Ankle Deformity Secondary to Acquired Fibular Segmental Defect in Children

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Background: The authors report the long-term effect of acquired pseudoarthrosis of the fibula on ankle development in children during skeletal growth, and the results of a long-term follow-up of Langenskiold's supramalleolar synostosis to correct an ankle deformity induced by an acquired fibular segmental defect in children.

Methods: Since 1980, 19 children with acquired pseudoarthrosis of the fibula were treated and followed up for an average of 11 years. Pseudoarthrosis was the result of a fibulectomy for tumor surgery, osteomyelitis of the fibula and traumatic segmental loss of the fibula in 10, 6, and 3 cases, respectively. Initially, a Langenskiold's operation (in 4 cases) and fusion of the lateral malleolus to the distal tibial epiphysis (in 1 case) were performed, whereas only skeletal growth was monitored in the other 14 cases. After a mean follow-up of 11 years, the valgus deformity and external tibial torsion of the ankle joint associated with proximal migration of the lateral malleolus needed to be treated with a supramallolar osteotomy in 12 cases (63%). These ankle deformities were evaluated using the serial radiographs and limb length scintigraphs.

Results: In all cases, early closure of the lateral part of the distal tibial physis, upward migration of the lateral malleolus, unstable valgus deformity and external tibial torsion of the ankle joint developed during a mean follow-up of 11 years (range, 5 to 21 years). The mean valgus deformity and external tibial torsion of the ankle at the final follow-up were 15.2° (range, 5° to 35°) and 10° (range, 5° to 12°), respectively. In 12 cases (12/19, 63%), a supramalleolar corrective osteotomy was performed but three children had a recurrence requiring an additional supramalleolar corrective osteotomy 2-4 times.

Conclusions: A valgus deformity and external tibial torsion are inevitable after acquired pseudoarthrosis of the fibula in children. Both Langenskiöld supramalleolar synostosis to prevent these ankle deformities and supramalleolar corrective osteotomy to correct them in children are effective initially. However, both procedures can not maintain the permanent ankle stability during skeletal maturity. Therefore any type of prophylactic surgery should be carried out before epiphyseal closure of the distal tibia occurs, but the possibility of a recurrence of the ankle deformities and the need for final corrective surgery after skeletal maturity should be considered.

Keywords: *Fibula, Pseudarthrosis, Valgus deformity, Ankle, Children*

In 1972, Wiltse[1] reported a gradual valgus deformity of the ankle joint after a fibular resection in children younger than 10 years of age, and Paluska and Blount[2] reported

the upward migration of the lateral malleolus after a resection of the fibula for bone grafts in children. Hsu and associates[3,4] and Moon et al.[5] reported that a fibular defect in children retards normal growth of the ankle joint, resulting in a valgus ankle deformity that is caused by the upward migration of the distal fibula and the early secondary closure of the lateral part of distal tibial physis through the concentration of body weight. Langenskiold[6] advocated a distal tibiofibular synostosis as a method to

stabilize and avoid the progressive ankle valgus. However, there is paucity of periodic long-term follow-up results of acquired fibular segmental defects and Langenskiold's operation in children.

This study reviewed retrospectively the effect of acquired fibular segmental defect on the development of the ankle joint in children and reviewed the literature on the mechanism of upward migration of the fibula, valgus deformity and external tibial torsion of the ankle joint. The long term results of the Langenskiold's metaphyseal synostosis between the distal tibia and fibula are also reported.

METHODS

Nineteen children with acquired fibular pseudoarthrosis were treated from 1980 to 2006. Their mean age was 6 years (range, 1 to 15 years) and the male to female ratio

was 14 to 5 (Table 1).

Fibular pseudoarthrosis was caused by a fibulectomy for a tumor mass in 10 cases, acute or chronic osteomyelitis in 6 cases and a compound fibular shaft fracture with segmental bone loss in 3 cases. In all cases, the distal physis of the tibia and fibula appeared intact in the initial radiographs. Initially, to prevent further ankle deformities, a Langenskiold's operation and fusion of the lateral malleolus to the distal tibial epiphysis were performed in 4 (case 1) and 1 case (case 2), respectively. Ankle development of the other 14 cases was observed during skeletal growth. All cases showed variable degrees of valgus deformity and external tibial torsion of the ankle during skeletal growth and 12 cases (12/19, 63%) underwent a supramalleolar corrective osteotomy to correct these deformities. The remaining 7 cases (7/19, 37%) still had mild ankle deformities and are currently under observation (case 3). The severity of the valgus deformity,

