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Case report

Periosteal ostoesarcoma of the femur treated with modified capanna procedure: A case report

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ABSTRACT

Introduction: Periosteal osteosarcoma is a rare type of primary bone tumor. A vascularized fibula graft incorporates this revolutionary approach with a traditional massive allograft to reconstruct large femur and tibia defects during oncological resection. A structurally competent reconstruction with improved vascular and osteogenic capacities with the ability to achieve lower rates of fracture, infection, and non-union is obtained by integrating the benefits of the separate components.
Method: A 16-year-old female diagnosed with periosteal osteosarcoma of the left shaft femur. We performed neoadjuvant chemotherapy, limb salvage surgery consists of surgical resection and reconstruction, followed by adjuvant chemotherapy post operatively. We used Capanna procedure to salvage the femur.
Result: Post-operative evaluation showed stable fixation clinically and radiologically. There is no complications observed during recovery, as both distal motor and sensory are normal eventhough the patient were still limited in the motion of the hip and knee at the time due to post-operative pain.
Discussion: Cappana procedure has been known as a novel surgical method that could decrease the risk of complications results from classic reconstruction method, such as fracture, non-union, and infection.
Conclusion: Modified Cappana procedure which introduce the use liquid nitorgen-recycled autograft from the resected affected bone as a peripheral shell supporting a centrally placed vascularized fibular graft to fill the massive bone defect left by surgical resection, had successfully performed in our patient whom previously diagnosed with periosteal osteosarcoma of femoral shaft.

1. Introduction

Periosteal osteosarcoma is a rare type of primary bone tumor that occurs as surface lesion. It affects young patients as intermediate-grade osteosarcoma. The most common locations are tibia (40%) and femur (38%), followed by humerus and ulna (5-10%). Age, gender and tumor position are multiple factors that have been identified to determine the predilection of periosteal osteosarcoma. Age has been shown to be a major factor impacting periosteal osteosarcoma incidence. The peak occurrence of periosteal osteosarcoma is in the age range of 15-30 years [1,2].

Swelling followed by pain or discomfort is the most common clinical symptoms of periosteal osteosarcoma. Tumor signs may be present years prior to the first surgical intervention due to slow progressivity. The predilection of periosteal osteosarcoma is in the long bones of the limbs, namely femur, tibia, humerus, radius, fibula, ulna and phalanx. The lesion is predominantly metaphyseal and occasionally diaphyseal. Bertoni et al. identified the primary location of periosteal osteosarcoma as the middle upper and lower diaphysis of the tibia and femur. Due to closely related histologic or radiological findings, periosteal osteosarcoma can be confused with Ewing's, central cancellous, parosteal or central cancellous carcinomas [1,2].

A broad-based surface soft tissue mass has been documented in the radiological appearance of periosteal osteosarcoma, leading to extrinsic erosion of the thickened diaphysal cortex along with the periosteal reaction that invades the soft tissue portion. The periosteal reaction to the

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diaphyseal cortex is primarily perpendicular. Magnetic resonance imaging (MRI) is performed to detect the presence of a soft tissue mass attached to the cortex, any associated aberrations in the medullary canal, cortex thickening, cortical scalloping, periosteal reaction perpendicular to the diaphysis and extending through the soft tissue mass [3,4].

For the management of the tumor, proper surgical intervention is indispensable. If prompt and sufficient surgical care is conducted, the risk of survival is increased and the risk of metastasis is significantly reduced. Treatment of periosteal osteosarcoma is mostly surgical procedure, such as amputation, limb salvation, and resection, which may or may not be followed by chemotherapeutic management. To support surgical procedures, adjuvant chemotherapy may be recommended for pharmacological support. An adjuvant regimen of cisplatin and doxorubicin has sometimes been prescribed, but the result remains inconclusive [1,5,6].

Neoadjuvant chemotherapy has revolutionized the management of primary bone tumors over the last three decades. Long-term survival rates for osteosarcoma have increased from 20% to over 60%, and lower-limb salvage surgery has improved as well. Many patients have avoided amputation while on chemotherapy. Huge allografts are commonly used in long bone reconstruction. Nonetheless, they can cause serious issues like non-union, infection, and fracture. The vascularized fibula graft is the most commonly used vascularized bone graft in intercalary femur and tibia reconstructions. While the transferred fibula hypertrophy strengthens the repair, it is susceptible to early fracture and requires prolonged immobilization of the affected limb [1,7,8].

