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Gigantic secondary pelvic chondrosarcomas treated with pelvic resection type I and III: A case report

Muhammad Wahyudi^{*,1}, Andrian Astoguno Bayu Prakurso

Department of Orthopaedic and Traumatology, Fatmawati General Hospital, Jakarta, Indonesia

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ABSTRACT

INTRODUCTION: Patients with osteochondromatosis have a higher risk of malignant transformation into secondary chondrosarcoma. Chondrosarcoma at the pelvic region tends to present late and therefore pose a significant challenge for orthopedic surgeons because of the large tumor size, local extension, and complex anatomy with proximity to major neurovascular structure, intestinal and urinary tract.

PRESENTATION OF CASE: A 44-year-old male presented the chief complaint of 15 years growing lumps on his left buttock and right groin, presenting with pain. Plain radiography revealed popcorn calcification at the left iliac wing and right superior pubic rami. Multiple exostoses were also visible. MRI showed a larger tumor diameter at the left iliac wing by 33 cm and right pubic rami by 13 cm. The histopathological result from the biopsy suggested low-grade chondrosarcoma.

RESULT: The patient underwent pelvic resection type I and III in two-stage surgery. About one month after the first surgery, there was a postoperative infection. Debridement and antibiotic therapy resulted in a desirable functional outcome with an MSTs score 27 and no local recurrence sign during a one-year follow-up.

DISCUSSION: Low-grade chondrosarcomas are not sensitive to radiation and chemotherapy; wide surgical resection is the mainstay of treatment. Chondrosarcoma at the iliac wing can be treated by pelvic resection type I, and further reconstruction needed to prevent pelvic tilting. Chondrosarcoma at pubic rami can be treated by pelvic resection type III.

CONCLUSION: Proper patient selections, preoperative planning, and wide surgical margins with reconstruction provide desirable local control and clinical outcomes following pelvic resection.

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1. Introduction

Hereditary multiple exostoses (HME) or Osteochondromatosis presents with multiple exostoses or osteochondromas, benign cartilage-capped bone tumors growing outward the metaphysis of long bones. Most lesions of osteochondromatosis stagnate and ossify when skeletal growth is complete, but occasionally one might grow more aggressively or reactivate as chondrosarcoma [1]. The most frequently observed pelvic sarcoma is chondrosarcoma, followed by osteosarcoma. The Iliac is the most common location for chondrosarcoma at the pelvic region [2].

These tumors also tend to present late and therefore pose a significant challenge for orthopedic oncologists because of the large

tumor size, local extension, and complex anatomy with proximity to major neurovascular structures, intestinal, and urinary tracts. Subsequently, these are associated with less favorable prognosis and survival than other extremity tumors [3]. Even today, pelvic sarcomas treatment remains one of the most predominant challenges for orthopedic oncologists due to the proximity of visceral organs and neurovascular structures [4].

Previously, the definitive treatment of malignant pelvic tumors was external hemipelvectomy (hindquarter amputation), but with new development in surgical techniques and chemotherapy and radiation therapy, limb salvage procedures (internal hemipelvectomy) have also emerged as feasible modalities. Internal hemipelvectomy involves resectioning the lesion with part or all of the hemipelvis while preserving the ipsilateral lower extremity. In patients whose tumor yet to invade major neurovascular structures, wide resection is possible, and therefore internal hemipelvectomy can be considered without affecting the limb function. Since an adequate and tumor-free resection margin is of great importance for the long-term oncological outcome, hemipelvectomy remains as a curative approach [5–7]. With adequate management, internal hemipelvectomy do not pose higher recurrence rates compared to amputations by external hemipelvec-

* Corresponding author at: Department of Orthopaedic and Traumatology, Fatmawati General Hospital, Jl. RS Fatmawati Raya no 4, Cilandak, South Jakarta 12430, Indonesia.

E-mail addresses: dr.m.wahyudi@gmail.com (M. Wahyudi), andrianastoguno@gmail.com (A. Astoguno Bayu Prakurso).

