



First Case of Medullary Osteogenic Sarcoma of the Pelvis: 12-Year Follow-up of Reconstruction with Hemipelvis Allograft after Resection

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Abstract

A 45-year-old male patient with low-grade central osteosarcoma (LGCO) in the periacetabular region underwent wide resection, fresh frozen hemipelvis allograft reconstruction, and total hip prosthesis. To the best of our knowledge, this case is the first example of low-grade osteogenic sarcoma in flat bones. Aseptic loosening of the acetabular cup was observed 44 months after the operation, and it was revised with a constrained acetabular cup. Recurrent subluxation due to constraint ring failure and cup malposition was observed at 89 months after the revision surgery. Revision operation was performed with cage + non-constrained cup. Twelve years after the first operation, he is in exceptionally functional and disease-free condition. He can walk unlimited distances without pain. Radiographs show complete union at the junction of the allograft and disease-free bone.

In conclusion, fresh frozen hemipelvis allograft is one of the best reconstructive options following hemipelvectomy because of its potential durability and capacity to restore pelvic stability and preserve functionality.

Keywords

low-grade osteogenic sarcoma, low-grade central osteosarcoma, intramedullary, rare subtype

INTRODUCTION

Osteosarcoma, characterized by osteoid production by atypical, malignant osteoblasts, is the most common primary malignant bone tumor.^[1] Osteosarcomas represent a heterogeneous group of tumors with different biological behavior and treatment. Besides the most common conventional intramedullary high-grade osteogenic sarcomas, there is a group of low-grade tumors that have been described as a histological variant of osteosarcomas. This group consists of low-grade intramedullary osteosarcoma and various forms of surface osteogenic sarcomas.

Low-grade intramedullary (central) osteogenic sarcomas (LGCO) represent less than 2% of all osteosarcomas.^[2]

Although pelvic tumors constitute 5%-10% of all malignant bone tumors, the most common ones are chondrosarcoma, Ewing sarcoma, and osteosarcoma.^[3] The prognosis and survival of patients with bone sarcomas in this region are worse compared to extremity sarcomas.^[4] However, LGCO's localize to long bones, and to the best of our knowledge, they have not been reported in flat bones. Treatment of LGCO is wide resection unless dedifferentiation is detected.

Reconstruction of defects in the bony pelvis is one of the most complex issues in orthopedic surgery. The mor-

bidity of pelvic resection and reconstruction is very high and should only be performed in the presence of adequate technical and physical equipment, as well as a team capable of providing an experienced oncologic multidisciplinary approach. Various reconstruction methods have been described in the literature after pelvic resections. These are flail hip, sacral transposition, iliofemoral and ischiofemoral arthrodesis and pseudoarthrosis, and massive implants (pelvic allograft or allograft prosthetic composite, autoclave or irradiated autograft, pelvic prosthesis, and saddle type prosthesis).^[5-7]

In this study, follow-up findings of hemipelvis allograft applied to a 45-year-old male patient with low-grade (medullary) osteosarcoma in the periacetabular region were reported. To the best of our knowledge, this case is the first case of medullary osteosarcoma located in the pelvis in the literature. The patient was informed that the data related to the case would be published, and consent was obtained.

CASE REPORT

A 45-year-old male patient presented with the complaint of right hip pain. Initially, right groin pain was observed. Over time, his complaints increased. He stated that he also had pain at night and at rest. On physical examination, there was antalgic gait on the right side and a tender mass on the right ilium. Radiographs, magnetic resonance imaging (MRI), and computed tomography (CT) revealed a large, destructive lesion of 9×8×4 cm extending from the dome of the acetabulum to the ilium in the right hemipelvis (Fig. 1).

