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Case Report

Physal Distraction During Surgical Treatment of Osteosarcoma of the Femur

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Abstract

Introduction

First report of an acute physal distraction in preserving the uninvolved metaphysis in osteosarcoma patient.

Case presentation

We treated a 9-year-old female, Asian heritage patient with osteosarcoma of the diaphysis and metaphysis of her left femur which left an intact epiphysis and growth plate and was not suitable for the Cañadell technique of physal distraction. After neoadjuvant chemotherapy, she received tumor resection, acute physal distraction (1-2mm/min), the involved femur inactivation and re-implantation sequentially. 4 cycles of adjuvant chemotherapy were taken in this patient. The whole surgery lasted for about 3 hours, with a total blood loss of less than 600 ml. The epiphysis was separated at 35 minutes. The wound healed well. During the following up of 38 months, her limb discrepancy reached 10 cm with no tumor relapse or metastasis. The revised Musculoskeletal Tumor Society score increased from 8 of preoperative value to 23 postoperatively. The main problem is bone healing, limitation of knee joint function and involved limb shortage.

Conclusion

Physal distraction during surgery is an alternative treatment of Cañadell technique in given patients with satisfied clinical result except for remarkable limb discrepancy.

Keywords: Case Report; Osteosarcoma; Physal Distraction; Limb Saving Surgery; Growth Plate; External Fixators; Fixation

Introduction

Osteosarcoma is a primary, malignant, bone tumor that occurs in young, growing patients [1]. The most frequently affected parts are the distal femur and proximal tibia [2]. With improvements in adjuvant chemotherapy, the survival rates of patients with osteosarcomas have increased from 20% to 60%–80%. In recent years, amputation can be avoided in 80%–90% of patients owing to the availability of various limb-saving procedures, such as tumor prosthesis implantation and biologic reconstruction [3], which can adapt to the long-term functional demands of these patients.

In some patients, osteosarcoma occurs before the growth

plate has fulfilled its function of lengthening the bone. In these patients, tumor excision and prosthetic replacement may result in considerable limb-length discrepancy because the epiphysis is sacrificed [4]. To avoid the loss of limb function and length, surgeons have attempted physal distraction to save the uninvolved epiphysis and used biologic reconstruction with allografts or autografts to repair the defect resulting from tumor resection [5]. Jose Cañadell [6] first developed this innovative technique in 1994 - using external fixation devices, controlled distraction of the epiphysis from the growth plate permits iatrogenic separation of the epiphysis from the affected region, achieving a safe margin. Favorable clinical outcomes with excellent growth and normal function of the affected limb have been obtained after

such treatments [7,8].

In patients in whom the tumor fills the entire medullary cavity of a long bone but does not involve the physis and epiphysis, amputation is not a good choice. However, conventional physal distraction cannot be performed because the distraction nail passing through the metaphysis or epiphysis can contaminate the surrounding normal tissue with tumor cells, which may cause tumor relapse after limb-sparing surgery.

To resect the tumor en bloc and save the uninvolved epiphysis, we used a new method for intraoperative physal distraction and obtained acceptable results. Here, we describe the method and present our findings.

Case Presentations

General information

Our patient was a 9-year-old female, Asian heritage patient. She had pain and swelling of the left thigh for 4 months. Magnetic resonance imaging (MRI) showed tumor invasion most of the diaphysis and metaphysis of her left femur with an intact epiphysis and growth plate. The closest distance between the tumor and the growth plate was 0.5 cm. She underwent a core needle biopsy, and a pathologic diagnosis of osteosarcoma was obtained. The Enneking stage was IIB.

She was treated with two cycles of neoadjuvant chemotherapy with high-dose methotrexate (12 g/m²), ifosfamide (10 g/m²), doxorubicin (90 mg/m²) and carboplatin (400 mg/m²). Another MRI examination was performed before operation to evaluate the results of chemotherapy. The imaging data showed significant reductions in tumor size and diameter (Figure 1).



Figure 1. Magnetic resonance imaging (MRI) performed ① before and ② after neoadjuvant chemotherapy. The tumor has reduced in size and diameter.

Surgical Technique

Two weeks after the neoadjuvant chemotherapy, the patient underwent tumor resection, and femur bone inactivation and re-implantation.

Position and incision: The patient was placed in the dorsal decubitus position, and general anesthesia was induced. An anterolateral incision was made on the left thigh, from the anterior superior iliac spine to the lateral femoral condyle.

Tumor resection: The femur was separated from the surrounding normal muscles, but a 2–3-cm layer of normal soft tissue covering the tumor was left intact. The femoral vessels and sciatic nerve were identified and protected during the operation. Next, a V-shaped subtrochanteric osteotomy was performed (Figure. 2a). The separated femur was packed with 3M protective coating and bandages. The operative field was also covered with the protective coating (Figure. 2b).

