence indicates if the deformity recurred on both sides or only one side.

and 95.2° to 87.8° (P < .01), respectively at the final follow-up (Table 2). In the valgus knees, the mean preoperative MPTA (n = 12) and mean preoperative LDFA (n = 11) improved from 97.6° to 92.7° (P < .05) and 77.5° to 89.4° (P < .01), respectively at the final follow-up (Fig. 4, Table 2). The MPTA corrected at a mean annual rate of 0.38° in varus knees and 0.29° in valgus knees (P = .08) and the LDFA corrected at a mean rate of 0.6° in valgus knees and 0.51° in varus knees (P = .19).

The change in the JLCA following HE was analyzed in 53 varus knees (20 bilateral; 1 unilateral; 6 bilateral repeat HE) and 18 valgus knees (6 bilateral, 4 unilateral; 1 bilateral revision HE). The preoperative JLCA improved from  $6.4^{\circ}$  to  $3.4^{\circ}$  (P < .05) and from  $11.4^{\circ}$  to  $7.4^{\circ}$  (P = .11) in the varus and valgus knees, respectively, at the final follow-up (Table 2).

The overall pre-HE JLCA improved by an average of  $5.3^{\circ}$  (P < .05). The preoperative JLCA ranged from  $0.5^{\circ}$  to  $30.5^{\circ}$ , and we analyzed the correlation between the preoperative JLCA and rate of deformity correction of tibia and femur to assess if coronal plane intra-articular malalignment impacted the outcome of HE in SkD. The improvement of deformity was more pronounced in varus knees (P < .05) than in valgus knees (P = .11) regardless of the severity of joint instability. The

pre-HE JLCA did not impede the rate of femoral (r = -0.22) or tibial (r = -0.21) corrections (Table 3).

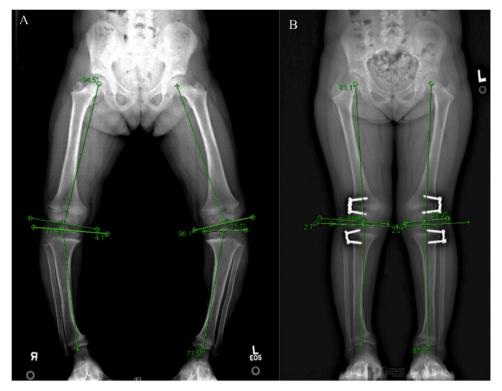
The presence or absence of patient-reported pain was documented in 23/32 (72%) patients preoperatively. Eighteen of these 23 patients (78%) reported having pain, with 9/18 (50%) specifying pain with activity. At final postoperative follow-up (mean follow-up: 26.4  $\pm$  13.5 months), patient-reported pain was documented in 24/32 (75%) patients. Of these, 8/32 (25%) experienced activity-related pain.

#### Discussion

This retrospective study of patients with SkD who underwent HE includes a cohort with a wide range of disorders with achondroplasia being the most common. The advent of HE has revolutionized deformity correction in growing children [6,13]. It has proven to be an efficacious modality in correcting both coronal and to some degrees, even sagittal plane deformities [5,14–17]. Given its proven track record in deformity correction the indication for HE has been extended to include children with SkD. However, multiple studies have reported higher rates of failure of HE and recurrence of deformity after hardware removal in association with "sick physis," excessive weight and in older children [18–20]. Despite these limitations, HE has shown promising results in some children with SkD.

In our cohort, the rate of correction was similar in both varus and valgus knees. The slower rate of correction of tibial and femoral deformities and the relatively high proportion of undercorrection in our series are comparable with the previous reports [5,14,16]. Undercorrection is commonly reported for HE in children with SkD. Kitoh et al. reported that 14% to 20% deformity persisted despite HE in SkD [5]. Similarly, a dual-center study on 29 patients pegged the number of failed HE in SkD at 23% [21]. Trisolino et al. noted that only 55% of coronal plane knee deformities in multiple osteochondroma were corrected compared to nearly 90% successful correction in idiopathic knee deformities [16]. Coronal plane knee deformity did not fully correct in a third of the patients in our series. This phenomenon is generally blamed on the "sick physis," advanced bone age at the time of HE; therefore, it is difficult to predict the exact patients where the deformity is likely to persist, undercorrect, or recur [21,22].

