Hemicortical Resection and Biological Reconstruction using Allograft for Parosteal Osteosarcoma of the Distal Femur. A Case Series

Olga D Savvidou (olgasavvidou@gmail.com)
National and Kapodistrian University of Athens, ATTIKON University General Hospital

Stavros Goumenos
National and Kapodistrian University of Athens, ATTIKON University General Hospital

Ioannis Trikoupis
National and Kapodistrian University of Athens, ATTIKON University General Hospital

Angelos Kaspiris
University of Patras

Dimitra Melissaridou
National and Kapodistrian University of Athens, ATTIKON University General Hospital

Panagiotis Gavril
National and Kapodistrian University of Athens, ATTIKON University General Hospital

Jimmy Georgoulis
National and Kapodistrian University of Athens, ATTIKON University General Hospital

Panayiotis J Papagelopoulos
National and Kapodistrian University of Athens, ATTIKON University General Hospital

Research Article

Keywords: parosteal osteosarcoma, surface osteosarcoma, biological reconstruction, allograft, hemicortical resection, limb salvage

Posted Date: December 2nd, 2021

DOI: https://doi.org/10.21203/rs.3.rs-1119930/v1

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Abstract

**Background:** Parosteal osteosarcoma (PAO), which is a surface osteosarcoma, can be treated with wide excision and endoprosthetic or allograft application. However, due to the low local recurrence and metastasis rate, when it appears in the posterior surface of the distal femur, can be managed with hemicortical wide resection and biological reconstruction with hemicortical allograft.

The purpose of this study was to evaluate the oncological and functional outcomes of patients with parosteal osteosarcoma (PAO) of the posterior cortex of the distal femur who underwent biological reconstruction after hemicortical resection.

**Methods:** Eleven patients who underwent wide tumor resection and defect reconstruction of the posterior surface of the distal femur using hemicortical allograft were studied retrospectively. Local recurrence, metastasis, complications and the functional outcome using the Musculoskeletal Tumor Society scoring system (MSTS Score) were evaluated.

**Results:** The average postoperative follow-up period was 53.64 months (range, 30 to 84 months). At the latest follow up, all patients were free of the disease without appearing any metastases. A patient with local recurrence underwent revision surgery with fibula autograft reconstruction. The mean MSTS score was 93.45 ± 3.56.

**Conclusions:** In patients with PAO of the posterior aspect of the distal femur the treatment of hemicortical resection and allograft reconstruction has satisfactory oncological and functional outcomes and low complication rates.

**Background**

Osteosarcomas are high grade intramedullary bone tumors that require wide excision and endoprosthetic reconstruction for limb salvage surgery. Parosteal osteosarcomas (PAO), that belong to surface osteosarcomas, compose a rare distinct clinicopathological category of osteogenic tumors rather than a subtype of intramedullary conventional osteosarcomas[1].

PAO are the most common surface tumor, representing 6% of all osteosarcomas and have a predominance for the second, third and fourth decade of life as well as in female gender. The posterior cortex of the distal femur is the most commonly affected site while it can also be detected in the metaphysis of other long bones [2].

Wide tumor resection and endoprosthesis or biological reconstruction are the most commonly used therapeutic strategies [2]. PAO are low-grade tumors with low risk for metastasis and local recurrence, hence PAO of the distal femur can be managed with hemicortical wide tumor excision and biological reconstruction, reducing the risk of endoprosthetic reconstruction complications [3].
The purpose of this study was to evaluate the oncological and functional outcomes of hemicortical excision and biological reconstruction using allograft for the treatment of PAO of the posterior aspect of distal femur.

**Methods**

The study was approved by the ethical committee of ATTIKON University General Hospital with the reference number of AD 232/19-04-2021. All patients agreed to participate in the study and provided written consent prior to publication. The research complies with the 1964 Helsinki Declaration and its later amendments.