Physal distraction: Two Steinmann pins were inserted through the epiphysis and one through the metaphysis under radiographic monitoring with a C-arm X-ray machine such that the growth plate was left intact. The outer parts of the Steinmann pins were coated with anhydrous alcohol for 10 min, and the pins were fixed with half-ring external fixators. The physis was distracted at a rate of 1–2 mm/min until the epiphysis was separated (Figure. 2b, 2c). The remaining soft tissue around the physis was then separated, and the affected bone was removed from the operative field. The histological study showed the margin is free of tumor during and after operation.

Inactivation and re-implantation of the femur: On another operating table, the tumorous cortex and medullary cavity were separated from the rest of the femur (Figure. 2d). The femur was then fixed with four interlocked, bended plates that followed the shape of the greater trochanter and lateral femoral condyle, at both ends (Figure. 2e). The bone was placed in anhydrous alcohol for 40 min and then washed with a large amount of physiological saline. Autologous fibular bone grafts harvested from the same side of leg were used to fill the femoral cavity. The inactivated bone was then reduced and fixed with the femoral head and femoral condyle (Figure. 2f). Shavings of the iliac crest were used to fill the junction between the separated bones. The wound was then closed.

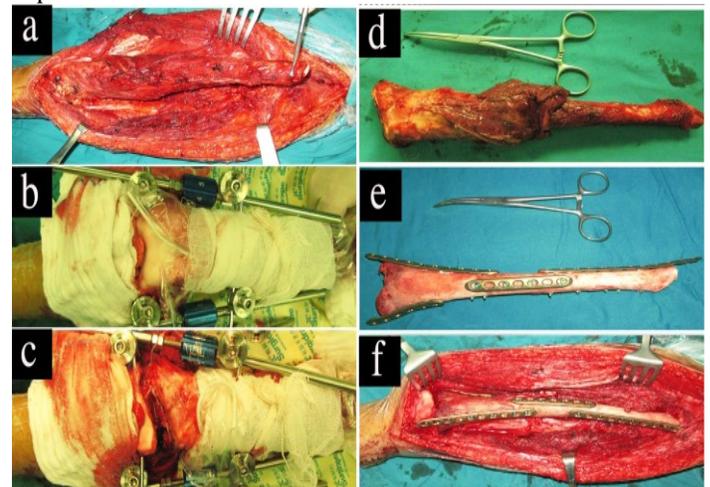


Figure 2. Physal distraction and femur bone inactivation and re-implantation.

① The femur is separated with a subtrochanteric osteotomy. The femur ② before and ③ after distraction. ④ Completely separated femur with a layer of normal tissue. ⑤ The femur is fixed with interlocked, bended plates. ⑥ The inactivated bone has been re-implanted.

The whole procedure lasted for about 3 hours, with a total blood loss of less than 600 ml. The epiphysis was separated at 35 minutes.

Postoperative management and follow-up

Postoperatively, the patient received intravenous ciprofloxacin as standard antibiotic prophylaxis until all drains and catheters were removed. Radiographs obtained 1 week later showed excellent reduction and fixation of the femur (Figure. 3b) according to preoperative radiographs (Figure. 3a). Pathological examination of the surgical specimen showed no tumor cells at both ends of the separated bone. After wound healing, she received another four cycles of chemotherapy with the same 4 agents.

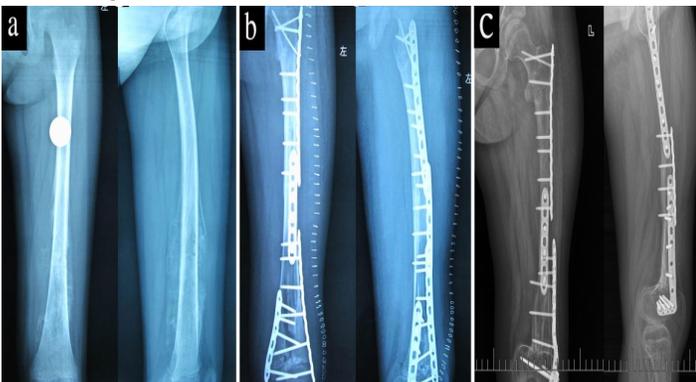


Figure 3. Radiographs of the involved femur ① before, ② after, ③ 38 months after the operation. Anatomical reduction and strong fixation of the femur bone are observed.

The patient was followed up every 3 months after the surgery. At each visit, radiographs of the chest and the local site were taken, and the patient was examined. At the following up of 38 months, no tumor relapse or metastasis occurred according to clinical symptoms, pulmonary CT and whole body ECT. Limb discrepancy was 10 cm and the range of knee joint movement reached 90° in flexion and 0° in extension at the last visit (Figure. 4a, 4b).

