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Original Article

Progressive valgus deformity of the donor-site ankle after extraperiosteal harvesting the fibular shaft in children. Treatment with osteotomy and synostosis at one session

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ABSTRACT

After extraperiosteal harvesting of the fibular shaft in children, progressive valgus deformity of the donor-site ankle may pose a serious problem. We present three illustrative case-histories: three children became functionally impaired and required surgery. Pathogenesis, natural history and surgical options are discussed. A supramalleolar osteotomy combined with a distal fibular-tibial synostosis is a good option to correct valgus and eliminate instability in one surgical session.

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1. Introduction

In children a valgus tilting of the joint line of the ankle is frequently seen as a consequence of harvesting the fibular shaft for revascularized transposition.^{1–3}

As the preparation of such a graft has to be extraperiosteal, the shaft does not regenerate and an atrophic pseudarthrosis will persist at the harvest site. Thus, the remaining lateral malleolus lacks support by continuity. In consequence children are prone to develop gradually on weight bearing a symptomatic hind foot valgus deformity with typical lateral

wedging of the tibial epiphysis and shortening of the lateral malleolus. This can become symptomatic and may cause arthritic changes in the ankle joint.

We present three case-histories of children with such a valgus deformity of the ankle. The children became functionally impaired and were treated surgically: two of them with a supramalleolar osteotomy combined in one session with a distal fibular-tibial synostosis and one adolescent with only a definitive synostosis. The combined surgical technique is described.

Pathogenesis, natural history and surgical options are discussed.

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2. Case 1

This boy, known with type 1 neurofibromatosis, was seen for regular checkups after a successful revascularized transposition of the left fibular shaft for reconstruction of a congenital tibial pseudarthrosis on the right at the age of six years. At the time of harvesting no surgical measures were taken to prevent valgus deformity of the ankle joint. Two years later some heel valgus was still easily corrected into varus through on-toe-standing. A standing antero-posterior (AP) radiogram already showed a joint line in 15° valgus; some lateral wedging of the distal tibial epiphysis and some lateral shift of the talar dome; shortening of the lateral malleolus and inward tilting of the remnant distal fibula; in the lateral part of the distal tibia trabecular and cortical hypertrophy is evident

while the bone density in the remnant fibula is less (Fig. 1a). However any discomfort was denied, orthotic footwear was discarded off.

Six years after harvesting the fibular shaft he presented with some pain and a feeling of instability on weight bearing. On-toe-standing did elevate the medial foot arch well but the heel valgus was not corrected at all. The lateral malleolus was proximal to the medial one; the remnant distal fibula could be tilted somewhat in the frontal and sagittal planes as the ankle mortise allowed for lateral translation. Range of motion in the ankle, subtalar and Chopart joints was normal. A standing AP ankle radiogram revealed progression of the deformity with a joint line in 27° valgus, upward sloping of the lateral part of the growth plate and evident lateral shift of the talar dome (Fig. 1b).

Fig. 1 – a. Standing AP ankle view of an 8-year-old boy, 2 years after a free fibular graft showing valgus tilt, lateral wedging of the tibial epiphyses, lateral shift of the talar dome, shortening and inward tilting of the lateral malleolus. **b.** Standing AP view 6 years after harvesting the fibular shaft shows progression of valgus tilt, lateral wedging of the tibial epiphyses and lateral shift of the talar dome. **c.** Intraoperative AP view showing correction of the valgus deformity by a supramalleolar closed wedge osteotomy and fibula-tibial synostosis. **d.** Standing AP view 6 years after correction of the valgus deformity shows no relevant relapse into a valgus deformity.

Surgical correction was obtained combining a supramalleolar closed wedge osteotomy with a definitive fibulotibial synostosis. At surgery the remnant fibula proved to be stable in vertical direction, aligned well after the osteotomy and showed no relevant tilting mobility anymore (Fig. 1c). Thus packing with cancellous bone was sufficient to achieve solid healing within 12 weeks. Afterwards on weight bearing there was no discomfort anymore.

At the end of growth, six years after correction of the valgus deformity, no relevant relapse into deformity was seen, the ankle joint was clinically and radiologically stable without any signs of osteoarthritis (Fig. 1d).

