Increasing the Knee Arc of Motion in Patients With Arthrogryposis: Minimum 2-year Follow-up

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Background: Surgery for knee flexion contractures in patients with arthrogryposis multiplex congenital (AMC) have achieved extension to redirect the arc of motion and improve ambulation but has not demonstrated maintained increases in total range of motion (ROM). This study aimed to review the clinical outcomes of combined posterior knee release, proximal femoral shortening, and nerve decompression in patients with arthrogryposis.

Methods: A retrospective chart and radiographic review were performed on patients with AMC who underwent treatment for knee flexion deformities ≥ 30 degrees. ROM, ambulation status, and orthotic use were reviewed and analyzed. Complications were recorded.

Results: Twenty-nine patients with 51 knees and a mean age of 5.7 years were included. The mean follow-up was 36.9 months. The mean ROM increased from 49 to 80 degrees between pre-op and latest follow-up ($P < 0.0001$). The mean final follow-up flexion deformity was 10 degrees ($P < 0.0001$). Preoperative ROM was moderately correlated with final ROM ($r_s = 0.51$). The percentage of ambulatory patients improved from 39% to 93%. Five limbs experienced a fracture either intraoperatively or postoperatively, and 5 limbs required a return trip to the operating room.

Conclusions: Improved ambulation and knee ROM can be maintained in patients with AMC at a minimum 2-year follow-up. Prospective investigation and longer follow-up are required to validate these findings.

Level of Evidence: Level IV—therapeutic.

Key Words: arthrogryposis, contracture, knee, shortening, motion, flexion, femur, posterior, release, AMC, multiplex, congenita arthrogrypotic

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None of the authors received financial support for this study.
The authors declare no conflicts of interest.

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Supplemental Digital Content is available for this article. Direct URL citations are provided in the HTML and PDF versions of this article on the journal's website, www.pedorthopaedics.com.

DOI: 10.1097/BPO.0000000000002826

(*J Pediatr Orthop* 2025;45:e179–e185)

Arthrogryposis multiplex congenital (AMC) is a congenital condition with diverse etiology, characterized by joint contractures in 2 or more joints.[1] An incidence of 1 in 3000 to 5000 live births has been reported.[2] Patients undergo hip and knee surgery in hopes of improving ambulation ability.[3]

Surgical interventions for severe knee flexion deformities have included posterior capsular releases, hamstring lengthening, anterior hemiepiphysiodesis, distraction and extension with external fixation, and distal femoral extension osteotomies.[4–12] These interventions have been able to achieve knee extension and redirect the arc of motion, but not increase the total range of motion (ROM).[4–12] While a patient's ambulatory function may improve, their ability to sit may be compromised due to loss of flexion. The deformity often recurs, particularly in skeletally immature patients.[2,5,6,13]

In 2021, a novel approach for treating flexion deformities in patients with AMC was published and included peroneal nerve decompression, posterior capsulotomy, hamstring lengthening, and proximal femoral shortening (fixed with an intramedullary rod and small plate and screws); the entire operation was performed through a long lateral incision and small medial incision (Fig. 1) (a description is available in the electronic appendix, Supplemental Digital Content 1, http://links.lww.com/BPO/A794).[14] This improved ROM and ambulation at 6-month follow-up.[14] Nevertheless, there exists a high rate of recurrence in AMC[2,5,6,13] and continued monitoring is required to demonstrate efficacy. The purpose of this paper is to evaluate the knee ROM measurements (between time intervals and within the classification), ambulation status and complications following surgery at a minimum 2-year follow-up. The authors hypothesized that the improved motion and ambulatory status would be maintained.[14]

METHODS

After obtaining Institutional Review Board approval (IRB #2021-029, MetroWest Medical Center), a retrospective chart review was performed Orthoped on all patients with AMC presenting to the authors' institution from January 2015 to June 2023. Patients treated through June

FIGURE 1. Preoperative knee flexion contracture (A) and postoperative image (B) demonstrating improvement of the flexion contracture following sciatic/peroneal nerve decompression, posterior capsulotomy, and femoral shortening.