The revised Musculoskeletal Tumor Society score increased to 23 postoperatively, from its preoperative value of 8. In order to avoid epiphyseal arrest, we took out the screws fixed in the epiphysis at 12 months after surgery. At the following up of 18 months, nonunion occurred at the subtrochanteric junction (Figure. 5a). We changed the screw position and re-implanted with autogenous iliac bone. Bone healing was observed at 24

months (Figure. 5b). Another bone resorption and displacement happened at the distal medial metaphysis from plain films at 24 months after operation (Figure. 6a). Autologous fibula and allograft bone was implanted with replacement of internal fixation. Bone union obtained at 38 months of following up (Figure. 3c, 6b).



Figure 4. Appearance and X-ray film of involved limb at the following up of 38 months. ① appearance of the involved limb. ② 10 cm of limb discrepancy shows by plain film.



Figure 5. Radiographs of the subtrochanteric junction at 16 and 24 months after surgery. ① Nonunion occurred at the subtrochanteric junction at 16 months. ② Bone healing was observed after autogenous iliac bone implantation and screw position adjusted at 24 months.



Figure 6. Radiographs of the metaphysis junction at 24 and 38 months after surgery. ① Bone resorption and nonunion happened at the distal medial metaphysis. ② Autologous fibia and allograft bone was implanted with replacement of internal fixation and bone union was obtained.

Discussion

In growing children, progressive limb-length discrepancy following the removal of the growth cartilage is a major problem in the long-term management of osteosarcoma. In 1994, Cañadell et al. [8] first described an innovative solution to this problem: increasing the resection margin while preserving the epiphysis in young patients with metaphyseal tumors close to the growth plate. From 2007 to 2011, we have used this method in six young patients and obtained excellent postoperative limb function and encountered few complications, with a discrepancy of only 1–3 cm between the two lower limbs [6].

However, this method cannot be used in patients in whom the tumor has invaded most of the long bone or filled most of the medullary cavity. The insertion of pins into the diaphysis, which is required during the above method, can contaminate the surrounding normal soft tissues with tumor cells and possibly lead to tumor recurrence. In addition, tumor has the safe distance from growth plate, osteoarticular allograft or tumor prosthesis implantation is not an acceptable alternative, because this may be associated with great limb-length discrepancy or prosthesis loosening after the operation. We therefore used physal distraction to separate the tumor and spare the growth plate. The epiphysis was separated in about half an hour; the growth plate was intact, and tumor cells did not contaminate the epiphysis.

We used a novel method of epiphysiolysis that has not been reported by Cañadell et al. [8], which was easy manipulation, no tumorous or infectious contamination of the nail channels and low risk of injury to the vessels and nerves during distraction. In Cañadell's opinion, distraction is through the physis at a low rate of 1–1.5 mm/day [9], the growth plate is disrupted at the degenerative layer of cells [9,10]. But in this case, a much stronger force and high rate of 1–2 mm/min is exerted on the growth plate, and this is associated with the latent distroy of growth plate. In addition, nail insertion into the bone area involved by the tumor may contaminate the operative field. Therefore, meticulous protection of the normal tissue and anhydrous alcohol treatment of the nails that inserted into the diaphysis are critical steps in our method of physal distraction. In this patient, we chose to inactivate and re-implant autologous bone instead of using allografts because of two reasons. First, the involved bone had an intact cortex with no tumor invasion. Second, matching allografts are difficult to obtain in children. During the following up, the main shortage is bone resorption and nonunion of the subtrochanteric and metaphysis junctions which need bone re-implantation. Long time of

arthrorisis may also caused ankylosis and limited movement of knee joint. Another shortage is limb discrepancy with poor epiphyseal growing function caused by distroy of growth plate. Since our study involved a single patient and a short follow-up, we cannot determine whether our novel method was as safe as the distraction technique reported by Cañadell et al [8]. However, in some circumstances, this new method may be a feasible alternative treatment.

Conclusion

Physal distraction during surgery is an alternative treatment of Cañadell technique in given patients with satisfied clinical result except for markable limb discrepancy.

Consent

All authors have reviewed the final version of the manuscript and approve it for publication; Neither this manuscript nor one with substantially similar content by the authors has been published elsewhere or is being considered for publication elsewhere; The manuscript has been submitted with the full knowledge and approval by all the authors.

Written informed consent was obtained from the patient's legal guardian(s) for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Competing interests

There was no conflicts of interest or funding sources of this study.

Authors' contributions

All authors have participated sufficiently in this work to take responsibility for it.

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No.

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