3. Case 2

A 9-year-old boy was referred for a symptomatic hind foot valgus and flexible flatfoot on the left, four years after harvesting the fibular shaft for revascularized reconstruction of a radical resection site of an osteosarcoma of the ipsilateral proximal humerus. At the time of harvesting nothing was done to prevent future valgus of the ankle joint. In spite of orthotic footwear he experienced discomfort, particularly a feeling of instability in his ankle on weight bearing. On-toe-standing elevated the medial arch well and corrected the heel valgus just to neutral. The lateral malleolus was at level with the medial one but was rather mobile and even tilted on peroneal contraction with an unstable ankle mortise as a result. Other physical findings were normal. A standing AP ankle radiogram showed a joint line in 17° valgus; lateral wedging of the distal tibial epiphysis; shortening of the lateral malleolus and inward tilting of the remnant fibula. A lateral shift of the talar dome was not visualized (the ankle mortise was not well positioned). Bony hypertrophy was less apparent but significant loss of bone density in the remnant fibula catches the eye (Fig. 2a).

At surgery the remnant fibula indeed appeared to be very mobile and tended to proximalize on closing the medial wedge. Hence it was fixed in 'distal' position with a proximal setscrew and a distal lag screw (Fig. 2b). Poor quality of bone required the use of a small tubular plate instead of washers. The interosseous space was packed with cancellous bone. Healing was uneventful with complete remodelling. Discomfort on weight bearing disappeared. Remineralization of the remnant distal fibula is remarkable (Fig. 2c).

At the age of fifteen years, 5 years after surgical correction of the valgus deformity, he was still without any complaint. At physical examination the position of the hind foot appeared almost normal (Fig. 2d). On-toe-standing did correct the heel into varus. But the position of the lateral malleolus was proximal to the medial one and in the ankle mortise some lateral translation was possible. A standing AP ankle radiogram did show a relapse into valgus; progressive shortening of the lateral malleolus; a lateral position of the talar dome in a widened ankle mortise and a tendency to upward sloping of the lateral tibial growth plate; there are no signs of osteoarthritis. On the sound side the tibial growth plate starts closing medially, indicating the end of growth is nearing (Fig. 2e). In future a definitive supramalleolar correction may be necessary.

4. Case 3

A 14-year-old adolescent was referred for a feeling of instability in his left ankle on weight bearing, three years after harvesting the fibular shaft for revascularized reconstruction of a radical resection site of an Ewing sarcoma of the ipsilateral femoral shaft. In contrast to the previous two cases in this case the ankle mortise was temporary fixed with a supramalleolar setscrew at the time of harvesting (Fig. 3a). Progressive weight bearing with crutches was allowed after 12 months.

Physical examination did not reveal any significant heel valgus. The medial foot arch appeared normal. The lateral malleolus was at a normal level, a broken screw was palpable in the subcutaneous tissue beneath the scar. The remnant distal fibula could be tilted somewhat, particularly lateral translation in the ankle mortise was marked. On-toe-standing corrected the heel even into varus, subtalar mobility was free. Other physical findings were normal.

A standing AP ankle radiogram showed a joint line in 7° valgus and minimal wedging as compared to the other side (3° valgus); almost unaltered position of the lateral malleolus and no significant lateral shift; some inward tilting of the remnant distal fibula, less bone density and a broken setscrew. Note the considerable hypertrophy of the lateral tibial cortex (Fig. 3b).

As the valgus was minor and the distal tibial growth plate already closing, there was no need for supramalleolar correction. Thus, at surgery an autologous corticocancellous graft was fitted into the supramalleolar interosseous space and the ankle mortise was fixed as in case 2. The feeling of instability on weight bearing disappeared after solid healing.

At the end of growth, two years after creating a tibiofibular synostosis, there was neither discomfort nor any increase of valgus. Remodelling was complete, bone density restored and the ankle mortise deepened (Fig. 3c).

5. Surgical technique

5.1. Preoperative assessment

Standing radiograms, AP and lateral ankle plus foot, are required to measure the degree of valgus tilt and assess the width of the ankle mortise, the position and quality of the remnant distal fibula as well as the tarso-metatarsal relations.

5.2. Approach

The metaphysis of the remnant distal fibula is exposed in front subperiosteally through a straight lateral approach – at first the level of the growth plate is verified with the image intensifier as the perichondral ring should not be damaged. Following the interosseous membrane, the tibial metaphysis is exposed and the bony surface roughened. The remnant distal fibula is more or less mobile: it can be tilted in the frontal and sagittal plane, some vertical mobility is possible too. The metaphysis of the distal tibia is exposed through a curved anteromedial approach subperiosteally in a similar way after the level of the growth plate is identified.

Fig. 2 – a. Standing AP ankle view of a 9-year-old boy, 4 years after harvesting the fibular shaft showing valgus tilt, lateral wedging of the tibial epiphyses, shortening and inward tilting of the lateral malleolus. Note demineralization in the remnant distal fibula. **b.** Intraoperative AP view showing correction of the valgus deformity by a medial closed wedge osteotomy and fibula-tibial synostosis. **c.** A standing AP view one year after surgical correction shows remarkable remineralization of the remnant distal fibula. **d.** Posterior view of the ankles 5 years after surgical correction of the valgus deformity. The position of the hind foot appeared almost normal. **e.** Standing AP view 5 years after correction of the valgus deformity reveals a relapse of the ankle into valgus. Note the shortening of the lateral malleolus, lateral position of the talar dome in a widened ankle mortise and upward sloping of the lateral tibial growth plate. The tibial growth plate starts closing medially, indicating the end of growth.

