Treatment of severe early-onset Blount’s disease by simultaneous medial hemiplateau elevation and metaphyseal osteotomy using the Ilizarov fixator

Mohamed Othman, Ahmed M Abdelwahab and Mohamed Abdelfattah Sebaei

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Abstract
The purpose of this study was to evaluate the outcome of simultaneous medial tibial plateau (MTP) elevation and metaphyseal osteotomy using Ilizarov fixator in severe early-onset Blount’s disease.

Patients and methods: Between 2012 and 2017, we treated 10 cases (14 limbs) of severe early-onset Blount’s disease by double tibial osteotomy procedure using Ilizarov fixator. Their mean age was 10.3 years (9 – 13). All were Langenskiold’s stages V /VI with MTP-depression > 30°.

Results: The mean follow-up period was 18 months (12 - 32 months). Radiographically; (a) The tibio-femoral angle (TFA) changed from -35° (-25 to -49°), preoperatively to +3.1° (-1 to +9°), postoperatively (P=0.00), and to 0.9 (-5° to + 3°) finally (P=0.06). (b) The medial proximal tibial angle (MPTA) changed from 52° (44 – 63°) preoperatively, to 90° (90 to 95°) postoperatively (P=0.00), and to 87°.8 (83 – 90°) finally (P=0.23). (c) The medial tibial plateau depression angle (MTPDA) changed from 38°(32 – 57) preoperatively, to 6° (0 -8) postoperatively (P=0.00), and to 7° (2 -9°) finally (P=0.21). (d) The mechanical lateral distal femoral angle (mLDFA) was 86°(83 – 88°) and showed no significant change till finally. The outcome according to Schoenecker’s criteria was good in 12 limbs and medium in two. Complications included pin tract infection (10 limbs), temporary foot drop (one limb), and distal osteotomy delayed union (two tibias).

Conclusions: This procedure is reliable in treating severe early-onset Blount disease, in terms of joint congruence, limb axes and limb length discrepancy.

Keywords: Blount disease, double osteotomy, plateau elevation, tibial osteotomy

Introduction
The treatment of early-onset Blount’s disease varies with the age of the patient and the disease stage \(^{[1-2]}\). In the early stages of the disease, the process is reversible and a correction of the mechanical axis by a metaphyseal osteotomy can restore normal physeal growth. This is probably why a single valgus osteotomy before the age of four years is successful in 60% to 75% of patients \(^{[1-3]}\). However, in late-presenting infantile Blount’s disease, the pathology becomes advanced, with a premature medial physeal arrest, due to a postero-medial bony bridge formation, leading to multi-planar abnormalities of the proximal tibia \(^{[3-5]}\). These consist of, intra-articular deformity due to medial plateau depression (MTP); causing joint instability and increasing the overall varus deformity and extra-articular metaphyseal deformities (varus, internal tibial torsion and procurvatum), plus leg length discrepancy (LLD); in unilateral and asymmetrical bilateral cases and distal femoral varus in some cases \(^{[5-8,10]}\). These changes (if left without treatment), may lead to progressive LLD, gait abnormalities and premature osteoarthritis \(^{[6-8]}\). Here, conventional metaphyseal osteotomies, are not enough and recurrence is common. Therefore, a more complex method of deformity correction is required. This involves a comprehensive correction of all pathological elements; through simultaneous or staged medial hemiplateau elevation, correction of tibial metaphyseal deformity, lengthening, with or without fibular epiphysiodesis \(^{[2,6-14]}\).

Historically, 1964, Langenskiold and Riska \(^{[4]}\) were the first to propose elevation of the MTP for late-presenting infantile Blount’s disease. This procedure has since then been reviewed,
assessed, and modified [6–7]. In addition to the MTP elevation, the varus of the tibia is corrected by a valgus tibial osteotomy, either simultaneously or in a separate operation [8–12]. The purpose of this study was to evaluate the outcome of simultaneous medial hemiplateau elevation and metaphyseal osteotomy using the Ilizarov fixator for treatment of severe early-onset Blount’s disease.