¹ Mailing address: Department of Orthopaedic and Traumatology, Fatmawati General Hospital, Jl. RS Fatmawati Raya no 4, Cilandak, South Jakarta 12430, Indonesia.



Fig. 1. The clinical presentation showed lumps on the left buttock and right groin.



Fig. 2. A plain radiograph revealed popcorn calcification.

tomy [8] and, in contrast, lead to better functional results in many cases [9]. However, in cases of neurovascular invasion and where wide margin excision is unmanageable, external hemipelvectomy remains the preferred option [10]. Extensive dissections can compromise the muscle flaps viability, causing a large dead space, leading to the formation of collections, and consequently, surgical site infection [2].

This report presents a rare case of gigantic secondary chondrosarcomas of the iliac wing and pubic bone. A thorough investigation evaluated the oncologic outcome, functional outcome, and complications following pelvic resection type I and III. This case report had been written according to the SCARE guideline [11].

2. Case report

A 44-year-old man complained lumps on his left buttock and right groin for 15 years. The lumps grew slowly, and there was pain related to the lump. On physical examination, the lumps were around 30 cm and 10 cm in diameter with the normal overlying skin. The lumps were firm and non-tender on palpation (Figs. 1–6).

The patient underwent some investigations to confirm the diagnosis, including a blood test, plain X-ray, and pelvic MRI. The alkaline phosphatase level was 162 U/L; other laboratory findings were within normal ranges. Pelvic X-ray revealed popcorn calcification on the left iliac wing and right pubic bone. Sessile

osteochondromas were also visible on both femoral necks. Pelvic MRI indicated the tumor size at the iliac wing was around 15.3 × 16.5 × 25.9 cm, and at the right pubic rami was around 8.9 × 12.59 × 6.26 cm. T1 and T2 weighted MRI images demonstrate a heterogeneous signal intensity of the lesion.

The histopathological result from the biopsy suggested low-grade chondrosarcoma. The patient was planned for a two-stage surgery. The first surgery was pelvic resection type I, and the second surgery was pelvic resection type III.

The patient underwent pelvic/resection type I for iliac wing chondrosarcoma. He was placed on the lateral decubitus position with an anterior tilt to allow posterior access. The surgical approach followed a utilitarian pelvic incision procedure. Abdominal muscles and some abductor muscles were detached from the tumor. Osteotomies were done near the sacroiliac joint and 1 cm superior from the acetabulum. Reconstruction was done using a reconstruction plate and bone cement to prevent pelvic tilting. The surgery took 10 hours. The largest tumor diameter which had been resected was 33 cm. The amount of bleeding during Pelvic resection Type I was around 2200 cc. About one month after the first surgery, there was a postoperative infection. Debridement and antibiotic therapy were given to solve a postoperative infection. This complication can eventually be solved.

The second surgery stage was pelvic resection type III for right pubic chondrosarcoma, performed five months after the first