From 2010 to 2018, 11 patients with PAO of the posterior cortex of the distal femur underwent wide hemicortical resection and reconstruction with hemicortical strut allograft in our Department. There were 7 female and 4 males. The mean age and BMI of the patients was 29 ± 7.46 years and 25 ± 2.76 kg/m$^2$, respectively. All patients were diagnosed with well differentiated PAO of the distal femur, involving less than 60% of the cortex circumference, and based on computer tomography (CT) studies, magnetic resonance imaging (MRI) and histopathological results, intramedullary extension was not detected. None of the patients had distant metastatic disease.

**Operative technique.**

The operation performed under general anesthesia and a pneumatic tourniquet was used on the proximal thigh. After patients were set on prone position, a midline longitudinal incision at the posterior aspect of the distal thigh and knee was performed. The fascia and the popliteal space were carefully dissected, and the peroneal and tibial nerve, as well as the femoral artery and vein were identified and preserved. The posterior aspect of the distal femur was exposed and removed en-block with the tumor, using osteotomes. The mean cortical resection was 14.64 ± 5.5 cm. The posterior cortex of a distal femur allograft was prepared to match the cortex defect of the host. In 3 patients two cancellous compression screws in a posteroanterior direction were used for prophylactic fixation (Figures 1A, B-6A, B), while in 9 patients anatomic distal femoral plate was used to secure the allograft on the host bone (Figures 7A, B-10A, B). A drain suction was used and the wound was closed in layers. Postoperatively a posterior splint and later a brace was applied until radiographical union was demonstrated. Partial weight-bearing was started when more than 50% union of the transverse and longitudinal osteotomies appeared in the radiographic evaluation.

None of the patients were lost during the follow-up. The average postoperative follow-up was 53.64 months ± 16.94 months (range, 30 to 84 months). None of the patients received neoadjuvant chemotherapy.

All surgical procedures, postoperative evaluation and assessment were performed by the same surgical team. The primary endpoint of our study was the achievement of satisfactory disease control. Secondary
endpoints were: functional outcomes during the last follow-up, time to graft incorporation and assessment of postoperative complications.

As for the primary endpoint assessment, all patients underwent sequential imaging staging (MRI) and computer tomography (CT) scan for local and systematic disease progression and a CT scan of the chest every four months in the first two years, and every six months thereafter. Regarding the secondary endpoints of our study, functional results were evaluated using the Musculoskeletal Tumor Society Score (MSTS score) [4] and measuring the knee range of motion.

The time of the allograft incorporation was established based on radiographic findings of union. The patients were examined every month until the osteotomies had consolidated, and then after every six-month. Bridging across three of four cortices in biplanar radiographs was considered evidence of consolidation.

The postoperative complications were classified as mechanical (type 1,2,3), and non-mechanical (type 4 and 5) according to the classification system regarding the failure of limb salvage after biological reconstruction described by Henderson et al. [5].

**Results**

Wide excision of the lesion with clear surgical margins was achieved in all patients based on histopathological examination. At the follow-up examination no distant metastases were detected. All patients were free of disease at the latest follow-up evaluation (primary endpoint).

The mean MSTS score was 93.45 ± 3.56. All patients were ambulatory without any postoperative pain or restriction of daily activities at the latest follow-up. The mean range of knee flexion was 105.1° ± 7.66.

Radiologic findings showed successful union within the first postoperative year. The average time of allograft incorporation was 7.64 ± 1.8 months.

As far as the postoperative complications, there were no soft tissue (type 1), hardware failure (type 3) or implant related infections (type 4) based on Henderson criteria. Only one patient had local recurrence (case no 6) and underwent revision surgery with wide excision and reconstruction using fibula autografts (Figures 11A, B-16A, B). This patient remains free of the disease five years postoperative. (Table 1).

