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ORIGINAL ARTICLE
ORTHOPEDICS AND TRAUMATOLOGY

CORONAL PLANE GROWTH MODULATION FOR GENU VALGUM IN SKELETAL DYSPLASIA

MODULAÇÃO DO CRESCIMENTO DO PLANO CORONAL PARA GENU VALGUM NA DISPLASIA ESQUELÉTICA

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ABSTRACT

Objective: To investigate the efficiency and rates of correction by hemiepiphysiodesis using 8-plate to manage genu valgum deformity in children with skeletal dysplasia. Methods: Eleven children with skeletal dysplasia (three female, eight male; mean age = 10.5 years; age range = 7–13) who underwent temporary hemiepiphysiodesis using 8-plates for genu valgum deformity were retrospectively reviewed. There were nine bilateral cases and two unilateral cases. The mean follow-up time from the index surgery to the final follow-up was 45 (ranging from 24 to 72) months. Radiographical assessment including preoperative and final follow-up measurements of joint orientation angles and mechanical axis deviation (MAD) were conducted. Results: Deformities were completely corrected in nine lower extremities (45%) and partially corrected in seven extremities (35%). In four extremities of two children with Morquio syndrome, MAD worsened. The correction rate of MAD was 1.25 ± 1.62 mm/mo. Conclusion: Though hemiepiphysiodesis using 8-plate requires a longer treatment period, it seems to be an effective treatment for correction of genu valgum in children with skeletal dysplasia. ***Level of Evidence IV, Case Series.***

Keywords: Bone Dysplasia. Genu Valgum. Growth Plate. Epiphyses. Retrospective Studies.

RESUMO

Objetivo: Investigar a eficiência e as taxas de correção da hemiepifisiodese usando placa-8 no tratamento da deformidade de geno valgo em crianças com displasia esquelética. Métodos: Foram avaliadas retrospectivamente 11 crianças com displasia esquelética (três meninas e oito meninos; idade média = 10,5 anos; faixa etária = 7-13) que foram submetidas à hemiepifisiodese temporária com placa-8 devido à deformidade do geno valgo. Havia nove casos bilaterais e dois casos unilaterais. O acompanhamento médio desde a cirurgia de implante até o acompanhamento final foi de 45 (variação de 24 a 72) meses. Foi feita avaliação radiográfica incluindo medidas de acompanhamento pré e pós-operatórias dos ângulos de orientação da articulação e desvio mecânico do eixo (MAD). Resultados: As deformidades foram completamente corrigidas em nove extremidades inferiores (45%) e parcialmente corrigidas em sete (35%). Em quatro extremidades de duas crianças com síndrome de Morquio, o MAD piorou. A taxa de correção do MAD foi de 1,25 ± 1,62 mm/mês. Conclusão: Embora a hemiepifisiodese com placa-8 necessite de um período de tratamento mais longo, a técnica parece ser um tratamento eficaz para a correção do geno valgo em crianças com displasia esquelética. ***Nível de Evidência IV, Série de Casos.***

Descritores: *Displasias Ósseas. Geno Valgo. Lâmina de Crescimento. Epífises. Estudos Retrospectivos.*

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INTRODUCTION

Genu valgum is a common condition which occurs mostly physiologically as part of normal developmental changes in lower limb alignment in the growth phase (from two to 11 years old). This development-related change usually resolves itself spontaneously.[1,2] In turn, pathological genu valgum, which may be idiopathic or secondary to congenital disorders such as skeletal dysplasia, often leads to progressive displacement of the mechanical axis, with pain and functional limitation – thus requiring surgical intervention.[3]

Skeletally immature children can avoid major surgical procedures, including osteotomy and external or internal fixation, by growth modulation using hemiepiphysiodesis. Hemiepiphysiodesis application can be permanent or temporary.[4] Whereas permanent hemiepiphysiodesis must be used carefully in proper surgical timing to prevent under- or over-correction,[5] temporary hemiepiphysiodesis using several implants has expanded for correcting deformities without the risks from the permanent surgery. Because of its simplicity and ease of application, the 8-plate system for temporary hemiepiphysiodesis has recently replaced transphyseal screws and staples.[6]

All authors declare no potential conflict of interest related to this article.