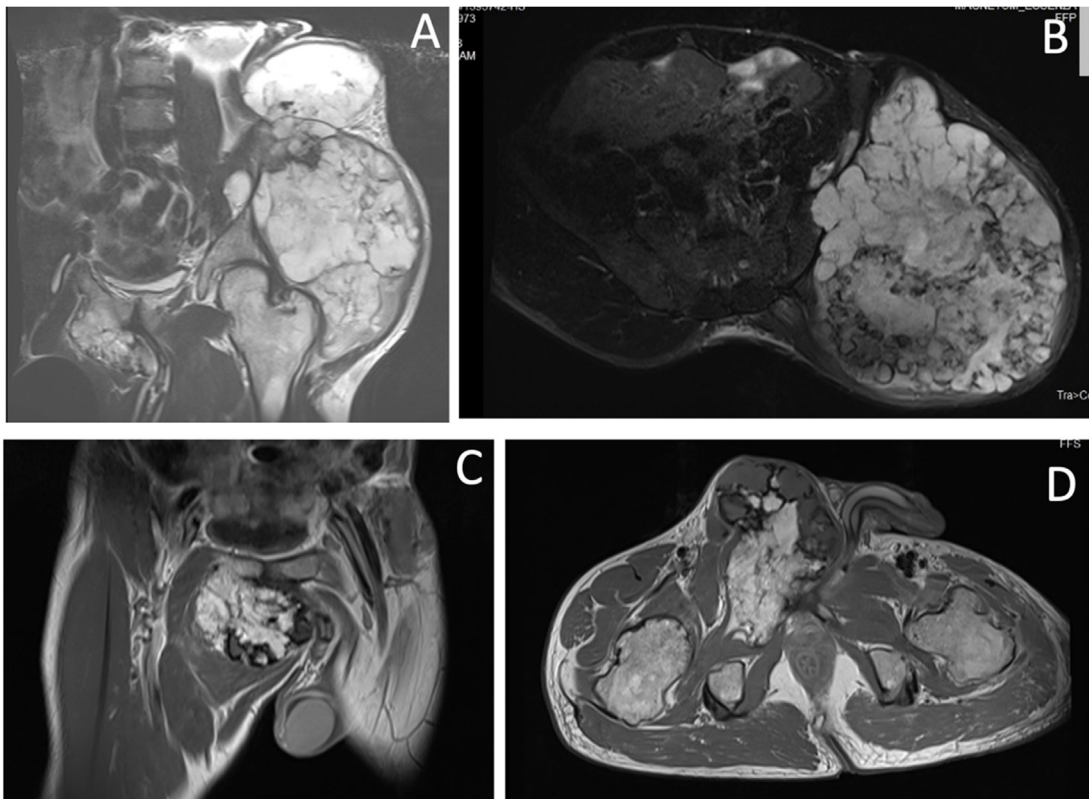


Fig. 3. (A and B) MR images showed a tumor on the left iliac wing. (C and D) MR images showed a tumor on the right pubic.

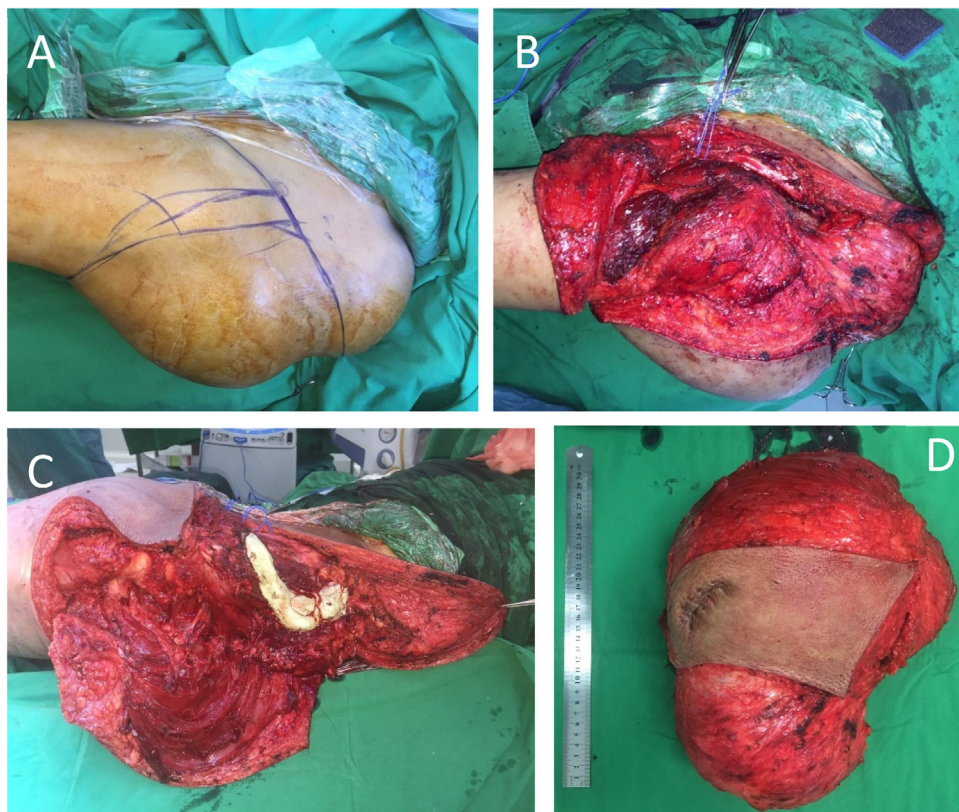


Fig. 4. (A) Utilitarian pelvic incision. (B) External iliac artery and vein identification, abdominal muscle detachment from the tumor. (C) Reconstruction after pelvic resection type I. (D) Tumour measurement.

