Deformity progression in congenital posteromedial bowing of the tibia: a report of 44 cases.

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Abstract

Background

Congenital posteromedial bowing of the tibia (CPMBT) is an ultra-rare defect present at birth, characterized by shortened bowed leg, and ankle deformity. We described a single institution experience in the management of CPMBT

Methods

We identified 44 CPMBT in 44 children. The age at presentation was 5.5 ± 5.6 years and the mean age at the final review was 10.1 ± 4.8 years. Radiographic evaluation included the antero-posterior and lateral inter-physeal angle (AP-IPA and L-IPA), the limb length discrepancy (LLD), the morphology of the distal tibia and the lateral distal tibial angle (LDTA). During the study period, 26 children underwent surgical treatment.

Results

The estimated curves showed a progressive spontaneous correction of both AP-IPA and L-IPA during growth, but a progressive increase of the LLD. The L-IPA showed a more predictable behaviour while the AP-IPA showed a scattered correction, with a wider variation of the estimated final angle. The final LDTA was 83.4° ± 5.2° and was correlated with the L-IPA. Among the 26 children which underwent surgical treatment, 23 cases had limb lengthening, 1 case had contralateral epiphysiodesis, 1 child underwent tibial osteotomy, 1 patient was treated by hemiepiphysiodesis of the distal tibia to address ankle valgus deformity.

Conclusions

To date, we reported the largest case series of CPMBT. Nevertheless, further studies are needed to understand which is the best strategy to address this ultra-rare deformity during childhood.

Background

Congenital posteromedial bowing of the tibia (CPMBT) is an ultra-rare defect, firstly fully described in 1949 by Heyman and Herndon [1]. It has been generally considered a benign, self-solving condition, in contrast to the anterolateral bowing associated with congenital pseudarthrosis of the tibia, and the anteromedial bowing associated with fibular hemimelia [2].

CPMBT is present at birth with bowed and shortened leg, associated with various degrees of ankle valgus deformity [2–5]. Although the cause of CPMBT is not completely known, a potential role of amniotic strains has been hypothesized [6]. This condition is generally unilateral and not associated with any other abnormality [1,7].

Several authors reported a spontaneous improvement of the bowing of the tibia, within the first 3–4 years of life. Conversely, the limb length discrepancy (LLD) increases with age, until it reaches 4–7 cm at maturity [2,3]. Moreover, the amount of the bowing (especially in the frontal plane) has been related to the shortening [2,3,8]. Finally, a potential residual valgus deformity of the ankle joint has been reported [2], but the incidence and the relationship with the leg deformity have not been established. Currently, many therapeutic options have been proposed for CPMBT, but the treatment of choice remains controversial.

Therefore, the purpose of the present study is to investigate a case series of children affected by CPMBT treated at a single institution. The study aims to explain the behaviour of CPMBT during growth and the relationship between the tibial bowing, the leg shortening and the ankle deformity. These aspects could be useful in order to suggest a possible rationale of treatment.

Methods

Case series

The present study is a retrospective analysis of medical charts and radiographs of children affected by CPMBT, which were admitted at the Department of Pediatric Orthopedics and Traumatology from 1972 to 2016. Our institution is a tertiary referral center for pediatric orthopedics and traumatology, highly specialized in the treatment of complex deformities of the lower limb. All the charts and radiographs were analysed by independent observers, which were not involved in the decision process about treatment and surgical management of the patients.

During the study period, 44 CPMBT in 44 children were identified. There were 27 boys and 17 girls. All children had unilateral involvement and no cases were excluded from the present study. The right side was affected in 25 children whereas the left side was affected in 19. The age at presentation was 5.5 ± 5.6 years (range 0–15) and the mean age at the final review was 10.1 ± 4.8 (range 0–16). 26 patients underwent surgery during the study period, while for the remaining 18 patients the follow-up is still ongoing and surgery not yet planned.

Radiographic evaluation

The following variables were measured on serial sequential radiographs in order to assess the initial deformity and the spontaneous remodeling:

1) The anteroposterior interphyseal angle (AP-IPA) and the lateral interphyseal angle (L-IPA), which are the angles measured between a line perpendicular to the proximal physis and a line perpendicular to the distal physis, on a true anteroposterior and a true lateral view of the leg respectively [2]. Positive AP-IPA indicates medial bowing, while negative AP-IPA indicates lateral bowing; Positive L-IPA indicates posterior bowing, while negative L-IPA indicates anterior bowing;

2) The limb length discrepancy (LLD) measured on long standing radiographs. The difference was expressed as crude length (LLD_{cm}) and as percentage shortening as compared with the opposite side

($LLD_{\%}$);

3) The level of the distal fibular growth plate was graded according to the method of Malhotra et al. [9], while the extent of the wedging of the distal tibial epiphysis was graded according to the method of Shapiro et al. [10].