Patients and Methods
A prospective study was executed at our institution, between October 2012 and March 2017, on simultaneous medial hemiplateau elevation and metaphyseal osteotomy using the Ilizarov fixator for treatment 10 cases (14 limbs) with severe early-onset Blount’s disease, after authorization by the local Ethical Committee. None of them was lost to follow-up. Six cases were females and four were males. Their mean age at the time of surgery was 10.3 years (range: 9 – 13 years). They were troubled by the cosmetic appearance of the deformity, awkward gait (in-toeing and/or lateral thrust) and exertional knee pain. Three cases had undergone a previous metaphyseal valgus osteotomy, but the deformity recurred. The remaining cases had no previous treatment. All cases were classified as stages V and VI according to Langenskiöld and Risma classification [4]. We included cases with a MTP– depression angle (MTPDA) >30°. We excluded cases less than stage V, that with a MTP slope angle < 30° and cases with other aetiologies. Five cases had bilateral Blount disease. One case had a MTPDA <30° and therefore treated with only metaphyseal valgus osteotomy; hence, this limb was not included. Four cases were treated by the index procedure bilaterally, with an interval of 3 to 4 weeks.

Operative technique
Similar to the techniques described by previous authors [2, 89-11], the principal steps were:

a) Mounting the frame: the preassembled frame consisted of three full tibial rings and 5/8 ring proximally.

b) A 3-cm diaphyseal fibular graft was harvested.

c) MTP elevation osteotomy: using a curved anteromedial tibial incision. A Schanz screw is inserted in the depressed MTP, about 1.5 cm below and parallel to the joint line, to be used for elevation of the MTP. Osteotomy was performed using a curved osteotome under optical control and fluoroscopy, starting from below the metaphyseal beak and moving towards the tibial intercondylar eminence, short of the subchondral bone, without damaging the joint cartilage, which was to be used as hinge. Elevation was performed progressively, using the subchondral Schanz screw, with the aid of a lamina spreader, till the MTP reach the level of the lateral plateau, thus restoring tibial plateau alignment. Elevation was then supported by the excised part of the fibula and by advancing the subchondral medial Schanz screw into the lateral tibial plateau. The fibular graft was cut into two segments and the taller one was inserted in the posterior part of the osteotomy for correction of posteromedial slope and the shorter one anteriorly.

d) The preassembled frame was fixed proximally, about 1.5 cm below the joint line, through the 5/8 ring by 2 olive wires and two half pins. The next ring is fixed to the proximal tibia, between the two osteotomy sites by 1 to 2 half-pins. The 3rd and 4th rings are fixed to the tibia distal to the distal osteotomy. Olive wires were inserted from medial to lateral in the middle ring perpendicular to tibia, to allow laterization of the distal tibial segment during correction.

e) Proximal tibial osteotomy was then performed at the metaphyseal-diaphyseal junction below tibial tuberosity.

f) The internal rotation deformity was corrected acutely. The varus, procurvatum and shortening were corrected gradually, by distraction osteogenesis.

Postoperative care
1. After a 7-day latency period, lengthening was performed for 1 to 2 weeks, followed by angular correction (with additional lengthening, if needed).

2. It was important to maintain knee and ankle ROM within the limitations of the frame, with the aid of a physiotherapist

3. The families and children were taught the basics of physical care and cleanliness of the fixator and its pins.

4. Partial weight bearing with crutches was allowed as the patient can tolerate.

5. After satisfactory correction was obtained, the fixator was locked and left in place until the osteotomy was consolidated. Evidence of consolidation was determined by radiographs. If 3 of 4 cortices were identified, on AP and lateral views then consolidation was considered adequate for removal of fixator [13]. The patient was then placed in a weight-bearing long leg cast for 6 weeks.

Preoperative and postoperative assessment
Clinical assessment
Patients were evaluated preoperatively and postoperatively, using the following clinical parameters: limb alignment, LLD using a tape measure and blocks, the range of knee movement (ROM), knee stability and tibial rotation (by measuring the thigh-foot angle), gait (in-toeing and lateral thrust), pain, activity level and satisfaction of patients/parents [2, 15, 16].