The study was conducted at Istanbul University, Istanbul Faculty of Medicine, Department of Orthopedics and Traumatology.
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Genu valgum is one of the common orthopedic manifestations of skeletal dysplasia, which is a complex group of conditions that affect bone development.[7] Evidence shows that 8-plates have been effective in correcting idiopathic genu valgum deformity.[1,8,9] However, the literature on the efficacy of this technique in children with skeletal dysplasia is still incipient. The surgery of the pathological physis is also concerning due to potential rebound or risk of permanent growth arrest.[6]

This study sought to determine the efficiency, correction rates, and complications of hemiepiphysiodesis using 8-plate to correct genu valgum deformity in children with skeletal dysplasia.

METHODS

After obtaining approval from the institutional review board, we retrospectively reviewed the medical records of 13 children with several types of skeletal dysplasia who underwent temporary hemiepiphysiodesis using 8-plates (Eight Growth Plate™, TST medical®, Istanbul, Turkey) for genu valgum deformity from 2010 to 2018. Based on the eligibility criteria (Table 1), two patients were excluded (one lost to follow-up; one underwent a corrective lower limb surgery) and the other 11 children (20 extremities; nine with bilateral and two with unilateral genu valgum) were included in the study. Parents were informed that their medical records would be used for scientific purposes only and their written informed consent was collected at the final visit.

The diagnoses of skeletal dysplasia were established by the Department of Medical Genetics (Table 2) and all children were managed by the Department of Pediatric Endocrinology and Metabolic Diseases at our institution.

Radiographic outcome measures

To analyze the genu valgum deformities, anatomical tibiofemoral angle (aTFA), mechanical axis deviation (MAD = 3–17 mm), anatomical and mechanical lateral distal femoral angles (aLDFA = 79–83° and mLDFA = 85–90°, respectively), medial proximal tibial angle (MPTA = 85–90°), and lateral distal tibial angle (LDTA = 86–92°)[10] were measured on the immediate preoperative period and on the final follow-up using orthoroentgenography (anteroposterior full-length weight-bearing radiography).

All measurements were taken by an experienced orthopedic surgeon who was blinded to the clinical information of subjects, thus eliminating inter-observer variability but not intra-observer variability. Orthoroentgenograms were obtained by the well-established, standard technique of using three radiographic exposures centered over the hip, knee, and ankle joints and combining these images into a single film to minimize magnification error.[11] The correction rate was defined by dividing the angular correction value (degree) by the period of treatment with the 8-plate in months.[12] Correction amounts and rates were estimated based on MPTA at the proximal tibia and on LDFAs at the distal femur. Furthermore, the correction rate of MAD was estimated by dividing the amount of MAD correction (mm) by the treatment period in months.[4]

Table 1. Eligibility criteria for inclusion and exclusion of the study participants.

Inclusion criteria	Exclusion criteria
· A diagnosis of skeletal dysplasia; · Radiologically-confirmed genu valgum deformity; · Treated for HES using 8-plates; · A minimum of 12 months of follow-up after the index surgery; · Willingness to participate in the study.	· Lost to follow-up; · A history of lower limb correction surgery; · Unwillingness to participate in the study.

HES: temporary hemiepiphysiodesis.

Table 2. Demographic characteristics of the study participants.

Number of patients (extremities)	11 (20)
Bilateral involvement **Unilateral involvement**	9 2
Gender	8M, 3F
Types of skeletal dysplasia	Achondroplasia→3 X-linked hypophosphatemic rickets→3 Ellis-Van Creveld syndrome→1 Morquio syndrome→4
Mean age at surgery (year)	10.5 (ranging 7–13)
Treatment period (month)	35 (ranging 12–60)
Implant removal	+ => 11 limbs of 6 children – => 9 limbs of 5 children
Follow-up duration (month)	45 (ranging 24–72)

Surgical technique

Surgery was performed based on a standardized procedure after preparation under general anesthesia. After a two-to-three cm incision to center the growth plate, a careful surgical dissection was conducted in the submuscular plane. Anatomic positions for the 8-plates (2-hole 4.5 mm titanium plate) relied on joint orientation angles comprising LDFA, MPTA, and LDTA. Except in one child, the plates were attached to both the tibia and femur since both LDFA and MPTAs were abnormal. In one child with only abnormal LDFA (case 4; Table 2), the plate was attached to the femur only. To attach the plates, a 1.2 mm K-wire was first placed into the physis under the image intensifier; then, the 8-plate (2-hole 4.5 mm titanium plate) was inserted extraperiosteally. Next, 1.6 mm guidewires were carefully introduced into the epiphysis and metaphysis to avoid damaging the periosteum and the perichondrial ring. At the final stage, the plate was fixed to the bone using 4.5 mm fully threaded self-tapping cannulated screws over the guide wires.

