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RESEARCH

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How do lesions affect limb lengthening in children with Ollier's disease?

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Abstract

Purpose Ollier's disease (multiple enchondromatosis) can cause severe lower limb length discrepancy and deformity in children. Osteotomy and limb lengthening with external fixation can correct the lower extremity deformity. There may be lesions in the osteotomy part (OP), and the internal fixation part of the external fixation(FP). This study aimed to evaluate: (1) whether lesions in OP and FP influence the lengthening length, speed, and deformity correction; (2) the number of intact sides of the OP and FP that are necessary to provide enough stability to achieve the lengthening correction aim.

Methods Fifteen children with Ollier's disease underwent treatment of 21 lower limb segments using distraction osteogenesis. All osteotomies were performed at the center of rotation and angulation, resulting in a total of fourteen OP intralesionally and nineteen FP intralesionally. The lengthening length, speed, and correction of angular deviation were compared in different groups (lesions vs. non-lesions in OP /FP).

Results Full correction of the deformity and full restoration of length were achieved in all cases. There were no significant differences between intralesional (14 cases) and extralesional (7 cases) distraction groups in new bone formation speed (OP). Although 19/21 of the FP were inserted intralesionally, all the wires and half-pins were well stabilized throughout the external fixation period. There were no significant differences between these groups in the incidence of complications, such as infection, pathological fractures, and early consolidation.

Conclusions In Ollier's disease, the stability provided by newly formed callus and the external fixation were sufficient to lengthen and correct lower limb deformities successfully, even when OP and FP were performed intralesionally with / without intact sides.

Keywords Ollier's disease, Multiple enchondromatosis, Osteotomy, Lengthening, External fixation, Intralesionally, Healing speed, Cortex intact

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Introduction

Ollier's disease, also known as multiple enchondromatosis, is a rare non-hereditary skeletal disorder [1], with a prevalence of about 1:100,000 [2]. This disease is attributable to abnormal endochondral ossification. The affected bones show numerous islands of cartilage in close proximity to the physis, which is linked to bowing of the long bones, and results in deformities and distortions in the longitudinal bone growth, mainly in the legs, such as genu varus, broadening of the metaphysis, and limb length discrepancy (LLD) [3–8].

Ollier's disease causes limb length discrepancy and angulation, especially of the lower limbs. The inheritance patterns for Ollier's disease are unclear; mutations in two genes (PTHR1 and IDH1) are thought to be responsible [3, 4, 8], but this needs further study. Long bone deformity resulting from Ollier's disease represents a treatment challenge. The lesions are widely distributed in the extremities in Ollier's disease. Therefore, traditional methods for treating bone tumors, such as excision and bone grafting, are not possible to completely remove or treat the lesions [6]. There is no consensus for the optimal surgical technique and implants for correction of limb deformities in patients with Ollier's disease [9–11].

There is no effective treatment to cure Ollier's disease itself. Because the lesions are extensive, complete curettage of the enchondromas is practically impossible [8]. Multiple areas of abnormal bone and difficulty obtaining normal bone for grafting are other important problems that need to be addressed [12]. The presence of pathological bone and enchondromas has been shown to preclude internal fixation at the osteotomy site; the poor quality of the bone, the integrity of the fixation, the high risk of complications, and the high possibility of recurrence are well-known problems encountered in these cases [7, 13, 14].

Over the past few years, the use of external fixation systems, including circular fixation (such as Ilizarov fixation), Taylor Spatial Frame (TSF), and monolateral fixation (such as OrthoFix fixation), has become a popular treatment option for correcting angular deformities and LLD [15]. Osteotomy and limb lengthening techniques with external fixations can be performed as many times as necessary to correct the deformities. Because the lesions in the long bones are extensive, the osteotomy part (OP) and the inserted bone part of the external fixator (FP) (such as wires and half-pins) may sometimes be intralesional in situation; sometimes the bone cortex of OP or FP are eroded by lesions and the cortex cannot be maintained intact. The bone may be fragile and bone stabilization is difficult to attain using internal devices [7, 13].

In this study, we wanted to address some lingering questions about intralesional treatment for lower limb deformity due to Ollier's disease, such as (1) whether lesions in OP and FP influence the lengthening length, speed, and deformity correction and (2) the number of cortex intact sides of the OP and FP that are necessary to provide enough stability to achieve the lengthening correction aim.

