Femoral Non-elongating Rodding in Osteogenesis Imperfecta – The Importance of Purchasing Epiphyseal Plate

Wei-Chun Li, Hsuan-Kai Kao, Wen-E Yang, Chee-Jen Chang[1], Chia-Hsieh Chang

Background: Corrective osteotomy and intramedullary rodding are widely used in children with osteogenesis imperfecta (OI), but revision rodding is often required. This study aims to investigate the effect of purchasing distal femoral epiphysis on the longevity of fixation using non-elongating rod.

Methods: We investigated children with Sillence type III or IV OI who received antegrade femoral Rush rod fixations at age between 4 and 10 years in our institution. The fixations were classified into group A in which the rod reached distal femoral epiphysis and group B in which the rod stopped at femoral metaphysis. Failure of fixation is defined as rod cutting out of the cortex or when revision surgery was performed. Calculation of longevity of each rod fixation and Kaplan–Meier survival analysis were performed and compared between the two groups.

Results: Eighteen children had the first femoral rodding at a mean age of 6.9 years and received a total of 61 femoral roddings with a mean follow-up of 11.4 years. Group A included 38 roddings performed at a mean age of 7.1 years and group B included 23 roddings performed at a mean age of 6.6 years. Group A had less revision rate (58% vs. 87%), more chance of survival > 3 years (74% vs. 43%), and longer median survival time (80 months vs. 33 months) than group B.

Conclusions: Less revision rate and better 3-year and 5-year survival rate were proved in the roddings that purchased epiphysis. We emphasize on using the precise implant length to purchase distal femoral epiphysis when non-elongating rod is the only available implant for fixation in children with OI.
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At a Glance Commentary

Scientific background of the subject

Revisions were often required after non-elongating rod fixation for fracture or osteotomy in children with OI. Several technical tips have been described as expert's opinions, rather than scientific evidence.

What this study adds to the field

Less revision rate, more chance of survival >3 years, and longer median survival time were proved in femoral roddings that purchased epiphyseal plate in a case series study controlling OI severity and operation age.

Key words: epiphyseal plate, intramedullary rodding, osteogenesis imperfecta

Osteogenesis imperfecta (OI) is a common inherited disorder of connective tissue that primarily affects the skeleton. Patients with OI show considerable phenotypic variability, but most have brittle bones and joint hypermobility. A major problem for patients with OI is the tendency to sustain multiple fractures with progressive bowing of their long bones.[1] The bowing of long bones would cause further bending stress and repeated fractures.

Since Sofield and Miller reported corrective osteotomy and intramedullary rod fixation in 1959, the surgeries have been widely used to treat bowing of the long bones and prevent repeated fracture in OI.[2-6] However, rod fixation in children with brittle bone is associated with high rate of complications, including outgrown nail, nail migration, and re-fracture; therefore, revision surgeries are often required.[7,8] To increase the longevity of nail fixation, various designs of elongating nail have been developed with variable clinical results.[9-16]

From the Department of Pediatric Orthopedics, Chang Gung Memorial Hospital at Linkou, Chang Gung University College of Medicine, Taoyuan, Taiwan; [1]Graduate Institute of Clinical Medical Science, Chang Gung University, Taoyuan, Taiwan
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Correspondence to: Dr. Chia-Hsieh Chang, Department of Pediatric Orthopedics, Chang Gung Memorial Hospital at Linkou. 5, Fusing St, Gueishan, Taoyuan 333, Taiwan (R.O.C.). Tel: 886-3-3281200 ext. 2423; Fax: 886-3-3289582; E-mail: chiahchang@gmail.com
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In countries where elongating intramedullary nails are not available, non-elongating Rush rod is the implant of choice to fix fracture and osteotomy in children with or without OI.[17,18] We observed that in femoral fixation, if the tip of Rush rod fixation penetrates distal femoral epiphysis, it can last longer before revision. The purpose of this study is to investigate the effect of rod purchasing distal epiphysis on the longevity of femoral fixation.

METHODS

We performed a retrospective review of patients with OI who had received antegrade femoral intramedullary rodding using Rush rod in authors' hospital from January 1992 to June 2010. To decrease the confounding factors from phenotype variability in subsequent analysis, this study only included Sillence type III and IV patients. Patients who had the first operation at age before 4 years or after 10 years were excluded, because of very weak bone in the younger age group or mild cases of OI in older patients.

Twenty-four patients received this kind of surgery from January 1992 to June 2010. However, four patients older than 10 years and two patients younger than 4 years were excluded from our analysis for the control of age factor. Finally, 18 patients were included in this study.