4) The lateral distal tibial angle (LDTA) measured on long standing radiographs in children approaching the skeletal maturity. [11,12]

Statistical Analysis

Data were entered in Excel, SPSS and nlme package in R. Continuous variables were expressed as mean ± standard deviation (SD), while dichotomous or ordinal variables were expressed as percentages and 95% confidence interval (CI). Exploratory univariable and multivariable analysis was performed to assess the relationship between the parameters of CPMBT.

Normality was tested using the $\chi 2$ test for categorical variables and the Kolmogorov-Smirnov test for continuous variables. The Spearman's Rho correlation test was used to investigate the relationships between continuous variables. The differences between groups were determined using the Fisher exact test for categorical variables and the independent sample t-tests for continuous variables with normal distribution. Variables with skewed distributions (Kolmogorov–Smirnov test, $p < 0.05$) were tested with the Mann– Whitney U-test.

In order to predict the spontaneous progression of both the deformity and the length discrepancy among patients over time, generalized linear mixed models, fitted by the maximum likelihood, were performed (for each parameter a separate model), using the subject ("patient") as a random factor. The best fitting model was chosen using the ANOVA R function, which compares the AIC, BIC and logLik values. A p-value of < 0.05 was considered statistically significant, and all reported p-values are 2-sided.

Results

The estimated curves showed a progressive spontaneous correction of both the AP-IPA and the L-IPA (Fig. 1a-b), and a slight decrease in $LLD_{\%}$ (Fig. 1c), but a progressive increase of the LLD_{cm} (Fig. 1d), until a final estimated discrepancy of 4.3 cm (95% CI 3.7–5.0).

Considering the prediction model of the spontaneous correction, we found that the best fitting curve was log-linear for the AP-IPA and LLD_{cm}, exponential for the L-IPA and linear for the $LLD_{\%}$. Yet, the L-IPA showed a more predictable behaviour while the AP-IPA showed a scattered correction, with a wider variation of the estimated final angle.

We found an almost perfect correlation between AP-IPA and L-IPA (eta-square 0.81; $p < 0.0005$), a substantial correlation between AP-IPA and $LLD_{\%}$ (eta-square 0.69; $p < 0.0005$). With the cases available,

we did not find any relationship between the Malothra score, the Shapiro score and the LDTA. An illustrative case is showed in Fig. 2.

Twenty-six patients (mean age: 12.8 ± 2.7 years; range 5.3–15.7) underwent surgery during the study period.

The first case of CPMBT was diagnosed in our hospital in 1972 and was treated in 1978, at the age of 6 years. The patient underwent tibial lengthening by unilateral Wagner external fixator, then he had plate fixation after two months. The patient further developed delayed union treated with autologous bone graft and a subsequent fracture of the regenerated bone, treated by intra-medullary nailing.

Two patients had intermediate surgical treatment during the study period.

In one patient, a severe tibial deformity persisted at the age of 5.8 years, with an AP-IPA of 26°, a L-IPA of 6° and a LLD 5 cm (18%) (Fig. 3a-c). The patient was treated by corrective tibial osteotomy at the apex of the deformity, stabilized with two Kirschner wires (Fig. 3d). At the latest follow up, 3.5 years after surgery, the patient had a residual deformity with an AP-IPA of 9° a L-IPA of 5° a residual LLD of 5 cm (16%) and an ankle valgus deformity of 70° (Fig. 3e). The patient is waiting for final correction and lengthening of the leg.

One patient had medial unilateral screw hemiepiphysiodesis to treat ankle valgus (LDTA = 79°) at the age of 10.4 years. At the latest follow-up visit, 2 years after the operation, the ankle axis was restored (LDTA = 90°) and the patient is waiting for final lengthening to address a residual LLD of 4 cm (11%).

One patient, which presented with a residual LLD of 2.2 cm (7%) and no relevant deformities of the tibial and ankle axis, was treated at the age of 11.4 years with contralateral epiphysiodesis. The patient achieved limb length equality 30 months after surgery, with no further complications.

