Periosteal stripping and periosteal division for leg length discrepancy after proximal femoral intertrochanteric osteotomy for Perthes disease

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Research Article

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Abstract

Background: Periosteal stripping and periosteal division (PSPD) promotes growth of the long bone of children with leg length discrepancy (LLD). We performed PSPD when LLD was observed at the time of implant removal surgery after proximal femoral osteotomy for Perthes disease. This study aimed to clarify the efficacy and safety of PSPD for acquired LLD related to Perthes disease.

Methods: This retrospective study enrolled 10 patients treated with PSPD and 6 control patients who declined the PSPD for LLD associated with Perthes disease. The lengths of the femur, tibia and entire leg were measured in the full-length standing radiographs at baseline and final follow-up. Baseline was defined as the time of the last preoperative observation. LLD and changes in LLD (ΔLLD) were measured. The correlation of ΔLLD with age at time of surgery, follow-up period, and extent of PSPD was investigated.

Results: Patients' mean age and LLD at baseline were 9.4 years and 20.5 ± 4.6 mm in the PSPD group and 10.2 years and 11.5 ± 10.0 mm in the control group. With a mean follow-up period of 4.3 years, the PSPD group showed a mean ΔLLD decrease of 13.9 mm, which was significantly greater than that of the control group at 3.2 mm with a mean follow-up period of 5.4 years. Logistic regression analysis revealed that age at the time of surgery was a significant factor for obtaining >10 mm ΔLLD with PSPD and the cutoff value by the receiver operating characteristic curve was 9.6 years (sensitivity, 0.83; specificity 0.83).

Conclusion: PSPD seemed to be a safe and effective surgical option for LLD associated with Perthes disease. The age at the time of surgery negatively correlated with the amount of LLD correction. Obtaining >10 mm LLD correction is more likely if the patients are <10 years of age.

Trial Registration: Present study was registered in original registration system in Chiba Children's Hospital. Registration number was 2021-074 and registration date was 14th May 2022.

Background

Leg length discrepancy (LLD) can cause various clinical problems, including scoliosis and, gait abnormalities, and early osteoarthritis [1][2]. The causes of LLD include lower limb hypoplasia, unilateral hypertrophy due to neoplastic lesions, growth disturbances due to trauma or infection, slipped capital femoral epiphysis (SCFE), and Perthes disease [1] [3][4].

LLD reaching up to 7 cm can occur in Perthes disease [5][6][7]. The causes of LLD associated with Perthes disease include spread of the femoral head lesion [8][9][10], premature closure of the proximal femoral growth plate [11][12][13][14], disuse bone atrophy due to orthotic unloading [16], and proximal femoral intertrochanteric osteotomy [8][16][17].

Surgical treatments for LLD include acute shortening, acute or gradual bone lengthening, growth plate suppression, and PSPD [18]. Acute shortening may cause transient limp due to relative muscle length shortening, and acute lengthening may cause traction neuropathy [19]. Gradual bone lengthening with external fixation has a risk of complications, including knee and ankle joint contracture due to soft tissue elongation, traction neuropathy, pin site infection. Additionally, the psychological aspect of long-term treatment needs to be considered [20]. Growth plate suppression targets the growth plate of the longer side of the leg. Temporary growth suppression using the eight-plates has been widely used recently due to its simple and low invasiveness [21]. Nevertheless, several complications, such as loosening or breaking of the screws, premature closure of the growth plate [22], and changes in proximal tibial bone morphology [23], are reported. The suppression effect is difficult to predict.

PSPD is a technique for promoting growth of the long bone, as reported by Limpaphayom et al. in 2011 [18]. They reported 11 cases with the mean preoperative LLD of 60 ± 38 mm and mean age of 9.0 ± 2.5 years. PSPD was reportedly performed on the entire length, and correction of LLD was achieved in eight of 11 patients in an average of 25 ± 17.2 months and was maintained throughout the follow-up period [18]. PSPD is less burdensome to patients as only one surgery is needed; however, predicting the amount of leg length correction is difficult in PSPD. Moreover, there have been no other reports on the clinical results of PSPD.