Patients and methods

From September 2018 through September 2024, 21 sgments in fifteen patients with Ollier's disease were treated at our institution (Table 1). The mean age at surgery of the patients was 6.9 years (range, 2.8–14.0 years). Monosegmental lengthening involved the femur in fourteen cases and the tibia in seven. One patient underwent two lengthening procedures in the same tibia and one in the femur, one patient underwent two lengthening procedures in the same femur, and three patient underwent two lengthening procedures, one in the femur and one in the tibia. The study was conducted in accordance with the Declaration of Helsinki (as revised in 2013). This study was retrospective, it was approved by Ethics Committee of our hospital ([No.(2022) 150]) and informed consent for this retrospective analysis was waived.

Femoral shortening ranged between 2.9 and 7.8 cm and tibial shortening ranged between 2.3 and 6.7 cm. Preoperative assessment included clinical and radiological evaluation (clinical evaluation of the LLD and the range of motion of joints and a standing view radiograph to measure the magnitude of the deformities and the mechanical axis deviation).

Ring external fixators were used in nine segments (two femurs, seven tibias), and monolateral external fixators were used in twelve segments (twelve femurs). All external fixators were installed using standard techniques. The center of rotation and angulation (CORA) method was used to evaluate the deformity [16]. According CORA, we decided the osteotomy part. According the osteopomy part, we installed the external frame, inserted wires and half pins at both ends of the osteotomy. Usually, in circular ring fixator, two circular rings were placed on one end of the osteotomy, and two wires are crossed on each circular ring; in monolateral fixation, or install three half-nails were installed at one end of the osteotomy, and the half-nails are connected by monolateral rod. All osteotomies were performed at the CORA level, using the circular ring fixator or monolateral fixation; wires and half pins of the fixations were inserted into the segments according to the osteotomy position, without considering intralesionality. Osteotomy was carried out via a small incision; multiple drill holes were made, and an osteotome was used to complete the osteotomy. After a latency period of 5–8 days, gradual distraction histogenesis was achieved at a rate of nearly 1 mm/day (one third-turn of the nut every 8 h).