Twenty-two patients were treated at skeletal maturity (13.7 ± 1.8 years). The pre-operative radiographic data are summarized in Table 1. All these patients received leg lengthening by circular external fixation. Bifocal lengthening was used in one case (Fig. 4), in which deformity correction was achieved at the distal osteotomy with further lengthening at the proximal osteotomy, to limit time in the frame, due to the better healing potential of a metaphyseal osteotomy. In this group, the final limb equality was achieved in all patients, but we experienced 11 complications in 8 patients. According to Lascombes et al. [13], there were 2 grade IIa complications (2 operations to change or modify the frame) and 9 grade IIIa complications (1 delayed bone healing; 3 fibular nonunion; 1 knee joint stiffness; 2 achilles tendon shortening; 2 malalignment with residual anterior bowing of the tibia). Moreover the final LDTA was 83.3° ± 5.2° (range 72° − 90°), with 9 patients out of 14 showing a residual LDTA < 85°.

Table 1
pre-operative radiographic characteristics of the 22 patients that underwent leg lengthening by Ilizarov external fixation.

Radiographic parameter	Mean ± SD (range)
AP-IPA	6.8 ± 4.6 (0–20)
L-IPA	4.8 ± 3.8 (0–11)
LLD (%)	12.9 ± 3.0 (9–22)
LLD (cm)	4.6 ± 0.9 (3–6.2)
LDTA	86.1 ± 3.0 (78–90)

Discussion

To the best of our knowledge, this study describes the largest series of children with CPMBT. To date, about 200 cases of CPMBT have been reported in the available literature [1–5,7,8,14–18]. For years, CPMBT has been considered a benign self-solving condition, due to the virtually absent risk of fracture or pseudarthrosis and the natural tendency to the spontaneous resolution, with minimal residual deformity. Nonetheless, we noticed that the spontaneous correction of the bowing is sometimes incomplete, the LLD is frequently wide and a residual ankle valgus may persist at the end of growth. We found some interesting correlation between the AP-IPA and the L-IPA, and between the AP-IPA and the LLD%. In other words, the greater is the angular deformity at birth, the wider will be the LLD at skeletal maturity. This finding is confirmed by previous reports [2,3,8,18].

The treatment of CPMBT is still controversial. Given the tendency to the spontaneous correction of the deformity, some authors suggest conservative treatment consisting of manipulation, serial casting, orthoses and shoe lifts; then, a limb equalization is proposed during late childhood, if needed [4,7,14]. Currently, there is no evidence that the use of braces and orthoses may improve the angular correction in CPMBT, since it occurs spontaneously and independently by the use of orthoses. The main goal of braces and orthoses is to aid walking and balance, while the patient is too young for the surgical treatment.

There are three reasons for surgical intervention in CPMBT: equalize the LLD, correct the ankle valgus and correct the residual bowing of the tibia [2]. Nonetheless, there are no clearly defined guidelines for the surgical treatment of CPMBT.

Regarding the LLD, we found that the final discrepancy at skeletal maturity averaged 4.3 cm, corresponding to 13% of the length of the unaffected leg; this percentage, remains rather constant during growth, as confirmed by the majority of the previous reports [2–5,18]. In our series, all the children who reached skeletal maturity underwent limb lengthening by circular external fixator while only one child underwent contralateral epiphysiodesis. Whether contralateral epiphysiodesis has been recommended in

