CASE REPORT

Limb lengthening for radiation-induced growth arrest of the pelvis and femur

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INTRODUCTION

Aggressive treatment, including surgery and high-dose irradiation, is essential in the treatment of soft-tissue tumors. However, these techniques are not without sequelae. Irradiation of bone in the developing skeleton can lead to significant limb-length discrepancy (LLD) and deformity from growth arrest.[1–3] The source of deformity and LLD may not always be unifocal or completely accounted for in the long bones. Irradiation and surgical treatment of pelvic tumors can lead to significant growth disturbances at both the proximal femur and the pelvis.[4] A scanogram or other radiographic modality that measures hip to ankle length will not accurately depict the entire LLD. Correction of deformity and LLD is paramount in these patients because it can result in gait deviation as well as degenerative changes in the joints of the lumbar spine and lower extremity.[4–6] Long-bone limb-length equalization may not result in complete correction of the deformity leading to patient discomfort. Contributions of the hemipelvis to limb length must be taken into account. Complete evaluation of the lower extremity using clinical examination and full-length standing anteroposterior radiographs is necessary to fully assess deformity and appropriately plan correction.

In this report, we describe the evaluation and treatment of a unique pattern of growth arrest secondary to irradiation. Each patient in this report was informed and consented to the submission of data for publication contained within this report.

CASE 1

A 22-year-old man presented to our institution with complaints of pain in the lower lumbar spine, left hip and thigh, in addition to limited physical activity. The patient reported continuously worsening pain for the past few years and was dissatisfied with conservative measures. He did not wish to use orthoses. The patient had a history of LLD of his left lower extremity, secondary to treatment for rhabdomyosarcoma. The tumor was excised from his left pelvic region at 3 years of age, and the patient later had a recurrence at the age of 5.5 years for which he received radiation, chemotherapy, and autologous bone marrow transplant. Since the completion of his treatment, the patient had not experienced any relapses. Further records regarding previous treatment modalities were not available to the treating surgeon.

On physical examination, the patient walked with a Trendelenberg gait, had visible malorientation of the pelvis, limb-length shortening, and a visible defect of the left iliac crest (Figure 1,A). Examination of the left hip joint yielded 0–120° of flexion, 10° internal rotation, 50° external rotation, 50° abduction and 30° adduction. The left knee flexed from 0–130° with no evidence of instability. Motor power of the left lower extremity was 4/5 hip flexors, 5/5 hip extensors, 4/5 hip abductors and 5/5 knee flexors/extensors. The patient felt comfortable standing on a 3.0-cm block under the affected limb, which leveled the pelvis and spine (Figure 1,B).

A 51-in full-length standing anteroposterior radiograph was obtained with the patella facing forward, using the wooden block under the affected limb (Figure 1,C). LLD was analyzed as described by Paley (Table 1).[6–8] A full extension lateral radiograph yielded no evidence of procurvatum or recurvatum deformity (Figure 1,D).

This patient experienced growth arrest in his pelvis and proximal femur as a result of irradiation before the cessation of skeletal growth. The LLD, including contribution of the pelvis, was about 3.0 cm with the pelvis accounting for approximately 1.2 cm of shortening (see Table 1). This was confirmed by measuring the pelvic height distance (Figure 2). At the proximal femur, there was shortening of the femoral neck, but the neck-shaft angle was normal. Deficiency of the ilium, abductor muscles, and coxa breva contributed to his Trendelenburg gait. With a shoe lift to correct the LLD, his gait improved but not entirely. It was determined that lengthening of the left femur was most appropriate. However, leveling of the pelvis would require over-lengthening of the femur in this patient to account for the pelvic deformity. Clinically the patient was comfortable with a 36-mm block, and it was decided that the goal of lengthening would be the same. The lengthening and plating (LAP) technique was chosen to decrease time required in an external fixator.[9]