The inborn error in collagen synthesis is not just restricted to endochondral ossification but can affect ligaments as well. Children with SkD can present with a wide spectrum of ligamentous laxity ranging from true joint hyperlaxity to stiff and deformed joints [2,3,23,24]. Severe coronal plane intra-articular malalignment of the knee can lead to joint ligament and capsule complex incompetence due to overstretching. The lateral thrust in severe Blount's disease is an excellent example of this phenomenon [22]. Higher failure rates of TBP-assisted HE in patients with severe coronal plane deformities and in children with high Body Mass Index and "pathological physis" indicate the adverse effect of the altered limb biomechanics [19]. Despite these reports of the adverse effect of the altered biomechanics secondary to the coronal plane malalignment [25], minimal literature exists that examines the effect of the mediolateral instability on HE. The delayed growth rate of the physis in SkDs is blamed for the relatively slower rate of correction following HE [14,22,26]. To assess if coronal plane intraarticular malalignment contributed to this slower rate of correction in SkD, the correlation between JLCA and the rate of correction was calculated. There was a weak negative correlation that was statically nonsignificant between coronal plane intra-articular malalignment and the rate of correction of distal femoral and proximal tibial deformities. Thus indicating that slower correction rate following HE in SkD may not be related to any coronal plane intra-articular malalignment. We sought to examine if intra-articular malalignment affects the outcome of HE in SkD. Theoretically, coronal plane intra-articular malalignment is expected to increase the compression across the tensioned near hemiphysis and increase the distraction tension at the far hemi-physis in a post eight-plate HE scenario, thus stimulating growth of the far hemi-



**Figure 2.** (A) Preoperative radiograph of a coronal plan knee deformity and (B) final follow-up radiograph displaying a lateral distal femoral angle (LDFA) correction (\*) and proximal medial tibial angle (MPTA) correction (+).

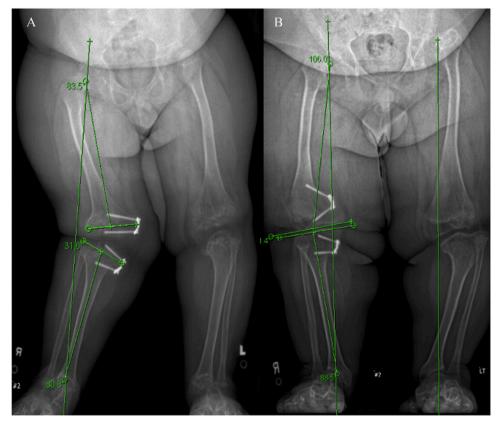


Figure 3. (A) Early postoperative radiograph of a coronal plan knee deformity and (B) final follow-up radiograph displaying a lateral distal femoral angle (LDFA) correction (\*) and proximal medial tibial angle (MPTA) correction (+).

Table 2
Preoperative and postoperative mean differences in MPTA, LDFA, and JLCA.

Parameters	Pre-HE	Final follow-up	Difference	P value
Varum knees				
Mean MPTA $(n = 45)$	79.4°	85.2°	5.8°	< .01
Mean LDFA $(n = 32)$	95.2°	87.8°	7.4°	< .01
Mean JLCA $(n = 53*)$	$-6.4^{\circ}$	$-3.4^{\circ}$	3°	< .05
Valgus knees				
Mean MPTA $(n = 12)$	97.6°	92.7°	4.9°	< .05
Mean LDFA $(n = 11)$	77.5°	89.4°	11.9°	< .01
Mean JLCA $(n = 18*)$	11.4°	7.4°	4°	.11

MPTA, medial proximal tibial angle; LDFA, lateral distal femoral angle; JLCA, joint line congruence angles; HE, hemiepiphysiodesis.

physis and accentuate the correction [27,28]. However, weight bearing on mediolaterally unstable knee joints with misaligned JLCAs will lead to the appearance of malalignment despite corrected bony deformities as measured by the MPTA and LDFA.