Open biopsy was reported as low-grade sarcoma. Histological examination revealed that parosteal osteosarcoma and low-grade osteosarcoma (well differentiated, medullary) could not be differentiated. Metastatic staging study was performed with thoracic and whole abdomen tomography and body technetium-99m bone scan. It was determined as IA according to Musculoskeletal Tumor Society (MSTS) staging.^[8]

The multidisciplinary committee consisting of medical oncology, radiation oncology, radiology, and pathology special-

ists and orthopedics decided that neoadjuvant chemotherapy and radiotherapy would not be beneficial because the lesion was low grade and the lesion should be resected. Wide margin resection (type I-II resection) was performed. The abductor mechanism and the sartorius and rectus femoris muscles were dissected from the pelvis to expose the ilium and acetabulum. Under fluoroscopic guidance, the hemipelvis was cut at the level of the triradiate cartilage with a saw. A cut was made with a saw, parallel to the articular line with the sacrum, medial to the superior ilium, and some disease-free ilium was preserved lateral to the sacrum. The remaining soft tissue appendages were also separated and the hemipelvis containing the tumoral tissue was removed as a block. Although the acetabulum and most of the upper portion of the hip capsule were removed by resection, a significant portion of the native acetabulum below the level of the triradiate cartilage remained intact. The resulting defect was reconstructed with a fresh-frozen osteoarticular allograft (irradiated allograft for sterilization) from an adult right hemipelvis. The iliac wing and acetabulum of the allograft were trimmed to fit the patient's defect and allow proper coverage of the femoral head in a stable position. The graft was fixed with a screw and 2 plates from the upper part of the allograft to the remaining natural ilium in the superomedial region. In addition, the graft was fixed to the remaining natural inferior acetabulum and ramus with 2 plates inferiorly. A cemented acetabular cap was placed. Total hip prosthesis was applied and abductor mechanism was repaired. Total operation time was 370 minutes and estimated blood loss was 2000 mL. Histopathological examination of the specimen revealed low-grade (central) osteosarcoma. Surgical margins were clean. There were no neurovascular complications in the post-operative evaluation. There was no brevity. No skin healing problem was observed (Fig. 2).

At six months, it was observed that the graft was united (Fig. 3). His MSTS score was 85.^[9]

The patient presented with right inguinal pain at the 44th month of follow-up. Radiographs showed acetabular loosening between the allograft and cement interface of hip arthroplasty. A revision operation was performed with a constrain cup (Fig. 4).

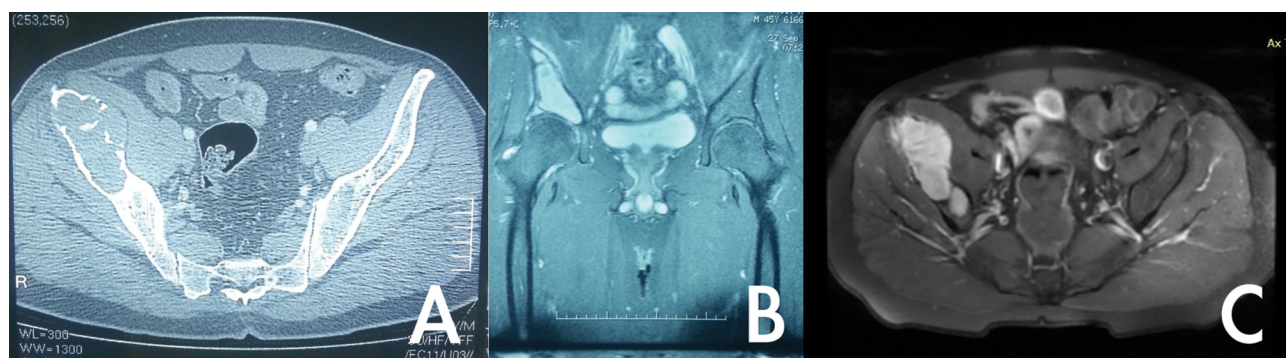


Figure 1. A. Preoperative axial computer tomography of a 45-year-old patient with right periacetabular osteogenic sarcoma. B, C. Intramedullary osteogenic sarcoma in the right periacetabular region in coronal and axial T2 contrast-enhanced magnetic resonance images of the same patient.



Figure 2. A. Fresh frozen hemipelvis allograft; B. Type I, II resection with the diagnosis of stage 1a osteosarcoma and post-operative radiograph after reconstruction with hemipelvis allograft; C. No wound healing problem was observed in the post-operative period.