Capanna and colleagues have lately identified a novel approach to limb salvage. A vascularized fibula graft incorporates this revolutionary approach with a traditional massive allograft to reconstruct large femur and tibia defects during oncological resection. A structurally competent reconstruction with improved vascular and osteogenic capacities with the ability to achieve lower rates of fracture, infection, and non-union is obtained by integrating the benefits of the separate components [1,9–11]. This case report has been reported in line with the SCARE Criteria [12].

2. Case illustration

We presented a case of a 16-year-old female complained of worsening pain in the left thigh in the last 2 months. The patient started to feel pain since 13 months prior to admission that increase in severity progressively. On physical examination, we observed a palpable solid and immobile mass on the middle part of the left thigh with tenderness VAS 3-4. No sign of venectation and no distal edema (Fig. 1). Gait disturbances were observed despite the ability to walk without using aid. Both active and passive range of motion of the hip, knee, and ankle was normal.

Radiological examination was conducted with left femoral x-ray that showed an expanding well defined lytic geographic lesion on the medial mid-diaphyseal area, intact opposite cortex, narrow transitional zone, and calcification matrix (Fig. 2). Chest x-ray examination showed no pulmonary metastasis. Further examination with MRI showed a well-defined margin heterogenous hyperintense lesion on the medial cortex of the femur, without the involvement of the surrounding neurovascular structure (Fig. 3). Laboratory examination showed slight increase in ESR with normal serum ALP level. An open biopsy was performed from the anteromedial side of the left thigh and the histopathology report showed pieces of connective tissue with spindle-shaped cells accompanied by the formation of matrix chondroid matrix, and visible osteoid at the edge of the chondroid zone with hemorrhagic areas. It was clinically supportive to the diagnosis of periosteal osteosarcoma.

The treatment modality on this patient including series of neoadjuvant chemotherapy, limb salvage surgery consist of surgical resection and reconstruction, followed by adjuvant chemotherapy post operatively. Patient had four cycles of neoadjuvant chemotherapy in the

Fig. 1. Clinical Pictures of the left thigh region on preoperative Examination. The picture was taken post open biopsy procedure and 4 cycles of neoadjuvant chemotherapy.

spent of 4 months with the regimens of Ifosfamid, Adriamisin, and Mesna. Following neoadjuvant chemotherapy, patient underwent intercalary resection of the tumor and reconstruction using the modified Cappana procedure, where we used a liquid nitrogen-recycled autograft as peripheral shell supporting a centrally placed micro-vascular fibular graft from the ipsilateral cruris to fill the bone defect left post removal of the tumor.

The patient was placed on supine position. Medial longitudinal femoral approach was conducted while keeping the continuity of the biopsy tract. The interval between sartorius and vastus medialis were opened, exposing the superficial femoral artery and vein along with saphenous nerve. Intercalary resection was done about 1 cm distal and proximal from the tumor, with a cuff of vastus medius and intermedius surrounding the tumor. We excised approximately 14 cm of the affected segment, removal of soft tissue, curettage the tumor, and soak it with liquid nitrogen for 20 min. Then, it was thawed in room temperature for 15 min and thawed using sterile saline for 10 min. We incorporated the vascularized fibular graft harvested from the ipsilateral cruris and fixated it using 3,5 mm cortical screw. The graft was then anastomosized; 1 graft each to branch of superficial femoral artery and vein. The recycled segment then was put back to the remaining healthy femur and fixated using a 4,5 mm broad LCP. The preservation of the surrounding neurovascular bundle was achieved during the surgery (Fig. 4). It took us 7 h to finish the operation with a total of 1600 cc estimated blood loss.

Post-operative evaluation showed stable fixation clinically and radiologically (Fig. 4A). No complications observed during recovery, as both distal motor and sensory are normal eventhough the patient were still limited in the motion of the hip and knee at the time due to post-operative pain. The patient able to perform non-weight bearing ambulation using bilateral crutch on day 7 after the surgery.

The histopathology examination from resected tumor showed fragments of bone trabeculae with a small amount of chondromyxoid matrix accompanied by the formation of hemosiderophage. There were also

Fig. 2. A) Preoperative Femoral X-Ray, 2B) Preoperative Femoral MRI; A) T1-weighted axial imaging, B) T2FS-weighted axial imaging, C) T1-weighted sagital imaging, D) T2FS-weighted coronal imaging.