At our hospital, we consider PSPD for cases with LLD at the time of implant removal surgery after proximal femoral intertrochanteric osteotomy for Perthes disease, and we perform the surgery if the patients and their family desire so. The present study aimed to clarify the efficacy and safety of PSPD for acquired LLD related to Perthes disease. Additionally, we sought to examine the correlations between the leg length correction effect, age at the time of surgery, and the extent of surgery to predict the amount of leg length correction with PSPD. We hypothesized that the younger the patient at the time of surgery and the larger the area of surgery, the greater the leg length correction effect.

Methods

In this retrospective study, we followed-up patients treated with perthes disease to evaluate outcomes. The study was conducted at the Chiba Children's Hospital, Japan, under the supervision of Dr. Takashi Saisu.

Patients

From August 2012 to August 2020, we have performed PSPD in 10 children with LLD related to Perthes disease. The inclusion criteria were as follows: 1) patients with unilateral Perthes disease; 2) LLD of > 20 mm before implant removal surgery after proximal femoral intertrochanteric osteotomy or the attending physician predicted LLD of ≥ 20 mm at the end of growth based on the clinical course of the patient 3) the patient was at the age when the growth plate is still present and PSPD is expected to promote bone overgrowth; 4) the parent and guardian agree to the surgical risks and benefits; 5) radiographs of the entire leg were taken before and after the surgery; and 6) patients were followed up for at least 1 year after PSPD. The exclusion criteria were as follows: 1) patients with infection of the surgical sites; 2) nonunion of the osteotomy; 3) neurological disorders; and 4) skeletal dysplasia. During the study period, 10 cases met the above inclusion criteria. During the same period, 12 patients with unilateral Perthes disease underwent proximal femoral intertrochanteric osteotomy followed by implant removal surgery without PSPD. To clarify the natural course of LLD after the proximal femoral osteotomy, the patients who did not undergo PSPD were set as a control group. The inclusion criteria of the control group were as follows: radiographs of the entire leg were taken before and after surgery and the patients were followed up for at least 1 year after surgery. Of these, five cases without preoperative LLD measurement and one case without postoperative LLD measurement were excluded; the remaining six cases met the inclusion criteria.

This research has been approved by the IRB of the authors' affiliated institutions. Patients' informed consent was obtained using the opt-out method.

Surgical procedures

PSPD was performed as described elsewhere [18]. The patient is placed in a lateral position, and the location of the epiphyseal line is confirmed by fluoroscopy. A longitudinal skin incision is made laterally on the femur, antero-medially on the tibia, and laterally on the fibula. A longitudinal incision is made on the periosteum, and the periosteum is dissected circumferentially (Fig. 1A). A circular incision is made in the periosteum at the proximal, middle, and distal parts of the diaphysis of the femur (Fig. 1B). The same technique is used for the tibia and fibula. A drainage tube is placed on the treated bone to prevent compartment syndrome.

Radiological assessments

The entire leg length was defined as the distance from the center of the hip joint to the center of the ankle joint. The femoral length was defined as the distance from the center of the hip joint to the center of the knee joint. The tibial length was defined as the distance from the center of the knee joint to the center of the ankle joint. The center of the joint means both the horizontal and vertical widths of the joint gap. The distances (mm) were measured by PACS in full-length standing frontal radiographic view of the entire leg. The length difference between the healthy and affected sides was defined as LLD (mm), and ΔLLD (mm) was the change obtained by subtracting the LLD at the last observation from the preoperative LLD.

Statistical analysis

As statistical studies, preoperative and postoperative LLD were analyzed by performing the Wilcoxon-signed rank test, and the difference in the mean LLD between groups was analyzed with the Mann−Whitney U test. The Pearson's correlation coefficient was calculated between ΔLLD and age at the time of surgery or follow-up period. The Spearman's correlation coefficient was calculated between the ΔLLD and the extent of PSPD. Logistic regression analysis was performed to determine whether the ΔLLD effect of ≥ 10 mm could be obtained using age at the time of surgery as a predictor variable. A p-value < 0.05 is considered to be significant. Statistical analyses were performed using JMP pro ver. 15 (SAS Institute Inc., Cary, NC, USA).