Ragiographic assessment
Patients were evaluated preoperatively and postoperatively (after frame removal and at the final follow-up). Preoperatively, they were assessed for Langenskiöld staging [4], joint congruence, limb alignment and LLD. Postoperatively, they were assessed for correction of components of the 3-D deformity and LLD. We used:

a) Lateral radiographs of knee and proximal tibia to assess anterior bowing (procurvatum).

b) Weight bearing- (AP) long leg radiographs of the lower extremities taken with the patellae forward; to calculate: (1) The femoral shaft – tibial shaft angle (TFA: normally is about 5° of valgus) [9,15]; (2) The medial proximal tibial angle (MPTA: formed by a line connecting both apices of the tibial plateau to the axis of the tibia, (N. = 85 - 90°) [17]; (3) The MTP-depression angle (MTPDA) following Schoenecker et al. [6]; between a line approximating the proximal MTP and a line parallel to the lateral tibial plateau); (4) The mechanical lateral distal femoral angle (mL DFA: formed by a line drawn through and parallel to the distal surface of the femoral condyles and the mechanical axis of the femur [N.=87°], to assess concomitant femoral deformity [17].

Schoenecker’s criteria for assessment of the results [6];

Good: absence of pain, instability or lower limb length discrepancy, normal angle between femoral and tibial mechanical axes (±5);
Fair/medium: occasional pain, moderate instability, 5—10 deviation from theoretical femorotibial angles;

Poor: frequent pain, severe instability, major axis abnormalities and final LLD >3 cm.

Statistical analysis
Results were expressed as means ±SD (standard deviation). The differences between pre- and post-operative data were analyzed by Paired T test. A statistical significance was set at p<0.05

Results
The mean correction time was 6 weeks (4 – 8 weeks) and the mean consolidation time was 6 weeks (range: 4.5 – 9 weeks). The external fixator was removed after a mean of 11.8±3.2 weeks (range: 10–14 weeks). The patients were followed-up postoperatively for a mean of 18 ± 6.2 months (range: 12 - 32 months).

Clinical results: (fig1M &2K)
At the final follow-up, the alignment was neutral to slight varus in 10 / 14 knees, and slight valgus in four. The mean shortening improved from a preoperative value of 3 cm (2.5 – 4cm), after a (1 to 3 cm) preemptive overlengthening, to a final value of 1cm (0 -1.5 cm). There were no restriction of movement of knees. All knees were stable except two knees, which showed a moderate coronal instability. The gait showed a significant improvement. There was notable improvement in pain. All patients returned to a higher activity level. All patients/parents were satisfied with the final outcome.

Radiographic results: (Table 1, Fig. 1 & 2)
a) Both osteotomies healed completely in all patients.
b) Improved radiographic parameters:
1. The tibio-femoral angle (TFA) improved from -35°±5.5 (-25 to -49°), preoperatively to +3.1°±2.8 (-1 to +9°), after frame removal (P=0.00**), and at the final follow-up, it became 0.9±2.7° (-5° to + 3°) (P=0.06).
2. The medial proximal tibial angle (MPTA) improved from 52°±9.5 (44 – 63) preoperatively, to 90°±5.5 (90 to 95) after frame removal (P=0.00**), and at the final follow-up, it became 87.8±6.1 (83 – 90) (P=0.23).
3. The medial tibial plateau depression angle (MTPDA) improved from 38°±10.8 (32 – 57) preoperatively, to 6°±2.6 (0 -8) after frame removal (P=0.00**), and at the final follow-up, it became 7°±2.4 (2-9) (P=0.21).
4. The mechanical lateral distal femoral angle (mLDFA) was 86°±4.5 (83 – 88) and showed no significant change throughout the treatment course.

Table 1: Radiologic results

<table>
<thead>
<tr>
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<th>Preoperative</th>
<th>After frame removal</th>
<th>Final</th>
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<tr>
<td>TFA</td>
<td>-35°±5.5 (-25 to -49°)</td>
<td>+3.1°±2.8 (-1 to +9°)</td>
<td>0.9±2.7° (-5° to + 3°)</td>
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<tr>
<td>MPTA</td>
<td>52°±9.5 (44 – 63)</td>
<td>90°±5.5 (90 to 95)</td>
<td>87.8±6.1 (83 – 90)</td>
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<tr>
<td>MTPDA</td>
<td>38°±10.8 (32 – 57)</td>
<td>6°±2.6 (0 -8)</td>
<td>7°±2.4 (2-9)</td>
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<tr>
<td>mL DFA</td>
<td>86°±4.5 (83 – 88)</td>
<td>NS change</td>
<td>NS change</td>
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[* (-) indicates varus; (+) indicates valgus]; NS: non-significant.
Fig 1: (A) Preoperative clinical photo (B) Standing radiographs showing bilateral Blount disease and preoperative radiographic MTPDA Rt 30° and Lt 18° (C) Long-leg radiographs (D) proposed osteotomy sites (E-H) Fluoroscopic views; (I-K) in external fixator; (L) clinical photo after correction of Rt. Side : (M) after bilateral correction. (N,O) Final radiographs of Rt side
The results according to the Schoenecker’s criteria [6]
The outcome was good in 12 limbs, medium or fair in two, but no poor results.