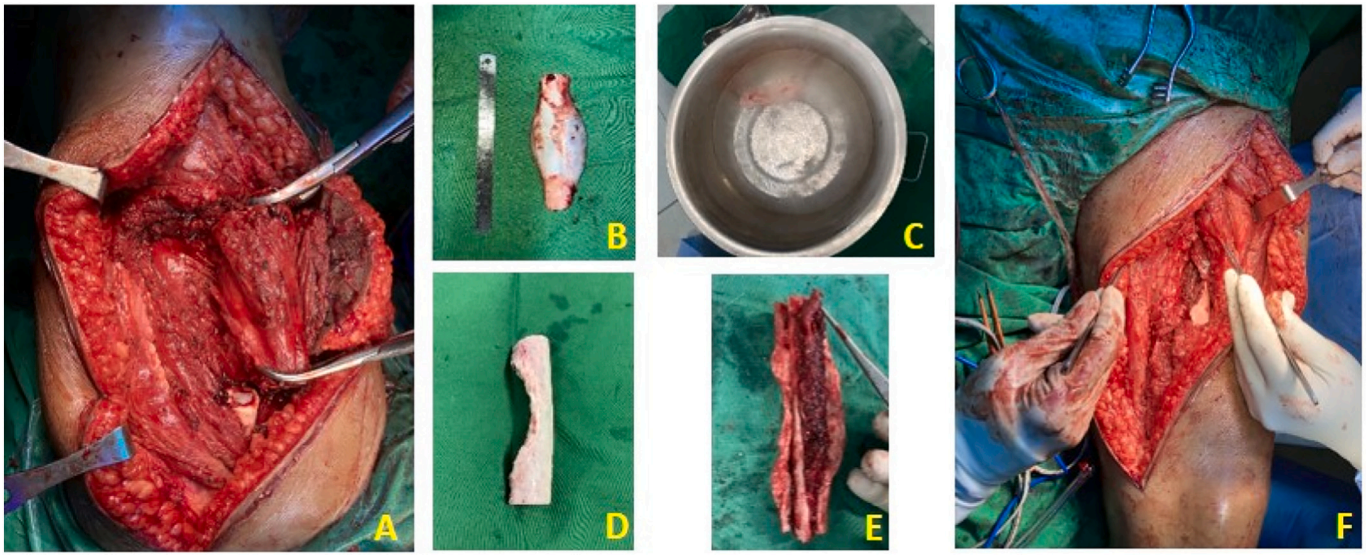

Fig. 3. Modified Cappana Procedure Performed in Our Patient; 3A) Intercalary resection, 3B) Post removal of surrounding soft tissue and tumor curretage, 3C) Affected segment was soaked in liquid nitrogen, 3D) Recycled autograft, 3E) Incoroporation of free-vascularized fibular graft to the autograft, 3F) Post graft fixation and vascular anastomoses.

neoplastic areas consisting of mild to moderate atypical spindle cells. At the peripheral area, the cells arranged lobulated and mitotic activity can be found (Fig. 5).

On 2 months post-operative follow up, the patient reported pain with superficial surgical site infection. The infection was resolved after surgical debridement and antibiotic administration as the histopathology sample from the procedure showed a section consisted of fat surrounded with septum of connective tissue, and there were also necrotic and connective tissue with infiltration of mononuclear and polimorfonuclear inflammatory cells. The conclusion that it is a secondary infection instead of recurency. No complication recorded on the donor site on the ipsilateral cruris. The patient then continued adjuvant chemotherapy protocols using the same citostatic agent used pre-operatively for 6 cycles. Seven months post surgery, the patient able to perform painless full weight bearing on the affected limb. The patient also underwent physiotheraphy and muscle strengthening exercise during rehabilitation. A