Follow-up protocol

All children were encouraged to ambulate immediately post-operation as much as pain allowed. No child received specific physical therapy. Children were monitored every three months and assessed by standing x-rays. Treatment aimed to convert MAD to its opposite side.

The deformity correction was divided into three categories according to Boero, Michelis and Riganti:[9] no correction, partial correction (if MAD improved but did not reach neutral alignment), complete correction (if MAD reached neutral or slight varus alignment). Intra and postoperative complications were observed.

Statistical analyses

The IBM SPSS Statistics software version 20.0 (IBM Corp., Armonk, New York, NY, USA) was used for statistical analysis. A $p < 0.05$ was considered statistically significant. Normality tests were conducted using the Shapiro–Wilk test and histograms. Data were presented as "minimum", "maximum", "range", "arithmetic mean", "standard deviation", and "percentage". Preoperative and postoperative values were compared using a Paired-samples t-test. Correction amounts and rates were compared for distal femoral and proximal tibial physes using the Mann-Whitney U test.

RESULTS

The mean period of treatment with the 8-plate was 35 (ranging from 12 to 60) months. The mean age at the time of surgery was 10.5 (ranging from 7 to 13) years. The mean follow-up time from the index surgery to the final follow-up was 45 (ranging from 24 to 72) months. The mean age at the final follow-up was 13.6

(ranging from 10 to 17) years. The mean follow-up time after plate removal was 14 ± 7 months (tables 2 and 3).

Table 4 presents measurements of joint orientation angles (aTFA, MPTA, aLDFA, mLDFA, LDTA) and MADs. Only the decrease in aTFA and MAD was statistically significant ($p = 0.005$ and $p = 0.024$, respectively); all other angles improved, albeit insignificantly ($p > 0.05$). Figure 1 shows the box and whisker plot indicating the distributions of angular correction rates at the distal femur (aLDFA, mLDFA) and versus proximal tibia (MPTA).

Deformities were completely corrected in nine lower extremities (45%) and partially corrected in seven extremities (35%) (Figure 2). However, in four extremities of two children diagnosed with Morquio Syndrome, MAD worsened and deformities remained uncorrected (Figure 3). Corrective osteotomy was then performed in these children.

Four limbs of two children (patients no. 8 and 9) with no correction were excluded in correction rate assessment. The correction rates of aLDFA and mLDFA at the distal femur were 0.384 ± 0.5 degree/

Table 3. Demographic and radiographical data of the study participants including preoperative and final follow-up measurements of joint orientation angles and mechanical axis deviation.

Type of Skeletal Dysplasia	Patient No.	Type of Skeletal Dysplasia	Age at Treatment Onset (years)	Site of Plate Insertion: Femur/Tibia	Duration of 8-plate Treatment (months)	Removal of implants (+/–)	Follow-up period (months)	Side	Joint Orientation Angle (°) Angle Initial Angle Final Δ	Mechanical Axis Deviation*** (mm) Initial MAD Final Δ
Achondroplasia	1 – U*	Achondroplasia	7	Both	R = 24	+	24	R	MPTA 100 88 12 aLDFA 73 76 3 mLDFA 82 90 8 LDTA 84 93 9	62 – 10 72
	2 – B**	Achondroplasia	10	Both	R = 36 L = 36	+	48	R	MPTA 95 92 3 aLDFA 83 81 2 mLDFA 90 84 6 LDTA. 89 97 8	35 22 13
								L	MPTA 94 80 14 aLDFA. 83 87 4 mLDFA 87 90 3 LDTA 82 97 15	42 – 21 63
	3 – B	Achondroplasia	9	Both	R = 60 L = 60	+	72	R	MPTA 94 88 6 aLDFA 79 107 28 mLDFA 86 113 27 LDTA 94 95 1	21 – 44 65
								L	MPTA 92 83 9 aLDFA 85 108 23 mLDFA 92 111 19 LDTA 92 87 5	12 – 60 72
X-linked Hypophosphatemic Rickets	4 – U	X-linked Hypophosphatemic Rickets	4.5	Femur	R = 24	+	60	R	MPTA 89 90 1 aLDFA 75 88 13 mLDFA 87 94 7 LDTA 103 79 22	22 8 14
	5 – B	X-linked Hypophosphatemic Rickets	12	Both	R = 60 L = 60	+	72	R	MPTA 96 90 6 aLDFA 85 86 1 mLDFA 90 86 4 LDTA 92 76 16	14 10 4
								L	MPTA 88 90 2 aLDFA 77 79 2 mLDFA 84 85 1 LDTA 91 89 2	19 10 9
	6 – B	X-linked Hypophosphatemic Rickets	9	Both	R = 24 L = 24	-	24	R	MPTA 97 81 16 aLDFA 67 84 17 mLDFA 78 89 11 LDTA 81 84 3	49 – 19 68
								L	MPTA 100 83 17 aLDFA 62 83 21 mLDFA 70 84 14 LDTA 87 75 12	54 – 9 63
Ellis-Van Creveld syndrome	7 – B	Ellis-Van Creveld syndrome	10	Both	R = 12 L = 12	+	48	R	MPTA 100 95 5 aLDFA 59 58 1 mLDFA 76 72 4 LDTA 92 90 2	47 21 26
								L	MPTA 92 91 1 aLDFA 79 87 8 mLDFA 92 94 2 LDTA 98 97 1	5 4 1