CPMBT, due to the lower risk of complications [3], compared to limb lengthening, aesthetical issues can raise due to the loss of body height. Moreover, recent concern has mounted regarding the potential risk of compromising the morphology of the proximal tibia, when a large, congenital LLD must be addressed [19,20]. Therefore, we suggest to reserve this treatment only for children in which the LLD% at 10–12 years is less than 10%, corresponding about 2 to 3 cm. Regarding the tibial bowing, the majority of cases improved spontaneously by the end of growth. Our behaviour consisted in a “waiting strategy”, using braces and orthoses until skeletal maturity, then, correcting in a single stage the length and the potential residual bowing. In our opinion, this strategy should reduce the risks for the patient and the costs for the health service. Nonetheless, a more pronounced reduction of the angular deformity was noticed during the first six years of life; thereafter, the rate of spontaneous correction showed a marked reduction. Other authors reported that, in CPMBT, the greatest rate of correction is observed during the first year of life, and rapidly decreases until the age of four [2–4,15,18]. This aspect may have practical implications, because an extreme bowing of tibia in a school-age child might hamper even the possibility of using braces to aid walking. In this scenario, some authors suggested early corrective osteotomy at the apex of the deformity, by the age of 3 to 6 years [2–4,15,16]. It has been argued that the intense periosteal activity at this age allows for early bone healing of the diaphyseal osteotomy; furthermore, the overgrowth of the tibia due to the physeal stimulation and the tibial straightening could potentially contribute to the leg length equalization [15]. We treated only one case by early corrective osteotomy of the bowed tibia: although we achieved a rapid healing of the osteotomy and a perfect alignment of the tibia, we observed a progressive partial relapse of the bowing, an important ankle valgus, while the LLD remained unchanged. These finding are consistent with those reported by Johari et al. [4], suggesting that the early tibial osteotomy should be proposed only in case of severe, disabling bowing, as an intermediate treatment, to avoid complex bracing and allow walking with simple foot orthosis or shoe lift. Another possible reason to recommend early tibial osteotomy, is given by the possibility to perform intramedullary lengthening by telescopic nails at skeletal maturity [21]. This technique has been reported as safe, effective and more tolerated by the patients, in comparison with external fixation. Nevertheless, the procedure is more simple, safe and effective in a straight, rather than a bowed tibia. Yet, simultaneous correction of the bowing and lengthening by circular external fixation is not simple, requires high compliance by the child and the parents, high proficiency with the technique and it has high risk of complications. In our series, we experienced a relevant rate of moderate and severe complications, in line with other reports: Kaufman et al.[5] reported 17 mild to severe complications in 11 CPMBT treated by external fixation; Johari et al.[4] described complications in all the 6 cases treated by external fixation; Wright et al.[18] reported 16 complications in 17 children treated by external fixation. Moreover, we suggest to perform the lengthening procedures closer to the skeletal maturity, since we did not experience any recurrence of the limb length inequality, in contrast with other authors, which reported recurrence of the limb length inequality, if the lengthening procedure was performed during growth [2–5,18].

Finally, in our series about one third of children with CPMBT presented a valgus ankle by the end of growth (LDTA < 85°). This issue has been reported previously [2,4]. Although the normal range of the LDTA has been established [11,12], an exact cut-off to define a pathologic deformity has not been clearly

defined, with proposed values varying from 5° to 10° of valgus [20–23]. In our series, only one patient underwent distal tibial hemiepiphysiodesis to treat ankle valgus. Nonetheless, it is our opinion that, if at age of 10–11 years, the valgus inclination of the distal tibial articular surface persists, a medial distal tibial hemiepiphysiodesis may be performed, when sufficient growth potential is still present [18–19]. This simple procedure can effectively realign the ankle, minimizing the complications [22–27] and avoiding demanding realignment procedures (double or triple corticotomies, complex external fixation constructs) at the end of growth.

Although our study describes the largest series of CPMBT in the available literature, some limitations must be underlined. We conducted a retrospective analysis of cases collected across more than 40 years, sometimes with incomplete information and missing data; moreover, 12 cases out of 44 had no sufficient radiographic follow-up available for the evaluation of the progression of the deformity. In addition, the cases were not collected uniformly at birth and 16 children had the first radiographic evaluation when they were older than 5 years. Finally, almost a half of cases did not reach the definitive treatment. These issues, were reported in most of the previously published case series describing CPMBT [2–5,18], emphasizing the difficulty to achieve complete information about ultra-rare developmental pathologies, in which at most one case per year can be collected, even in the highly specialized referral institutions. In order to address this issues and minimize biases, we used mixed effect models, a complex statistical approach that allows to maximize the prediction power from small and heterogeneous groups of subjects with missing data; nonetheless, we are aware of potentially biased results as well as a loss of statistical power and precision.

Conclusions

In conclusion, despite its supposed benignity, CPMBT is a complex deformity, in which the spontaneous correction of the angular deformity can be inconstant and incomplete, the LLD is generally wide, and a substantial ankle valgus can be observed by the end of growth. A combination of surgical treatments (osteotomies, epiphysiodesis or hemiepiphysiodesis, leg lengthening) in a staged multistep surgical process, should be tailored on the development characteristic of the deformity; This approach may accomplish the correction of the deformity, minimizing complications and failure, and helping surgeons and parents in the decision process. Further studies are needed to understand which is the best strategy to address this ultra-rare deformity during childhood.

Abbreviations

CPMBT: congenital posteromedial bowing of tibia; AP-IPA: anteroposterior inter-physeal angle; L-IPA: lateral inter-physeal angle; LLD: limb length discrepancy; LDTA: lateral distal tibial angle; SD: standard deviation; 95% CI: 95% confidence interval; ANOVA: Analysis of Variance; AIC: Akaike Information Criterion; BIC: Bayesian Information Criterion.