We enrolled 18 children (10 males and 8 females) in this study. There were 168 intramedullary Rush rod fixations in total during the study period; among them, 61 fixations were performed in the femurs. The mean age of the first femoral rodding was 6.9 years. The mean follow up was 11.4 years after the first rodding. Fourteen patients had bilateral femoral roddings and the other four patients had unilateral femoral roddings with 32 femurs having Rush rod fixation in this study. Since there were unilateral or bilateral femur cases, we describe the surgeries by the femurs instead of by the patients [Table 1].

Four patients received bisphosphonate (Fosamax) therapy during the study period, but they were equally distributed between two groups. They had the usual dosage of 70 mg administered orally per week. Five femoral roddings were performed during the period of bisphosphonate therapy and the other 56 femoral roddings were performed without bisphosphonate therapy.

The 61 femoral roddings were classified into two groups by the location of the Rush rod tip. In group A, the Rush rods penetrated physis and entered distal femoral epiphysis [Figure 1]. In group B, the Rush rods did not penetrate physis and stopped at distal femoral metaphysis [Figure 2]. There were 38 roddings in group A and 23 in group B. To study the longevity of each femoral rodding, we defined failure of fixation when revision surgeries were performed or when the Rush rod tip was cut out of the metaphyseal cortex of the distal femur, but revision was not scheduled for clinical reasons [Figure 3]. Longevity of fixation, revision rate, and other background characteristics were compared between the two groups using the Student's *t*-tests for continuous variables and Chi-square test for categorical variables. Kaplan–Meier survival analysis was used to compare the longevity of

Table 1: Outcomes of the femoral roddings in groups A and B

	Group A (tip in epiphysis)	Group B (tip in metaphysis)	*p* value
No. of femoral roddings	38	23	
Age at surgery (years)	7.1	6.6	0.32
Follow-up (months)	98.4	67.0	0.10
Revision	22 (58%)	20 (87%)	0.02
Longevity>3 years	28 (74%)	10 (43%)	0.02
Median survival time (months)	80	33	<0.01

Figure 1: Right femur Rush rod fixation in a girl aged 9 years and 8 months. The rod tip was in distal epiphysis (group A).

Figure 2: Left femur Rush rod fixation in a boy aged 9 years and 3 months. The rod tip was in metaphysis (group B).

femoral roddings in the two groups. The analyses were performed using SPSS (version 17.0, SPSS Inc., Chicago, Illinois).

RESULTS

Eighteen OI patients had Rush rod fixation in 32 femurs, including 14 Sillence type III and 4 type IV children. They were equally distributed between the two groups. Among the 32 primary femoral roddings, 24 roddings had been revised after a mean of 40.3 months. Five of the 24 revision roddings had the second revisions 44.2 months after the first revisions. In the re-revision cases, all five patients received surgery due to outgrown rod. Among these five re-revision surgeries, there was no infection, physeal bony bar formation, and other perioperative complications such as fracture. Besides, there was no difference between revision and re-revision cases with regard to surgical time, blood loss, infection, and union time for osteotomy site. In summary, 32 primary roddings and 29 revision roddings were performed in the 18 patients. The indications of revisions included outgrown rod in 25 roddings, proximal rod migration in 2 roddings, and fracture in 2 roddings.

In group A, there were 38 femoral roddings performed between 1997 and 2008. The mean age at surgery was 7.1 years (range, 4.2-9.9 years). When the rod tip still purchased epiphyseal plate, it rarely displaced anteriorly within the distal femoral epiphysis. Once the rod tip withdrew to distal femoral metaphysis, it gradually displaced and even penetrated metaphyseal cortex [Figure 3]. Twenty-two of the 38 roddings (58%) had undergone revision after a mean of 49.4 months (range, 7-128 months). In the other 16 roddings that are still in place, they have been observed for a mean of 98.4 months (range, 33-157 months). Five of the 16 roddings were regarded as failure in this study because of cortex penetration, though revision had not been performed. No transphyseal bony bar formation or deformation of femoral condyles was noted in the group A cases.

In group B, there were 23 femoral roddings performed between 1992 and 2008. The mean age at surgery was 6.6 years (range, 4.0-10.0 years). Twenty of the 23 roddings(87%) had undergone revision after a mean of 34.9 months (range, 16-79 months). The other three roddings have been observed for a mean of 67.0 months (range, 50-91 months). One of the three roddings was regarded as failure because of cortex penetration.