Table 3. Demographic and radiographical data of the study participants including preoperative and final follow-up measurements of joint orientation angles and mechanical axis deviation.

Type of Skeletal Dysplasia	Patient No.	Type of Skeletal Dysplasia	Age at Treatment Onset (years)	Site of Plate Insertion: Femur/Tibia	Duration of 8-plate Treatment (months)	Removal of implants (+/–)	Follow-up period (months)	Side	Joint Orientation Angle (˚) Angle Initial Angle	Final Angle	Δ	Mechanical Axis Deviation*** (mm) Initial MAD	Final Δ
Mucopolysaccharidosis type IVA (Morquio syndrome)	8 – B	Mucopolysaccharidosis type IVA (Morquio syndrome)	10	Both	R = 36 L = 36	-	36	R	MPTA	98	104	6	60 91 – 31
									aLDFA	78	59	19	
									mLDFA	83	63	20	
									LDTA	92	80	12	
								L	MPTA	102	106	4	29 85 – 56
									aLDFA	66	56	10	
									mLDFA	71	63	8	
									LDTA	86	78	8	
	9 – B	Mucopolysaccharidosis type IVA (Morquio syndrome)	12	Both	R = 48 L = 48	-	48	R	MPTA	106	110	4	49 37 12
									aLDFA	81	80	1	
									mLDFA	94	86	8	
									LDTA	74	70	4	
								L	MPTA	88	89	1	16 – 11 28
									aLDFA	80	90	10	
									mLDFA	77	94	17	
									LDTA	91	90	1	
	10 – B	Mucopolysaccharidosis type IVA (Morquio syndrome)	13	Both	R = 24 L = 24	-	24	R	MPTA	87	88	1	22 – 16 38
									aLDFA	79	88	9	
									mLDFA	75	91	16	
									LDTA	89	89	0	
								L	MPTA	100	103	3	26 40 – 14
									aLDFA	75	76	1	
									mLDFA	83	80	3	
									LDTA	60	70	10	
	11 – B	Mucopolysaccharidosis type IVA (Morquio syndrome)	8	Both	R = 24 L = 24	+	36	R	MPTA	102	94	8	27 – 10 37
									aLDFA	84	85	1	
									mLDFA	91	93	2	
									LDTA	65	64	1	
								L	MPTA	92	106	14	11 32 – 21
									aLDFA	85	70	15	
									mLDFA	87	78	9	
									LDTA	98	74	14	

U*: unilateral genu valgum; B**: bilateral genu valgum; Δ: difference between preoperative and final follow-up measurements; MAD: mechanical axis deviation; MPTA: medial proximal tibial angle; mLDFA: mechanical lateral distal femoral angle; LDTA: lateral distal tibial angle.

Table 4. Preoperative and final follow-up measurements of joint orientation angles and mechanical axis deviations.

