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Knee Arthritis in Congenital Short Femur after Wagner Lengthening

Changhoon Jeong, MD[,†]; Muharrem Inan, MD[*,‡]; Eric C. Riddle, MHS, PA-C[*]; Peter G. Gabos, MD[*]; and J. Richard Bowen, MD[*]*

Knee anomalies are common in patients with congenital short femurs who require lengthening to correct limb-length discrepancies. We retrospectively reviewed the incidence of knee arthritis and the factors influencing its occurrence after femoral lengthening using the Wagner method. Twenty-three patients with congenital short femurs treated with the Wagner method were followed up until skeletal maturity (minimum, 5 years postoperatively). The mean age of the patients at lengthening was 10.8 years (range, 8.4–14.5 years). The mean leg-length discrepancy at the time of surgery was 9.7 cm (femur, 7.6 ± 3.7 cm; tibia, 2.1 ± 1.8 cm). Femoral lengthening (mean, 7.9 cm) was performed in 17 patients. Femoral lengthening and tibial lengthening were performed simultaneously in six patents (mean, 11.8 cm). The mean age of the patients at the last followup was 16.8 years (range, 14–20.3 years). Eighteen patients had arthritis at followup. Nine patients had severe arthritis develop, seven of whom had knee instability preoperatively and temporary subluxation during the lengthening procedure. Seventy-eight percent of patients had arthritis develop in the knee after lower-limb lengthening using the Wagner method for congenital short femurs. Patients who had an unstable knee before surgery had temporary knee subluxation develop during the lengthening procedure, and patients who had simultaneous lengthening of the femur and tibia had a high association with degenerative arthritis changes in the knee.

Level of Evidence: Retrospective study, Level IV (case series). See the Guidelines for Authors for a complete description of levels of evidence.

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From the [*]Department of Orthopaedic Surgery, Alfred I. duPont Hospital for Children, Wilmington, DE; the [†]Department of Orthopedic Surgery, Holy Family Hospital, The Catholic University of Korea, Bucheon, Korea; and the [‡]Center of Orthopedic and Traumatology, Malatya, Turkey.
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Correspondence to: J. Richard Bowen, MD, Alfred I. duPont Hospital for Children, 1600 Rockland Road, PO Box 269, Wilmington, DE 19899. Phone: 302-651-5723; Fax: 302-651-5951; E-mail: jrbowen@nemours.org.
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Congenital short femur is a rare condition that may result in a severe leg-length discrepancy.[22] An anticipated discrepancy greater than 4 to 6 cm often is treated with femoral and/or tibial lengthening.[16,29] However, stress from extensive lengthening can cause adverse effects on the adjacent joints.[4,10,13,28] Complications include stiffness,[32] subluxation, and dislocation during or immediately after lengthening procedures.[10,13]

The combined effects of knee anomalies in patients with congenital short femurs include hypoplasia of the lateral femoral condyle, genu valgum, hypoplastic patella, and the absence of an anterior cruciate ligament.[12,24] Limb-lengthening procedures undoubtedly increase the risk for having early knee arthritis develop.[12,24] Nakamura et al[17–19] and Fink et al[8] reported intraarticular cartilage injury and meniscal damage after lower-limb lengthening in rabbits. Stanitski[26] found dogs that had a femoral lengthening of 30% or greater had changes associated with early arthritis, including gross cartilage fibrillation, loss of proteoglycan staining, and frank cartilage necrosis. Although articular cartilage changes that could lead to arthritis have been shown experimentally after limb lengthening,[8,17–19,25–27] we are unaware of any articles describing predisposing factors that could lead to knee arthritis after limb-lengthening procedures in patients with congenital short femurs.

We asked whether there would be a substantial risk of arthritis in patients with congenitally short femurs lengthened using the Wagner method. We also asked if there were predisposing factors in patients with congenital short femurs lengthened by the Wagner method that would lead to a risk of arthritis.