5.3. Medial closed wedge osteotomy

The level and size of the wedge osteotomy is determined according to the preoperative planning and marked with a chisel. The rotation is secured with antero-posterior Kirschner-wires on both sides. Then the osteotomy is performed with an oscillating saw leaving intact the lateral cortex to serve as a hinge. Thereupon the osteotomy is closed and fixed with two staples perpendicular to each other. See Fig. 2b.

5.4. Synostosis

By closing the osteotomy the remnant fibula will be aligned and far less mobile. In case of relevant mobility the ankle

mortise is fixed proximally with a setscrew to maintain the malleolar height and the interosseous distance and distally with a lag screw to close the ankle mortise. If the bone is osteoporotic, instead of washers a small tubular plate should be used. Then the cancellous wedge is crumbled into small particles with which the space between the remnant distal fibula and the tibial metaphysis is packed tightly. See Fig. 2b.

5.5. Postoperative care

A below-knee-cast is applied for two periods of 6 weeks, in the second period full weight bearing is allowed. Healing is checked by radiograms at 6 and 12 weeks. Thereafter no orthotic footwear is needed.

Fig. 3 – a. Standing AP ankle view of an 11-year-old boy treated with a free fibular graft. The ankle mortise was temporary fixed with a supramalleolar setscrew to prevent valgus deformity of the ankle. b. Standing AP ankle view 3 years after surgery shows a joint line valgus, minimal wedging, an almost unaltered position of the lateral malleolus, no significant lateral shift and some inward tilting and less bone density of the remnant distal fibula. Note the hypertrophy of the lateral tibial cortex and the broken setscrew. c. Standing AP view of the tibiofibular synostosis, performed 2 years after primary surgery, shows normal bone density and deepening of the ankle mortise.

6. Discussion

The case-histories of these young children do illustrate and help to understand the typical pathogenesis and natural history of valgus ankle deformity following extraperiosteal harvesting a fibular shaft for revascularized transposition.

Few studies dilate upon this typical donor-site phenomenon. Their number of cases is small and in most of these series some kind of surgical prevention is already introduced at the time of harvesting, thus assuming this phenomenon as a problem with a high incidence and a high risk of functional impairment too.^{4–6} Actually this fact was well-known long before revascularized fibula transposition became accepted as a state of art option for reconstruction of large bone defects.⁷ Any chronic fibular defect with lack of continuity – whatever the origin – may trigger this deformity in children with sufficient growth remaining.

In biostatic models the load carried by the lateral malleolus varies between 7%⁸ and 17%⁹ of the axial load on the ankle in neutral position, increasing in dorsiflexion and decreasing in plantar flexion. The intact fibula transmits this load to the tibia through the syndesmosis, interosseous membrane and proximal fibula-tibial joint. In dynamic studies the position of the lateral malleolus changes continuously during gait as regards rotation and vertical shifting. At dorsiflexion the lateral malleolus follows the increasing width of the anterior talar dome into external rotation. At plantar flexion the fibula glides slightly downward pulled by contraction of the muscles originating (in part) from her surface and at the same time against the tibia by tightening the interosseous membrane, thus deepening the mortise and enhancing stability.^{9,10} This load transmission as well as this subtle interaction between loading

and positioning becomes disturbed in case of a chronic fibular defect.

After extraperiosteal harvesting a fibular shaft the axial loading of the distal remnant is grossly reduced^{8,9} the lateral part of the distal tibia has to take over. According to Volkmann's law this increased lateral loading causes inhibition of appositional growth in the lateral part of the epiphysis as well of longitudinal growth in the lateral part of the physis resulting in a valgus deformity at first with a wedged epiphysis and at last with even an upward slope of the growth plate. And, according to Wolff's law this increased lateral loading will cause trabecular and cortical hypertrophy in the lateral part of the tibial metaphysis and shaft, while the decreased loading of the remnant distal fibula causes loss of bone density. These adaptive phenomena are quite apparent in our cases (Fig. 1b, 2a, 3b). Whether the growth in the distal fibular physis is also altered by the decreased and asymmetric loading, remains uncertain.