	Preoperative	Final follow-up	Δ	P
aTFA˚	27.4 ± 10	12.6 ± 20	14.8 ± 17.7	*0.005**
MPTA˚	95.6 ± 5.39	92.5 ± 9.10	– 3.5 ± 8.23	0.140
aLDFA˚	76.8 ± 7.76	81.4 ± 13.6	4.65 ± 12	0.076
mLDFA˚	83.8 ± 7.18	87 ± 12.6	3.45 ± 11.4	0.35
LDTA˚	87 ± 10.6	83.7 ± 10.1	– 2.7 ± 9.39	0.177
MAD (mm)	30.5 ± 16.5	8.95 ± 37	22.1 ± 38.1	*0.024**

Δ: difference between preoperative and final follow-up measurements; aTFA: anatomical tibiofemoral angle; MPTA: medial proximal tibial angle; aLDFA: anatomical lateral distal femoral angle; mLDFA: mechanical lateral distal femoral angle; LDTA: lateral distal tibial angle; MAD: mechanical axis deviation. *Significance was defined at $p < 0.05$.

mo and 0.395 ± 0.39 degree/mo, respectively. The correction rate of MPTA at the proximal tibia was 0.297 ± 0.38 degree/mo. Figure 4 presents the box and whisker plot of the distributions of angular correction amounts at the distal femur (ΔaLDFA, ΔmLDFA) versus proximal tibia (ΔMPTA). The correction rate of MAD was 1.25 ± 1.62 mm/mo (Figure 5).

Hardware was removed from six children (ten extremities). Correction with plates was maintained in five children (nine extremities) with open growth plates. No clinical complications such as implant failure, loosening, or infection were observed during follow-up except for undercorrection or overcorrection of the angular deformity.

DISCUSSION

Pathological genu valgum, a well-known component of skeletal dysplasia, causes both cosmetic problems and premature osteoarthritis and must be corrected.[13] Although osteotomies have traditionally been the basis of deformity correction,[3] these procedures have potential drawbacks, including prolonged immobilization, extensive soft-tissue dissection, infection, delayed union, nonunion, and malunion.[3,14] To overcome these drawbacks, temporary hemiepiphysiodesis using the 8-plate has been successfully used in recent years to correct idiopathic genu valgum.[1,8,9] However, to our knowledge, the literature on the correction of genu valgum secondary to skeletal dysplasia with 8-plate is still incipient. Furthermore, since skeletal dysplasia is a rare and heterogeneous group of disorders that particularly affect epiphyseal growth plates, the efficacy of this surgical method in such disorders raises concern.

One major concern is whether the 8-plate treatment can correct genu valgum in children with a pathologic physis as seen in skeletal dysplasia. Our review of the literature shows that few studies[3,4,9,13] involving children with different types of skeletal

dysplasia have reported different correction rates in the knee. Boero, Michelis, and Riganti[9] compared idiopathic versus dysplasia/syndrome patients and suggested early operation with the 8-plate in the group of children with skeletal dysplasia since this treatment has a longer correction period. Table 4 shows that comparing these studies is difficult due to different definitions of correction rates based on the joint orientation angles (MPTA, LDFA) or MAD as well as heterogeneous patient series and different types of deformity (varus, valgus, and windswept). Nonetheless, our findings seem to corroborate other studies, indicating that this method allows significant correction even though the correction rate would be slower in a pathologic physis than in an idiopathic deformity.

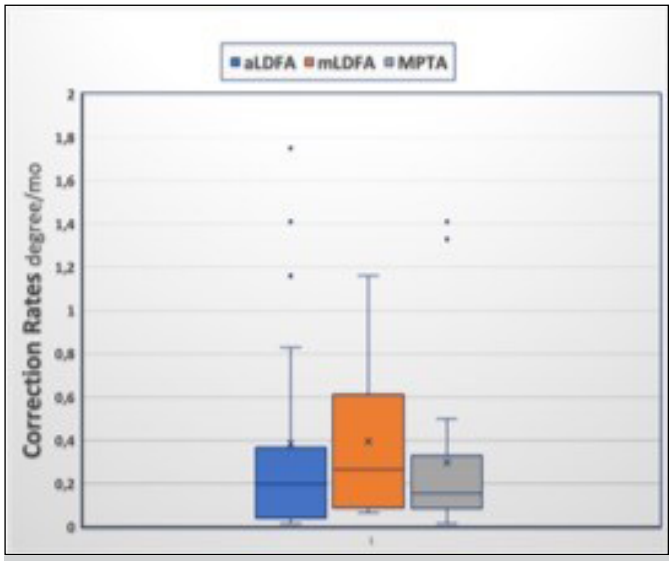

Figure 1. Box and whisker plot showing the distributions of angular correction rates at the distal femur (aLDFA, mLDFA) and versus proximal tibia (MPTA). Horizontal bars represent the median whereas boxes and whiskers show the interquartile ranges and data ranges, respectively.