MATERIALS AND METHODS

We retrospectively reviewed the medical records and radiographs of 23 consecutive patients (23 limbs) treated for congenital short femurs by the senior author (JRB) from 1981 to 1990. The patients were divided into one of three groups by severity of arthritis at followup. Fourteen patients were boys and nine were girls. All patients were diagnosed with congenital short femurs, had femoral lengthening (with or without simultaneous tibial lengthening) using the Wagner lengthening method,[31] and were followed up until skeletal maturity. The mean age of the patients at the time of surgery was 10.8 years (range, 8.4–14.5 years). The 17 patients who had femoral lengthening alone had a mean leg-length discrepancy of 7.9 cm. Femoral and tibial lengthening were performed simultaneously in the other six patients with a mean leg-length discrepancy of 11.8 cm. The mean duration of followup was 8.2 years (range, 5–16 years), and the mean age of the patients at the last followup was 16.8 years (range, 14–20.3 years).

The patient demographics were similar in all three groups (Table 1). The patients in Groups 1, 2, and 3 were of similar ages at the time of limb lengthening: 10.6, 10.6, and 11.2 years, respectively. Their mean ages at followup also were similar; 15.8, 16.3, and 17.9 years, respectively. The mean preoperative leg-length discrepancy was 9.7 cm (range, 4.2–17.7 cm). The mean discrepancy of the affected femur was 7.6 cm (range, 3.6–17.3 cm), and the mean discrepancy of the affected tibia was 2.1 cm (range, 0.3–7.9 cm). The percentages of lengthening were similar in Groups 1 and 2: 15.8% ± 4.7% and 15.4% ± 4.7%, respectively, whereas the percentage of lengthening in Group 3 was 19.5% + 5.4%.

TABLE 1. Patient Data According to Group

Parameters	Group 1 (n = 5)	Group 2 (n = 9)	Group 3 (n = 9)
Mean age at lengthening (years)	10.6	10.6	11.2
Mean age at followup	15.8	16.3	17.9
Congenital short femur			
Type I	4	6	6
Type II	1	3	3
Fibular hemimelia	3	5	3
Hypoplasia of lateral femoral condyle	3	3	2
Hypoplasia of tibial spine (Types 0, I, II)	0, 4, 1	0, 4, 5	4, 4, 1
Knee instability	1	2	7
Subluxation of knee during lengthening	0	3	7
Percentage of lengthening (percent)	15.8	15.4	19.5
Simultaneous femoral and tibial lengthening	0	2	4
Mean mechanical axis deviation (mm)	9.6	0.6	8.2
Mean lateral distal femoral angle (degrees)	76.4	76	78.7
Mean medial proximal tibial angle (degrees)	89	86.6	87.1

Group 1 = no arthritic changes; Group 2 = mild arthritic changes; Group 3 = severe arthritic changes

The Wagner technique included a middiaphyseal transverse osteotomy. The osteotomy was created by multiple drill holes in the transverse plane perpendicular to the long axis of the bone at a chosen location. An osteotome was placed parallel to and at the level of the drill holes to complete the osteotomy. The long bone was distracted 1.5 cm immediately after the osteotomy. The iliotibial band and lateral intramuscular fascia were incised. The Wagner fixator was applied to allow correction of any preoperative varus or valgus deformity at the knee.

The lengthening procedure was started on the third postoperative day. The femur (and tibia when indicated) was distracted at a rate of 1.5 mm per day. The children were hospitalized during the lengthening process and received physical therapy for a minimum of 2 hours per day. Additionally, the children received a minimum of 1 hour of aqua therapy per day. Lengthening was stopped if there was knee subluxation. The patients were treated with physical therapy until the knee subluxation resolved and limb lengthening then was restarted. When the desired lengthening was obtained, the femur (and the tibia when indicated) was fixed internally with a Wagner plate, and the distraction gap was filled with autologous iliac crest bone graft. Immediately after internal fixation of the femur (and the tibia when indicated), the patient was permitted to ambulate with partial weightbearing using crutches.

We evaluated the following clinical parameters: age of the patients at lengthening and followup, type of congenital short femur using the classification of Kalamchi et al (Type I in 16 patients and Type II in seven patients),[14] types of fibular hemimelia using the classification of Achterman and Kalamchi (Type IA in seven patients, Type IB in one patient, and Type II in three patients),[2] and anterior knee instability before lengthening using the anterior drawer test.