At the same time the lack of fibular continuity does eliminate the relative 'fibular descent': as the growth rate of the distal fibula is 5% less than that of the distal tibia and the growth rate of proximal fibula 5% more than that of the proximal tibia normally the lateral malleolus keeps up with the tibial epiphysis through a proximal to distal 'thrust' of the fibula^{11,12} while in case of a chronic fibular defect it will 'shorten' i.e. lag behind. This phenomenon is also clearly illustrated in our cases (Fig. 1b, 2a, 3b).

As deformity progresses the resulting valgus and secondary flatfoot may be compensated into a neutral position through subtalar inversion. This kind of compensation can delay, minimize or even prevent discomfort. It is obvious to explain the discomfort expressed as 'pain' on weight bearing as overloading of the lateral part of the ankle joint but a feeling of 'instability' may be more prominent and seems to be

connected with shifting of the talar dome and tilting of the remnant distal fibula on weight bearing, such as in our cases. Due to lack of fibular continuity the load on the lateral malleolus is transmitted to the distal tibial epiphysis through the syndesmosis and in stead of a slight distal glide at plantar flexion during stance the lateral malleolus will be pushed in a proximal glide by the vertical component of the loading force⁹ whereas the lateral component may result in some lateral tilt and peroneal contraction may bring about even some sagittal tilt. These forces are now restrained just and alone by ligaments - the syndesmosis and the collateral ankle ligaments - and not by bone anymore. Thus, while at the same time valgus deformity progresses, 'shortening' reduces the articular facet of the lateral malleolus and probably 'widens' the ankle mortise, in the long run this extra strain may cause some elongation of the ligaments resulting in increased joint laxity. As the ankle joint becomes less and less constrained, talar shifting and malleolar tilting will become symptomatic with a definite feeling of 'instability'.

Whether the appositional growth of the distal tibial epiphysis is disturbed by the increased traction transmitted by the syndesmosis, remains uncertain.

The younger the child at the origin of the defect, the more growth is still ahead to develop a serious deformity.

Prevention of this donor-site phenomenon through a supramalleolar setscrew at the time of harvesting as suggested in some studies^{2,6,12} is not likely to be successful in the long term as such a screw will not normalize load distribution⁸ nor accelerate distal fibular growth rate and inevitably will fail by fatigue or loosening through the strain of the load transmitted and the unequal growth in the distal lower leg (Fig. 3b).

A definitive supramalleolar synostosis¹³ at the time of harvesting^{1,2,4,14} will most likely improve albeit not normalize load distribution. Even in our cases, years after harvesting, load sharing proves to be considerable as bone density invariably improves and remodeling is seen (Fig. 1d, 2c, 3b). Consequently it will stimulate a more normal growth of the ankle mortise, thus valgus deformity will at least be delayed and fairly less severe⁵ also significant loss of constraint will be prevented – even in the younger child, although studies are few, numbers small and except for one study⁴ follow-up is short.

As the synostosis is created with the ankle joint in slight dorsiflexion and afterwards the lateral malleolus invariably will 'shorten', gait will not be impaired. Furthermore in the long run supramalleolar synostosis seems not to cause any significant problem in adults.¹⁵

With early reconstruction of the large fibular defect we have no experience. In some studies this option is mentioned as a side issue.^{1,3} For sure if continuity is restored, it will prevent any deformity. Although this may be successful in smaller defects,^{7,16} in the larger defects reconstruction is rather difficult with a high failure rate and morbidity, thus not acceptable as a preventive method⁶

Correction of an established symptomatic valgus deformity may be achieved gradually with a temporary hemiepiphysiodesis or at once with a supramalleolar osteotomy.

Temporary hemi-epiphysiodesis is less appropriate, because with a median 0.6° pro month¹⁷ it takes too long a time before sufficient correction is gained and moreover joint laxity causing a feeling of 'instability' will persist.

In such cases a supramalleolar closed wedge osteotomy combined with a distal fibular-tibial synostosis as described above will solve both the problem of valgus and joint laxity in one surgical session by improving load distribution and joint constraint. This is followed by only one period of non-weight bearing. Depending on the growth remaining some overcorrection is advised. Metaphyseal bone and cancellous graft will heal within six weeks and remodel on weight bearing in a below-knee walking cast. As the intervention is quite feasible, morbidity will be low. If pubertal growth causes symptomatic relapse into valgus, a definitive supramalleolar correction may be necessary at or after the end of growth.

On the basis of these facts and considerations on pathogenesis and natural history we recommend to combine a distal synostosis with a supramalleolar osteotomy as the treatment of choice for an established donor-site ankle deformity. Furthermore to prevent or at least delay and minimize such a deformity in children a distal fibular-tibial synostosis done at the time of harvesting seems to be the most appropriate measure, as was recommended earlier in particular by Omokawa et al 1996.⁴

Conflicts of interest

All authors have none to declare.

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