Postoperative complications
1. Pin tract infection occurred in 10 limbs, but recovered without residuals on antibiotics and local pin care in 8 limbs, plus 1 to 2 pin replacement in 2 limbs.
2. Transient foot drop occurred in one limb, but recovered completely on conservative treatment.
3. Delayed union of the distal osteotomy occurred in two tibias (in two patients). This required compression of the osteotomy site and prolongation of time in frame. Eventually, both became well-united. No cases of deep infection or septic knee arthritis, or significant recurrence of the deformity were encountered in any of the patients.

Discussion
Patients with late-presenting infantile Blount’s disease have severe and resistant deformities [2]. These include combined intra-articular deformity due to MTP depression and extra-articular metaphyseal deformities (varus, internal tibial torsion and procurvatum), and LLD and distal femoral valgus in some cases [6-11]. These changes (if left without treatment), may lead to progressive LLD, gait abnormalities and premature osteoarthritis [10, 11, 18]. Here, metaphyseal valgus osteotomies, do not solve the whole problem and recurrence is common and many patients required multiple tibial valgus osteotomies [4, 6, 13, 15]. Therefore, a more comprehensive correction of all pathological elements is required; through the double tibial osteotomy technique [2, 8-10, 14]. It consists of one osteotomy to elevate the depressed MTP and another osteotomy to correct the rest of the deformity [6, 8, 15]. The elevation of the depressed MTP allows restoration of the joint architecture, corrects part of the overall deformity and improves any medial ligamentous laxity [19].

This study demonstrates: that this combined osteotomy procedure can achieve a satisfactory in most cases. Clinically, there was a remarkable improvement in limb alignment, knee stability, gait and no significant LLD. Radiologically, also, there was a remarkable postoperative improvement of radiographic parameters of joint congruence and limb alignment, without significant change at the final follow-up. The outcome was good in 12 limbs, medium in two, but no poor results according to the Schoenecker’s criteria [6].

These results are consistant with: The reports of many authors using the combined osteotomy procedure for managing similar groups of Blount disease. In the study of van Hyyssteen et al, [8], on 34 knees in 24 children The MTPDA improved from 49˚ (40˚ to 60˚), preoperatively to 26˚ (20˚ to 30˚), postoperatively, which was maintained at follow-up. The mean pre-operative mechanical varus of 30.6˚ (14˚ to 66˚) was corrected to 0˚ to 5˚ of mechanical valgus in 29 knees. In five knees, there was an undercorrection of 2˚ to 5˚ of mechanical varus. In the series Fig 2: (A) Rt severe Bount disease; (B) Preoperative standing radiograph; (C) Long-leg radiograph; (D-F) intraoperative photos of elevation osteotomy; (G-I) while in fixator; (J) Final radiographic correction; (K) final clinical correction
of Bar-On et al. [2], formed of four patients, the mechanical axis was corrected from 23° (13 – 30) preoperatively to 0° in three patients and 6° in one, post-operatively and at the final follow-up, it was 0° in two and 7° varus in two tibias were lengthened by 1 – 4 cm and finally, over-lengthening was diminishing as planned. In a retrospective study executed by McCarthy et al. [9] on 16 patients (22 limbs), The mean TFA improved from 29° varus to 3° of valgus, the mean MPTA improved from 55° to 86° and the mean MTPDA improved from 37° to 12°. However, varus recurrence at the metaphyseal osteotomy occurred in five cases. Fitoussi et al. [10], treated a similar series formed of eight knees (six patients), using an Ilizarov external fixator. After a mean 48 months’ follow-up, the mean hip-knee-ankle (HKA) angle improved from 151, preoperatively to 179.6 postoperatively and the mean MTPDA improved from 42 preoperatively to 5.4 postoperatively. The mean final LLD was 11 mm. The outcome according to Schoenecker’s criteria [6] was good in six cases, medium in one and poor in one. Gkiokas and Brilakis [10], used acute correction of both osteotomies for treating eight children (9 limbs). At the latest follow-up [10 years (range 5–15 years)], there was no observable restriction of knee ROM and no signs of instability or lateral thrust. All patients had returned to a higher activity level. Leg-lengthening surgery was performed in one child, but LLD was already present preoperatively. No other complications were noticed. All the angles measured on X-rays had been corrected, and this correction was retained until the final follow-up. Edwards et al. [13], treated Seven patients (eight limbs) using an external fixator. The MAD improved from 4.85 cm to 1.88 cm, MPTA from 43.58° to 75.46° and MTPDA from 43.41° to 20.71°. Recurrence was noted in three patients.