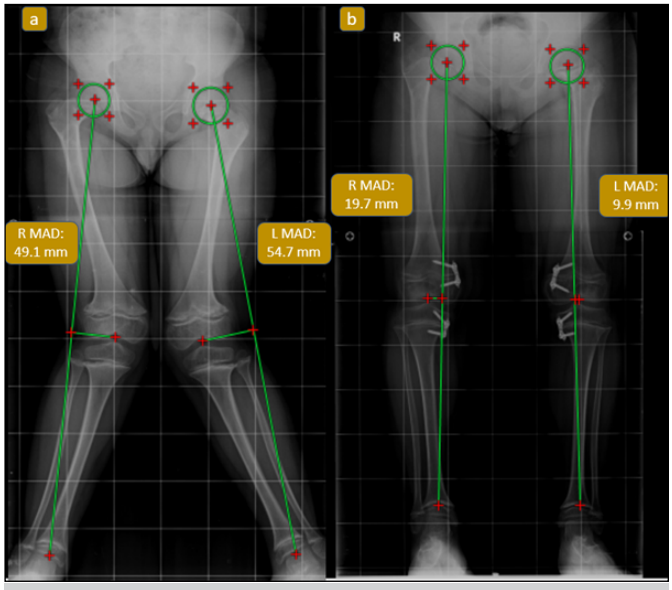

Figure 2. Preoperative (a) and final follow-up (b) orthoroentgenograms of case no. 6 (see Table 3). Observe the complete correction on both sides, with neutral mechanical axis deviation on the left side and slight varus alignment on the right side.

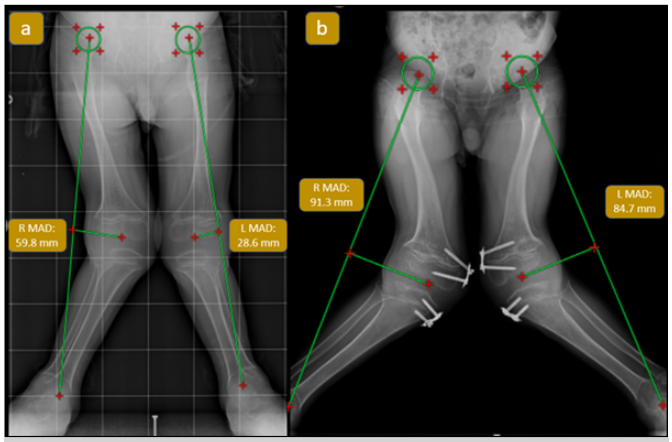

Figure 3. Preoperative (a) and final follow-up (b) orthoroentgenograms of case no. 8 (see Table 3). No correction on both sides, with a worsening of mechanical axis deviations.

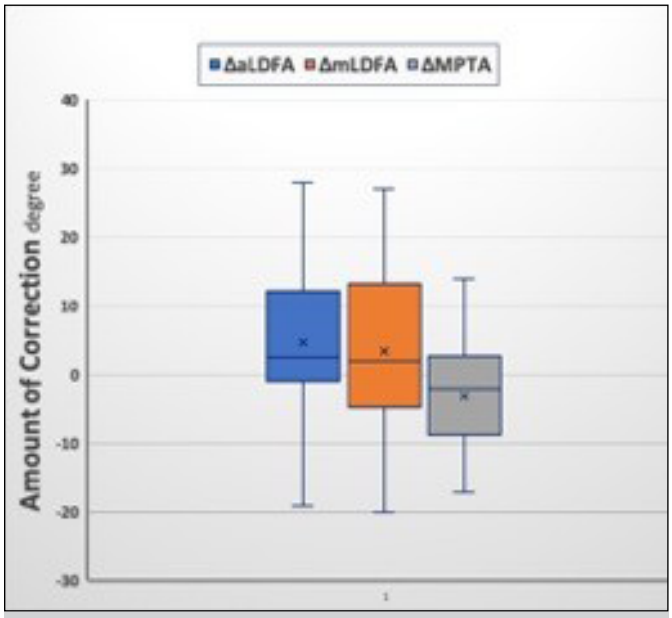

Figure 4. Box and whisker plot showing the distributions of angular correction amounts at the distal femur (ΔaLDFA, ΔmLDFA) versus proximal tibia (ΔMPTA). Horizontal bars represent the median whereas boxes and whiskers show the interquartile ranges and data ranges, respectively.