Declarations

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Author's contribution

Conception and study design, analysis and data interpretation: GLDG, SS, GT. Data acquisition: GG, EAMV, CR. Manuscript drafting and revision: GLDG, EO, LMR, GT. All authors read and approved the final manuscript.

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Availability of data and materials

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Competing interests

The authors declare no conflicts of interest related to any aspects of the presented manuscript.

Ethics approval and consent to participate

Ethics approval was sought and obtained from the IOR Ethical Committee (PG nr. 01-13 12/19/2017). The study was conducted in accordance with the Helsinki declaration and all patients gave informed consent in writing to participate. Parents provided written consent for the inclusion of the patients in this study, since all the patients were minors (age less than 18) at the time of participation in the study.

Consent for publication

Not applicable.

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Figures

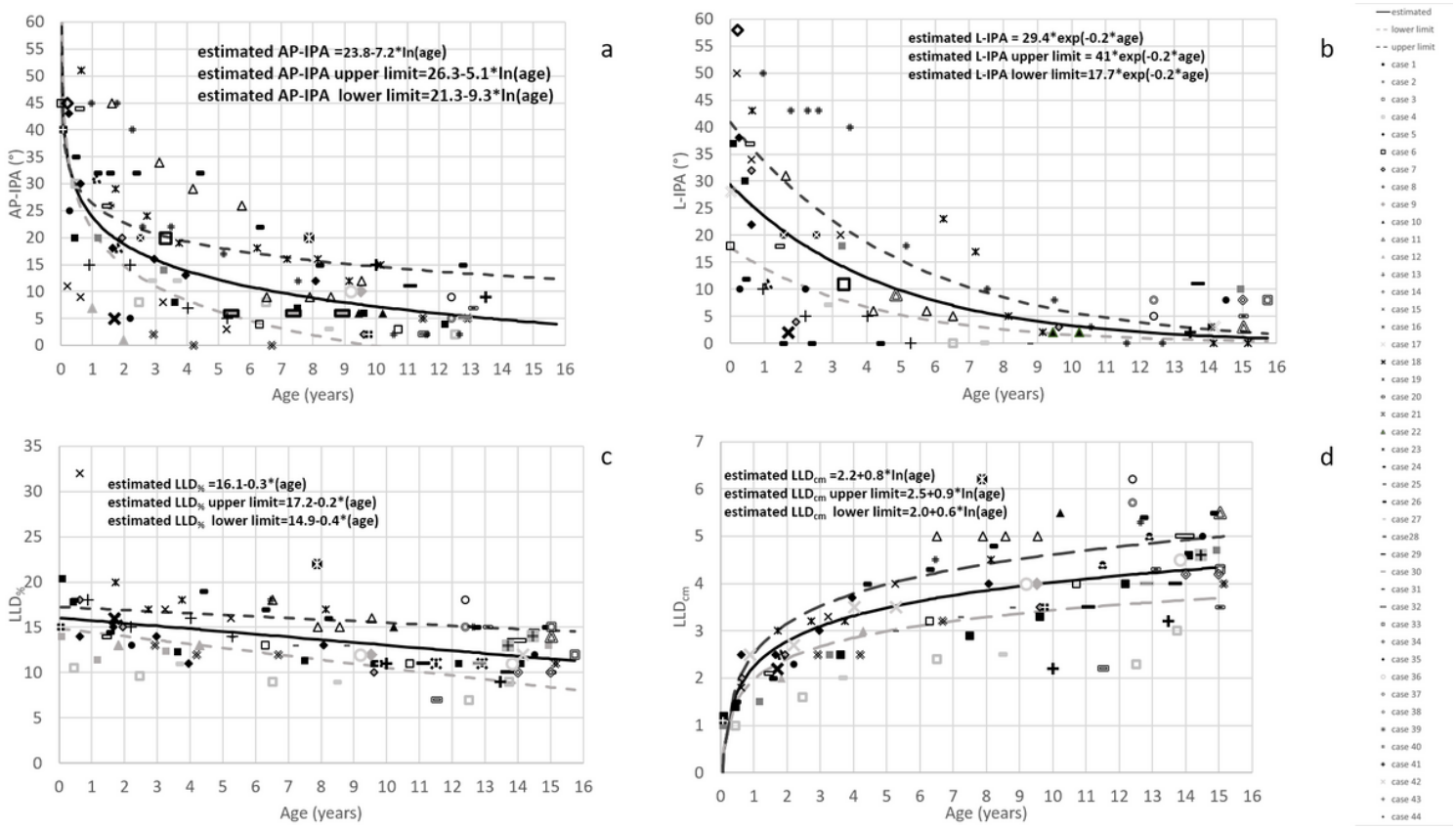

Figure 1

Graph illustrating the estimated (black straight line) spontaneous variation over time and the 95% confidence interval upper (dark grey dashed line) and lower (light grey dashed line) limits of the anteroposterior interphyseal angle (AP-IPA: figure 1a), the lateral interphyseal angle (L-IPA: figure 1b), the limb length discrepancy expressed as percentage shortening, as compared with the opposite side (LLD%: figure 1c), and as crude length (LLDcm: figure 1d).

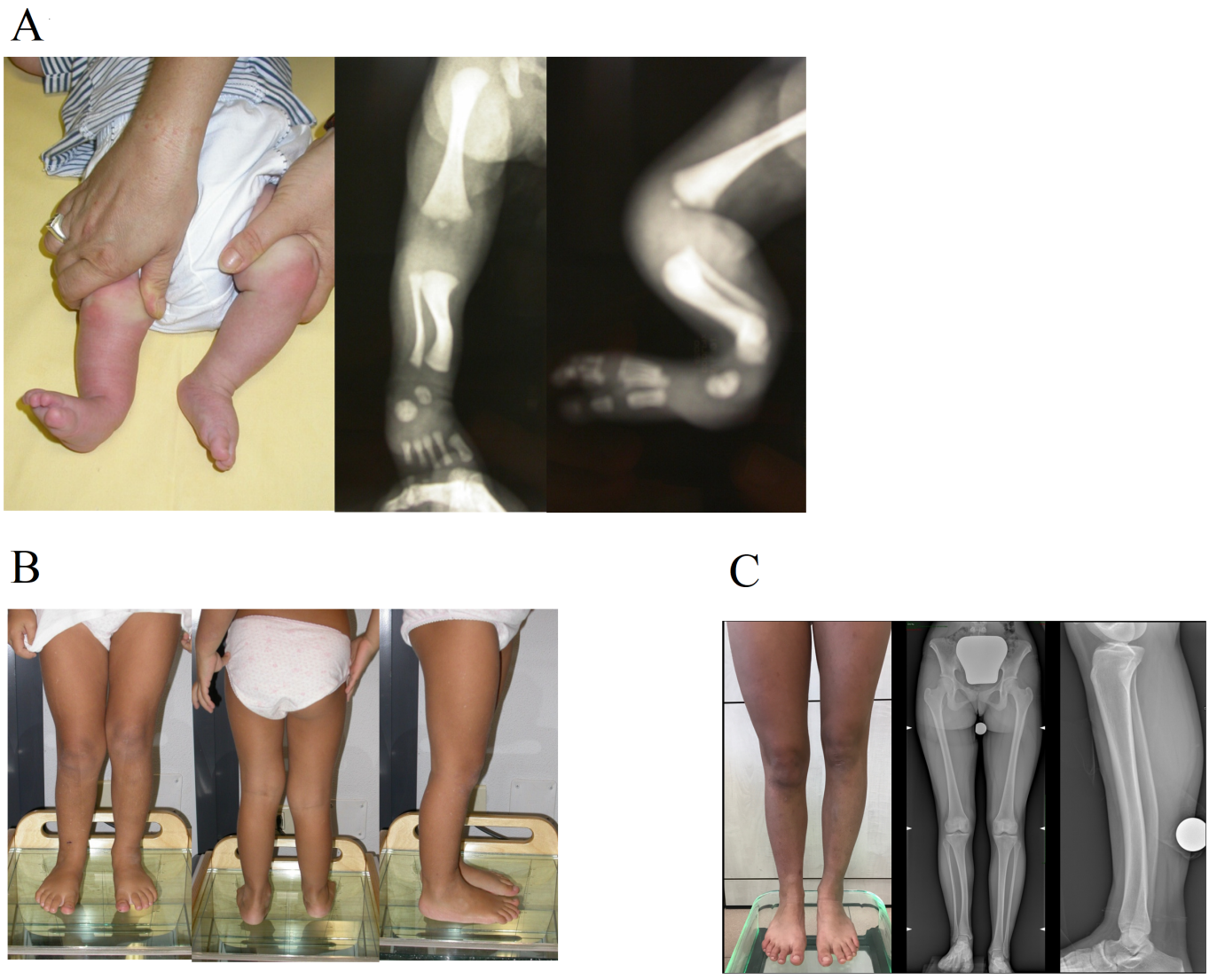

Figure 2

Clinical and radiograhic case showing the spontaneous evolution of the CPMBT. Figure 2a: at 6 months of age; figure 2b: at 3.5 years of age; figure 2c at 15 years of age.