Table 1. Clinical Data of the Patients

Case	Age/Sex	Cause of pseudoarthrosis	Initial management	Follow-up (yr)	SCO	Amount of correction at SCO (°)	Valgus deformity at last follow-up (°)
1	1/F	OM	Observation	6	-		20
2	2/F	OM	Fusion	15	O	25 (age15)	10
3	3/M	OM	Langenskiold + SCO	21	O	23 (age18)	7
4	3/F	Osteofibrous dysplasia	Observation	9	-		25
5	2/F	OM	Observation	15	O	25 (age16)	8
6	5/M	Graft	Langenskiold	11	O	21 (age15)	9
7	6/M	Trauma	Observation	8	-		35
8	7/M	Graft	Observation	12	O	25 (age14)	17
9	9/M	Osteochondroma	Observation	13	-		22
10	6/M	Fibrous dysplasia	Observation	15	O	23 (age11)	19
11	7/M	Graft	Observation	10	O	25 (age12)	17
12	8/F	OM	Observation	11	O	22 (age16)	5
13	8/M	Graft	Observation	12	O	20 (age16)	12
14	7/M	Graft	Observation	11	O	25 (age15)	11
15	8/M	Graft	Observation	10	O	21 (age14)	15
16	5/M	Graft	Observation	11	O	20 (age13)	14
17	8/M	Trauma	Langenskiold	7	-		18
18	4/M	OM	Langenskiold	8	-		17
19	15/M	Trauma	Observation	5	-		7

SCO: Secondary chronic osteomyelitis, OM: Osteomyelitis.

181

Kang et al. Ankle Deformity Secondary to Acquired Fibular Segmental Defect in Children
Clinics in Orthopedic Surgery • Vol. 2, No. 3, 2010 • www.ecios.org

external tibial torsion and upward migration of the lateral malleolus was measured using the serial radiographs and the patients were followed up for an average of 11 years (range, 5 to 21 years). The valgus ankle, as an angle formed by a line crossing at the axis of the tibia and the tibial ankle surface, and the external tibial torsion by the thigh-foot angle were measured.

RESULTS

All cases showed a triangular shape of the distal tibial epiphysis, upward migration of the lateral malleolus, valgus deformity and external tibial torsion of the ankle joint. The mean valgus deformity and external tibial torsion of the ankle at the final follow-up were 15.2° (range, 50° to 350°) and 10° (range, 50° to 120°), respectively. In 12 cases (12/19, 63%), including 2 (40%) out of 5 cases

(initial Langenskiold & fusion) and 10 (70%) out of 14 cases (observation), the patients showed a severe valgus deformity (> 200) and ankle disability (pain, instability, weakness, limping, etc.), as well as a gradual progression of the deformity on the serial radiographs. A supramalleolar corrective osteotomy was performed to correct these deformities. However, three children had a recurrence requiring another supramalleolar corrective osteotomy for an average of 2.6 times (range, 2 to 4 times). The other children showed also valgus and external tibial torsional deformities that were mild and acceptable.

