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CASE REPORTS

Lengthening of a Free Fibular Graft after Sarcoma Resection of the Humerus

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We report a patient who had resection of the humerus for osteosarcoma, initial reconstruction with a free fibular graft of the humerus, and subsequent lengthening of the graft. A 9-cm (100% of free fibula length) lengthening was achieved to equalize the humerus length. A complication of regenerate fracture was treated successfully with plating. Four years after the initial surgery for lengthening, the patient presented with a new radial neuropathy.

Advances in the diagnosis and treatment of malignant bone tumors now allows limb-sparing rather than limb-sacrificing surgery.[7,9] Free fibular grafting is one of the standard salvage procedures for reconstructing a segmental bone defect after tumor resection.[3,4,10] However, when a child's growth plate is resected the graft does not provide longitudinal growth. Depending on the child's age, this may result in a progressive limb-length discrepancy (LLD) with functional and cosmetic ramifications.

Distraction osteogenesis is a well-established technique for bone lengthening.[5] There are no published reports regarding lengthening a free fibular graft after sarcoma resection with a monolateral fixator. We wondered whether successful bone lengthening of a free fibular graft could be achieved after a previous wide resection.

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Case Report

A 13-year-old boy presented with 9-cm right LLD discrepancy. At 4 years of age, he was diagnosed with osteogenic sarcoma of the right proximal humerus, which was his dominant arm. At the time of diagnosis he had tumor resection followed by shoulder hemiarthroplasty, which was revised 10 months later to an arthrodesis with internal fixation and vascularized fibular graft. This life-saving limb-salvage treatment controlled the tumor. With time, a progressive LLD developed. He became left-handed after this surgery. On physical examination his shoulder motion through the scapulothoracic joint was limited to 30° flexion and 20° abduction. Elbow and wrist range of motion (ROM) and neurovascular status were normal. The length of the right humerus was 18 cm and the free fibular graft site measured approximately 9 cm (Fig 1A).

The free fibular graft hypertrophied during the 9 years after implantation. We could not quantify the hypertrophy because the initial postoperative radiographs of the shoulder with the free fibula graft were unavailable for comparison.

We planned an osteotomy of the matured fibular graft area at its widest location with gradual 70-mm lengthening. This location was chosen because there was not enough native humerus to perform lengthening. We performed the osteotomy through a 0.5-cm incision using a multiple drill-hole technique and applied a monolateral pediatric external fixator (EBI, Parsippany, NJ) (Fig 1B–C) with hydroxyapatite (HA)-coated half pins. Distraction was started on Day 10 at 0.25 mm three times per day for 10 days, then changed to 0.25 mm two times per day. Compromised biology, which includes lack of normal soft tissue envelope and free fibula graft instead of native humerus, was a factor that influenced a decision

Fig 1A–C. Radiographs show the patient's (A) right humerus before the lengthening procedure and (B) after osteotomy and application of the external fixator. (C) A postoperative radiograph shows the patient after application of the monolateral external fixator.

for a slower distraction rate. We applied an EBI bone stimulator 3 weeks postoperatively to enhance bone healing.

The pin sites remained clean and dry, and the patient reported no pain. He took no pain medications or antibiotics during the treatment. Wrist and elbow ROM remained within normal limits, and there were no neurovascular problems (Fig 2). After achieving the planned 70 mm distraction, the patient still had a LLD but maintained full elbow ROM and normal neurologic function. The bone formation was good, but the patient wanted more length. We continued slow distraction of 0.50 mm per day for approximately 6 weeks.