Results

Patient's demographic data are shown in Table 1. The mean age at the time of PSPD was 9.4 years, and the mean follow-up period was 4.3 years. PSPD was performed to the proximal 1/2 of the femur in five patients, proximal 2/3 of the femur in four patients, and the entire length of the femur and lower leg in one patient. In the control group, the mean age and follow-up period were 10.2 and 5.4 years, respectively. All patients were boys.

Table 1
Demographic data of the patients.

PSPD							
Case	Age at operation (Y)	Extent of PSPD (mm)	Preoperative LLD (mm)	Follow up LLD (mm)	ΔLLD (mm)	Age at follow up (Y)	Follow up period (M)
P1	7	Prox. femur 1/2	18	−2	20	16	108
P2	8	Prox. femur 1/2	27	9	18	11	39
P3	10	Prox. femur 1/2	17	8	9	14	26
P4	11	Prox. femur 1/2	28	22	6	17	73
P5	11	Prox. femur 1/2	21	17	4	14	41
P6	6	Prox. femur 2/3	16	−4	20	9	40
P7	7	Prox. femur 2/3	14	−10	24	9	27
P8	9	Prox. femur 2/3	20	10	10	12	37
P9	13	Prox. femur 2/3	20	14	6	17	55
P10	12	Femur, tibia, fibula	24	2	22	19	90
Control							
Case	Age at implant removal (Y)		Preoperative LLD (mm)	Follow up LLD (mm)	ΔLLD (mm)	Age at follow up (Y)	Follow up period (M)
C1	7		10	−4	14	10	45
C2	8		12	11	1	14	80
C3	9		5	12	−7	19	117
C4	9		6	−6	12	17	101
C5	13		31	32	−1	16	34
C6	15		5	5	0	16	15

PSPD: periosteal stripping and periosteal division; LLD: leg length discrepancy; Prox.: proximal

All cases were boys.

Figure 2 shows the bone and leg length of the PSPD and control groups. At baseline, there were significant differences in the femoral and entire leg lengths between the affected and healthy sides in both the PSPD (p = 0.002, 0.002, respectively) and control groups (p = 0.031, 0.031, respectively), and there was a significant difference in tibial length in the PSPD group (p = 0.007). At the time of the latest follow-up, a difference in femoral length was still noted in the PSPD group (p = 0.049), but there were no significant differences in the entire leg length in the two groups.

Table 2 shows the amount of change in bone length. In the PSPD group, the change in the femoral and tibial lengths of the affected side was significantly greater than that of the healthy side, and the leg length also showed a significantly greater change in the affected side (p = 0.004, 0.043, 0.002, respectively). In the control group, the affected side also tended to show greater changes than the healthy side, but only the tibia length showed a significant difference in the amount of change (p = 0.047).

Table 2
Increase in bone length (mm).

	Increase in femoral length		**Increase in tibial length**		**Increase in whole leg length**	
	Healthy side	Affected side	Healthy side	Affected side	Healthy side	Affected side
PSPD	68.8 ± 31.5	77.3 ± 34.9*	53.1 ± 5.5	56.7 ± 26.2*	121.2 ± 55.7	135.1 ± 58.5*
Control	76.2 ± 49.5	77.3 ± 50.1	59.5 ± 40.1	61.7 ± 40.4*	134.3 ± 89.5	137.5 ± 91.1

PSPD: periosteal stripping and periosteal division

Values are presented as mean ± SD.

*: *P*< .05 between the sides in Wilcoxon signed rank test

Table 3 shows the difference in bone length between the affected and healthy sides. The baseline LLD was 20.5 ± 4.6 mm in the PSPD group, which was significantly greater than that of the control group at 11.5 ± 10.0 mm (p = 0.034). However, in the PSPD group, the bone length difference was reduced by 8.5 ± 8.5 and 3.6 ± 5.4mm in the femur and tibia, respectively, resulting in a LLD of 6.6 ± 10.0 mm at the final observation, and the effect of PSPD on ΔLLD was 13.9 mm. The ΔLLD of PSPD was significantly greater than that of the control group, which was 3.2 ± 8.1 mm (p = 0.034). There was no significant difference in the LLD at the final observation between the two groups.

Table 3
Difference in bone length between the affected and healthy side (mm).