Table 1 Demographics, features, and outcome of patients with Ollier's disease

Segment	Patient	Gender	Age	Segment	Method	OP lesions	OP number of intact side cortex	IP lesions	IP number of intact side cortex	LG (day)	CAD (°)	Angular deviation	DI (mm/d)	LL (cm)	L% (%)	LI (days/cm)	BHI (days/cm)	EFI (days/cm)	Compli-cations
1	1	Boy	5Y2M	Left Femur	OrthoFix	+	4(A, P,L, M)	+	4 (A, P,L, M)	8	12	Varus	0.9	5.0	20.7	10.6	19.6	31.8	None
2	2	Girl	4Y10M	Right Femur	OrthoFix	+	3 (A, L,M)	+	2(A, L)	5	16	Varus	0.9	5.9	22.1	11.4	14.2	26.4	Joint stiffness
3	3	Girl	3Y1M	Left Femur	OrthoFix	+	4(A, P,L, M)	+	1(P)	5	22	Varus	1.2	5.4	26.1	8.5	19.4	28.8	None
4	4	Boy	14Y	Right Femur	TSF	+	2 (P, M)	+	2(P, M)	7	20	Valgus	2.0	6.3	15.8	5.1	36.7	42.2	Patho-logical fracture
5	5	Girl	4Y9M	Left Femur	OrthoFix	-	4(A, P,L, M)	+	2(A, L)	5	8	Varus	0.8	5.0	23.4	12.0	26.6	39.6	None
6	6	Girl	8Y11M	Left Tibia	Ilizarov	+	4(A, P,L, M)	+	3(A, P,M)	8	6	Varus	1.0	6.7	29.1	9.8	25.3	36.1	None
7	7	Girl	7Y2M	Left Femur	OrthoFix	+	3(A, P,L)	+	4 (A, P,L, M)	7	0	/	1.0	5.5	18.9	9.8	46.8	57.9	None
8	8	Boy	9Y6M	Right Tibia	Ilizarov	+	3(P, L,M)	-	4 (A, P,L, M)	7	5	Valgus	0.8	3.2	13.4	13.3	25.9	41.4	None
9	8	Boy	11Y6M	Right Tibia	TSF	+	4(A, P,L, M)	-	4 (A, P,L, M)	6	25	Recurvate	0.7	4.8	15.2	13.5	40.8	55.6	Delayed union
10	9	Boy	5Y1M	Right Femur	TSF	+	2(A, P)	+	2 (A, P)	6	33	Valgus	0.9	3.0	12.2	10.7	25.7	38.3	None
11	9	Boy	5Y10M	Left Tibia	TSF	-	4(A, P,L, M)	+	1 (A)	5	9	Varus	0.6	5.5	29.3	16.9	38.0	63.3	None
12	10	Boy	4Y8M	Right Femu	OrthoFix	-	4(A, P,L, M)	+	2 (A, L)	7	15	Varus	0.9	5.5	22.8	11.6	11.5	24.4	None
13	8	Boy	14Y	Right Femu	OrthoFix	+	4(A, P,L, M)	+	4 (A, P,L, M)	7	18	Varus	1.7	7.9	22.0	6.0	15.2	22.0	None
14	11	Boy	6y7M	Right Femu	OrthoFix	+	4(A, P,L, M)	+	3 (A, P,L)	6	4	Varus	1.0	5.6	19.2	10.2	15.1	26.5	None
15	12	Girl	4Y1M	Right Femu	OrthoFix	-	4(A, P,L, M)	+	2 (A, L)	7	3	Valgus	1.1	5.3	25.7	9.01	10.5	20.6	None
16	1	Boy	7Y6M	Left Femur	OrthoFix	+	4(A, P,L, M)	+	4 (A, P,L, M)	6	16	Valgus	0.8	4.5	13.3	12.1	18.50	31.9	None
17	13	Girl	2y9M	Left Femur	OrthoFix	-	4(A, P,L, M)	+	0	7	12	Varus	0.8	5.6	35.0	13.0	17.3	31.6	None
18	13	Girl	3Y8M	Left Tibia	Ilizarov	-	4(A, P,L, M)	+	0	6	23	Varus	1.2	4.5	24.4	8.6	13.9	23.9	None
19	14	Girl	5y4M	Left Tibia	Ilizarov	-	4(A, P,L, M)	+	2(A, L)	7	8	Varus	1.5	2.3	8.7	6.6	62.0	71.7	None
20	5	Girl	8y	Left Tibia	Ilizarov	+	3(A, M,L)	+	2(M, L)	5	10	Valgus	0.9	3.9	15.4	11.0	12.6	24.9	None
21	15	Boy	4Y8M	Left Femur	OrthoFix	-	4(A, P,L, M)	+	4(A, P,L, M)	5	14	Valgus	0.8	2.9	11.0	12.0	28.8	42.5	None

OP: osteotomy part, IP: the inserted fixation part of the external fixation, LG: lengthening gap, CAD: correction of axial deviation, DI: distraction index, LL: lengthening length, L%: lengthening%, LI: lengthening index, BHI: bone healing index, EFI: external fixation index, A: anterior, P: posterior, L: lateral, M: medial

On X-ray, from anterior-posterior (AP) and lateral (LAT) views, we can visualize four aspects: anterior, posterior, lateral, and medial. We calculated whether there were lesions and the number of intact cortices (four sides) in the OP and the FP. If the cortex was incomplete in multiple places at the OP /FP, the smallest number of intact cortices was taken as the extended limb.

The clinical results were categorized into these objective outcomes [17]. Correction of axial deviation (CAD) is defined as the correction of axial deviation (°). Lengthening gap (LG) is defined as the time duration from osteotomy to beginning of lengthening (days). Distraction index (DI) is defined as the amount of lengthening length per day (mm/day). Lengthening length (LL) is defined as the total lengthening length (cm). Lengthening length percentage (L%) is defined as the percentage of lengthening length to initial length (%). Lengthening index (LI) is defined as the duration in the external fixation lengthening divided by the length gained (days/cm) Bone healing index (BHI) is defined as the duration until bony union in the frame divided by the length gained (days/cm). External fixation index (EFI) is defined as the entire duration of external fixation divided by the length gained (days/cm).

We used these above objective outcomes to evaluate our results (Fig. 1) [17].

After the desired length was achieved, the frame was retained until there was cortical continuity visible on three sides as seen on AP and LAT views of the regenerate [9].