**Discussion**

PAO is the most frequent surface osteosarcoma. It is a low-grade tumor with low risk for local recurrence and metastatic potential. The most typical location is the posterior surface of distal femoral metaphysis, followed by proximal tibia, fibula and humerus whereas involvement of the ulna, tarsal and carpal bones has been sporadically described. Symptoms include a painless mass with duration of at least one year.
As the mass increases, pain worsens. Decreased range of motion might occur if the lesion is closed to a joint [6,7].

Radiographically, PAO appear as a large lobulated mass with extensive, generally central bone formation and a thick mineralized stalk which looks to be “stuck” onto the bone surface, while the medullary cavities usually are spared. CT confirms the presence of the broad, of cortical density stuck and the cleft sign that separates the ground glass mass from the bone cortex. MRI can be used to visualize the overlying cartilaginous cap that frequently coexists.

The soft tissue component is isointense to hypointense on T1 weighted images, inhomogenous on fluid-sensitive MR images and shows inhomogenous enhancement on contrast-enhanced MR images; the cartilage cap is hyperintense on fluidsensitive MR images whereas the osteoid matrix is hypointense on both T1w and fluid-sensitive MR images [8].

Histologically, the tumor is composed of hypocellular areas of spindle cells arranged in fascicles in desmoplastic collagenous stroma with parallel trabeculæ of well formed woven bone (‘streamer pattern’). Spindle cells are characterized by minimal or, less frequently, moderate atypia and low mitotic activity. Its low-grade appearance and positivity for MDM-2 and CDK4 are key features in differentiating it from other surface sarcomas [8].

Differential diagnosis included fibrous dysplasia, myositis ossificans traumatic, osteochondral exostosis, developmental defect at insertion of adductor magnus or bizarre parosteal osteochondromatous proliferation, (BPOP), juxtacortical chondrosarcoma, high grade surface osteosarcoma and parosteal lipoma [8,9].

Wide resection of the tumor demonstrate 90% survival at 10 years follow-up [5]. Local recurrence may occur when wide resection is inadequate or in case of dedifferentiated PAO [10]. After wide tumor excision of the distal femur, the most common location of PAO, endoprostheses or different reconstruction techniques can be used. Due to the low grade of the tumor and to the young age of the patients hemicortical resection and reconstruction of the bone defect using polymethyl methacrylate (PMMA) or biological materials such as fibular autografts, allografts or pasteurized/autoclaved/irradiated host bones, with or without prophylactic fixation have been reported with good oncological and functional outcome [11-14].

Endoprosthetic replacements of the femoral defect have demonstrated long term survival and good functional outcome, following resection of PAO [15]. However postoperative complication rate is high and include infection, aseptic loosening, mechanical failure, fracture of the prosthesis or of adjacent bone [2,16-17]. Allograft allows mechanical and biological reconstruction after wide excision of tumour however, it is associated with high rates of complications, including fracture, non-union, transmission of disease and infection [18].
Campanacci et al. in 1984 was first described [19] the hemicortical resection with unicortical window of the distal femur in the early stage of PAO of the distal femur. Later this hemicortical resection method was established as safe therapeutic procedure [11]. However, due to the rarity of the disease, there are no studies with a large number of patients and data on functional outcomes. Lewis et al. reported very good oncological and functional outcomes with no complications using this technique in 6 patients with PAO of distal femur [12]. Deijkers et al., in one of the largest cohort studies, analysed 22 allograft hemicortical reconstruction procedures, showing very satisfying oncological and functional outcome. All patients had good or excellent MSTS score, low complication rates and the allograft incorporation rate thirty months postoperative was 100% [13]. Agarwal et al. reviewed 10 patients who underwent hemicortical excision of the distal femur and reconstruction with autograft or allograft. In both methods, functional outcome was optimal and there were no major surgical complications or local recurrence [3]. Liu et al. reported very good oncological and functional outcome with relative low complication rates in 13 patients diagnosed with PAO and managed with wide excision and reconstruction with pasteurized hemicortical autograft and internal fixation. The authors stated that the technique although demanding is safe and effective in selected patients [20].