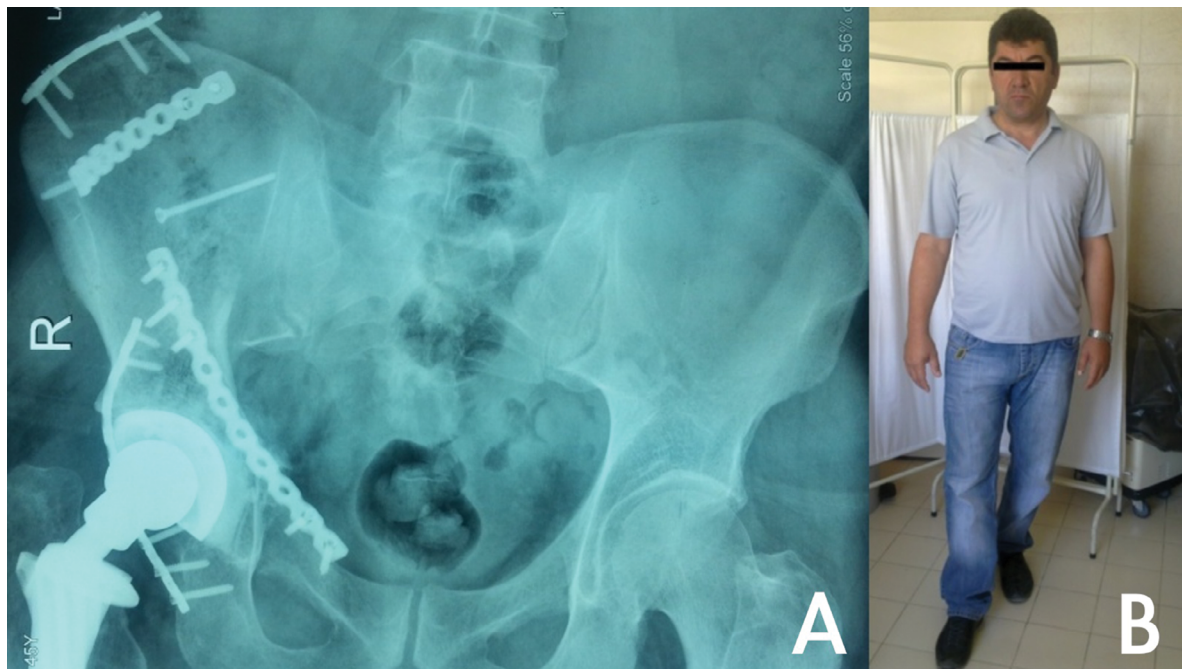


Figure 3. A. Union of the junction between the allograft and disease-free bone is seen in the postoperative control radiograph at 6 months; B. Excellent functional result was obtained with 85% MSTTS score. The patient walks without any assistive device.

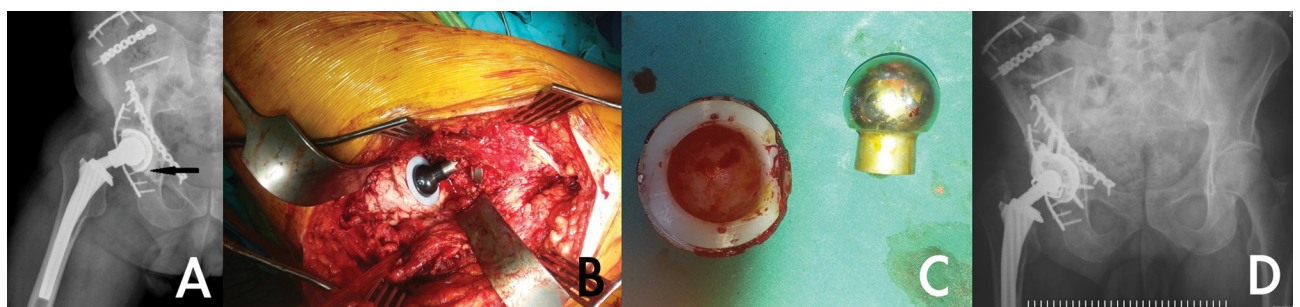


Figure 4. A. Aseptic loosening complication at the allograft and cement interface of hip arthroplasty at 44 months of follow-up of our patient who underwent type I + II resection; B, C. Intraoperative images; D. Revised with constrained muller cup.