He achieved 90 mm distraction (50% of preoperative length of the humerus and 100% of the free fibula length) within 6 months followed by 6 months fixation during consolidation. However, delayed regenerate formation was observed during last 2 cm of distraction despite the slow rate. The middle of the regenerated section was thin (0.5 cm diameter) and failed to thicken despite applied compression forces in the frame and weightbearing exercises (Fig 3A–C). External fixation index (time in a frame in months divided by cm of lengthening) was 1.33. We removed the frame and applied a coaptation plaster splint. At the first postoperative visit, the radiographs showed a low-energy oblique fracture through the central part of the regenerate—the narrowest region. We then applied a Sarmiento brace and 1 week later there was angular deformity and minimal shortening (Fig 3D). The patient had open reduction and internal fixation (ORIF) through an

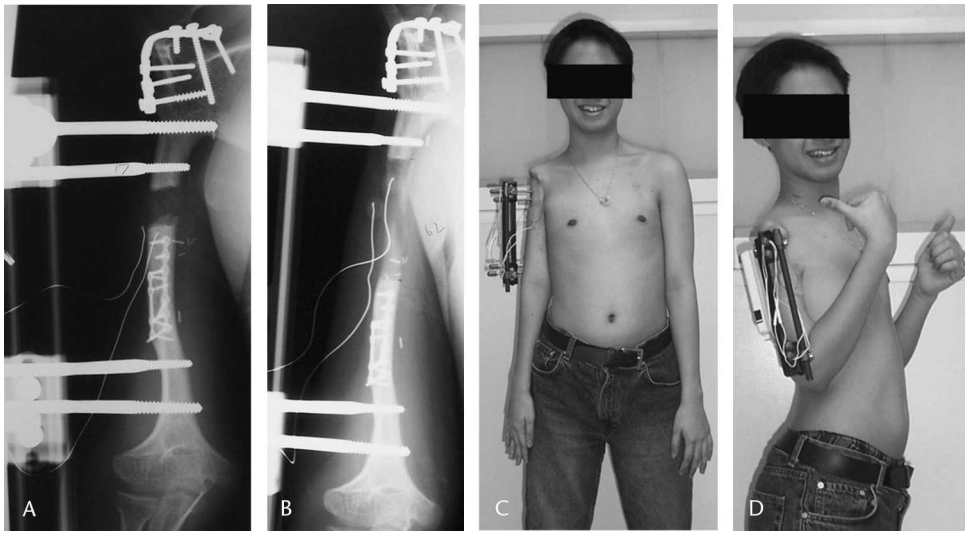

Fig 2A–D. Radiographs show the patient's (A) right humerus at 1 month and (B) at 3 months of distraction. Photographs show the patient during (C) the distraction phase with improved LLD and (D) the patient's elbow flexion during the distraction phase.