Statistical analysis

The statistical analysis was carried out using SPSS 17 software (SPSS Inc., Chicago, IL, USA). Quantitative data were expressed as median and range (min-max), and qualitative data were expressed as frequency and percentage. Descriptive statistics (central tendency measures and data spread) were applied. Statistical significance was determined using the independent Student's t-test. $P<0.05$ was regarded as statistically significant.

Results

Deformity correction followed by lengthening was performed in fifteen segments and limb-lengthening in 21 segments. All osteotomies were performed by the CORA method; wires and half pins were inserted into the segments according to the osteotomy position. Therefore, nine osteotomies were performed intralesionally followed by distraction osteogenesis, and nine FP were inserted intralesionally.

The mean amount of length gained was 49.7 mm (range, 22.6–78.5 mm), and the mean angular correction was 13.3° (range, 0°–33.0°). The mean external fixation period was 175 days (range, 97–320 days), and the mean EFI was 36.5 days/cm (range, 20.6–71.7 days/cm).

After the removal of the external fixator, the lengthened limb was protected by plaster slab at least 2 weeks (almost 2 weeks to 4 weeks) with no weight bearing. We tried to continue to follow up to 18 years, at least six months follow-up. In study, the mean follow up was 36.1 months (range, 8.3–34.6 months).

Three cases met complications. One case (4.7%, Segment 3) met joint stiffness, after removal the external frame, the knee can only bend 0 to 90 degrees. One case (4.7%, segment 4) met pathological fracture at the proximal femur fracture, and was added another half pins to fix the fracture at the proximal end of the TSF frame. One case (4.7%, segment 8) met delayed union.

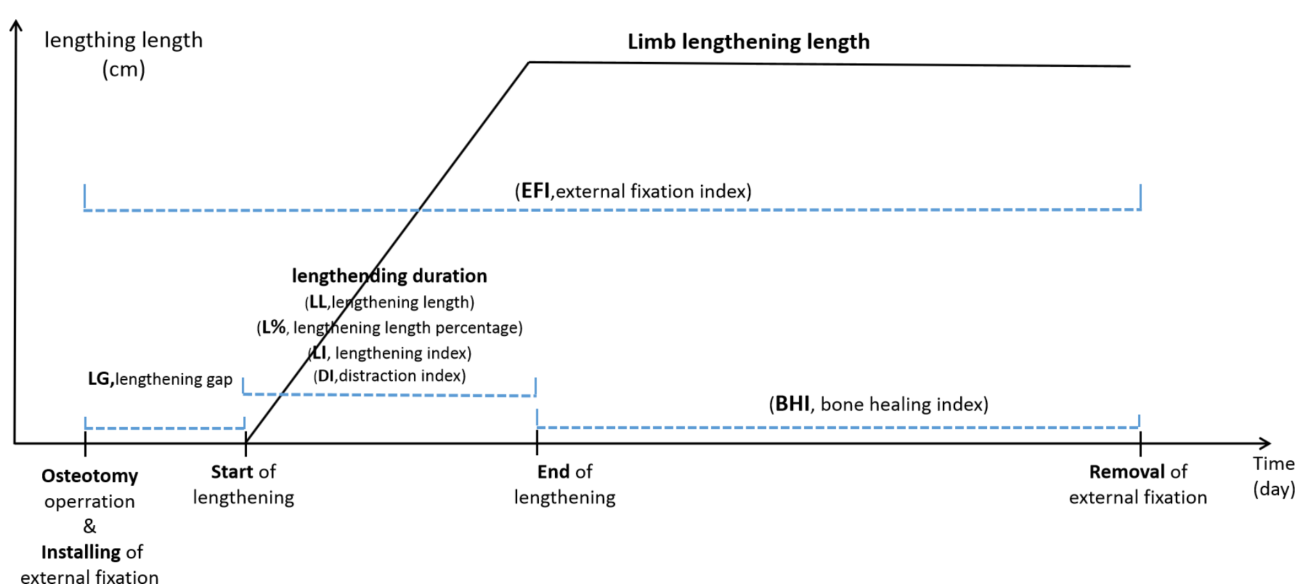

Fig. 1 Objective outcomes according to lengthening duration [17]