	Difference of femoral length			Difference of tibial length			Difference of leg length		
	baseline	Follow-up	changes	baseline	Follow-up	changes	baseline	Follow-up	changes
PSPD	15.4 ± 3.0	6.9 ± 10.3	8.5 ± 8.5	4.1 ± 3.8	0.5 ± 5.7	3.6 ± 5.4	20.5 ± 4.6*	6.6 ± 10.0	13.9 ± 7.6*
Control	9.7 ± 7.2	8.5 ± 10.9	1.2 ± 6.8	2.3 ± 3.1	0.2 ± 3.5	2.2 ± 2.1	11.5 ± 10.0	8.3 ± 13.8	3.2 ± 8.1
Values are presented as mean ± SD.									
*: *P* < .05 between PSPD and Control in Man–Whitney U test									

The PSPD group showed a significant LLD correction effect, but the effect varied in each case (range, 4–24 mm). There was no correlation between the amount of change in LLD and follow-up period (p = 0.53), a significant correlation was observed between the amount of change in LLD and age at the time of surgery (r = − 0.69, 95% confidence interval: −0.92 to − 0.1; p = 0.03, Fig. 3A). Especially, there was a strong correlation between the change in femoral length difference and age at the time of surgery (r = − 0.87, 95% confidence interval: −0.97 to − 0.52, p = 0.0012, Fig. 3B). Cases P1, P6, and P7 were operated on at the age of ≤ 7 years, and there was a reversal of the LLD, with the affected side being longer at the last observation. A LLD correction effect of 22 mm was obtained in patients who underwent PSPD on both the femur and lower leg at the age of 12 years. However, there was no significant correlation between the extent of PSPD performed on the femur (1/3 or 2/3) and the LLD correction effect (p = 0.38). Contrarily, the change in LLD in the control group varied from − 7 to 14 mm, but there was no significant correlation observed with the follow-up period or age at baseline (p = 0.83 and p = 0.32, respectively).

In the PSPD group, the LLD correction effect of ≥ 10 mm was obtained in six cases. A logistic regression analysis was performed to determine whether the ΔLLD effect of ≥ 10 mm could be obtained by the age at the time of surgery. It revealed that age at the time of surgery was a significant factor (p = 0.009, odds ratio of 3.32), and the cutoff value by the receiver operating characteristic curve was 9.6 years (sensitivity, 0.83; specificity, 0.83).

There were no complications related to PSPD, such as vascular or nerve injury, infection, fracture. During the course after PSPD, five patients had temporary genu valgum of the coronal plane on the affected side (cases 1, 2, 3, 5, and 7). Thereafter, the alignment gradually improved, and the alignment at the final observation showed improvement without significant differences between the healthy and affected sides in all the patients (Fig. 4). Similarly, two patients showed a varus knee deformity (cases 4 and 9) but both cases showed improvement at the final observation.

Discussion

This study revealed the efficacy and safety of PSPD in improving LLD after proximal femoral intertrochanteric osteotomy for Perthes disease in children. In the control group, a spontaneous correction of LLD was also observed, but the correction was only approximately 28% of the baseline LLD. In contrast, the PSPD group showed a mean correction of 13.9 mm, which was 68% of the baseline LLD, suggesting that the effect of surgery was significant. Additionally, the amount of leg length correction in PSPD was related to the age at the time of surgery.

The younger the age at the time of surgery, the greater the LLD correction, and it is more likely that a leg length correction of > 10 mm can be achieved if the procedure is performed in patients < 10 years of age.

The cause of overgrowth of the long bone due to PSPD include changes in the hemodynamics and the external force applied to the growth plate. Yabsley et al. reported that periosteal stripping caused a blockage of blood flow to the cortical bone and increased blood flow to the growth plate [24]. Warrell et al. suggested that the development of growth plate is mechanically controlled by the tension of the periosteum at both ends, and that a transverse incision of the periosteum reduces the tension of the periosteum on the growth plate, leading to overgrowth [25].