The patient was taken to the operating room for application of EBI Biomet Multiaxial Correction Frame (EBI Biomet, Parsippany, NJ) and osteotomy of the distal femur. The patient achieved 3.0-cm lengthening over 30 days. Use of the frame was discontinued at the request of the patient because he was satisfied with the results. The frame was removed from the patient within 2 months, and a distal femoral locking plate was inserted (Figure 1,E). Concurrently, an iliac crest bone marrow harvest was performed using the technique described by Hernigou *et al*.[10] and injected into the distraction gap to promote consolidation. Clinical healing, defined as callous bridging between 3 of 4

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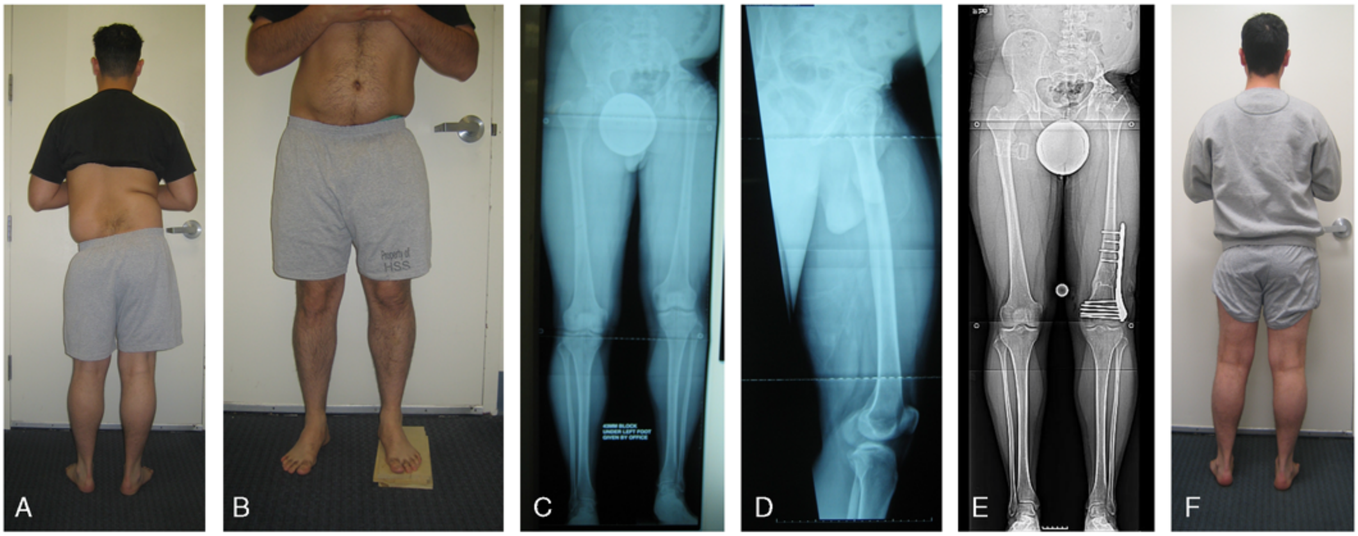

FIGURE 1. (A and B) Preoperative clinical photographs. Note level of spine. (C) Preoperative full-length standing anteroposterior radiograph. (D) Lateral radiograph of hip-thigh. (E) Postoperative radiograph showing frame removed and intact distal femoral plate. (F) Post-treatment photograph from behind. Note leveling of spine.

cortices, was achieved at 4 months. Approximately 13 months later the plate was removed to improve knee range of motion and hardware-related discomfort.

At the latest follow-up of 24-months after treatment, the patient's leg lengths were equal (Figure 1,F). A 51-in standing anteroposterior radiograph was obtained. There was no evidence of sagittal plane deformity on lateral radiographs. Range of motion of the knee was 0–130° of flexion with no instability. There was no change in range of motion or motor power about the hip. The final LLD was 2.0-mm with the left side shorter (see Table 1.) The patient's pain decreased markedly, and there was marked improvement in gait by clinical examination. He was satisfied with the final results.