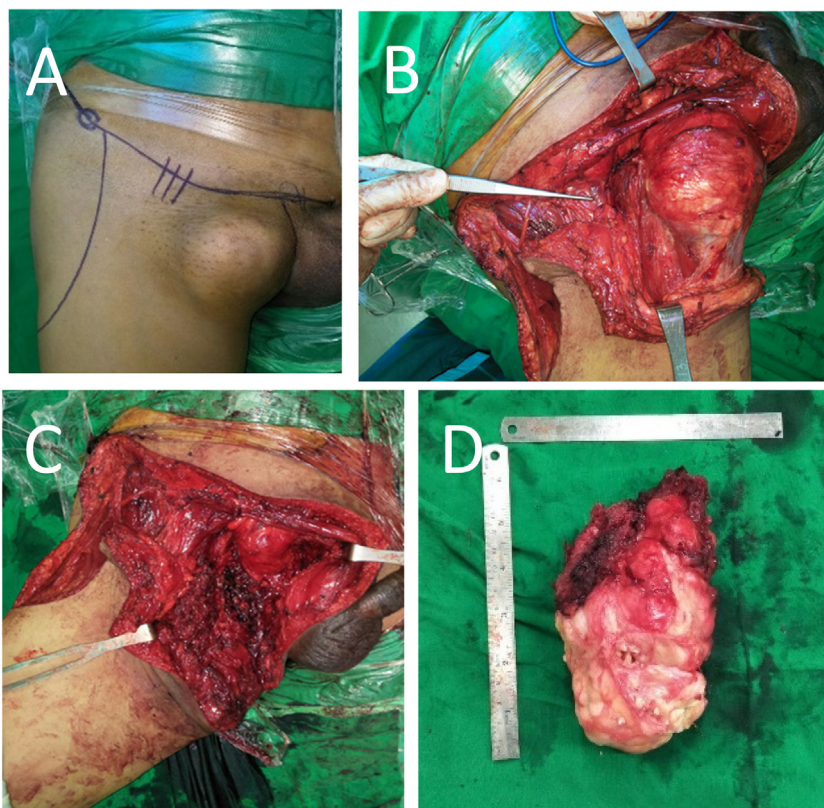


Fig. 5. (A) Utilitarian pelvic incision. (B) Neurovascular and spermatic cord identification. (C) After pelvic resection type III. (D) Tumour measurement.



Fig. 6. X-Ray of the pelvic had been taken after pelvic resection type I, III, and reconstruction.

surgery. The patient was placed on the supine position on the operating table. The Surgical approach followed the utilitarian pelvic incision procedure, which consists of ilioinguinal incision to permit retroperitoneal exploration and mobilization of the femoral artery, vein, and nerve. The other incisions are longitudinal incision, which follows the perineal crease, and longitudinal incision from the anterior superior iliac spine. Abdominal muscles were detached from the superior pubic bone. Pelvic floor muscles and adductor muscles were detached from the tumor and ischium. Osteotomies were performed at superior pubic rami, ischium, and symphysis pubis. The tumor size in pubic rami was 13 cm. Blood lost during pelvic resection type I amounted to 1500 cc. The surgery took 6 hours.

The histopathology examination from the resected tumor of the iliac wing and pubic showed cartilage matrix and hypercellular chondrocyte. The chondrocytes were atypical, larger in size, vary in shape, with hyperchromatic nuclei. There were a large number of binucleation, with apparent mitosis, and focally present stromal myxoid changes. The surgical margins were free from the tumor cell. The Conclusion from the histopathology examination was chondrosarcoma grade II.