No significant difference was noted in the age at surgery and duration of follow-up between the two groups. Group A had significantly less revision rate (58% in group A vs. 87% in group B, $p = 0.02$) and more chance to have implant fixation longer than 3 years (74% in group A vs. 43% in group B, $p = 0.02$). [Table 1] Kaplan–Meier survival analysis showed a significantly better survivorship in group A. Group A had better 3-year survival rate (0.76 vs. 0.44) and 5-year survival rate (0.53 vs 0.16) than group B. The median of estimated survival time is 80 months in group A and 33 months in group B ($p = 0.001$) [Figure 4].

DISCUSSION

The study offers scientific evidence that femoral non-elongating rod fixation in children with OI could sustain longer when the rod tip purchases distal femoral epiphysis. Several techniques were recommended to prevent complications in using Rush rods, including anatomic reduction, adequate diameter and length of implants, and dual rodding fixation.[14,17,19] Placement of the rod in the center of medullary cavity of the bone is essential to minimize the risk of extracortical migration.[14] Purchasing distal femoral epiphysis to enhance fixation stability

Figure 3: Rod was outgrown and penetrated through anterior cortex.

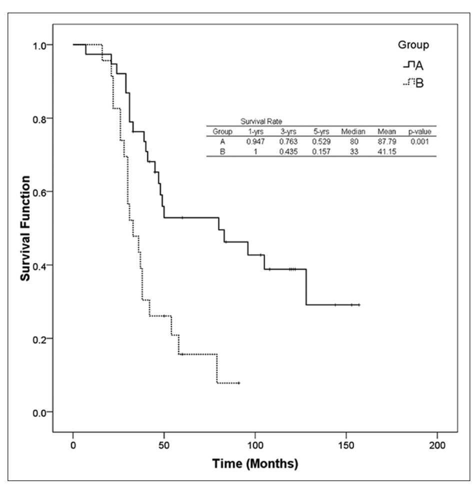

Figure 4: Kaplan–Meier survival curve for groups A and B.

has been described in literature before. Tiley and Albright mentioned this technical tip when they emphasized central placement of the rod.[17] This study emphasizes on choosing proper rod length for distal epiphysis purchasing when using non-elongating rods, which are still the implants of choice in several countries.

Antegrade rodding purchasing the distal femoral epiphyseal plate could offer better fixation stability than the rodding only reaching metaphysis. Rod tip rarely displaced on coronal or sagittal plane when purchasing the epiphysis. The cartilaginous epiphyseal plate has better holding of fixation than the osteoporotic cancellous bone in metaphysis because chondrogenesis at the growth plate and epiphyseal cartilage are not involved in OI. In this study, no femur fracture or progressive angulation deformity occurred in these patients when the rod tip still remained in epiphysis. In group A of this study, the rod tip stayed in epiphysis for 6-18 months and osteotomy or fracture site had united before the rod tip withdrew to metaphysis. Once the rod tip withdrew to metaphysis because of bone growth, re-fracture and rod tip impingement against metaphysis cortex occurred gradually [Figure 3].

Regarding the bisphosphonate therapy in our study, 10 patients received bisphosphonate therapy after the age of 10 years, 4 patients received it between 4 and 10 years of age (our inclusion criteria), and the other 4 patients did not receive bisphosphonate therapy. Five femoral rodding surgeries were performed during the therapy and their survival longevities were 33, 40, and 79 months in group A and 38 and 58 months in group B. There was no very strong evidence to support bisphosphonate therapy to improve the longevity of this kind of surgery.

Limitations of this study include its retrospective design, small number of cases studied, and not following up to skeletal maturity in all cases. We mixed the first roddings for corrective osteotomy and subsequent roddings for re-fracture or outgrown rod in the study subjects and compared the longevity between groups. Though the composition of first and subsequent roddings in both groups is comparable, a study including first rodding only and adequate case number would be of higher research quality. The impact of better bone quality after using bisphosphonate on the longevity of fixation requires further study. This study concludes the clinical significance of purchasing distal femoral epiphysis in antegrade femoral non-elongating rodding in children with OI. Less revision rate, longer intervals before revision operations, survival increasing to more than 3 years, and better survival curve have been proved after controlling for disease severity, age, and location of rodding. No growth tethering or bony bar formation was noted after rodding through the epiphyseal plate. We emphasize on choosing the precise implant length to reach distal femoral epiphysis when non-elongating rod is used for fixation in children with OI.

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