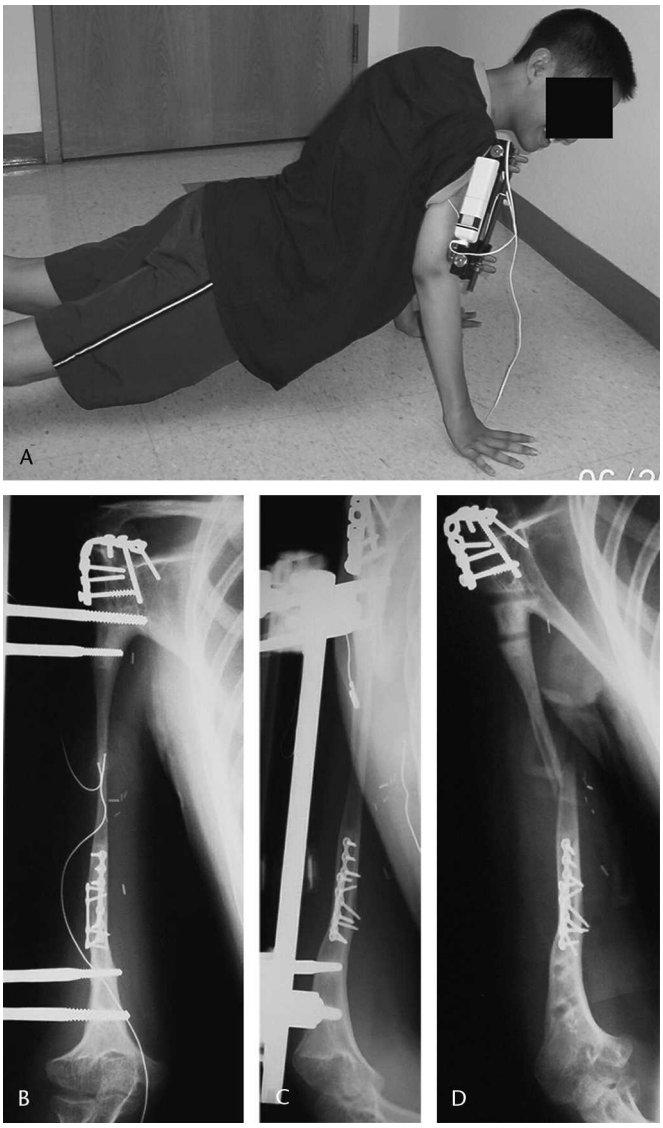

Fig 3A–D. (A) A photograph shows the patient doing weight-bearing exercises during the consolidation phase. (B) Anteroposterior and (C) lateral radiographs show the patient's right humerus before removing the external fixator. (D) A radiograph of the patient's right humerus after frame removal shows a fracture through midportion of the regenerate.

anterolateral approach with a 12-hole locked small fragment plate (Synthes, Paoli, PA). Four screws were placed in the proximal fragment and three in the distal fragment, along with 5 cc Grafton crunch (Osteotech, Eatontown, NJ), a demineralized matrix bone graft substitute. The alignment was corrected, but 7 mm of length was lost. The patient also had an incomplete median neurapraxia develop secondary to compression from the postoperative coaptation splint. At 3 months followup the neurapraxia resolved and radiographs showed a completely healed fracture and widening of the previously narrow regenerate (Fig 4). He had no infection. His right elbow ROM was 0° to 150° extension-flexion, 90° pronation, and 90° supination. Right wrist ROM was 80° flexion and 70° extension. His right elbow, wrist, and hand motor strength were normal (Medical Research Council [MRC] Motor Grading Scale–5/5). Three years after the lengthening procedure, the patient expressed satisfaction with the cosmesis and improved reach. Radiographs showed well-remodeled regenerate and no signs of bone resorption (Fig 5).

Four years after the initial surgery, the patient presented with a radial neuropathy. This was a new finding and was not associated with any trauma. Magnetic resonance imaging (MRI) demonstrated radial neuritis characterized by a long segment of generalized thickening of the radial nerve. There was no evidence of recurrent tumor. Electromyography (EMG) revealed right radial mononeuropathy with axonal degeneration with the lesion in the mid to distal arm. Radial nerve decompression is planned.

DISCUSSION

Experimental and clinical data regarding healing of a distraction regenerate and the intrinsic and extrinsic factors influencing bone formation have been published.[1,5,6] Humeral lengthening has resulted in good bone formation, improved function, and improved cosmetic appearance.[2,6] However, we are unaware of any report on lengthening a vascularized free fibular graft after sarcoma resection. Limb reconstruction with free fibular transfer after resection of bone tumors has become one of the standard procedures.[3,4,10] However, growing patients may need additional surgeries to equalize LLD when there is growth plate involvement. There are no guidelines available regarding potential bone healing of the graft, rate and rhythm of distraction, amount of lengthening, or the best time to perform this procedure. We did not meet this patient until 8 years after he had free fibula surgery. We think the best time to do this is after the end of growth so that the lengthening goal is clear. Also, it seems best to do this after maximal hypertrophy of the fibula.

Our patient had a substantial LLD and a fibular graft had substituted ½ the original humeral length. He also had a shoulder arthrodesis and lacked muscle coverage on the shoulder girdle. The old fixation plate was embedded in the bone while bridging a junction of a native bone and a graft. We planned a 70-mm lengthening through a matured free fibular graft just above the plate because of the short amount of native bone below the plate at the elbow, the amount of lengthening we deemed possible, and the desire to preserve elbow function. We did not have information regarding the location of the nutrient vessel, although it could be a crucial issue to know the distribution of intra-