2021 were identified and classified into 3 types as seen in Table 1.

Patients classified as type 1 or type 3 who underwent surgical treatment for their knee flexion deformity were included in this study. The standardized surgical treatment for knee contractures was utilized for all type 1 and 3 patients, was described in full detail in the previous study,[14] and can be found in the electronic appendix, Supplemental Digital Content 2, http://links.lww.com/BPO/A794. Additional inclusion criteria included preoperative, postoperative and final follow-up ROM ambulatory status; and a minimum follow-up time of 24 months from the time of surgery. Patients diagnosed with Escobar syndrome (mutations in CHRNG or TMP2 genes) or Popliteal Pterygium syndrome (mutations in the IRF6 gene) were excluded from the study.

Ambulatory status, patient demographics and surgical history were recorded. ROM values were made with a goniometer and obtained from the office visits and physical therapy notes. The amount of femoral shortening was derived from the operative note. The use of knee-ankle-foot orthoses (KAFOs), ankle-foot orthoses (AFOs), and ambulatory status were recorded preoperatively and at latest follow-up. Hoffer classification determined ambulatory patients and nonambulatory patients.[15] Ambulatory patients were defined as independent community ambulators with or without aids and household ambulators who require aids, but may require a wheelchair in the community.[15] While nonambulatory patients were defined as nonfunctional ambulators who require a wheelchair but can ambulate during physical therapy/perform transfers and nonambulators who require wheelchairs and cannot perform transfers.[15]

Statistical analyses were performed using R Studio (R Core Team 2022, Vienna, Austria. https://www.R-project.org/). Fisher Exact test compared categorical variables. The knee parameters flexion deformity, maximum

TABLE 1. Arthrogryposis Knee Classification

Type 1 (flexion type)		Type 2 (extension type)		Type 3 (combined type)	
Lacking extension > 30 degrees with ROM > 45°		Full extension with < 30 degrees ROM		Lacking extension > 30 degrees with ROM < 45 degrees	
1A Quadriceps present*	1B Quadriceps absent†	2A Knee located	2B Knee dislocated	3A Quadriceps present*	3B Quadriceps absent†

*1 to 2/5 on manual strength testing.
†0/5 on manual strength testing.
ROM indicates range of motion.

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TABLE 2. Patient Demographics

Knee type	No. limbs	Sex	Age (y)
1A	26	14F/12M	4.7 (2.9)
1B	20	7F/13M	4.7 (5.4)
3	5	3F/2M	9.8 (3.1)
		$P=0.3954$	$P=0.0237$

Type 3 knees were combined due to low sample size.
F indicates female; M, male.

flexion, and range of motion were analyzed using a Friedman test and post hoc analysis was performed using the Conover test with Bonferroni correction. Effect size, a measure of the strength of a relationship in a given population, was calculated using Kendall W. Cohen interpretation (small: W<0.3, medium: $0.3 \leq$ W<0.5, large: W ≥ 0.5) was used for all effect sizes.[16] Differences between knee classification and knee parameters were analyzed using Kruskal-Wallis ANOVA and post hoc analysis was performed using a Dunn test with Bonferroni correction. Effect sizes were calculated with Pearson r. Wilcoxon Signed Rank test was used to determine whether there were differences in preoperative and follow-up knee parameters within each classification. Multiple Spearman Rho correlation coefficients were calculated to determine whether any correlation existed between the data points. A *P*-value of <0.05 was considered significant.

RESULTS

Twenty-nine patients (15 male/14 female) with 51 knees met the inclusion and exclusion criteria. The mean age at the time of surgery was 5.7 years ($\pm$3.2 y). The mean follow-up was 36.9 months (range 24.2 to 75.9 mo). Patient demographics and knee classifications are summarized in Table 2.