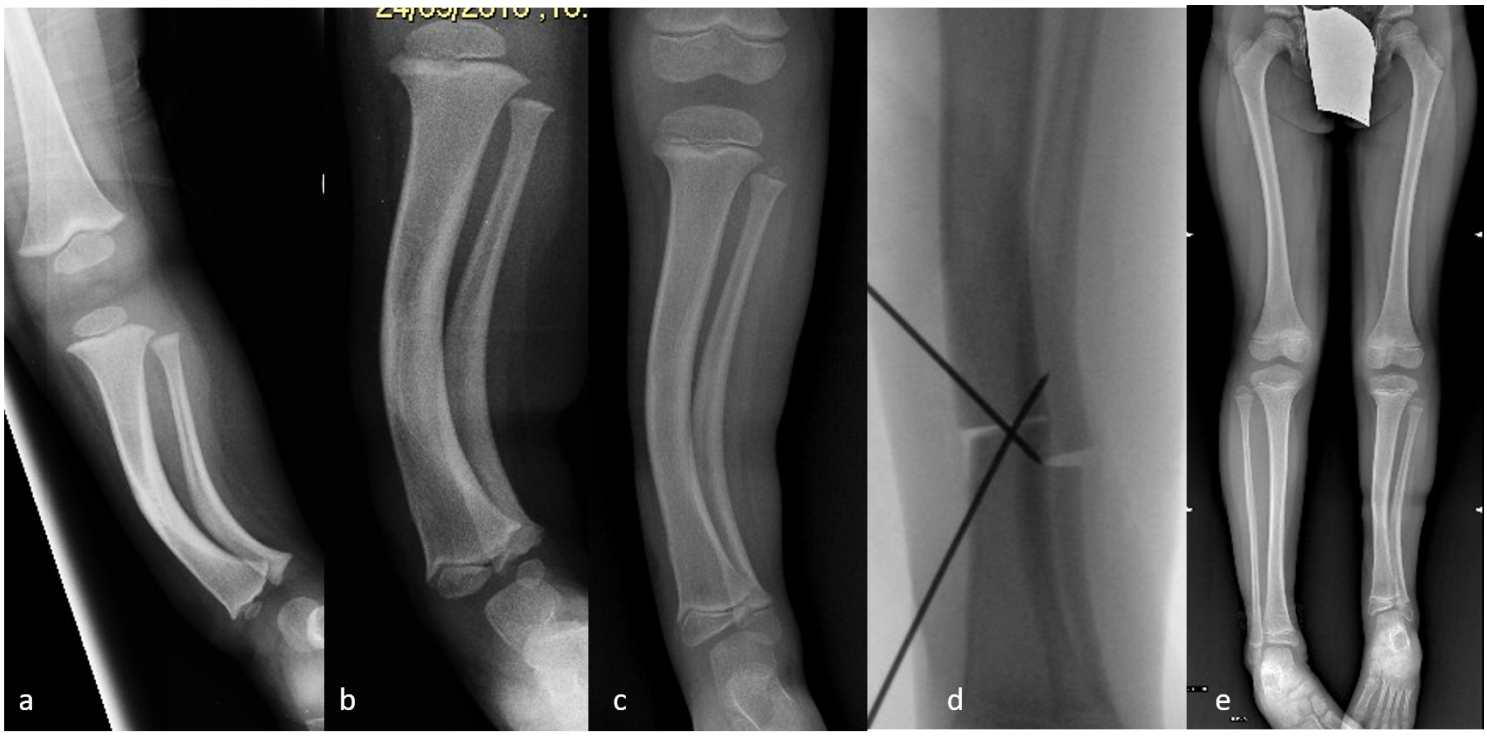

Figure 3

Illustration showing the radiographs of a child affected by CPMBT undergoing tibial osteotomy. 3a: anteroposterior view at 19 months of age. 3b: anteroposterior view at 3.5 years of age. 3c: pre-operative radiograph at 5.8 years of age, showing a residual AP-IPA of 26°. 3d: intra-operative view. 3e: long standing radiographs at 10 years of age. The patient had a residual deformity with an AP-IPA of 9° a L-IPA of 5° a residual LLD of 5 cm (16%) and a LDTA of 70°.