Figure 5. Box and whisker plot showing the distribution of correction rates of mechanical axis deviation.

Other major concerns of surgery for a pathologic physis include the rebound phenomenon and risk of permanent growth arrest.[6] Some authors[1,3,6] advocate an overcorrection of 5° to 10° (mild varus) in children with risk factors (dysplasia, obesity, etc.) to prevent a possible recurrence. Although some studies[3,9,13] included patients who underwent surgery due to recurrence of genu valgum, none of our patients sustained the rebound phenomenon. However, the short-term follow-up after hardware removal in some of our patients is insufficient to accurately report this phenomenon. Moreover, the pathologic physis was expected to show no tolerance against surgical intervention, with the risk of a permanent physeal arrest and irreversible injury.[15] Recent studies[3,4,9,13] on this issue showed a safe application of 8-plates in skeletal dysplasia. Our observations thus reaffirmed the safe use of the 8-plate for this condition.

We have consecutively performed this technique in children with genu valgum deformity secondary to skeletal dysplasia. Deformities were resolved completely in almost half of the cases (45%), but some achieved partial correction (35%) and required further osteotomies. Nonetheless, as Schroerlucke et al. suggest,[16] even partial correction can be considered a favorable outcome in such cases since it can lead to a technically easier osteotomy with a lower risk of complications.

In four lower extremities of two children with Morquio syndrome, no correction was achieved and MAD and joint orientation angles worsened. This poor outcome can result from several factors, including the timing of surgery. In our case series, cases with partial or no deformity correction presented a higher age at initial treatment than cases with complete corrections. The study by Boero, Michelis, and Riganti,[9] which sought to compare results of 8-plate use between idiopathic and pathological genu valgum, emphasized the importance of age at initial treatment. The authors began to treat pathological deformities earlier (2-13 years old) than idiopathic deformities (8-14 years old), obtaining successful comparable results. Our results support Boero, Michelis, and Riganti's finding that starting treatment at a very young age is reasonable since deformities are minor and correction is rapid.

Another risk factor for treatment failure is the type of skeletal dysplasia since distinct types are associated with different growth rates in the epiphyseal plate. Only two patients from our series presented treatment failure, caused by Morquie syndrome, in which bone growth typically decelerates quickly after the age of three and ends around 11 years old.[13] In a case series including 23 children with genu valgum deformities secondary to Morquio syndrome, Cooper et al.[13] determined that hemiepiphysiodesis using the 8-plate requires a longer time for deformity correction. Similarly to our observations, the authors stated that in children over 11 to 12 years old, deformity correction can fail due to insufficient growth capacity.

This study has limitations and strengths. The major limitations of the study were its retrospective nature, limited sample size, and short-to-mid-term follow-up. Another limitation was the lack of a control group with children with idiopathic genu valgum. Moreover, deformities were assessed only in the coronal plane using long anteroposterior radiographs. As a strength, however, based on the inclusion criteria, none of the children in our cohort had clinically problematic sagittal deformity that could impair the radiographic measurement of coronal alignment. Finally, another strength is that our study population consists of a heterogeneous group of children with several types of skeletal dysplasia. Despite its limitations, our study is one of the few[3,4,9,13] to present results of hemiepiphysiodesis with 8-plates in skeletal dysplasia.

CONCLUSION

Overall, temporary hemiepiphysiodesis using 8-plate seems to be an effective treatment to correct genu valgum in children with skeletal dysplasia, with low complication rates if applied at the right age. Despite having a longer treatment period, this technique can achieve a sufficient amount of correction at both the distal femur and proximal tibia in skeletal dysplasia if started early.

AUTHORS' CONTRIBUTIONS: Each author contributed individually and significantly to the development of this article. YS: study design, analysis and interpretation of data, drafting and critical revision of the manuscript; MD: data analysis, drafting and critical revision of the manuscript; AMY, FB, CS: study design, drafting and critical revision of the manuscript.

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