Descriptive statistics for knee parameters of the entire cohort at each time interval are shown in Table 3. Figures 2–4 demonstrate the changes and post hoc comparisons of the entire study cohort for the knee flexion deformity (large effect, W = 0.89), maximum knee flexion (small effect, W = 0.20) and knee range of motion (large effect, W = 0.53), respectively. Table 4 summarizes the median difference, CI and significance over the different time intervals for each knee parameter. Overall, there were significant improvements in the knee flexion deformity ($P<0.0001$) and knee range of motion ($P<0.0001$); however, overall maximum knee flexion was slightly worse compared with its measurement preoperatively ($P=0.0104$). The median amount of femoral shortening was 35 mm [interquartile range (IQR) 16.2 mm]. Knee motion improved 0.14 degrees/month of follow-up (IQR 0.60 degrees/month) and loss of extension occurred at 0.05 degrees/month of follow-up (IQR 0.31 degrees/month).

The knee parameters were compared between their classification types. Table 5 lists the medians/IQRs for each classification and comparisons with effect sizes. The final knee range of motion was significantly higher in the type 1A knees compared with type 1B ($P=0.0227$). The residual knee flexion deformity was significantly better in the type 1A knees compared with the type 1B knees ($P=0.0008$). There was no significant difference in the amount shortened between classification groups ($P=0.2840$).

Graphical representations of the within classification comparisons for flexion deformity, maximum flexion and range of motion can be found in the electronic appendix as Figure 1, Supplemental Digital Content 2, http://links.lww.com/BPO/A795, 2, Supplemental Digital Content 3, http://links.lww.com/BPO/A796 and 3, Supplemental Digital Content 4, http://links.lww.com/BPO/A797, respectively. Type 1A knees demonstrated a median improvement in flexion deformity of 40.0 degrees ($P<0.0001$, CI: 34.8-43.5 degrees), decrease in maximum knee flexion by 18.0 degrees ($P=0.0049$, CI: 7.1-32.4 degrees) and increase in knee range of motion of 32.0 degrees ($P=0.0005$, CI: 20.9-46.5 degrees). All type 1A changes had a large effect size (r = 0.88, 0.67, and 0.77, respectively). For type 1B knees, there was a significant improvement in the knee flexion deformity of 51.5 degrees ($P<0.0001$, CI: 38.4-65.7 degrees), decrease in maximum flexion by 22.5 degrees ($P=0.0300$, CI: 4.2-48.3 degrees), and improved in knee range of motion 30.0 degrees ($P<0.0001$, CI: 13.7-42.7 degrees). All changes had a large effect size (r = 0.88, 0.51, and 0.87, respectively). For type 3 knees, there were no significant changes.

Figure 5 is a correlation matrix for the preoperative and follow-up knee parameters, age and shortening amount. Inverse, moderate correlations were found between age at the time of surgery and both preoperative maximum knee flexion ($P=0.0011$) and preoperative knee range of motion ($P=0.0031$). The preoperative knee flexion deformity was also found to be inversely correlated with follow-up knee range of motion ($P=0.0021$) and positively correlated with shortening ($P<0.0001$). Finally, preoperative knee range of motion was moderately correlated with follow-up knee range of motion ($P=0.0001$).

Preoperatively, 11 patients were ambulators (37.9%) and 18 were nonambulators (62.1%). At follow-up, 27 were ambulators and (93.1%) and 2 nonambulators (6.9%). Of

TABLE 3. Knee Measurements of the Entire Study Cohort at Different Time Intervals

	Time interval		
	Pre-op	Post-op	Follow-up
Flexion contracture	46.0° (IQR 25.5°)	0.0° (IQR 3.5°)	10.0° (IQR 15.0°)
Maximum flexion	95.0° (IQR 32.5°)	80.0° (IQR 30.0°)	90.0° (IQR 15.0°)
Range of motion	49.0° (IQR 30°)	80.0° (IQR 35.0°)	80.0° (IQR 36.5°)