At the one year follow up, the patient was in good condition and could work normally. MSTS score evaluated functional outcomes, where the patient scored 27. Albeit presenting with a Trendelenburg gait, the patient did not complain of any pain. The patient also showed no sign of recurrence 12 months after the surgical procedure.

3. Discussion

Secondary chondrosarcoma is a distinctive type of tumor originating from a preexisting cartilaginous lesion. Histologically, it resembles primary conventional chondrosarcoma, which arises de novo in bone without a preexisting lesion [12]. About 10% of chondrosarcoma are secondary, most commonly arising in an osteochondroma, either in a solitary exostosis or in multiple osteochondromas [13]. Most authors agree that patients with solitary exostosis have a 1–2% risk of developing chondrosarcoma. This risk increases to between 5–25% for patients with multiple osteochondromas. [13–15]. Peterson et al. argued a higher chondrosarcoma risk for patients with multiple exostoses due to the greater number of lesions in these patients [16].

Secondary chondrosarcoma must be distinguished from dedifferentiated chondrosarcoma, which is characterized by high-grade sarcoma adjacent to low-grade cartilage tumors. Meanwhile, secondary chondrosarcoma is composed entirely of cartilaginous

tissue, and it usually a low-grade malignancy. In both, the tumor arises from a preexisting cartilage tumor [12].

The patient was diagnosed with secondary chondrosarcoma that arises from osteochondromatosis. Clinically patient had multiple lump and bilateral forearm deformity. The bone examination showed multiple exostoses, popcorn calcification on the left iliac wing and right pubic bone. Histopathology examination after surgery showed intermediate-grade malignant chondroid tissue. Meanwhile, a study by Ahmed et al. showed that most of the secondary chondrosarcoma was low-grade malignancy [17].

Histologic diagnosis of chondrosarcoma is difficult because of the borderline distinction between benign cartilage cells and low-grade malignancy. The presence of malignant chondroid tissue usually identifies sarcomatous transformation. Hallmarks include hypercellularity, binucleate cells, multiple cells in lacunae, atypical nuclei, and myxoid changes in the hyaline cartilage matrix [12]. It is expected that the histologic sections did not show evidence of osteochondroma in more than half the patients. Because of the abundance of cartilage associated with secondary chondrosarcoma, sections often do not show osteochondroma and chondrosarcoma. A few abnormal features in histopathologic findings also make it difficult to distinguish the cartilage of osteochondromas from low-grade chondrosarcoma. In these cases, clinical and radiographic findings are essential [12,17].

A study by Ahmed et al. showed that the iliac wing is the most common site for secondary chondrosarcoma that arise from exostosis. Meanwhile, the pubic bone is the second most common site. Therefore, malignant transformation of osteochondroma is a possible diagnosis with a growing lump and sudden onset of pain in the pelvic region [17].

Wide surgical resection was performed since it remains the mainstay of treatment for secondary chondrosarcoma, and marginal excision is at high risk for recurrence [12,18]. Chemotherapy and radiation are generally not practical for this tumor. As chondrosarcomas possess a low percentage of dividing cells within a poorly vascular matrix, they are relatively resistant to radiation and chemotherapy [19].

The Musculoskeletal Tumor Society has classified pelvic resections into three resection types: type I (iliac), type II (peri-acetabular), and type III (obturator) [20]. Advancement of surgical techniques and the development of implants have made limb-salvage procedures achievable with partial pelvic resections and reconstructions in selected cases, especially for benign tumors and low-grade malignancies [21]. Pelvic resection type I was performed with reconstruction for left iliac wing secondary chondrosarcoma, and then pelvic resection type III was performed too for right pubic secondary chondrosarcoma.

Treatment of pelvic chondrosarcoma remains technically difficult for orthopedic oncologists due to the proximity of visceral organs and neurovascular structures. Besides that, patients with this condition tend to present late and pose a major challenge because of the large tumor size [22,23]. Most chondrosarcomas of the pelvis are considerably larger than their appendicular counterparts, averaging 11 cm in size at diagnosis. The magnetic resonance imaging of this patient suggested a larger tumor diameter at the iliac wing by 33 cm, and pubic rami by 13 cm, larger than the average reported tumor size on previous investigations. [24–26].