CASE 2

A 36-year-old man presented to our institution with complaints of a right LLD in addition to discomfort in his lower back and buttocks region. The pain was worse with activity and was improved by lying down. The patient did not desire to use orthoses. At the age of 6 years, the patient was diagnosed with lymphoma in the pelvis. He subsequently was treated with irradiation and surgery. The patient received treatment at a prior institution for his discrepancy where he had an epiphysiodesis of the left distal femur at the age of 14 years. Further records regarding previous treatment modalities were not available to the treating surgeon.

On physical examination, the patient had a Trendelenburg gait on the right side, right pelvic tilt and atrophy of the right buttock. Examination of the right hip joint yielded 0–120° of flexion, 30° internal rotation, 30° external rotation, 50° abduction and 20° adduction. The left knee flexed from 0–140° with no evidence of instability. Motor power of the right lower extremity was 4/5 hip flexors, 5/5 hip extensors, 4/5 hip abductors and 5/5 knee flexors/extensors. Motor power about the ankle joint was 5/5 dorsiflexion, and 5/5 plantarflexion. The patient felt comfortable standing on a 2.0-cm block under the affected limb, which leveled the pelvis and spine.

The patient's deformity parameters were analyzed as before (Table 2.) A full extension lateral radiograph yielded no evidence of procurvatum or recurvatum.

This patient experienced growth arrest in the hemipelvis and proximal femur secondary to irradiation. In addition, there were degenerative and dysplastic changes noted in the right hip and acetabulum. The limb-length

TABLE 1. Summary of leg lengths, angles preoperatively and postoperatively, Patient 1*

Preoperative	Side	Femoral length (cm)	Tibial length (cm)	MAD (mm)	LDFA	MPTA
Hip to ankle LLD: 1.8 cm	Right	47.9	36.0	3 Med	88°	89°
Pelvic difference: 1.2 cm	**Left**†	**45.3**	**36.8**	**3 Med**	**89°**	**87°**
Total LLD: 3.0 cm	Difference:	2.6	−0.8			
Postoperative:	Side	Femoral length (cm)	Tibial length (cm)	MAD (mm)	LDFA	MPTA
Hip to ankle LLD: −1.0 cm	Right	47.9	36.0	3 Med	88°	89°
Pelvic difference: 1.2 cm	**Left**	**48.1**	**36.8**	**0**	**86°**	**86°**
Total LLD: 0.2 cm	Difference:	−0.2	−0.8			
Lengthening achieved: 2.8 cm						

*Negative values indicate the affected segment length is long relative to contralateral extremity.
†Bold characters denote affected extremity.
LDFA, lateral distal femoral angle; LLD, limb length discrepancy; MAD, mechanical axis deviation; MPTA, medial proximal tibial angle.

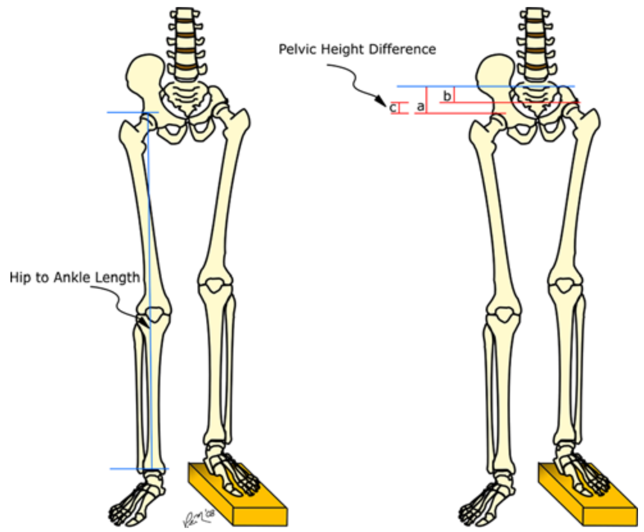

FIGURE 2. A patient with lower extremity shortening with contributions from the femur and pelvis. First, blocks are used to lift the affected leg until the spine straightens, effectively equalizing limb lengths. Hip to ankle length (left) is then demonstrated by drawing a line from the top of the femoral head to the mid-point of the plafond. The pelvic height difference (c, right) is measured by first drawing three horizontal lines at the top of the hip and spine (usually the sacro-iliac joint). Then, two perpendicular lines are drawn between the line at the spine and the hip. The pelvic height difference is determined by subtracting the difference between the two lengths of these perpendicular lines (a and b).