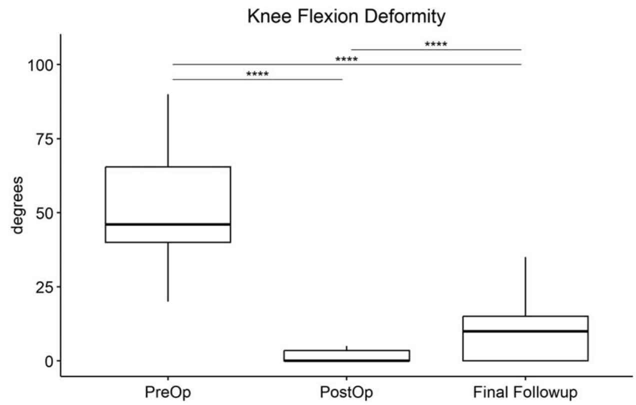

FIGURE 2. Boxplots demonstrating the median (horizontal line), interquartile range (box) and total range (whiskers) for the knee flexion deformity of the entire cohort compared at different time points. ****$P<0.0001$.

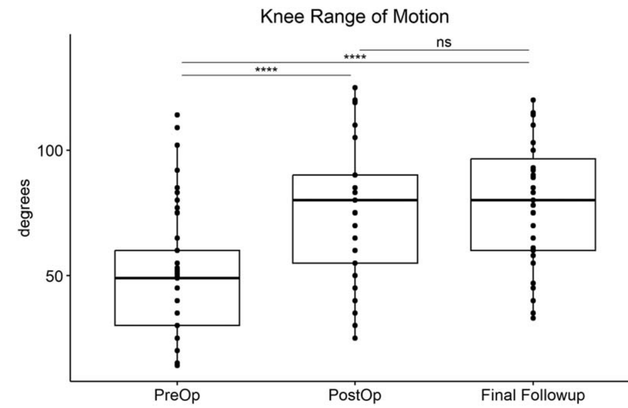

FIGURE 4. Boxplots demonstrating the median (horizontal line), interquartile range (box), and total range (whiskers) for maximum knee flexion of the entire cohort compared at different time points. ns indicates nonsignificant. ****$P<0.0001$.

the 27 ambulating patients, 15 patients were community ambulators and 12 were home ambulators. Nine patients used AFOS, 16 patients used KAFOS and 2 did not require an orthosis. Both nonambulating patients were classified as 1B. Comparison with knee classification demonstrated a significant increase in AFO usage over KAFO usage in the 1A group compared with the 1B group ($P=0.0294$) and odds ratio of 10.19 (CI: 1.72-113.61)

There were 5 patients/5 limbs that sustained an ipsilateral femur fracture. One femur fractured at the distal femoral physis during acute extension and was fixed with 3 wires. The wires were removed 3 weeks postoperatively. Two femurs fractured at 2 weeks postoperatively. One occurred early in the series before the use of an intramedullary device was part of the construct; this was taken back to the operating room for reduction and fixation with a plate and intramedullary rod. The other was treated non-operatively with a long leg splint as this femur already had a rod in place. Two femurs fractured at 1 month and 29 months postoperatively; both underwent revision fixation.

Six patients/7 limbs had temporary neuritis following surgery requiring gabapentin; this resolved on its own and no patient is currently on gabapentin. Four patients/5 limbs required a second operation, not related to a fracture. One patient/1 limb developed femoral anteversion and required a derotation osteotomy. One patient/2 limbs had recurrence of their bilateral knee flexion deformities and underwent revision posterior release without shortening. One patient/1 limb returned to the operating room for manipulation under anesthesia at 1 month postoperatively. Finally, 1 patient developed a distal femoral procurvatum deformity that was treated with a distal femoral osteotomy. There were no physeal arrests in this cohort, including the patient who had procurvatum. There was no wound breakdown or infections in this cohort.

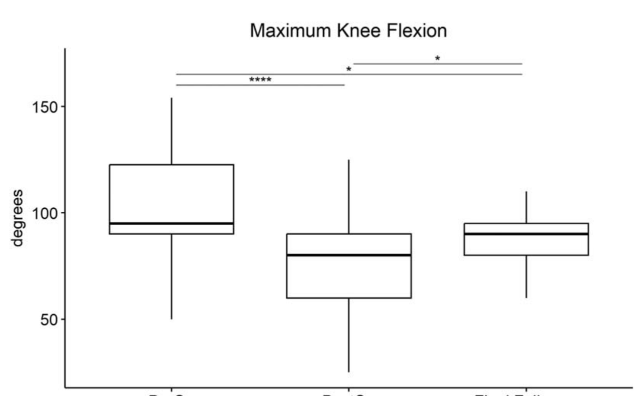

FIGURE 3. Boxplots demonstrating the median (horizontal line), interquartile range (box) and total range (whiskers) for knee range of motion of the entire cohort compared at different time points. ns indicates nonsignificant. *$P<0.05$, ****$P<0.0001$.