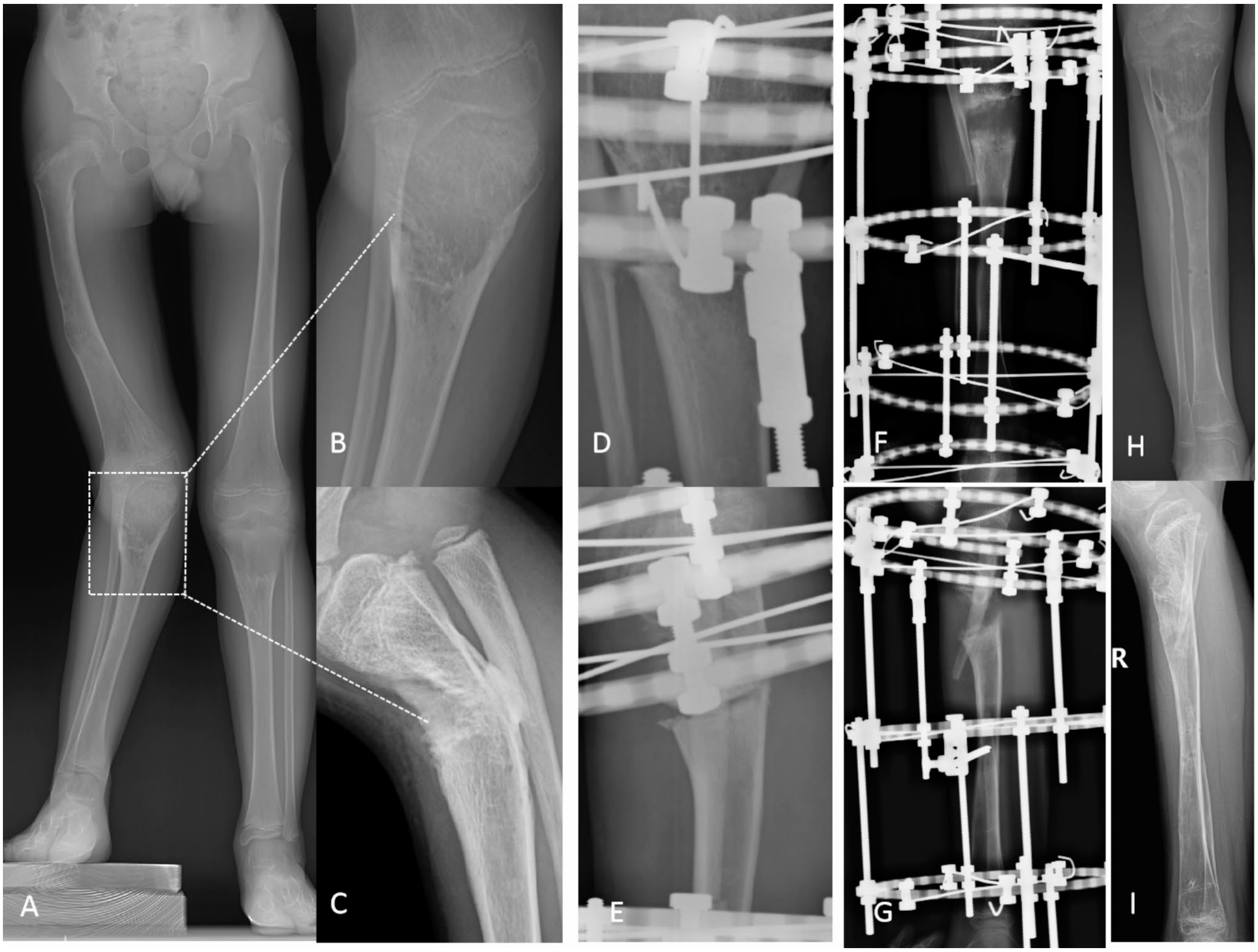

Fig. 2 9Y6M-old boy (case 8), right proximal tibia lesions with Ollier's disease. (**A**, **B**, **C**) anterior side of cortex was not intact (posterior, lateral, medial sides intact); (**D**, **E**) osteotomies were performed intralesionally at non-intact side; (**F**, **G**) and followed by distraction osteogenesis with LG 7 days, LL 3.2 cm, CAD valgus 5°;(**H**, **I**) new bone formed well

Table 2 Intralesional vs. extralesional distraction osteogenesis in OP

Distraction Osteogenesis(OP)	Segment	CAD (°)	DI (mm/d)	L% (%)	LI (days/cm)	BHI (days/cm)	EFI (days/cm)
Intralesional	14	14.5 ± 9.0	1.1 ± 0.4	18.2 ± 5.4	10.3 ± 2.4	24.6 ± 10.6	36.2 ± 11.0
Extralesional	7	11.0 ± 6.3	1.0 ± 0.3	24.4 ± 8.2	11.1 ± 3.4	24.7 ± 18.2	37.3 ± 18.2
P		0.381	0.651	0.050	0.519	0.984	0.865

Demographics, features, and outcome of patients with Ollier's disease are depicted in Table 1.