We evaluated patients' preoperative radiographs for leg-length discrepancy, tibial spine hypoplasia, and lateral femoral condyle hypoplasia. Radiographs taken during lengthening were evaluated for the percentage of lengthening and subluxation. Followup radiographs were evaluated by three observers (JRB, CHJ, MI) for mechanical axis deviation, joint orientation angles,[20] and arthritis.

Arthritic changes in the knee were graded according to the scale of Arsever and Bole[3] on anteroposterior (AP) radiographs taken with the patient in the standing position. Grade 0 (severe) was defined as complete joint space loss and/or severe subchondral sclerosis and cysts. Grade 1 (moderate) was defined as greater than 50% joint space narrowing and/or mild subchondral sclerosis and subchondral cysts. Grade 2 (mild) was defined as less than 50% joint space narrowing compared with the contralateral knee. Grade 3 was considered normal. Patients were divided into three groups: Group 1 included patients without arthritis (Arsever and Bole Grade 3), Group 2 included patients with mild arthritis (Arsever and Bole Grade 2), and Group 3 included patients with severe arthritis (Arsever and Bole Grades 0 and 1).

Femur and tibia lengths were measured using orthoroentgenograms. The percentage of lengthening was defined as the amount of lengthening/length of the limb being lengthened times 100. At the final followup, the mechanical axis deviation, the anatomic lateral distal femoral angle, and the anatomic medial proximal tibia angle were measured using 36-inch AP standing radiographs of both lower extremities with the patella oriented forward.

Tibial spine hypoplasia was classified by the number of tibial spines: Type 0 was a round, smooth tibial plateau without tibial spines, Type I had one tibial spine, and Type II had two tibial spines. Types 0 and I indicated hypoplasia of the tibial spine. Hypoplasia of the lateral femoral condyle and a shallow intercondylar sulcus were determined on the lateral radiographs of the knee.

Knee subluxation during lengthening was defined as posterior, lateral, or posterolateral. Posterior subluxation was defined as posterior translation of the tibia in relation to the femur (> ⅓ the AP width of tibial plateau on the lateral radiograph). Lateral subluxation was defined as lateral translation of the tibia in relation to the femur (> ⅓ of the width of the lateral tibia plateau). Posterolateral subluxation was defined as posterior subluxation with any component of lateral translation of the proximal tibia.

RESULTS

Eighteen of the 23 patients had degenerative knee arthritis develop after a Wagner limb-lengthening procedure. Nine patients had mild arthritis of the knee (Group 2) and another nine patients had severe arthritis (three patients had Grade 0 arthritis; six patients had Grade 1 arthritis) (Group 3). Only five patients (Group 1) had no clinical or radiographic evidence of knee arthritis. The patients in Groups 2 and 3 had similar clinical features. Both groups consisted of six patients, each with Types I and II congenital short femurs. Similarly, Groups 2 and 3 had an equal number of patients (three patients each) with Type II congenital short femurs. Groups 2 and 3 also had patients with fibular hemimelia. Group 2 had five patients with fibular hemimelia (Type IA in two patients, Type IB and Type II with one patient each, and Type III in one patient). Group 3 had three patients with fibular hemimelia (one patient with Type IA and two patients with Type II).

Simultaneous lengthening of the femur and tibia predisposed patients to having arthritis of the knee at followup. Two of nine patients in Group 2 and four of nine patients in Group 3 had simultaneous lengthenings of the femur and tibia. The patients in Group 1 had only femoral limb lengthenings. The mean percentage of limb lengthening was greater ($p = 0.05$) for patients in Group 3 (19.5%) compared with patients in Group 2 (15.4%) and in Group 1 (15.8%).

No patients in Group 1 had their limb-lengthening procedures complicated by knee subluxation. Four of these patients were classified as having Type I congenital short femurs and one patient had a Type II congenital short femur. Three of these five patients had an associated fibular hemimelia (two patients had Type IA and one patient had Type II). Knee anomalies included tibial spine hypoplasia (four patients with Type I; one patient with Type II) and three patients with lateral femoral condyle hypoplasia. One patient had clinical knee instability before lengthening. One patient had simultaneous femoral and tibial limb lengthenings. At followup, the mean mechanical axis deviation) was 9.6 ± 16.5 mm laterally, the mean anatomic lateral distal femoral angle was 76.4° ± 6.3°; and the mean anatomic medial proximal tibial angle was 89° ± 3.6°.