Case 1
A 24-year-old man underwent saucerization for chronic osteomyelitis of the fibula when he was 3 years old, which resulted in extensive segmental loss of the fibular shaft (Fig. 1A). He presented with a gradual valgus ankle deformity,
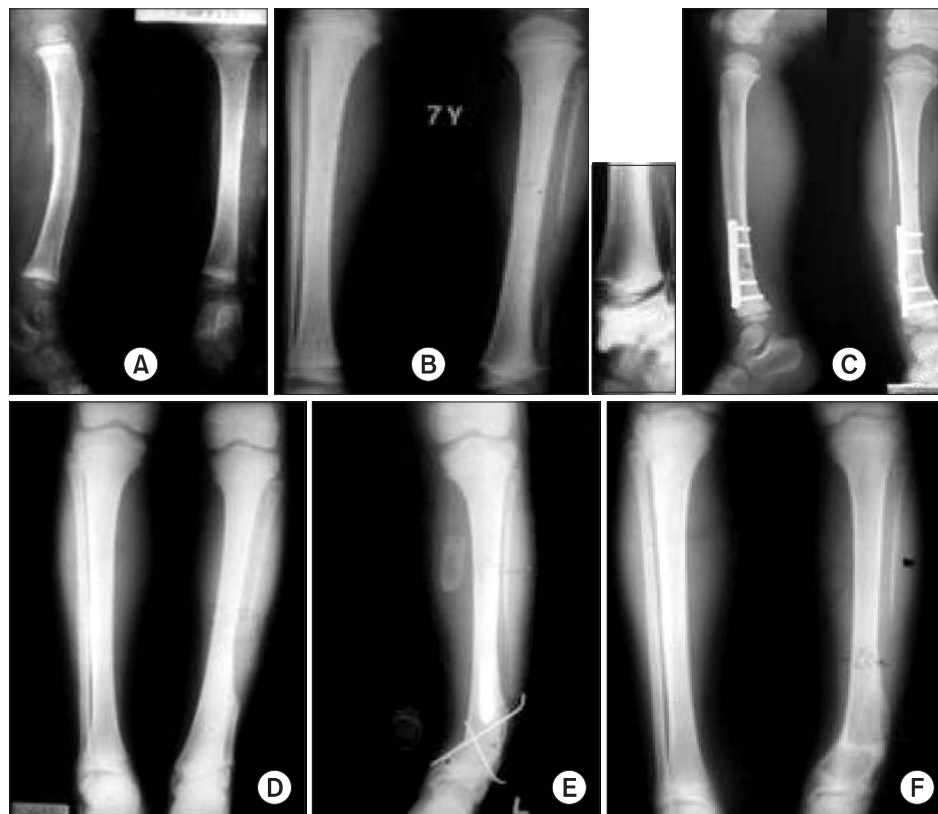

Fig. 1. (A) Radiographs of the lower leg at 3 years of age show a large segmental fibular defect by chronic osteomyelitis. (B, C) Four years later, serial radiographs show a valgus deformity of the ankle due to upward migration of the distal fibula and partial epiphyseal closure of the lateral part of the distal tibia, and a supramalleolar corrective osteotomy and Langenskiold's distal tibio-fibular synostosis was performed to correct the deformity. (D) At age 18, 11 years after the Langenskiold's operation, the radiographs show solid fusion between the distal tibia and fibula, but the valgus deformity had progressed by 23° and his leg was shortened by 1.2 cm. (E) Second supramalleolar corrective osteotomy: 8° of varus ankle position is shown to prevent the recurrence of the valgus deformity during skeletal growth. (F) At age 24, 6 years after the second supramalleolar osteotomy, the joint line between the ankle and knee was relatively parallel but the limb length discrepancy remained.

182

Kang et al. Ankle Deformity Secondary to Acquired Fibular Segmental Defect in Children
Clinics in Orthopedic Surgery • Vol. 2, No. 3, 2010 • www.ecios.org

some instability and a limping gait. The radiographs demonstrated upward migration of the lateral malleolus and partial growth arrest on the lateral part of the distal tibial physis. A supramalleolar corrective osteotomy and Langenskiold's tibiofibular synostosis had been performed just below the pseudoarthrosis when he was 7 year old (Fig. 1B and 1C). His ankle initially appeared fine without a gross deformity, but the valgus ankle had progressed. At age 18, 11 years after the Langenskiold's operation, the valgus deformity had increased by 23° and his leg had shortened by 1.2 cm (Fig. 1D). A second supramalleolar corrective osteotomy was performed to correct the valgus ankle deformity (Fig. 1E). At age 24, 6 years after the second supramalleolar osteotomy, the last follow-up radiograph showed a parallel joint line between the ankle and knee joint but the limb length difference remained (Fig. 1F).

Case 2

A 17-year-old girl suffered from chronic fibular osteomyelitis when she was 2 years old, which resulted in extensive loss of the fibular shaft (Fig. 2A). The remaining small lateral malleolus was fixed immediately to the distal tibial

epiphysis to maintain ankle stability. One year later, a 15° valgus deformity of the ankle developed but the shape with stability were ostensibly good (Fig. 2B). No further treatments were attempted. However, the valgus ankle deformity increased to 25° fourteen years later at age 16 (Fig. 2C). She complained of occasional dull pain around the ankle joint after standing or walking. Therefore, she underwent a supramalleolar corrective varus osteotomy and is currently under observation (Fig. 2D).