Regarding the complications associated with PSPD, Sola et al. reported pathological fractures in animal studies in 5 of 22 dogs (22%) and 3 of 10 monkeys (30%) after two repetitions of periosteal stripping [26]. Limpaphayom et al. used a cast for postoperative therapy and allowed weight-bearing walking after a 2-week unloading period [18]. In our study, the patient was allowed to walk with half-load from the day after the surgery, and full-load walking was gradually allowed according to the pain intensity, but none had pathological fracture. Houghton et al. reported a case of valgus tibia deformity observed when a semi-circumferential periosteal division on the medial side of the tibia was performed [27]. In our study, a transient valgus and varus knee joint deformity was observed in five cases after PSPD. In all cases, the leg alignment improved gradually. Imbalance of periosteal stripping between the medial and lateral sides of the bone may cause these temporal valgus or varus deformities, although a precise mechanism remains unknown. Other complications, such as superficial infection and hematoma, have been reported [28][29]. As for the scar, the wound is long because it covers the entire length of each bone. In the present study, all patients in the surgical group were boys, and none of them were concerned about the appearance of the wound at the time of the final observation (Fig. 5). However, careful explanation of the postoperative scar of PSPD before the surgery is necessary.

Regarding the relationship between the age at the time of PSPD and effect of bone growth, Limpaphayom et al. retrospectively reviewed 11 patients who underwent PSPD and reported that the mean age at the time of PSPD was 9 ± 2.5 years (5–13 years).[19] Our study revealed the significant correlation between the age at time of surgery and ΔLLD. Additionally, logistic regression analysis showed that a > 10-mm correction is expected if the patients are < 10 years of age at the time of surgery. In contrast, no correlation was confirmed between ΔLLD and follow-up length after the surgery. It is suggested that the age at the time of surgery of the patients, rather than the duration after the surgery, has greater influence on the amount of leg length correction. No correlation was confirmed between ΔLLD and extent of surgery (1/3 or 2/3 of femur) either. Nevertheless, one patient aged 12 years obtained ΔLLD of 22 mm after PSPD was performed on the entire femur, tibia, and fibula. This result suggested that, even at an older age, the wide extent of the PSPD is likely more effective. If PSPD was performed on the lower leg as well, the effect on ΔLLD can be expected to be significant. As it is difficult to accurately predict the effect of leg length correction, patients should be thoroughly followed up until their bone has completely matured.

There are several limitations to this study. First, the number of patients is small. The study participants were limited to patients who had undergone PSPD for LLD after proximal femoral intertrochanteric osteotomy for Perthes disease in order to have the same patient background. Clinical studies on more cases and other diseases are desirable. Second, the control group was not randomly selected and included those who had LLD after proximal femoral intertrochanteric osteotomy for Perthes disease but who did not undergo PSPD as per the request of patients or the LLD < 20mm. Given that the patient background, including LLD at baseline, was different from that of the PSPD group, the results were not strictly comparable. The present investigation is a level 4 case series study, and the purpose of having a control group is to show how much LLD is corrected in the natural course. Third, the extent of the surgery differed from patient to patient. One patient who had undergone PSPD of both the femur and lower leg was relatively old, but a leg length correction effect of 22 mm was obtained. However, the effect of the difference in the extent of PSPD performed on the femur was not as strong as the effect of age at the time of surgery on the leg length correction. Further research focused on the extent of PSPD to the entire leg is desirable.

Conclusions

PSPD provided a mean leg length correction of 13.9 mm at a mean follow-up duration of 4.3 years in patients with LLD after proximal femoral intertrochanteric osteotomy for Perthes disease. There was a significant correlation between the amount of change in LLD and age at the time of surgery. Obtaining > 10 mm limb length correction is more likely if the patients are < 10 years of age. No serious adverse effect occurred. PSPD is acceptable to patients because it allows leg-length correction on the affected leg. Although it is difficult to accurately predict the effect of leg length correction, PSPD is good adjunctive procedure when performing an implant removal surgery for LLD associated with Perthes disease.

List Of Abbreviations

PSPD
Periosteal stripping and periosteal division
LLD
leg length Discrepancy

Declarations

Ethics approval and consent to participate: This study was conducted after review and approval from the Institutional Ethics Review Committee of Chiba Children's Hospital. Informed consent was obtained from all the participants and their legal guardians., and all methods were carried out in accordance with relevant guidelines and regulations.

Consent for publication: All patients and their guardians gave consent for publication using the opt-out method.