To the best of our knowledge, this is the first study to investigate the effect of coronal plane intra-articular malalignment and how it affects HE in children with SkD. In the current study, we did not find a significant correlation between the preoperative joint instability and the rate of correction of tibial or femoral deformities in our coefficient models, suggesting that concurrent instability would impede deformity correction. However, we noted a significant improvement in the instability in the varus knees as well as a positive trend toward significance in the valgus knees. This improvement in joint instability may be attributed to improving overall limb alignment (ie, the correction of the mechanical axes and bone straightening), which may then lessen the impact of the intra-articular malalignment. However, whether HE impacts the soft tissues themselves remains unclear.

Of the available data from our cohort, 78% of patients reported preoperative pain with 50% of these patients having pain with activity. From the available postoperative data, 80% of patients reported pain. Despite patients having pain following HE, only 25% of these patients experienced activity-related pain. In comparison, a recent study reported that 65% and 59% of patients had not returned to their prior activity level and sports at 1-month post-HE due to pain, respectively [29]. This discrepancy may be explained by the longer follow-up

**Table 3**Correlation analysis between preoperative JLCA (laxity) and rate of deformity correction.

	R value	P value
Rate of MPTA correction	-0.22	.08
Rate of LDFA correction	-0.24	.15

JLCA, joint line congruence angles; MPTA, medial proximal tibial angle; LDFA, lateral distal femoral angle.

periods as over time and with continuous physical therapy, patients' pain levels with activity will ideally decrease. Another study demonstrated that prolonged pain existed among 35% of patients following HE procedures potentially attributed to plate position relative to physis and implant location [30]. However, given that in the present study pain status was measured subjectively, the limited data on the location (joint vs incision/procedural site) and frequency of pain, and no formal scale utilized to quantify pain levels, we are unable to accurately assess patient pain levels in relation to HE. Future studies should aim to further assess pain levels following HE procedures using validated patient-reported outcome measures.

This study has several limitations that need to be considered. First, this study was retrospective and can be subjected to inherent selection and ascertainment biases. Second, the study data were retrieved from a single large academic, urban setting, which limits the generalizability of these results to the broader SkD populations. Additionally, the variability in severity and direction of preoperative deformities (varus vs valgus), degree of joint instability (hyperlaxity vs overstretched and incompetent ligament due to angled joints), coexisting proximal femoral and distal tibial deformities, possible intra-articular deformities, and varied diagnoses further limits the generalizability of our findings. Despite all the long-leg x-ray films being standardized and the limbs being positioned with the patella facing anterior, some children with asymmetric bilateral deformity (Fig. 3A and B) required differential rotations to achieve the forward-facing position of the patella and the limb rotations were not always matched. Similar femoral rotational mismatch has been reported in other asymmetric deformities such as adolescent Blount's disease [10,31].

Another limitation is that the JLCA may not be an adequate measurement for determining the effects of intra-articular deformity and joint laxity. Paley and Tetsworth developed the malalignment test, which incorporated

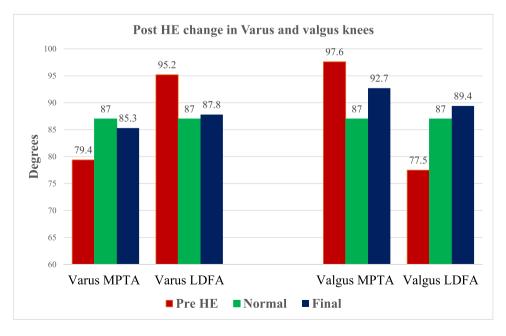


Figure 4. The preoperative and postoperative hemiepiphysiodesis change among varus and valgus knees as assessed by the medial proximal tibial angle (MPTA) and lateral distal femoral angle (LDFA). Numerical values in red represents preoperative hemiepiphysiodesis, green bars indicated a normal value, and blue bars demonstrate postoperative hemiepiphysiodesis values. HE, hemiepiphysiodesis.