discrepancy including pelvic contribution was about 2.3 cm short, including 1.0 cm of pelvic shortening. This was confirmed by measuring the pelvic height difference (see Figure 2). As most of the correctable discrepancy was found in the femur, the femur was chosen to be the site of lengthening. Clinically, this patient was comfortable with a 2.0-cm. lift, which would be the approximate goal of lengthening. Complete correction of the discrepancy created by arrest in the pelvis and femur would necessitate over-lengthening of the femur.

The patient was taken to the operating room where a distal femoral osteotomy was performed and an EBI Biomet Rail external fixator (EBI Biomet, Parsippany, NJ) was applied. After the completion of the distraction phase, the patient developed a severe knee extension contracture. He underwent a limited incision quadricepsplasty, which involved release of the vastus medialis fascia. The patient achieved 2.3 cm of lengthening (Figure 3,A). During lengthening the patient developed a slight stretch neuropraxia of the infrapatellar branch of the saphenous nerve that resolved. Clinical healing of the distraction gap was achieved at approximately 4.5 months (Figures 3,B and C). During this time, the patient developed a superficial pin track infection that resolved with antibiotic treatment. The frame was then removed at 5 months. Seven months after the removal of frame, the patient developed swelling and erythema in the thigh for which he received antibiotic treatment, and the symptoms resolved. It should be noted that during this time the patient was diagnosed with Hepatitis C, for which he was receiving Interferon and antiviral therapy for immunomodulation. Subsequently, 2 years after the original procedure, the patient developed osteomyelitis in the distal femur at the pin sites and was taken back to the operating room for debridement. The patient received 3-months of antibiotic treatment with no further complications.

At the latest follow-up of 36-months after treatment, the patient's leg lengths were equal (Figures 3,D–F). The patient maintained full range of motion and strength about the knee. There was no extensor lag. There was no change in range of motion or motor power about the hip. A 51-inch standing anteroposterior radiograph was obtained (see Table 2.) There was no evidence of sagittal plane deformity noted on the lateral radiographs. The patient's pain was markedly decreased to his satisfaction, and his gait markedly improved on clinical examination from previously.

DISCUSSION

There are a variety of etiologies that can result in growth arrest including radiation.[11,12] Irradiation may lead to growth arrest in more than one anatomical location causing a complex array of deformities such as LLD, varus or valgus deformity, and procurvatum or recurvatum deformity.[2] Patients receiving extensive radiation and excisional treatment for malignancy do not always undergo limb salvage procedures.[13,14] In these patients, the complete extent of deformity may not be entirely realized until skeletal maturity, presenting much later to the orthopaedist, often when patients are already experiencing severe discomfort.

Distraction osteogenesis using the Ilizarov method may prove to be useful in these complex cases. Distraction osteogenesis (the Ilizarov method) has been shown to be effective in the restoration of anatomical structure and length.[15–18] However, few have reported on the utility of this method in the treatment of growth arrest.[19–21] Deformity as a result of growth arrest is not always completely

TABLE 2. Summary of leg lengths, angles preoperatively and postoperatively, Patient 2*