Availability of data and material: The datasets used and/or analysed during the current study are available from the corresponding author on reasonable request.Competing interests: The authors declare that they have no competing interests.

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Authors' contributions:

Ayana Kitta: Data Curation, Investigation, Visualization, Writing-Draft. Takashi Saisu: Conceptualization, Data curation, Resources, Writing-review and editing. Jun Kakizaki: Project administration, Supervision. Yasuhiro

Oikawa and Yuko Segawa: Data curation. Ken Okazaki: Formal analysis, Methodology, Writing-review and editing.

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Figures

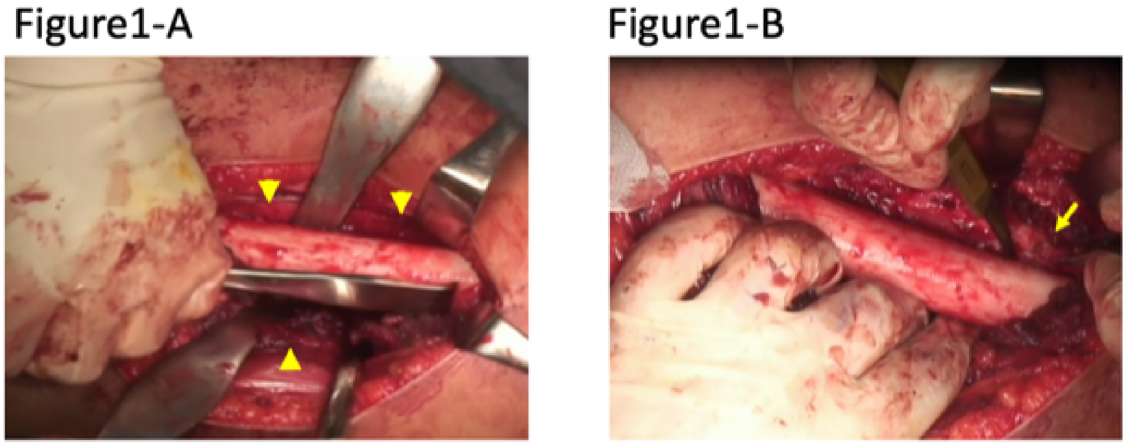

Figure 1

Intra-operative photographs during PSPD. A. Periosteal stripping of the femur. A longitudinal incision is made on the periosteum. Arrowheads: periosteum. B. Periosteal division. A circular incision is made on the periosteum. Arrow: incision on the periosteum.

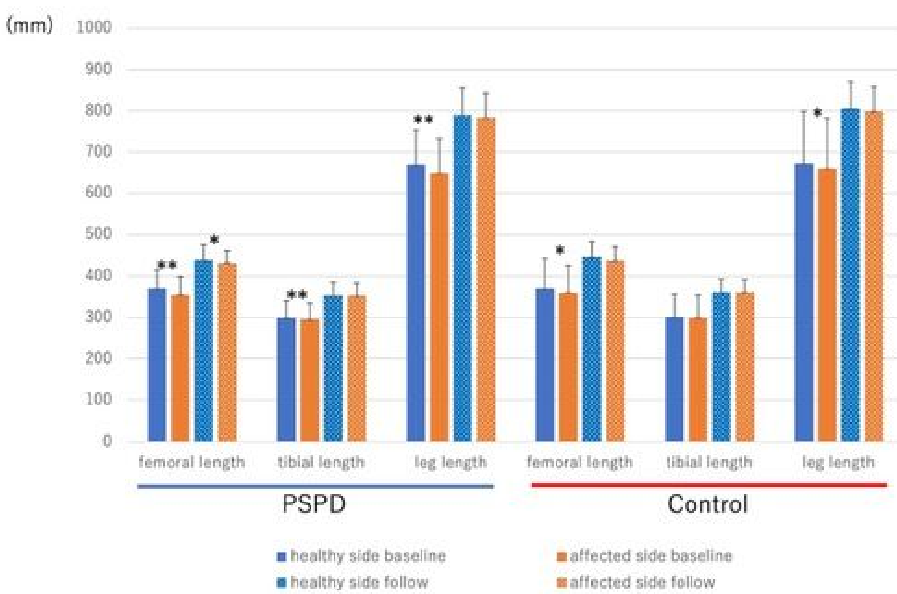

Figure 2

Bone and leg length of the healthy and affected sides at baseline and latest follow-up (mm). PSPD: periosteal stripping and periosteal division. *: $P < 0.05$, **: $P < 0.01$ in Mann−Whitney U test between the sides.