Recurrent subluxation due to constraint ring failure and cup malposition was observed 89 months after the revision operation. The plaque in the posterior colon was removed and revised with Cage + non-constrained muller cup (Fig. 5).

The patient is still under disease-free follow-up for 12 years. His MSTS score is 85. He walks pain-free and is satisfied with his life.

DISCUSSION

With the development of chemotherapy, imaging, and surgical reconstruction techniques, it was in the early 1980s that allografts adapted to limb-sparing procedures.^[10] Since then, the allograft reconstruction procedure has been increasingly used as an alternative to amputation and metallic implants in patients with bone tumors. Despite improved survival, internal hemipelvectomy for pelvic tumors poses one of the greatest technical challenges in orthopedic oncology. The difficulties brought by the complex anatomy of the pelvis and the large size of the pelvic tumors, which are usually detected late, make resection and reconstruction difficult, and it is not always easy to achieve local control. As in our patient, type II resection often results in disruption of the pelvic ring and requires reconstruction to restore pelvic stability. If the bone bridges the periacetabular region and the sacrum, the stability of the pelvic girdle is maintained. However, if continuity is impaired, the lower extremity may migrate proximally with weight-bearing, resulting in leg length discrepancy and pelvic pain. Patients with a stable, reconstructed pelvis have significantly better MSTS functional scores than patients without pelvic reconstruction.^[11] In the reconstruction of pelvic defects, surgery is performed with various methods such as iliofemoral and ischiofemoral arthrodesis, massive allografts, custom-made pelvic prostheses, saddle type, or modular pelvic prostheses. However, serious early and late complications have been reported in all of these methods. Flail hip is an ancient method associated with severe limb shortness and disability, located at the opposite extreme of the concept of pelvic stability. Although iliofemoral and ischiofemoral arthrodesis, which can be considered palliative, is a better option than amputation, it has become less preferred by patients and surgeons today.

In resections where the proximal part of the ileum can be preserved (type II, type III), reconstruction with saddle-type prostheses is possible, but severe instability and progressive bone resorption in the residual ileum cause poor long-term outcomes. Primary stability and long-term implant loosening are the main problems in pelvic prostheses. Reconstruction with pelvic massive allograft provides complete restoration of the anatomical architecture of the pelvis, preserves bone stock and limb length, and is a successful method in achieving satisfactory functional results when combined with the hip prosthesis in acetabular resections.^[5-7,12] In the current study, a hemipelvis allograft was reconstructed and a stable pelvis was obtained. There was no shortness or pain, and the patient had a highly satisfactory functional score.

The structural integrity of acetabular allografts depends on the union of the osteotomy lines and the ability of the graft to tolerate this load at the time of loading. Studies on revision hip arthroplasty have shown that structural allografts can fuse and incorporate into host bone. Wherever possible, the allograft should be stabilized with rigid internal fixation, and adequate protection against loading should be provided to accelerate union at the osteotomy sites.^[13] In this study, the anterior and posterior columns were stabilized with a plate each, while the ilium was also stabilized with a plate and screw. The complete union was seen at 6 months postoperatively.

If a periacetabular resection, i.e. type II resection, has been performed and hip arthroplasty will be required, the easiest and fastest method is bipolar arthroplasty. Using a large head also increases stability. If standard hip arthroplasty is to be performed, a cemented cup fixation that provides instant fixation is the best option. Cemented hip arthroplasty was performed in this study. Aseptic loosening was observed in the postoperative 44th month, and constraint ring failure and cup malposition were observed in the 89th month after the revision. The patient is satisfied with his condition after the last revision.

Although multimodal therapy, including chemotherapy and local control, is very important in improving the survival rate in patients with osteosarcoma, the treatment for localized medullary osteogenic sarcoma is wide resection. Chemotherapy or radiotherapy is not indicated. With appropriate treatment, LGCO shows a good prognosis with