There was a trend toward a positive correlation between preoperative clinical knee instability and/or the development of knee subluxation during the limb-lengthening procedure and degenerative arthritis present at followup. Clinical knee instability was present in only two of nine patients in Group 2, whereas seven of nine patients of the patients in Group 3 had an unstable knee before surgery. Unlike the patients in Group 1, knee subluxation occurred during the limb-lengthening procedure in three patients in Group 2 and in seven patients in Group 3 (five posteriorly and two posterolaterally).

The presence of tibial spine hypoplasia and/or hypoplasia of the lateral femoral condyle was present equally in all three groups (Table 1) and did not influence the development of degenerative arthritis of the knee after a Wagner limb-lengthening procedure. Knee anomalies characteristic of patients with congenital short femurs were present in patients in all three groups.[12,14,15,22,24] Hypoplasia of the lateral femoral condyle occurred in three patients in Group 1, three patients in Group 2, and two patients in Group 3. Type 0 hypoplasia of the tibial spine was present only in four patients in Group 3. Four patients in each group had Type I tibial spine hypoplasia. One patient had Type II hypoplasia of the tibial spine in each of Groups 1 and 3. Five patients in Group 2 had Type II hypoplasia of the tibial spine.

The mean mechanical axis of deviation assessed radiographically at followup was 9.6 ± 8.5 mm laterally in Group 1, 0.6 ± 14.3 mm laterally in Group 2; and 8.2 ± 17.2 mm laterally in Group 3. The mean anatomic lateral distal femoral angle was 76° and anatomic medial proximal tibial angle was 86.6° at followup. The mean anatomic lateral distal femoral angle was 76.4° in Group 1, 76° in Group 2, and 78.7° ± 5.1° in Group 3. The mean anatomic medial proximal tibial angle was 89° in Group 1, 86.6° in Group 2, and 87.1° ± 2.7° in Group 3.

DISCUSSION

Precautions must be taken when performing limb lengthening in patients with congenital short femurs to prevent the risk of knee arthritis. Seventy-eight percent of the patients had knee arthritis develop after limb lengthening by the Wagner technique. There seemed to be a predisposition to knee arthritis in patients who had an unstable knee before lengthening or if knee subluxation developed during the lengthening procedure. Although the number of patients in this study who had simultaneous limb lengthenings of the femur and the tibia by the Wagner technique was small (six patients); four of these patients had severe knee arthritis develop (Grade 0). Simultaneous lengthenings of the femur and the tibia predisposed patients with congenital short femurs to severe degenerative arthritis.

Our study has several limitations. It is a retrospective study with a small number of patients, and we have no patient satisfaction and function data. However, congenital short femur is a rare condition and we report the largest group of patients with congenital short femurs to have limb lengthenings by the Wagner technique. Although the population was small, clear and important trends were apparent. We lacked matched control subjects with similar knee dysfunction who did not have lengthening procedures. Also, we know of no study regarding the long-term natural history of the knee in patients with untreated congenital short femurs. Because of the retrospective study design and small numbers of patients, we are unable to comment on the degree of knee instability or knee subluxation that occurred during the limb-lengthening procedure and whether the severity of either relates to the ultimate severity of degenerative knee arthritis. The nature of the physical therapy treatments and the number of treatments required to correct the knee subluxation, which occurred during the limb-lengthening procedures of 10 patients, were unavailable. These questions need to be addressed in a prospective study.