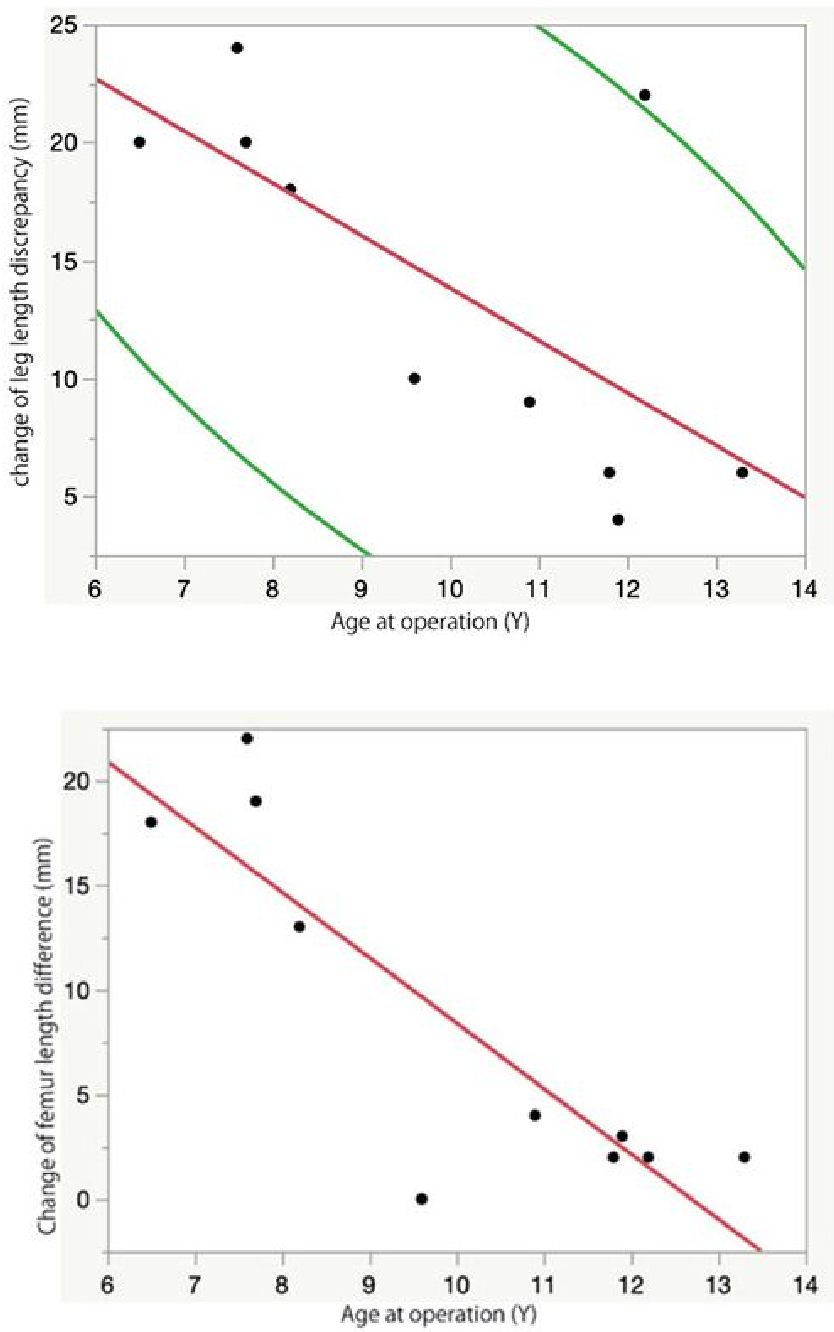

Figure 3

A. Correlation between the age at the time of surgery and change in the leg length discrepancy. B. Correlation between the age at the time of surgery and change in the femoral length difference.