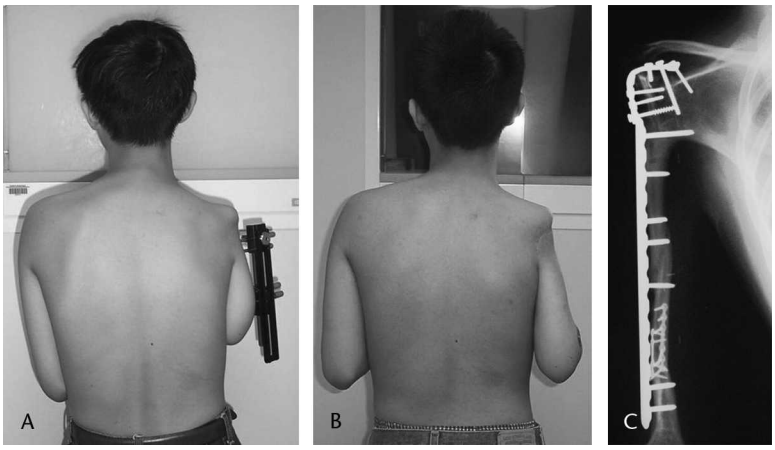

Fig 4A–C. Photographs show (A) the patient's LLD before lengthening and (B) after lengthening. (C) A radiograph shows the patient's right humerus 6 months after ORIF for the regenerate fracture.

medullary versus periosteal blood supply in a vascularized fibula. The osteotomy location was chosen at the widest area of free fibula (FF) and based on the available pin-site location.

Considering the situation, we predicted slower than usual bone formation. Therefore, the initial rate of distraction was only ¼ turn three times a day and then decreased after 10 days to two times a day. At this rate of ¼ turn two times a day, 70 mm of lengthening was achieved with radiographic signs of bone formation. However, the final 20 mm distraction resulted in a decrease in bone formation leading to narrowing of the regenerate midportion despite the slow distraction rate.

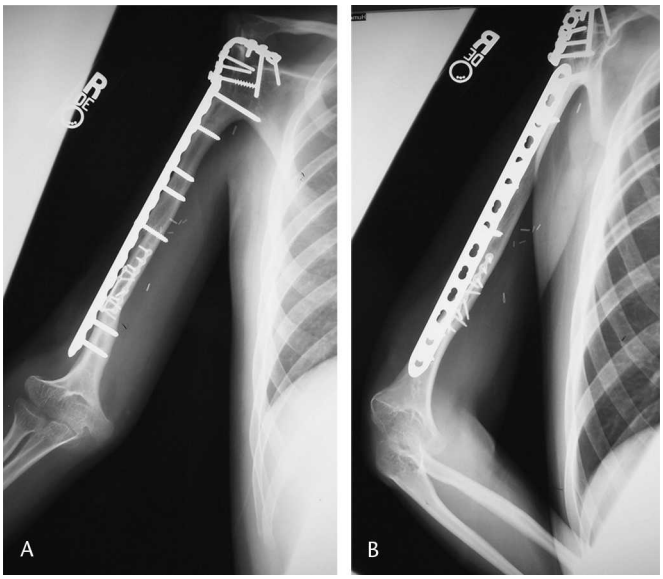

Fig 5A–B. (A) Anteroposterior and (B) lateral radiographs show the right humerus at 3 years followup.

The new problem of radial neuritis is concerning and could be the result of extreme lengthening (50%) in combination with an already compromised nerve. This presented 3 years after lengthening with no prior signs of radial nerve compromise.

Shaw et al reported 8 cm lengthening of the distal humerus through a native bone after osteogenic sarcoma resection and reconstruction with a vascularized fibular graft using a monolateral external fixator during 6 months with consequent persistent 20° elbow flexion contracture.[11] Lim et al reported a case of a free fibular graft lengthening in a child after neonatal osteomyelitis of the humerus using circular external fixation with a good result.[8] The transferred bone behaved like normal bone to distraction, and 6 cm lengthening was achieved over 3 months.[8] A circular fixator was used for 5 months with an additional 8 months in a splint to prevent stress fractures.[8]

We found distraction osteogenesis of a free fibular graft resulted in bone formation. However, this regenerate was not robust and at risk for refracture. We showed that successful lengthening can be performed using a monolateral frame, and a regenerate fracture can be treated successfully with open plating and bone grafting. However, we recommend a rate of distraction of 0.5 mm per day or less to sustain good regenerate formation and perhaps limit the amount of lengthening to 60 to 70 mm each treatment.

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