Compared to endoprosthetic reconstruction after wide resection of PAO of distal femur, many studies demonstrate, respectively good functional and oncological outcome. Funovic et al. compared 12 patients who underwent prosthetic reconstruction with 11 patients who underwent biological reconstruction. The authors concluded that oncological and functional outcome does not differentiate between the two reconstruction methods, but in the endoprosthetic reconstruction group significantly more revisions were needed (58%) compared to the biologic reconstruction cohort (18%) at an average of 10 years [21]. Wilke et al. compared 5 patients who underwent endoprosthetic reconstruction with 7 patients who underwent biological reconstruction. After long-term follow-up, patients’ group reported the same mean MSTS score (mean MSTS 23), range 16-30 in the endoprosthetic group and 18-26 in the allograft reconstruction group, while the rate of complications and reoperation was not differed between the two groups [2].

Although hemicortical allograft is a reliable biological reconstruction method after wide excision of PAO of the posterior cortex of distal femur, complications may occur. The most common complication is host bone fracture and local recurrence, followed by non-union and infection. Rarely allograft fractures can occur. These complications are associated directly to the size of bone defect after wide excision and most commonly require surgical reintervention [22]. None of our patients had a host bone fracture, allograft fracture, nonunion or infection. However, only one patient had local recurrence and was managed with revision and second time biological reconstruction with fibular autograft.

The results of our study are in accordance with that of the published literature that delves into the biological reconstruction after hemicortical resection of PAO of the posterior aspect of distal femur. We acknowledge that despite the satisfying results, our study has several limitations. It is an observational, retrospective study, which included a small number of patients, without a comparison group. However, all 11 patients were treated by the same oncology team, and were operated by the same surgeon (PJP). Randomized multicenter studies with large number of patients with PAO of distal femur are needed in
order to establish these good functional and oncological results and compared them to the other reconstruction techniques.

Conclusions

Parosteal osteosarcoma of the posterior surface of the distal femur with no intramedullary involvement can be treated with hemicortical excision and biological reconstruction with hemicortical allograft. This procedure is a safe a reliable treatment with good oncological and functional results and with low complication rate.

Abbreviations

PAO: Parosteal osteosarcomas; PMMA: polymethyl methacrylate; MSTS: Musculoskeletal Tumor Society Score

Declarations

Acknowledgements

Not applicable.

Authors’ contributions

ODS and JPJ: were responsible for the conception and design provided the study materials or patients; SG, IT, AK and DM: carried out the data analysis and interpretation and gave administrative support; PG, JP: did the collection and assembly of data and carried out the data analysis. All authors contributed in the manuscript writing. The authors read and approved the final manuscript.

Funding

No funding was received

Availability of data and materials

The datasets generated and/or analyzed during the current study are not publicly available because the data are confidential patient data, but are available from the corresponding author upon reasonable request.

Ethics approval and consent to participate
Ethics approval and consent to participate Ethical permission for this study was obtained from the Institutional Review Board of Attikon University Hospital, Athens, Greece (IRB Registration Number: AD 232/19-04-2021) and the written informed consents were acquired from all patients.

Consent for publication

Informed consents for publication were obtained by all the patients for this case series presentation

Competing interests

The authors declare that they have no competing interests.

References


Table

Table 1 is not available with this version.

Figures
Figure 1

A, B: Parosteal osteosarcoma (PAO) in a 16 years old female (no 6). Anteroposterior (A) and lateral (B) radiographs of the knee show a parosteal osteosarcoma as a large ossified opacity attached to the posterior cortex of the distal femoral metaphysis.
A, B: Sagittal computerized tomography (CT) reformatted images, soft tissue window (A) and bone window (C) exhibit with superior detail the thin separation between tumor and the intact femoral cortex (cleft sign) as well as the ossified thick stuck lytic areas are seen within the ossified mass which is surrounded by a thick hypodense rim representing cartilaginous tissue.