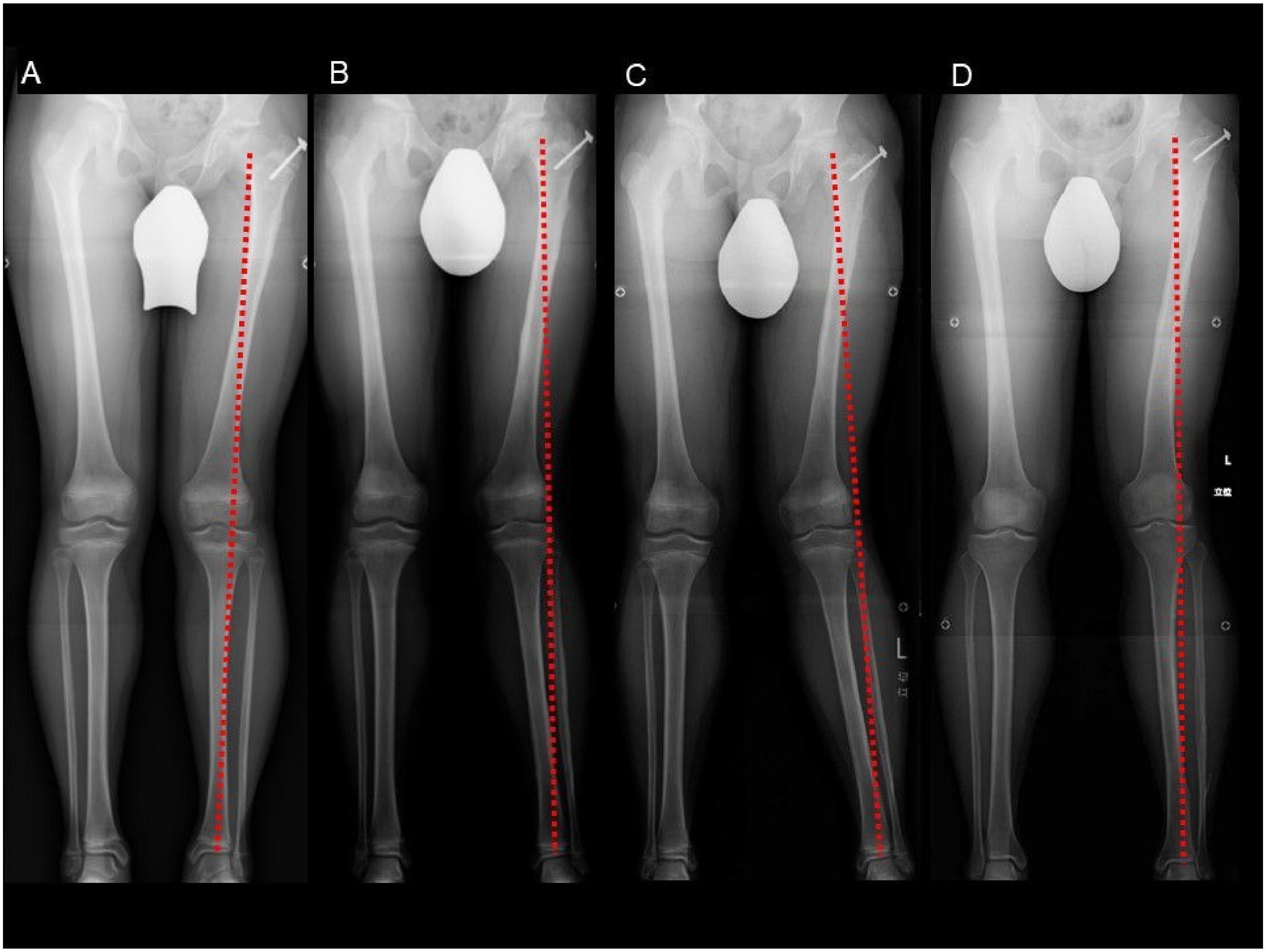

Figure 4

Whole-leg radiographs of a patient who showed a transient genu valgum. A: Before PSPD. B: At 1 year after PSPD. C: At 2 years after PSPD. D: At 3 years after PSPD.

Figure 5

Postoperative scars after PSPD. A, C : Scars of the femur, fibula and tibia at the early postoperative period. B, D : At 6 years after the operation. Scars improved at 6 years after the operation.