DISCUSSION

The knee is commonly affected in patients with AMC.[17] Knee flexion contractures are the most prevalent and interfere with ambulation.[18,19] Distal femoral extension osteotomies,[5,6,13,20] guided growth,[11] and external fixation[4,7,10,12] have effectively achieved extension to improve ambulation ability; however, the ability to improve total ROM has not been shown. In this patient series, the goal of surgery was to increase total ROM and improve ambulatory status. The previous study demonstrated improved ROM but had the majority of patients had <2-year follow-up.[14]

The results of this study demonstrated maintenance of near full extension (10-degree flexion deformity, Table 3) and an increased ROM (35.0 degrees°, Table 4) at median follow-up of 37 months (Fig. 6). At follow-up, 93% of patients were community or home ambulators. A previous study, utilizing distal femoral shortening in half of the patients and with 2-year minimum (average 11.9 y)

TABLE 4. Median Differences of the Knee Parameters at Each Time Interval With Their Associated Pairwise Comparison for the Entire Cohort

	Time intervals		
	Post vs. pre	Follow-up vs. pre	Follow-up vs. post
Flexion deformity	−45.0° (CI −48.2 to −40.6°)****	−40.0° (CI −43.9 to −34.2°)****	2.0° (CI −4.4 to 7.1°)****
Maximum flexion	−30.0° (CI −48.9 to −18.0°)****	−15.0° (CI −31.7 to −3.7°)*	5.0° (CI −4.4 to 10.2°)*
Range of motion	30.0° (CI 21.3–48.4°)****	35.0° (CI 27.3-45.0°)****	5.0° (CI −1.9 to 10.7°)

*$P < 0.05$.
****$P < 0.0001$.
For example, if the median postoperative knee flexion deformity was 0 degree and the preoperative knee flexion deformity was 48.5 degrees, the difference would be −48.5°.

follow-up, demonstrated overall improvements of flexion deformities from 53 degrees preoperatively to 11 degrees postoperatively to 27 degrees at the most recent follow-up but did not mention overall range of motion.[9]

This is the first study to demonstrate improvement of motion at a 2-year minimum follow-up. Increasing and maintaining motion is due to the postoperative bracing and physical therapy protocol following surgery. Patients are not placed in a cast following surgery. They are placed in a knee immobilizer and physical therapy for passive knee extension and flexion is initiated 2 to 3 days postoperatively. A custom molded KAFO is made at 2 weeks and patients began weightbearing at 3 to 4 weeks postoperatively. Families are encouraged to continue therapy until the child reaches maturity.

Femoral shortening has been described in previous articles discussing knee flexion deformities in AMC.[9,21] One study mentioned the amount that was shortened in a single patient, 1.5 cm.[21] The median amount of shortening in this study was 3.5 cm, and there was no difference found between knee types. There was a positive correlation between preoperative knee flexion deformity and the amount of shortening required. The amount of shortening is determined by the amount needed to achieve full extension. After making an initial osteotomy, the distal segment should be allowed to translate proximally, and the knee fully extended. The femur is shortened by the amount of overlap between the proximal and distal diaphysis when the knee fully extends without force.[14] It should be noted that many of the knees demonstrate posterior subluxation at maximum extension; long term effects are unknown.