Tsibidakis et al. [14], treated a similar group of 24 tibias in 16 children using TSF. Post-operative improvement of all measurements was observed. MPTA increased from 71.8°(58°- 79°) to 92.5°(90°- 95°), the TFA changed from 15.4°(10°- 25°) of varus to 5.9°(2°- 10°) of valgus.

### Postoperative complications:
In the current study included: pin tract infection in 10 limbs, temporary foot drop in one limb, and delayed union of the distal osteotomy in two tibias (in two patients). This is comparable to that reported by Hefny et al. [15] and Hegazy et al. [18], but less than that reported by others [9,13-16], (Table 2). Hill [20] suggested that knee stiffness is not a common problem after the index procedure, as the joint is not entered.

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<th>Table 2: Postoperative complications in different studies</th>
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<td><strong>No of knees</strong></td>
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<tr>
<td>Mc Carthy et al [9]</td>
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<td>Fitoussi et al [16]</td>
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<td>Tsibidakis et al [14]</td>
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<td>Edwards et al [13]</td>
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<td>Current study</td>
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The degree of MTP depression that mandates elevation: is controversial. According to McCarthy et al. [9], MTP depression should be corrected when slope exceeds 15°. However, according to Schoenecker et al. [6] and Fitoussi et al [16], it should be corrected when depression exceeds 30°. In the current study, we operated when MTPA was > 30°.

Acute versus gradual correction of deformity is a point of discussion and controversy: [22]. The correction could be done either acutely for both osteotomies—using internal fixation [4, 23], or external fixator [18], acutely in elevation osteotomy and gradually for the metaphyseal one, using external fixator [8, 11]; or gradually for both osteotomies, using external fixator [15]. Gradual correction has many advantages, that, it requires less invasive surgery, allows progressive and adjustable correction, permits bone lengthening if needed and achieves a more accurate correction [15, 22, 24-26]. However, it has the disadvantages of external fixator. [15]. Acute correction, has its proponents. However, many complications had been reported with it; that included peroneal nerve palsy, compartment syndrome, residual deformity, LLD, delayed union, and failure of fixation [14, 18, 27, 18].

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Alternative methods of stabilization: have been reported and they varied according to the method of correction whether acute or gradual, the surgery preference and experience and the need for LLD correction [13]. They include either: (a) A horizontal 1.6 mm Kirschner wire for the proximal osteotomy combined with two crossed 2.4 mm Steinmann pins for the distal osteotomy plus a long-leg cast, for acute correction [9]; (b) A horizontal K- wire for the proximal osteotomy combined with staples for the distal osteotomy [10]; (c) plates and screws as described by Schoenecker et al. [6] used for acute correction; or (d) external fixators, principally used for gradual correction [15], but were used for combined acute and gradual correction [10] and also with acute corrections [19].

External fixators used included monolateral fixation devices, Ilizarov Frame, and the Taylor Spatial Frame (TSF) [10, 18]. The advantages of external fixation are: (a) percutaneous application, without large surgical wounds, and no second surgery for removal of the implants, (b) gradual correction, (c) allow correction of multiplanar deformities and simultaneous leg lengthening (d) allow weight bearing during the period of treatment before full consolidation of bone (e) allow possibility of postoperative adjustment [8, 10, 16, 18]. The disadvantages of external fixation are: they are more expensive, more complicated and time consuming, depend on the cooperation of the patients and parents, associated with a high incidence of pin tract infection, daily care is required to avoid pin-site infection. More specifically, attention must be paid to avoiding premature consolidation of the proximal tibial osteotomy or the fibular osteotomy [8, 10, 18].