Among the 21 total external fixations, there were no significant differences in the limb lengthening length and correction deformities, such as CAD, DI, L%, LI, BHI, EFI between the two groups with and without lesions at the OP (group intralesional vs. group extralesional) (Fig. 2; Table 2).

In the OP group, LLD and angular deformity corrections were successfully achieved regardless of whether the four surfaces of the cortex were intact or incomplete, with no significant differences in CAD, DI, L%, LI, BHI and EFI (see Table 3).

Similarly, there were no significant differences in the relative data of limb lengthening and deformity correction between the two groups with and without lesions at FP (group intralesional vs. group extralesional) (Fig. 3; Table 4).

In the FP group, LLD and angular deformity corrections were successfully achieved regardless of whether the four surfaces of the cortex were intact or incomplete, with no significant differences in CAD, DI, L%, LI, BHI and EFI (see Table 5).

At least one intact cortex could afford enough stability to achieve the lengthening aim.

Table 3 Intact cortex vs. incomplete cortex intralesional distraction osteogenesis in OP

Distraction Osteogenesi(OP-Intralesional)	Segment	CAD (°)	DI (mm/d)	L% (%)	LI (days/cm)	BHI (days/cm)	EFI (days/cm)
Intact cortex	15	13.0 ± 6.8	1.0 ± 0.3	21.8 ± 7.4	10.7 ± 2.8	23.7 ± 13.4	35.7 ± 14.1
Incomplete cortex	6	14.0 ± 11.8	1.1 ± 0.4	16.3 ± 3.7	10.2 ± 2.8	27.0 ± 13.1	38.5 ± 12.1
P		0.807	0.641	0.100	0.710	0.618	0.678

Fig. 3 3Y1M-old girl (case 3), left distal femur lesions with Ollier's disease. (**A**, **B**, **C**) anterior, lateral, medial sides of cortex were not intact (only posterior side cortex was intact) at FP (**D**, **E**); (**F**, **G**) external fixation at FP with only one intact cortex could afford enough stability to achieve lengthening aim with LG 5 days, LL 5.4 cm, CAD varus 22

Table 4 Intralesional vs. extralesional distraction osteogenesis in FP

Distraction Osteogenesis(IP)	Segment	CAD (°)	DI (mm/d)	L% (%)	LI (days/cm)	BHI (days/cm)	EFI (days/cm)
Intralesional	19	13.2 ± 8.0	1.1 ± 0.3	20.9 ± 7.0	10.3 ± 2.7	23.7 ± 13.2	35.3 ± 13.2
Extralesional	2	14.9 ± 14.3	0.8 ± 0.0	14.3 ± 1.3	13.4 ± 0.2	33.4 ± 10.5	48.5 ± 10.1
P		0.783	0.221	0.210	0.123	0.334	0.189

Table 5 Intact cortex vs. incomplete cortex intralesional distraction osteogenesis in FP

Distraction Osteogenes(IP-Intralesional)	Segment	CAD (°)	DI (mm/d)	L% (%)	LI (days/cm)	BHI (days/cm)	EFI (days/cm)
Intact cortex	6	12.7 ± 9.1	1.0 ± 0.4	17.3 ± 3.8	10.9 ± 2.8	27.8 ± 13.0	40.1 ± 14.3
Incomplete cortex	15	13.6 ± 8.2	1.0 ± 0.3	21.4 ± 7.6	10.4 ± 2.8	23.4 ± 13.4	35.1 ± 13.2
P		0.839	0.794	0.221	0.742	0.498	0.451

Discussion

The most important treatment goals for lower extremity Ollier's disease are achieving normal mechanical realignment and equivalent lower limb length, which would enable normal walking and provide sufficient support to prevent pathological fractures [18].