The problems with massive tumors are blood loss during surgery and longer surgery time. The patient usually needs a large amount of blood transfusion and postoperative intensive care. Intraoperative blood loss was 2200 cc and 1500 cc for 10 h (600 min) and 6 h (360 min) of surgery, respectively. A study by Umer et al. showed that the mean surgery time in pelvic resection was 356 min and the mean intraoperative blood loss was 1900 cc. Meanwhile, that study also showed that 62.5% of the tumor diameter was less than 10 cm [3]. Therefore, honed surgical and intraoperative bleeding control

skills, which would result in less surgery time and blood loss, were substantial. A large amount of transfused blood and platelets should be prepared.

The ilium has two primary functions: First, it provides continuity between the acetabulum and the central skeleton through the sacroiliac articulation. Second, it provides a primary soft tissue attachment site for the abdominal, gluteal, pelvic floor, rectus femoris, sartorius, and the iliac muscles within the pelvis [27]. Chondrosarcoma at the iliac wing can be treated by pelvic resection type I, and further reconstruction needed to prevent pelvic tilting. After the type I pelvic resection in this patient, bone cement, reconstruction plate, and the screw was used to prevent pelvic tilting and necessary to enable force transmission from the lower extremity to the axial skeleton [27]. Most of the abductor muscles were resected due to their involvement in the tumor. In this case, Trendelenburg gait after surgery would happen. Meanwhile, resection of the pubic rami requires no skeletal reconstruction when acetabular stability can be maintained, and the majority of patients exhibit satisfactory functional outcomes [27,28]. This patient underwent pelvic resection type III surgery without further reconstruction.

There was no sign of recurrence after one year of follow up. Han et al. showed that the surgical margin was the factor most closely related to the local recurrence [29]. Several studies showed local recurrence rate 12.5–19% [3,30,31]. However, the patient may require further follow up. MSTS score in this patient was 27 points or 90%. Systemic review by Shao et al. for pelvic resection showed MSTS score ranged widely from 16% to 100%. In the studies of the last ten years, the mean MSTS scores were more congruent, ranging from 50%–70% [32]. The patient had a better MSTS score due to the acetabulum non-involvement. The tumor requiring pelvic resection type II tends to have difficult reconstruction and has lower functional outcomes compared to pelvic resection type I and III.

Complications after pelvic tumor resection are prevalent, including infection, hematoma, wound healing problem, nerve injury, bladder injury, bowel injury, extensive hemorrhage, thrombosis, and implant failure [29,33,34]. The patient presented with postoperative infection after the first surgery, resolved by antibiotic treatment and debridement. The infection rate after pelvic resection in several studies showed 21–29% [3,4,30]. Many factors increase infection risk after pelvic resection. During surgery, many adjacent muscles and soft tissues should be dissected to obtain a safe margin, sometimes leading to devascularization and create dead space. Prolong surgical time also increases the risk of infection.

4. Conclusion

Proper selection of patients, preoperative planning, and wide surgical margins with reconstruction provides excellent local control and clinical outcomes following pelvic resection. Patients with massive tumor need a large amount of blood transfusion and postoperative intensive care due to longer surgery time. The surgical site infection is a common complication after pelvic resection and can be managed with debridement and antibiotic therapy.

Declaration of Competing Interest

The authors declare that there is no conflict of interest regarding the publication of this paper.

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Ethical approval

The authors have no ethical conflicts to disclose.

Consent

Written informed consent was obtained from the patients 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.

Author contribution

1. Muhammad Wahyudi, MD. Contributed to performing the surgery, the study concept or design, data collection, analysis and interpretation, manuscript drafting, revising, validation of the article, funding acquisition, supervision, and final approval of the manuscript.
2. Andrian Astoguno Bayu Prakurso, MD. Contributed to making the study concept or design, analysis and interpretation, data collection, manuscript drafting, investigation, literature search, revising, writing the paper, contribution to the submission process, and approval for publishing.

Registration of research studies

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Guarantor

The sole guarantor of this study is Muhammad Wahyudi, MD.

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