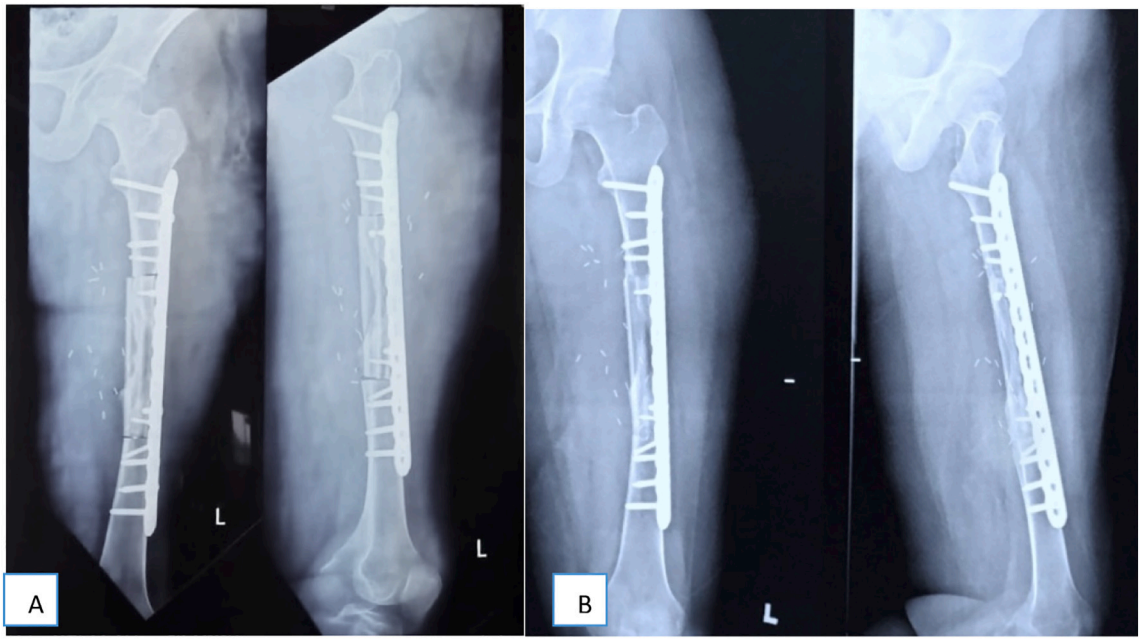

Fig. 4. Post operative radiograph of the Left Femur; 4A) Post-operative, 4B) 7-month post-operative follow up.

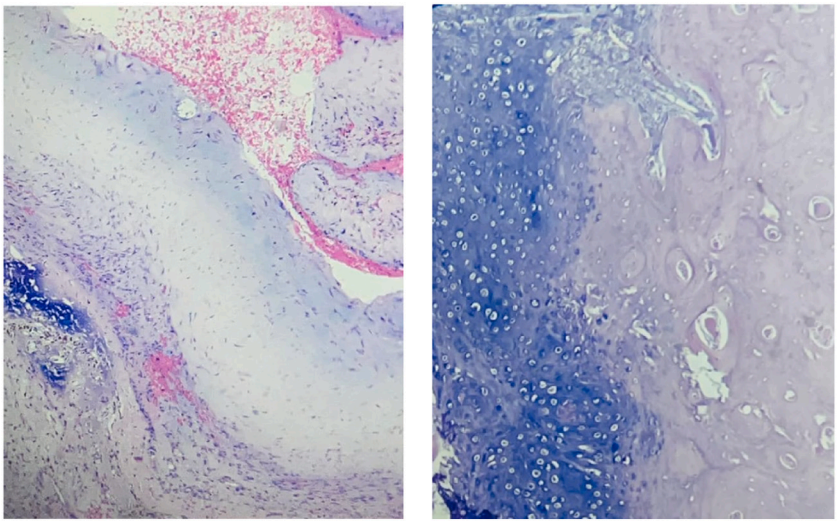

Fig. 5. Histopathology examination.

follow up radiograph examination showed incorporation and no evidence of recurrency (Fig. 4B). We also objectively measure the functional outcome of this procedure through The Musculoskeletal Tumor Society (*MSTS*) *scoring* system, which came out 11 (36%) pre-operatively and 23 (76%) at 7 months post-operatively (out of maximum score of 30).

3. Discussion

The classic surgical intervention which comprises of limb salvage and reconstruction using massive allograft has increased the long-term survival rate and allowed patients with periosteal osteosarcoma not to undergo amputation. Nonetheless, the convensional or some might call it traditional method could increase the risk of complications including non-union, fracture, and infection. Cappana procedure has been known as a novel surgical method that could decrease the risk of complications results from classic reconstruction method, such as fracture, non-union, and infection [1,11]. Hence, we decided to perform a modified version of the known Cappana procedure to treat our patient. The procedure main difference between the original and the modified version that the later used a recycled autograft instead of allograft to act as a peripheral shell supporting a harvested vascularized fibular graft. Vascularized bone graft have osteogenic property and high union potential. However, when a vacularized free fibula is used to reconstruct distal femur defects, many problems are caused. There is a significant mismatch between the size of large host bone and the slender donor bone. Poor contact between the host bone and the donor bone when the fibula is used for distal third femoral defects may compromise the chances of union. The use of merely a single fibula may not support adequate mechanical stability if the defect is large. The fibular graft is not strong enough and there are high chances of fracture. A double-barrel fibular graft has been suggested to solve this problem and to increase the strength of the graft. Nonetheless, when a double-barrel graft is utilised, the length of the defect that can be reconstructed becomes significantly lower. So, it