<sup>\*</sup> Including patients with recurrent deformity who underwent repeat HE.

the JLCA as one measure to identify malalignment disorders [32]. In this study, the JLCA was used as a surrogate for assessing joint laxity, with a threshold of  $\geq \pm 3^{\circ}$  indicating ligamentous instability of the knee. This threshold was selected as Paley suggested that a JLCA of  $\geq \pm 3^{\circ}$  was considered abnormal, signifying either ligamentous laxity or intra-articular pathology in malalignment disorders [12]. In other orthopaedic populations, the JLCA has been linked to knee soft tissue laxity. A review by Micicoi and colleagues identifies intra-articular deformity and surrounding soft tissue laxity as key factors influencing JLCA in high tibial osteotomies [33]. Furthermore, Lee et al. found a strong correlation between medial laxity and changes in JLCA both before and after surgery in patients undergoing openwedge high tibial osteotomy [34]. Collectively, these findings support the use of JLCA as an appropriate surrogate for assessing joint laxity and deformity. While MRI images would perhaps best identify any femoral or tibial epiphyseal abnormalities, MRI imaging was not a part of the standard preoperative assessment for Tension Band Plating-assisted HE. Thus, determining the JLCA was the most pragmatic option for assessing the effect of mediolateral joint instability, but it may not be an accurate representative of ligamentous instability and intra-articular deformity.

All the radiological measurements were done by a single observer, and therefore, the lack of interobserver and intraobserver reliability is a limitation of this study. However, this limitation should be interpreted in the context of the previous reports by Braun et al. and Akhmedov et al., which observed that some degree of interobserver error is inherent to the malalignment test [35,36]. Lastly, this study was conducted with a relatively small sample size, and 2 patients had short follow-up periods (1 patient was at 5 months follow-up and another had a mild deformity that corrected at the 5 months postoperative). However, considering the rarity of these conditions, prospective and larger studies are challenging to perform, and the current study is one of the larger published cohorts on this topic. Despite these limitations, this study provides valuable insights into the effectiveness of HE in correcting knee coronal plane deformities and its correlation with intra-articular malalignment for SkD patients.

Our study showed that the presence of intra-articular malalignment did not alter the rate or magnitude of correction with HE and that joint alignment (ie, JLCA) improved at the final follow-up. Further, HE effectively corrected deformity in SkD even in the presence of joint laxity and intra-articular deformity in this patient population. This study reinforces that HE should be considered earlier in childhood to compensate for the slower rate of correction among patients with SkD.

## Additional links

- Journal of Orthopaedic Science: Temporary Hemiepiphysiodesis with the Eight-Plate for Angular Deformities: Mid-term Results
- Journal of Pediatric Orthopaedics: Guided Growth for Angular Correction: A Preliminary Series Using a Tension Band Plate
- POSNA Study Guide: Hemiepiphysiodesis Technique

## Ethics approval and consent

The authors confirm that this study received Institutional Review Board Approval (REB#: 1000069403) with appropriate consents, permissions, and releases obtained from patients and/or guardians. Patient confidentiality was preserved during the study, and no patient-identifiable data was reported in the manuscript.

## **Author contributions**

**Prabjit Ajrawat:** Data curation, Formal analysis, Writing – original draft, Writing – review & editing. **Deeptiman James:** Conceptualization, Formal analysis, Methodology, Writing – original draft, Writing – review & editing. **Andrew Howard:** Writing – original draft, Writing – review & editing. **Maryse Bouchard:** Conceptualization, Formal analysis,

Investigation, Methodology, Project administration, Resources, Supervision, Writing – original draft, Writing – review & editing.

#### **Declarations of competing interests**

The authors declare the following financial interests/personal relationships which may be considered as potential competing interests. Dr Maryse Bouchard reports a relationship with OrthoFlexion Inc. that includes: board membership and equity or stocks. Dr Maryse Bouchard reports a relationship with OrthoPediatrics Corp. that includes: consulting or advisory. If there are other authors, they declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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