Bone grafting in tibial hemi-plateau elevation is controversial. Most authors [2, 8, 9, 14] used bone grafting to support the elevated MTP and enhance healing. This included either, the bony wedge removed from the valgus wedge osteotomy, tricortical iliac graft, the bony fragment removed from the precedent fibula osteotomy, or tricortical iliac allograft [9]. In our series, the bony fragment removed from the precedent fibula osteotomy was used. Others [15, 16] did not use bone grafting as they used rigid external fixator with gradual correction for both osteotomies. The system was sufficiently rigid to maintain elevation. The gap left under the MTP was progressively filled over a period of a few months. However, there is a need to wear the external fixator for longer periods [16].

Recurrence of the varus deformity after the index procedure is a critical concern. It may be due to an incomplete lateral epiphysiodesis [2], or to a mild distal femoral varus [8]. In a trial to avoid the recurrence, either concomitant epiphysiodesis of the lateral part of the proximal tibial physis, or overcorrection to a slight valgus should be performed [8, 11, 12, 28]. Many authors [4, 6, 29] advised that concomitant lateral proximal tibial and fibular epiphysiodesis should be performed when undertaking the double tibial osteotomy technique. However, it is not routinely advised by other authors [6, 8, 15, 30, 31], specifically, when the operation is performed close to skeletal maturity [10]. In the current series, epiphyseodesis was not performed. Instead, we performed an overcorrection. (5–15˚) of valgus; the younger the patient, the greater degree of overcorrection. No significant recurrence occurred in our cases till the final follow-up examination.

The issue of lower limb length, in such cases aroused much discussion in the literature. The aetiology of LLD, if any, is both the growth disturbance and the deformity [2]. In bilateral symmetric cases, LLD is not usually a problem and lengthening is not usually indicated. In unilateral or bilateral asymmetric cases, LLD is significant, especially in younger patients [2, 6, 10]. The expected LLD at the skeletal maturity, should be predicted from bone age and growth curves [2, 16]. Depending on the case, it could be prevented or lessened either by; a simultaneous epiphysiodesis of the contralateral proximal tibia and fibula [2, 9, 10] or an ipsilateral pre-emptive overlengthening according to the expected LLD [2, 11, 12, 16].

The minimum skeletal age for surgery should be seven years for girls and eight years for boys, to avoid marked LLD [9, 10]. In our study, the mean shortening improved from a preoperative value of 3 cm (2.5 – 4 cm), after a (1 to 3 cm) preemptive overlengthening, to a final value of 1 cm (0 -1.5 cm). Schoenecker et al. [6] addressed the problem of femoral deformity in infantile Blount’s disease. Four of the seven knees in their study had sufficient valgus (> 10˚) to warrant a femoral osteotomy, undertaken either before the tibial procedure or simultaneously. None of our patients showed such deviation.

The common peroneal nerve palsy: had been reported with such procedure [9, 20]. Hill [20] proposed that, common peroneal nerve is not particularly at risk during this procedure unless a lot of lengthening is being contemplated; however, the surgeon may wish to decompress the nerve at the start of the procedure. Moreover, the peroneal nerve is at risk at the time of fibular osteotomy and it is recommended to perform the osteotomy through an incision that permits sub-periosteal exposure and osteotomy under direct vision [13]. Transient foot drop occurred in one patient in the current study.

Alternative methods: for managing infantile Blount’s disease exist. Using a valgus metaphyseal osteotomy alone may be suitable for early cases, however, with the more advanced cases, it does not solve the intra-articular problem. Hence, a MTP-elevation should be added [8-10]. Restoration of growth plate function in the more advanced cases has been described by resection of the bony bridge and interposition of fat or silastic [32]. However, the results of these procedures are not always predictable, and in addition, they do not address the existing axis and joint deformity [2]. The limitations of this study include the small number of cases, but evolved Blount’s disease is rare and, a relatively short mean follow-up period.

Conclusions

Although this combined procedure is technically demanding, it gave satisfactory results in terms of restoration of articular surface congruence; correction the multiplanar deformity of the proximal tibia; solving the problem of associated LLD and minimizing or preventing the future recurrence. Therefore, it is a reliable solution for treating late-presenting early-onset Blount disease of stage V or greater.

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