Case 3

A 12-year-old girl was treated with segmental fibulectomy at age 3 for multiloculated osteofibrous dysplasia with anterior tibial bowing. Four years later, the radiograph showed pseudoarthrosis of the fibula with upward

Fig. 2. (A) Radiographs of the lower leg of a 2-year-old female show a large segmental fibular loss and the small remaining lateral malleolar epiphysis by chronic osteomyelitis. (B) One year later at age 3, radiographs show a mild valgus deformity (15°) of the ankle, but there is a relatively good ankle shape with stability. (C) Fourteen years later at age 16, the valgus ankle deformity was increased to 25°. (D) Radiographs of the supramalleolar corrective osteotomy.

Fig. 3. (A) A 3-year-old female was treated with a segmental fibulectomy by the multiloculated osteofibrous dysplasia on the tibia and fibula. Four years later, the radiographs show pseudoarthrosis of the fibula with anterior tibial bowing and upward migration of the lateral malleolus. (B) Two years later at age 9, the radiographs show gradual upward migration of the distal fibula and early closure of the lateral part of the distal tibial epiphysis, resulting in a valgus ankle deformity. (C) Two years later at age 11, valgus and external tibial torsion is progressing. (D) One year later at age 12, the last follow-up radiographs show more increased valgus ankle deformity of 25° and almost closed thelateral physis of the distal tibia.

183

Kang et al. Ankle Deformity Secondary to Acquired Fibular Segmental Defect in Children
Clinics in Orthopedic Surgery • Vol. 2, No. 3, 2010 • www.ecios.org

migration of the lateral malleolus (Fig. 3A). Early closure of the lateral part of the distal tibial epiphysis and valgus ankle deformities were observed at age 9 (Fig. 3B), which were still progressing at age 11 (Fig. 3C). At age 12, the last follow-up radiographs (post-operative 9 years) showed a further increase in the valgus ankle deformity of 25° and additional surgery is planned (Fig. 3D).

DISCUSSION

The fibula in humans is longer than the tibia and contributes partially to weight-bearing. In 1971, Lambert[7] using cadaveric ankles, reported that one-sixth of the static load of the leg was carried by the fibula. This force was generated by the articulation of the fibula with the talus and possibly by the inferior tibiofibular ligaments, and was applied to the proximal tibiofibular joint. A small load was transmitted by the interosseous membrane between the tibia and fibula, which only prevented bowing of the fibula. Embryologically, the ankle joint in newborns has a mild varus alignment and becomes parallel to the knee joint after 12. In adults, the fibula is responsible for approximately one sixth of the weight-bearing, and the lateral malleolar tip is located approximately 2 cm posteroinferior to the medial malleolar tip, and is mobile up and downward in the stance phase.[8] These anatomical characteristics of the fibula contribute to the ankle deformities that develop in the fibular segmental defect in this study.

Shortening of the fibula in children due to acquired segmental loss by trauma, osteomyelitis or congenital pseudoarthrosis results in upward migration of the distal fibula and valgus deformity with some instability.[9] Hsu and coworkers[3,4] reported that these phenomena are due to retarded longitudinal growth of the fibula through the loss of the physiological lateral thrust of the proximal fibula to the distal fibular physis, which leads to an upward migration of the fibula, lateral tibial epiphyseal wedging and valgus orientation of the ankle joint line.[1,10] It is believed that the tethering effect of the inelastic soft tissue contracture around the fibular segmental defect and the deranged interosseous membrane can also be another factor, particularly in patients with osteomyelitis. The degree of valgus deformity depends on the onset of the injury and the amount of skeletal growth remaining in the future. The valgus deformity should increase due to the loss of normal physiological loading from the fibular head to distal physis, and the upward dragging of the remaining distal fibula by the inelastic contracted soft tissue scar around the fibular segmental defect (tethering effect).
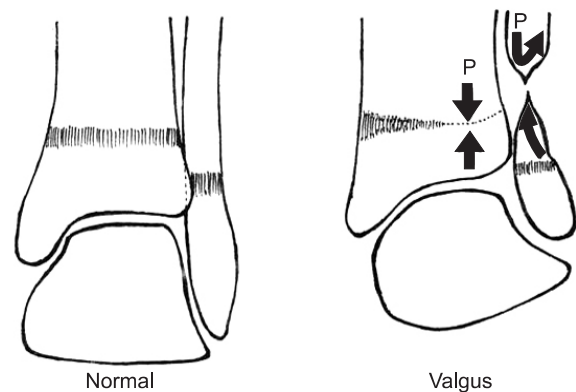