Preoperative	Side	Femoral length (cm)	Tibial length (cm)	MAD (mm)	LDFA	MPTA
Hip to ankle LLD: 1.3 cm	**Right†**	**50.5**	**41.6**	**8 Med**	**85°**	**85°**
Pelvic difference: 1.0 cm	Left	51.5	41.8	15 Med	86°	86°
Total LLD: 2.3 cm	Difference:	1.0	0.2			
Postoperative:	Side	Femoral length (cm)	Tibial length (cm)	MAD (mm)	LDFA	MPTA
Hip to ankle LLD: – 1.0 cm	**Right**	**52.8**	**36.0**	**13 Med**	**87°**	**83°**
Pelvic difference: 1.0 cm	Left	51.5	36.8	15 Med	86°	86°
Total LLD: 0.0 cm	Difference:	– 1.3	– 0.8			
Lengthening achieved: 2.3 cm						

*Negative values indicate the affected segment length is long relative to contralateral extremity.
†Bold characters denote affected extremity.
LDFA, lateral distal femoral angle; LLD, limb length discrepancy; MAD, mechanical axis deviation; MPTA, medial proximal tibial angle.

FIGURE 3. (A) Two months postoperative radiograph. Note partial consolidation. (B) Four months postoperative radiograph. Note complete consolidation. (C) Patient with frame at the completion of distraction. (D) Six months postoperative standing anteroposterior radiograph with frame removed. (E) Follow-up clinical photograph from front. (F) Follow-up clinical photograph from behind. Note atrophy of hip abductors and leveling of the spine.

accounted for in the long bones, creating a unique challenge for the orthopaedist and patient. In the traditional analysis of LLD of our two patients, a deficit would have clearly remained if the hip to ankle limb lengths were equalized (Figure 2). Despite limb equalization, the patients' would have still continued to experience the symptoms of LLD. In these situations we have found that it is even more imperative to correlate deformity parameters (especially pelvic height difference,) with the entire clinical picture. It should be noted that although not present in these patients, the source of LLD also can reside in the foot.

Typical radiographic measurements of limb length using the femoral head to tibial plafond are not adequate in the analysis of deformity in patients with pelvic contribution. In our experience, we have found that proper analysis of frontal plane deformity and discrepancy is best achieved through the use of a 51-inch standing bipedal anteroposterior radiograph. Recent reports have suggested that the standing bipedal anteroposterior radiograph is more effective in producing reliable results compared with a scanogram.[22] We have found that by using this technique we are able to see the deformity as it applies to the whole extremity. Through this method we are able to determine the contributions of deformity not only from the femur and tibia but the hemipelvis as well.

In this report, both patients' deformity analyses revealed LLD that was present not only in the long bones, but in the pelvis as well. Although the discrepancy may have been treated by orthoses, both patients did not desire this mode of treatment for cosmetic and personal reasons. In addition, this would not treat angular deformity and would result in different knee heights, an undesirable outcome. We have found that lengthening of this magnitude yields predictable results and is a good alternative. Although complications such as infection are possible, we have found this to be a rare occurrence. In our patient database of over 600 lengthenings, we have only experienced osteomyelitis in two patients which cleared after antibiotics or surgical debridement (unpublished data).

Normal bone physiology is essential for predictable healing potential. For this reason, we chose to lengthen the distal femur, a comparably healthy bone segment compared to the irradiated sites of the pelvis and proximal femur. Lengthening in irradiated bone should be avoided. Over-lengthening in both patients resulted in lower extremity lengths including the pelvis that were equal. Both patients achieved satisfactory results in limb functionality and gait. This suggests over-lengthening can be accomplished within the parameters of normal joint orientation with successful results and minimal complications.

In both of our patients, their gait markedly improved with correction of the LLD. However, their gait did not completely normalize because of abnormal abductor muscle function (Figure 3,F). This resulted from coxa breva and intrinsic compromise of the abductor muscle related to previous cancer treatment.

Overcorrection of deformity in the long bones is a strategy that can be effective in the treatment of patients with pelvic deformity contribution. Correction of pelvic deformity often is difficult and invasive.[23] We believe that distraction osteogenesis to overcorrect the long bones is less invasive and preferable. In addition, the concomitant use of lengthening and plating, or lengthening and then nailing will allow expedient recovery and functional improvement.[24]

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