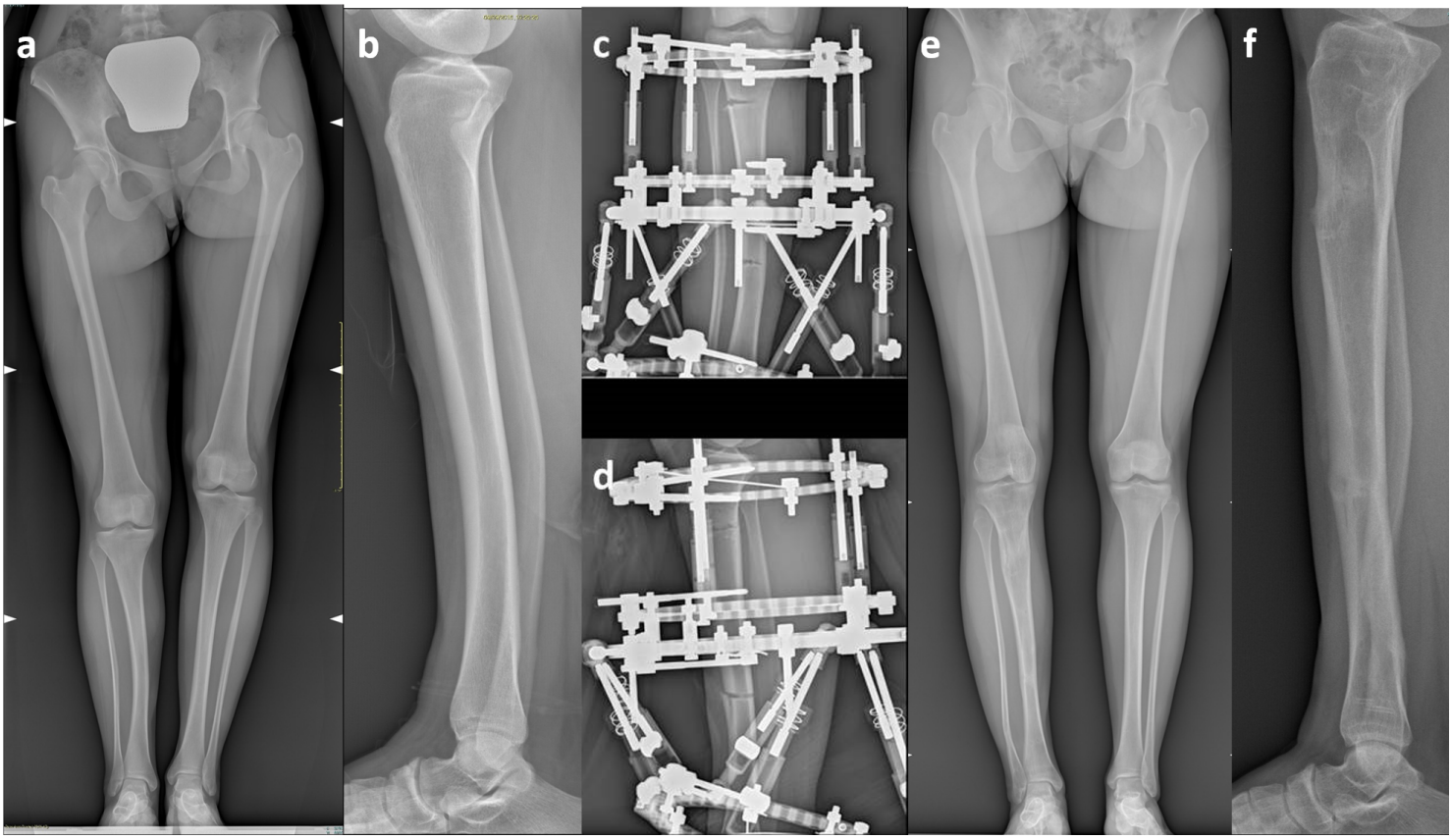

Figure 4

Illustration showing the radiographs of a 14-years girl affected by CPMBT undergoing bifocal lengthening by circular hexapod external fixation to equalize a LLD of 5 cm with a residual antero-posterior bowing of 8°. The deformity correction was achieved at the distal osteotomy with further lengthening at the proximal osteotomy.4a-b: pre-operative long-standing and lateral radiographs. 4c-d: early post-operative radiographs (antero-posterior and lateral view). 4e-f: postoperative long standing and lateral radiographs at the final follow-up one year after the external fixator removal.