Figure 5 demonstrates a positive correlation between preoperative knee range of motion and follow-up knee range of motion. This finding is reminiscent of total knee replacements: postoperative ROM is predicated on preoperative ROM.[22] The strongest correlation was between follow-up maximum flexion and follow-up knee range of motion ($r_s = 0.8$, $P < 0.0001$). The purpose of surgery is to decrease the knee flexion deformity; however, this finding supports the need to concentrate on maximizing flexion postoperatively too. Limited use of immobilization (KAFO/extension splint for 2 wk full-time and then only at night) and utilizing therapy to achieve maximum extension and flexion are recommended. Finally, preoperative knee flexion deformity is positively correlated with follow-up knee flexion deformity.

There was a difference between the knees based on classification at both the preoperative and final follow-up visits. Type 1B knees had significantly worse flexion deformities (68 degrees) preoperatively and this is related to the absence of quadriceps. Type 1A knees had more range of motion both preoperatively and at final follow-up compared with types 1B and 3. These findings suggest the classification may have utility for patients and surgeons.

Contracture recurrence has been described in the literature, mentioned in 46 of 68 studies providing a follow-up.[23] Using distal femoral extension osteotomies, DelBello and Watts[5] reported a loss of correction of 0.9 degrees per month and Moreira et al[6] reported a loss of 0.69 degrees/month. Yang et al[13] used external fixation and reported an

TABLE 5. Comparison of Knee Parameters Between Knee Classification

Comparison of knee parameters between knee types	Preoperative			Follow-up		
Type	Flexion deformity	Maximum flexion	Range of motion	Flexion deformity	Maximum flexion	Range of motion
1A	45 (10)	105 (30)	60 (28)	0 (10.0)	90 (24)	90 (27)
1B	68 (23.8)	90 (32)	35 (22.5)	15 (15.2)	90 (17.5)	68 (31.8)
3	45 (10)	60 (10)	15 (0)	10 (10.0)	80 (10)	70 (5)
Comparison						
1A vs. 1B	0.0087*	0.2080	0.0001†	0.0008*	0.4120	0.0227*
1A vs. 3	0.8560	< 0.0001†	< 0.0001†	1.0000	0.0289*	0.0699
1B vs. 3	0.0146†	0.0131†	0.2370	0.4810	0.3000	1

Median values with (interquartile ranges) are listed in the top section. The bottom section denotes the P-values for the comparisons and associated effect sizes.
*Moderate effect size.
†Large effect size.

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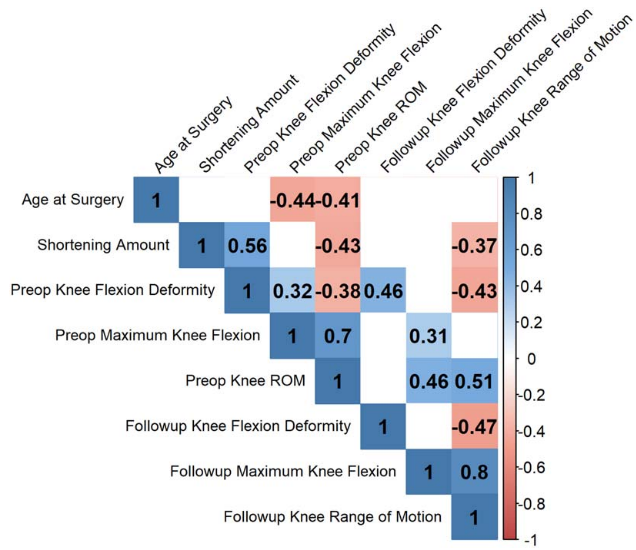

FIGURE 5. Correlation matrix comparing age at surgery, shortening amount and preoperative/final follow-up knee parameters. Spearman rho correlation coefficients (r_s) are labeled. All shown values are statistically significant. Blue shading/positive values denote positive correlation and red shading/negative values indicate inverse correlation.

increase in mean flexion contracture of approximately 20.8 degrees at 5 years. This equates to 0.35 degrees/month. The current study demonstrated an increase in knee motion of approximately 0.14 degrees/month and a loss of extension correction of 0.05 degrees/month.