Osteotomy and lengthening with external fixator could realize the aim of correction of angular deformity and lower limb length discrepancy. Curran et al. [19] found that poor bone regeneration in some pediatric patients with Ollier's disease who had undergone lengthening, required bone grafting and additional immobilization. However, Pandey et al. [20] noted that distraction osteogenesis through predominantly cartilaginous bone converted that into mature corticalized new bone rapidly. Jesus-Garcia et al. [13] found that conversion of the abnormal cartilage into histologically mature bone occurred in all patients using the Ilizarov technique. If there were lesions in OP, and cortex at OP was not intact, could osteotomy and distraction osteogenesis be performed successfully?

Normal or abnormal new bone generation could occur in OP by distraction osteogenesis [6, 7, 12, 13, 21]; however, experts have different opinions on this matter. Discussion of whether there are lesions in the newly formed bone is beyond the scope of the present study because pathological analysis is required to identify lesions, which would not pass ethical tests. We wanted to ensure that the strength of the new bone could support the weight without pathological fractures, after the removal of the external fixators.

If the cortical bone has been destroyed (at least how many faces intact), we wanted to ascertain whether the newly formed bone can maintain the weight without pathological fractures, and if the rate of new bone formation slowed down. Tables 2 and 3 show that there were no significant differences in the speed of new bone formation between lesions and non-lesions at OP (at least 2/4 cortex intact).

If there were lesions at FP, could external fixators provide sufficient stability to achieve the lengthening aim? Baumgart et al. [10] considered the bone in Ollier's disease to be relatively soft, so external fixator pins may cut out resulting in the premature removal of the fixator. However, according to Watanabe K [12], external fixators could provide enough stability to achieve the lengthening aim, without pathological fractures. However, the study did not discuss further whether there were differences in the lengthening speed between lesions and non-lesions at FP. If there are lesions at FP and the four faces are not intact, could external fixators still provide sufficient stability to achieve the lengthening aim, without pathological fractures? To the best of our knowledge, no study has considered this in depth.

In our study, Table 4 shows that there were no significant differences between lesions and non-lesions at FP in limb lengthening objective outcomes, such as CAD, DI, L%, LI, BHI, EFI. Table 5 shows that with lesions at FP, there were no significant differences between intact cortex and non-intact cortex in limb lengthening objective outcomes. External fixators could provide sufficient stability to achieve lengthening and correction program even if there was only one side cortex intact at FP.

Angelini et al. performed a systematic review of the literature from 1993 to 2017. Joint stiffness, infection, early consolidation, pathological fracture, deformity recurrence, delayed union, non-union, neurapraxia, and overlengthening were the reported complications with an overall rate of 27.9% [11]. In our study, the rate of complications was 27.3% (3/11, before 2021) in early operation, which is close to Angelini et al.'s findings. In later operation, because of rehabilitation training during the lengthening process, and plaster slab used for protection after removal of the external frame, the above complication did not occur, the rate of complications decreased to 14.3 (3/21, in total).

Our study, however, has some limitations. The retrospective nature of this study limited our ability to collect further data. Follow up of certain number of patients is only six month and therefore results section could be biased. We could not apply quality of life or other functional outcome scores due to difficulty in reaching some patients.

Conclusions

LLD and deformity caused by Ollier's disease can be treated successfully by distraction osteogenesis with external fixation. When osteotomy was performed intralesionally with non-intact cortex at OP, new bone formation could occur from the OP and could bear the weight. When non-intact cortex occurred at FP from lesions, external fixators could provide sufficient stability to achieve equivalent lower limb length and deformity correction without resulting in pathological fractures.

In summary, in this study, we established that intralesional osteotomy and intralesional FP is a valid treatment option with predictable outcomes in Ollier's disease. The position of the lesions should still be considered during surgery, and further clinical evidence is required to confirm these results.

Author contributions

Chunxing Wu: Manuscript preparation, performed measurements, and statistical analysis. Peng Huang, Yueqiang Mo, Dahui Wang: Performed measurements, cases collection, and statistical analysis. Bo Ning: Study design, supervision, and validation.

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Data availability

The datasets generated and/or analysed during the current study are not publicly available due to limitations of ethical approval involving the patient data and anonymity but are available from the corresponding author on reasonable request.

Declarations

Ethical approval

The study was approved by institutional ethics board of Children's Hospital of Fudan University [No.(2022) 150] and informed consent for this retrospective analysis was waived by institutional ethics board of Children's Hospital of Fudan University.

Competing interests
The authors declare no competing interests.

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