would be a major challenge to reconstruct defects of more than 12 cm [13–16].

Jayaramaraju D et al. had reported the result of modified capanna's technique using vascularized free fibular graft combined with allograft for post-traumatic long-segment defects of the lower end of the femur [13]. They reported that the bony union was achieved at mean of 6.6 months (range: 5-9). The mean of knee flexion was 90 degrees (range:45–110 degrees) and the mean of lower extremity functional score (LEFS) was 63 (range 46-72) [13]. Venkatramani H et al. also reported that the procedure of modified Capanna's technique resulted in graft union with average time of 6 months [15]. Those results are comparable to our result in which there was bony union at seven months follow-up. The patient in this current case also reported absence of pain and ability to perform full weight bearing. In the previous study by Jayaramaraju D et al., of the 19 patients who underwent modified capanna's technique using vascularized free fibula combined with allograft, superficial infection was reported in two patients. Infection with nonunion and graft resorption was reported in one patient. Three patients had delayed union and two patients had partial skin island necrosis of which one patient had deep infection [13]. In the current case, the patient also had superficial surgical site infection at two months follow up but the infection was resolved after surgical debridement and antibiotic administrastion.

Rahman MA et al. had performed reimplantation of the resected tumor-bearing segment after recycling using liquid nitrogen for osteosarcoma. The mean disease survival was 54 months (range 34–69 months) and no local or systemic recurrence was reported within a mean follow up of 54 months (range 34–69 months). After the procedure, the study group had a mean functional score of 82,4% (range 77-90%) measured using a modified system of the Musculoskeletal Tumor Society [17]. This result is comparable to the current result of this case in which on seven months follow-up, the patient did not reported any pain and able to perform full wight bearing. The graft united proximally and distally in all except one patient within six to ten months (mean of eight months) [17]. The results are in line with this current case in which radiograph examination showed signs of union and no evidence of recurrency.

After Cappana procedure, infections were more prevalent in tibial reconstructions (8.5%) than in femoral reconstructions (6%). Non-union rates were also more prevalent in tibial reconstruction (10.5%) than femoral resconstruction (6%). Conversely, allograft fractures occurred more in femoral reconstructions (18%) than tibial reconstruction (10.5%). An allograft nonunion occurred in Chang's series when the intramedullary fibula was too short to span the two osteotomies. Allograft nonunion merely occurred in Moran's series of pediatric patients, both in patients who underwent postoperative chemotherapy although the graft was well-vascularized intramedullary fibula grafts. Despite these problems, rates of limb salvage were excellent. In cases of large bony resection, the Capanna procedure has become a well-established means of long bone reconstruction and limb salvage [13,18–21]. In patients diagnosed with bony tumor of the limbs, Cappana procedure should be considered as limb salvage surgical method it has successfully treated our patient without causing any complications [5,13].

4. Conclusion

Modified Cappana procedure which introduce the use liquid nitorgen-recycled autograft from the resected affected bone as a peripheral shell supporting a centrally placed vascularized fibular graft to fill the massive bone defect left by surgical resection, had successfully performed in our patient whom previously diagnosed with periosteal osteosarcoma of femoral shaft. This Method provides reliable and relatively cheap reconstruction option, with minimal complications as recorded in our patient. More studies is still needed to evaluate the long term outcome, reliability, and complications of this procedure.

Ethical approval

Ethical approval was not required in the treatment of the patient in this report.

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Credit authorship contribution statement

Muhammad Zulhandani contributes to the study concept or design, data collection and writing the paper.

Muhammad Wahyudi, Oryza Satria, and Bonita Prawirohardjo contributes in the study concept or design, data collection, analysis and interpretation, oversight and leadership responsibility for the research activity planning and execution, including mentorship external to the core team.

Declaration of competing interest

None.

Acknowledgment

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Consent for publication

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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