A, B: A fat-suppressed T2w magnetic resonance image sagittal (A) and axial view (B) shows the densely ossified stuck centrally, the inhomogenous moderately T2 hyperintense mass in the middle, and the
hyperintense cartilaginous component in the periphery. There is no intramedullary extension of tumor.

Figure 4

A-F: (A) Hematoxylin-eosin stain (X200): hypocellular tumour composed of mildly atypical spindle cells arranged in fascicles in desmoplastic collagenous stroma. The morphological findings are consistent with parosteal osteosarcoma (PAO) (B) MDM-2 (X400) and (C) CDK4 (X400): the majority of the neoplastic cells show strong nuclear positivity. (D) Hematoxylin-eosin stain (X200): excision specimen of the lesion shown in A, B and C: parallel trabeculae of well-formed woven bone with spindle neoplastic cells in loose stroma. (E) Hematoxylin-eosin stain (X40): the neoplastic population is characterized by mild cellularity and mild cellular atypia. (F) hematoxylin-eosin stain (X40): foci of moderate cellularity and moderate cellular atypia in PAO.
Figure 5

A-E: A. Intraoperative image of the popliteal fossa after tumor resection, identification of the tibia nerve and the popliteal artery. Hemicortical resection B. the resected specimen C. distal femoral allograft D. part of the posterior femoral allograft match the dimension of posterior distal femoral defect, after tumor excision E. Two cortical screws were used to fixed the allograft to the distal femur.
Figure 6

A, B: Anteroposterior (A) and lateral (B) radiographs 6 months postoperatively.
Figure 7

A, B: Computer tomography (CT) of right femur in a 32-year-old male with parosteal osteosarcoma of distal femur. A. sagittal CT with multiple large ossified masses adjacent to the posterior surface of the distal femur. B. axial view, densely ossified stuck and the cleft sign that separated the ground glass mass from the bone cortex, with no evidence of medullary involvement.
Figure 8

A, B: Magnetic resonance imaging of the distal femur sagittal (A) and axial (B) view showing round mass of low signal intensity on T1 weighted imaging. The cortex appears intact along the deep surface of the lesion.

Figure 9

A-G: Intraoperative images showing (A) midline longitudinal incision at the posterior aspect of the distal femur and hemicortical resection. (B) Tumor resection. (C) Distal femur allograft. (D) Preparation of the
allograft. (E): Matching the allograft to the bone deficit. (F) Allograft fixation with four cortical screws in anteroposterior direction. (G) Anatomical distal femoral plate used as a bridging plate.

Figure 10

A, B: Anteroposterior and lateral radiographs nine months postoperatively showing incorporation of the allograft at the distal part; however, the proximal part of the allograft shows partial union.
Figure 11

A, B: Parosteal osteosarcoma (PAO) in a 16 years old female (no 6). Anteroposterior (A) and lateral (B) radiographs four years postoperative shows recurrence of PAO.
Figure 12

A-C: Intraoperative images with complete excision of the posterior-medial part of the distal femur, the excised specimen (B) and polymethyl-methacrylate (PMMA) was used to filled the osseous void.
Figure 13
A, B: Postoperative radiographs anteroposterior (A) and lateral (B) views showing the PMMA in the posterior-medial part of the distal femur.

Figure 14
A-C: A. Autograft from the contralateral fibula was used, and was cut in longitudinal axis. B. these two pieces of hemicortical fibula were inserted to the posterior-medial part of the distal femur after excision of the PMMA. C. Postoperative anteroposterior radiographs of the knee showing the fibula autografts in place.
Figure 15

A, B: Anteroposterior (A) and lateral (B) radiographs of the knee 13 months postoperatively showing incorporation of the fibula autografts with no signs of recurrence.
Figure 16

A, B: Magnetic resonance imaging of the distal femur sagittal (A) and axial view (B) showing the distal femur with the fibula autografts with no signs of recurrence, infection or autografts fracture.