The Wagner technique was first reported in the English literature in 1978.[28] It was a well-accepted technique for lengthening until Ilizarov and DeBastiani developed a less invasive method of distraction osteogenesis callus distraction (Callotasis).[6] The Wagner technique is not the preferred lengthening procedure because of its high complication rate.[1,11,13,32] Zarzycki et al reported the average complication rates were 45% for femoral lengthening and 40% for tibial lengthening using the Wagner technique.[32] They advocated immediate distraction and thought that a faster distraction rate might produce greater force on the surrounding joint surfaces, resulting in joint stiffness and motion limitations.[31,32] Several experimental studies have shown excessive limb lengthening may damage the articular cartilage in adjacent joints because of increased mechanical stress, which may predispose the joint to the development of early arthritis of knee.[17,19,26,27]

Gillespie and Torode[9] reported that knee instability is a common problem in patients with congenital short femurs. Our findings suggest instability before surgery and the development of knee subluxation during the lengthening procedure may predispose patients with congenital short femurs to early-onset degenerative knee arthritis. Seven of nine patients with severe arthritis (Group 3) had knee instability preoperatively and/or temporary subluxation during the lengthening procedure. Our study supports the work by Eldridge and Bell, who suggested that knee subluxation occurring during substantial limb lengthening may induce abnormal pressure on articular cartilage and result in degenerative changes.[7]

Our findings suggest simultaneous femur and tibia lengthenings should not be performed in children with congenital short femurs. Saleh and Burton made a similar recommendation for skeletally immature patients with achondroplasia.[23] Several authors have reported the rapid distraction characteristic of the Wagner method was associated with a high complication rate in the knee.[5,7,32] Dahl and Fishcher[5] reported knee subluxation occurred in five patients who had simultaneous femoral and tibial lengthenings and suggested avoiding simultaneous femoral and tibial lengthenings could minimize knee subluxation. Eldridge and Bell[7] reported rapid distraction, especially double-level lengthening procedures, resulted in knee contracture and subluxation secondary to the effects of gastrocnemius and hamstring muscle contracture. Four of our six patients who had combined lengthening had posterior knee subluxation, and all four patients ultimately had severe degenerative changes in the knee.

The percentage of limb lengthening also may predispose patients to a high complication rate.[6] Some authors have reported that limb lengthening in patients with congenital short femurs should not exceed 20% to 25% of the initial length during any one lengthening procedure.[6,9,15] Lengthening greater than 20% to 25% of the initial length of the limb results in a substantial increase in the complication rate.[6,9,15] In our study, the mean limb lengthening performed was less than 20% in all groups. The percentage of limb lengthening was 15.8% in Group 1, 15.4% in Group 2, and 19.5% in Group 3. The patients in Group 3, despite having limb lengthening less than 20%, experienced the greatest complication rate. Therefore, the percentage of limb lengthening alone may not be the most important factor influencing the outcome of limb-lengthening procedures.

Excessive mechanical axis deviation is also a risk factor for the development of early arthritic changes of the knee.[30] Lower-limb malalignment can produce relatively high contact pressures to the articular cartilage of the medial or lateral tibiofemoral joint surfaces during weight-bearing.[21] The high contact pressure may cause damage to the cartilage and ultimately lead to degenerative joint disease. The mechanical axis deviation was within normal range (9.7 + 6.8 mm), and there was no difference in mechanical axis deviation between groups.

The design of the Wagner external fixator does not allow the apparatus to span the knee during femoral lengthening. It has been shown in an animal model that ring fixators such as the Ilizarov device can be beneficial in protecting the knee during lengthening.[27] Stanitski et al developed a construct that spanned the knee, placing the hinges as close as possible to the axis of rotation of the knee.[27] Distraction for bone lengthening was performed at a rate of 0.25 mm three times daily for a 30% increase over initial bone length. In their limited canine series (n = 4), gross examination postmortem showed no evidence of pressure necrosis, loss of cartilage substance, or articular cartilage fissuring in the experimental limbs as compared with the contralateral control. Serial sagittal sections through the medial and lateral femorotibial condylar contact surfaces showed a loss of safranin O uptake, characteristic of proteoglycan depletion.[27] These findings suggest an improvement over previous constructs that did not span the knee.

Our findings suggest skeletally immature patients with congenital short femurs who have limb lengthenings by the Wagner method have a high incidence of degenerative knee arthritis at skeletal maturity. The presence of knee instability before surgery, the development of subluxation during the lengthening procedure, and simultaneous femoral and tibial lengthenings seem important factors which may predispose patients to early degenerative arthritis of the knee.

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