Fig. 4. Proposed mechanism for gradual development of the valgus ankle deformity due to acquired fibular pseudoarthrosis in growing children. In a normal ankle, even axial loading was applied to the physis of the distal tibia and fibula, and harmonious growth occurred. On the other hand, in pseudoarthrosis, the valgus deformity increased due to the loss of a normal physiological load (pressure) from the proximal fibula to the distal physis, and the upward dragging of the remaining distal fibula by the inelastic contracted soft tissue scar around the pseudoarthrosis (tethering effect), and asymmetrical suppression of epiphyseal growth. P: Pressure.

Moreover, the epiphyseal growth at the lateral distal tibial epiphysis is inhibited by eccentrical axial loading (Fig. 4). In Fig. 3, distal tibial epiphysis has valgus ankle alignment in a triangular shape for all due to asymmetrical suppression of the epiphyseal growth.[11] This is known as Heuter-Volkmann's law and Wolff's law for epiphyseal cartilage adaptation to excessive mechanical forces on the growth plate of the distal tibia.[9,12] In 1952, Strobino et al.[13] also reported that an excessive compression force over a physiological stimulus retards epiphyseal growth. Obtaining solid bony union between the tibia and fibula in acquired or congenital fibular pseudoarthrosis is difficult, and until now, the Langenskiold's operation (single leg bone procedure with distal tibio-fibular synostosis) has been the treatment of choice.[6,14] Cho et al.[15] reported good end results of Langenskiold's operation for congenital pseudoarthrosis of the fibula, but they did not follow-up the patients until skeletal maturity.[11,13] In the present study, various amounts of valgus deformity recurred during skeletal growth even after the Langenskiold's operation, which necessitated a secondary corrective osteotomy in several cases (Cases 1 and 2). The Langenskiold's method provided a good lateral strut initially but could not prevent the gradual upward migration of the remaining distal fibula and valgus deformity during skeletal growth. Therefore, the successful Langenskiold's operation cannot provide a proper growth stimulus to the remaining distal

184

Kang et al. Ankle Deformity Secondary to Acquired Fibular Segmental Defect in Children
Clinics in Orthopedic Surgery • Vol. 2, No. 3, 2010 • www.ecios.org

fibular physis. There are two surgical procedures besides Langenskiold's method, fibular osteosynthesis and distal tibial epiphysiodesis, but they were not carried out in this study. Fibular osteosynthesis[12] is indicated when 1) the ankle alignment is neutral, 2) the distal fibular fragment is large and long enough to be internally fixed, 3) the gap at the pseudoarthrosis can be approximated or filled with a bone graft at the time of internal fixation. Distal tibial epiphysiodesis[16,17] can be carried out on patients considerable growth potential, which can induce leg length discrepancy afterwards. However, distal tibial epiphysiodesis can cure a simple shortening problem more simply than a recurrent angular deformity problem. This study performed a long term follow-up of Langenskiold's operation even if there were a small number of cases. In 2 (40%) out 5 cases (Langenskiold's operation) and 10 (71%) out 14 cases (initially observation), supramalleolar corrective osteotomy was performed because of the gradual deterioration of the ankle joint deformities.

For an established valgus deformity of the ankle joint in children, the Langenskiold's operation or supramalleolar corrective osteotomy should be performed at the maximally angulated point as early as possible to maintain a normal functioning ankle mortise and prevent a recurrence of the deformity due to a late osseous deformity of the distal tibial and fibular epiphysis and talus. In addition, the possibility of a recurrence of the ankle deformities until skeletal maturity should be considered.

The lateral malleolus plays an important role in normal ankle growth in children. In acquired pseudoarthrosis in children, the valgus deformity and upward migration of the lateral mallolus are inevitable. Therefore, the Langenskiold's operation should be considered immediately, and a long term follow-up is essential for the early detection of ankle deformities during skeletal growth. Fibular osteosynthesis and distal tibial epiphysiodesis should be considered. However, further study on the two surgical procedures, such as a long term follow-up and a comparison, is needed.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

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