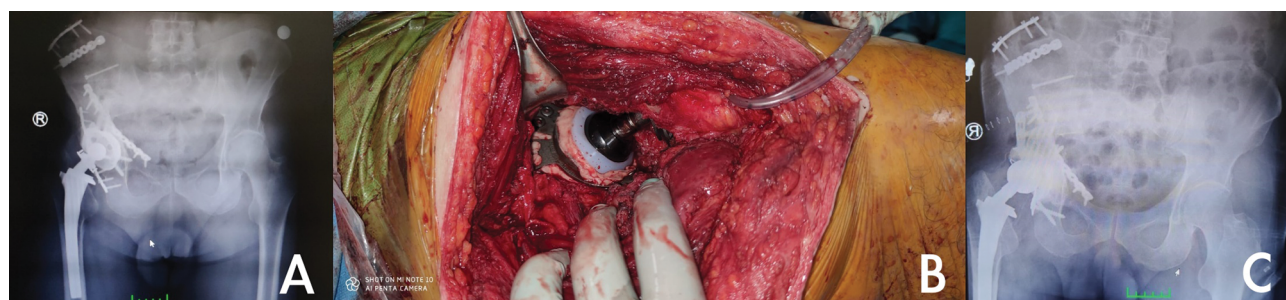


Figure 5. A. Pelvic ring failure and cup malposition 89 months after the revision operation; B. Revision intraoperative image with cage + nonconstrained cup; C. Post-operative radiograph.

5- and 10-year survival rates of 90% and 85%, respectively.^[1,14] If incomplete resection is performed, there is a risk of differentiation into a high-grade sarcoma. In addition, local recurrence and metastasis can be seen after inadequate resection. In the current case, the tumor was resected with wide margins, chemo-radiotherapy was not applied and it is still under disease-free follow-up for 12 years.

LGCO usually presents between 18 and 45 years, there is no male or female predilection.^[14] In our case, it was a 45-year-old male patient.

Pelvic resections and reconstructions in the same session are associated with long operative time and severe bleeding rates, even for experienced surgeons. In the literature, the average surgical time varies between 5-10 hours, depending on the type of reconstruction, and the average amount of blood loss varies between 2500 and 8000 mL.^[15] In this case, it is seen that the duration of surgery and the amount of bleeding were lower than in the literature. This may be due to the fact that all surgical procedures are performed through the same incision and without the need for additional assistance (general surgeon, plastic surgeon, spinal surgeon, vascular surgeon) and serious attention has been paid to hemostasis. At the same time, good preoperative planning and knowledge of anatomy may be the reason why our operation statistics are better than those reported in the literature.

Good function depends on the stability of the reconstruction and the function of the muscles involved to move the hip. Satisfactory functional results have been obtained in the reconstructions in which a good abductor, flexor, and extensor strength can be achieved in the hip. If we look at the functional scores after reconstruction with allograft: 62% in the series of Bell et al., 72% in the series of Donati et al., and 56.4% in the series of Langlais et al.^[5,7,15] In the present case, the functional score is 85, the patient can walk without support and is painless.

Reconstruction procedures performed after major resections of bone tumors have a high risk of complications, including reconstructions with allografts. The most common complications are fracture (19%), nonunion (17%), infection (11%), and instability (6%). Despite all the failures, the amputation rate due to tumor-related complications is only 7%. In various studies, acetabular complications were reported at a rate of 0-67%.^[5,7,15] In this case, aseptic loosening developed in a young active and overweight patient at the postoperative 44th month, and recurrent subluxation requiring revision was observed at the 89th month after the operation.

CONCLUSIONS

With the results obtained, despite the high complication rates specific to all pelvic resections, we recommend allograft reconstruction especially in young patients and tumor types that do not require adjuvant treatment as much as possible and have high survival rate compared to other

reconstruction options. Fresh frozen hemipelvis allograft is one of the best reconstructive options following hemipelvectomy because of its potential durability and capacity to restore pelvic stability and preserve functionality.

Author contributions

R.Ö. performed the surgery, drafted and designed the manuscript; A.E.K. performed the surgery, data collection and analysis; B.Ş.G. performed the surgery, and critically reviewed the manuscript

Conflict of Interest

The authors declare that they have no conflict of interest.

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Ethical approval

This article does not contain any studies with human participants or animals performed by any of the authors.

Informed consent

Written informed consent was obtained from the patient or first-degree relatives for publishing the individual medical records.