This intervention is not without complication. There were 5 limbs that fractured or loss of fixation during the perioperative/follow-up period. The intraoperative femur fracture led the authors to prophylactically fixate the distal femoral epiphysis to the metaphysis in a retrograde or antegrade fashion with 1.8 mm wires, to prevent fracture during the acute extension portion of the procedure. The wires are removed after extension. Care is taken during posterior dissection to not disrupt the periosteum near the distal femoral physis. Intramedullary devices are used in all primary cases in combination with a plate and screws. In addition, 2.0 or 2.4 mm plates and screws are used to minimize the stress-riser effect which was determined to be the cause of the other fractures. There were no cases of wound breakdown in this cohort, but the previous report had cases of incisional dehiscence and it is recommended to move the lateral incision slightly posterior to circumvent this complication.[14]

There were 4 patients/5 limbs, which required a second operation during follow-up. One patient required a femoral derotation for anteversion. Patients with AMC have hips that are affected as well. Rotation is assessed intraoperatively after fixation with the goal of a balanced rotational profile in extension. One patient/2 limbs underwent revision posterior release. One patient developed a distal femoral procurvatum deformity. This could be from an insult to the posterior distal femoral physis, an occult fracture during manipulation in the operating room or therapy. This patient went on to require a distal femoral osteotomy, but no physeal bar was identified and the distal femur has continued to grow symmetrically.

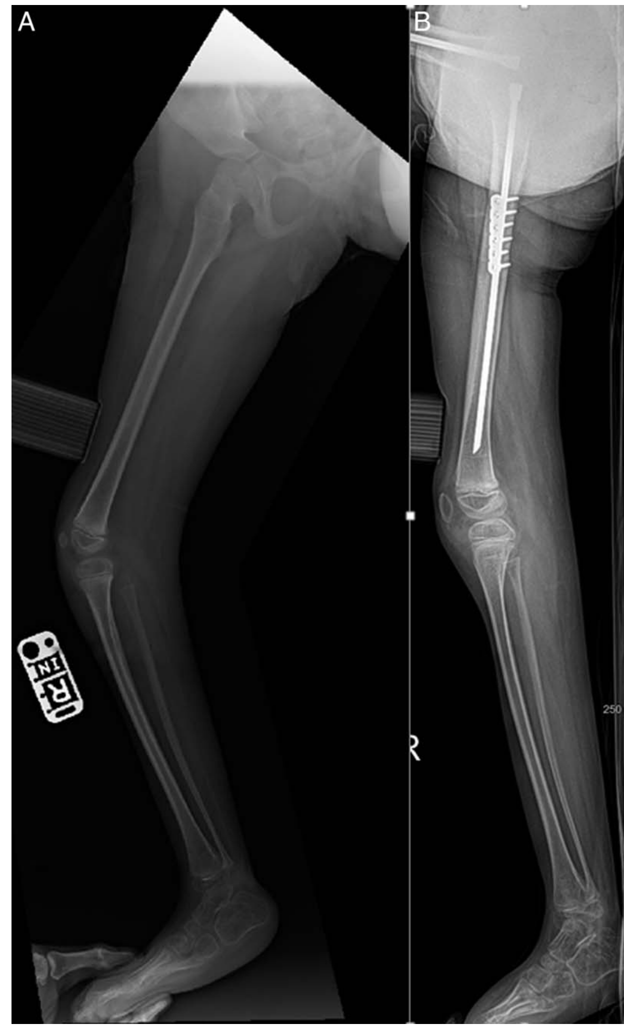

FIGURE 6. Preoperative (A) and most recent follow-up (B) long leg lateral radiographs in maximum extension following nerve decompression, posterior capsulotomy, and femoral shortening.

This study is not without its limitations. As a retrospective study, the data are derived from what is available in the medical records. In addition, there are no patient-reported outcomes. Although ambulatory ability is presented, this does not replace functional tests or patient-reported outcomes. A prospective study with longer follow-up is needed to validate the outcomes and support the findings associated with this specific treatment.

CONCLUSIONS

Single-stage treatment for arthrogrypotic knee flexion deformities demonstrates improved ambulation and ROM with minimum 2-year follow-up. The current study demonstrates promising results considering the literature and difficulty in treating these patients. Ambulation and ROM improvement were able to be achieved without further significant intervention.

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