REFERENCES

1. Öztürk R. Current Therapeutic Approaches for Osteosarcoma. In: Amarasekera H, editor. Recent Advances in Bone Tumours and Osteoarthritis. IntechOpen; 2021. Available from: <https://www.intechopen.com/chapters/77019>
2. Antonescu CR, Huvos AG. Low-grade osteogenic sarcoma arising in medullary and surface osseous locations. *Am J Clin Pathol* 2000; 114:90–103.
3. Shin KH, Rougraff BT, Simon MA. Oncologic outcomes of primary bone sarcomas of the pelvis. *Clin Orthop Rel Res* 1994; 304:207–17.
4. Kawai A, Healey JH, Boland PJ, et al. Prognostic factors for patients with sarcomas of the pelvic bones. *Cancer* 1998; 82:851–9.
5. Bell RS, Davis AM, Wunder JS, et al. Allograft reconstruction of the acetabulum after resection of stage IIB sarcoma. Intermediate term results. *J Bone Joint Surg Br* 1997; 79:1663–74.
6. Renard AJ, Veth RP, Schreuder HV, et al. The saddle prosthesis in pelvic primary and secondary musculoskeletal tumors: functional results at several postoperative intervals. *Arch Orthop Trauma Surg* 2000; 120:188–94.

7. Donati D, Di Bella C, Frisoni T, et al. Alloprosthetic composite is a suitable reconstruction after periacetabular tumor resection. Clin Orthop Relat Res 2011; 469:1450–8.
8. Enneking WF. A system of staging musculoskeletal neoplasms. Clin Orthop Relat Res 1986; 204:9–24.
9. Enneking WF, Dunham W, Gebhardt MC, et al. A system for the functional evaluation of the reconstructive procedures after surgical treatment of tumors of the musculoskeletal system. Clin Orthop 1993; 286:241–61.
10. Enneking WF. Transplanting allografts. J Am Coll Surg 2005; 201:5–6.
11. Riff AJ, Gross CE, Foucher KC, et al. Acetabular osteoarticular allograft after Ewing sarcoma resection: a 15-year follow-up: a case report. JBJS Case Connect 2016; 6:89.
12. Apffelstaedt JP, Karakousis CP. Partial and complete internal hemipelvectomy: complications and long term follow-up. J Am Coll Surg 1995; 181:43–8.
13. Paprosky WG, Martin EL. Structural acetabular allograft in revision total hip arthroplasty. Am J Orthop 2002; 31:481–4.
14. Narang NC, Diwaker P, Narang S, et al. Low-grade central osteosarcoma: report of two unusual morphologic variants. Indian J Surg Oncol 2018; 9:74–8.
15. Langlais F, Lambotte JC, Thomazeau H. Long-term results of hemipelvis reconstruction with allografts. Clin Orthop Relat Res 2001; 388:178–86.

Первый случай медуллярной остеогенной саркомы таза: 12-летнее наблюдение за реконструкцией аллотрансплантатом Hemipelvis после резекции

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Резюме

Больному 45 лет с центральной остеосаркомой низкой степени злокачественности (LGCO) в периацетабулярной области выполнена широкая резекция, реконструкция аллотрансплантатом свежемороженой половины таза и тотальное протезирование тазобедренного сустава. Насколько нам известно, этот случай является первым примером остеогенной саркомы низкой степени злокачественности в плоских костях. Через 44 месяца после операции наблюдалось асептическое расшатывание вертлужной впадины, которое было исправлено на сжатие вертлужной впадины. Рецидивирующий подвывих из-за несостоятельности ограничительного кольца и неправильного положения чашки наблюдался через 89 месяцев после ревизионной операции. Ревизионную операцию выполняли с использованием кейджа + неограниченная чашка. Через 12 лет после первой операции пациент находится в исключительно функциональном и безболезненном состоянии. Он может проходить неограниченное расстояние без боли. Рентгенограммы показывают полное сращение в месте соединения аллотрансплантата и здоровой кости.

В заключение, свежемороженный аллотрансплантат Hemipelvis является одним из лучших реконструктивных вариантов после гемипельвэктомии из-за его потенциальной долговечности и способности восстанавливать стабильность таза и сохранять функциональность.

Ключевые слова

остеогенная саркома низкой степени злокачественности, центральная остеосаркома низкой степени злокачественности, интрамедуллярный, редкий подтип