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Case report

Acetabular reconstruction with total hip replacement and femoral head autograft following pelvic resection of malignant bone tumour: A case report

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ABSTRACT

Introduction: Pelvic tumors greatly impact survival and quality of life of the patient. Reconstruction following resection of neoplasms involving the acetabulum remains one of the most challenging procedures for orthopaedic surgeons. We reported an 18-year-old female with Hodgkin's lymphoma of the left iliac wing.

Presentation of case: A 18-year-old female presented left hip pain since one year before admission. Pelvis X-ray demonstrated lytic lesion on the left iliac crest with moth-eaten pattern. However, the contrast-enhanced MRI showed the true extent of the tumour which engulfed the iliac crest and extended to the anterior border of the acetabulum. The acetabulum was reconstructed using femoral head autograft and total hip replacement. At six months of follow-up, CT scan of the pelvis demonstrated no tumour. No complications occurred during 14 months of follow-up. However, the patient died 28 months post surgery.

Discussion: In pelvic sarcomas, the utilization of this technique remains limited, as the complex anatomy and the bulk of tumour growth often limits the choice of what procedure can be conducted. Reconstruction techniques have also advanced, albeit difficult and laden with complications, especially when the lesion involves the acetabulum.

Conclusions: The choice of implant for pelvic resection in the developing country remains challenging due to the high cost of implants. However, in cases of pelvic sarcomas, the utilization of this technique remains limited, as the complex anatomy and the bulk of tumour growth often limits the choice of what procedure can be conducted. Reconstruction techniques have also advanced, albeit difficult and laden with complications, especially when the lesion involves the acetabulum.

1. Introduction

Pelvic reconstruction after periacetabular tumour resection is technically challenging and associated with significant complications and functional limitations. Frequently used methods used include hip transposition, arthrodesis, autograft with recycled tumour-bearing bone, allograft-prosthetic composite, and custom-made or modular pelvic prostheses [1]. Pseudarthrosis and arthrodesis elicit stable long-term effects but may cause hip function problems and leg length discrepancy [2,3]. Endoprosthetic replacement after pelvic tumour resection is associated with a high risk of complications and functional restrictions [1,4–6]. Limb salvage surgery with endoprosthetic replacement, or biological reconstruction is favoured by patients and surgeons over classic hemipelvectomy. Biological reconstruction with autografts, allografts, iliofemoral and ischiofemoral arthrodesis are limited by the extent of bone and soft tissue defects which subsequently results in higher complication such as infection, dislocation, non-union, periprosthetic fracture and poor limb function [7–11].

Nowadays, improvements in imaging modalities and multidisciplinary approach leads to prolonged patient survival, and thus, limb-sparing procedures have become the treatment of choice, particularly with respect to low patient acceptance of hindquarter amputation [12,13]. Although biologically-based reconstructions such as allografts have potential advantages in those undergoing pelvic tumour resections who have better oncologic prognosis, autograft pelvic reconstruction after periacetabular tumour resection has rarely been reported [14–16]. The main objective of the procedure is to local control the tumour by complete resection with secondary goal of restoring a functional and

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stable hip joint [17,18]. We reported a case of combined biological and mechanical reconstruction following resection of a malignant pelvic tumour using total hip replacement. The work has been reported in line with the SCARE criteria [19].

2. Case report

An 18-year-old female presented with left hip pain since one year before admission. The pain was aggravated by walking or sitting, and made her difficult to walk. She also complained about the enlarging mass at her left hip. The mass was ill-defined and tender. She had limited hip range of motion (Fig. 1A). An X-ray of the pelvis and contrast-enhanced MRI was ordered along with standard laboratory panel for tumors which included CBC serum ALP, LDH, liver and renal function tests.

Pelvis X-ray demonstrated lytic lesion on the left iliac crest with moth-eaten pattern. However, the contrast-enhanced MRI really showed the true extent of the tumour which engulfed the iliac crest and extended to the anterior border of the acetabulum (Fig. 1C-D). owing to its difficult location and vulnerability to penetrate visceral organs, we elected to do a CT-guided core biopsy (Fig. 2).

Radiography examination demonstrated primary bone tumour and the characteristic was malignant that came from her iliac wing extending to the acetabulum with differential diagnosis of osteosarcoma. It was suspected as osteosarcoma seeing from the clinical appearance, the incidence at her age, the predilection site, and from the imaging appearance. Then we asked to do an MRI and confirmed it using CT-Guided core biopsy (Fig. 3).

The biopsy result was discussed in a clinicopathological conference and the diagnosis of conventional osteosarcoma was established. The patient underwent 3 cycles of neoadjuvant chemotherapy with Doxorubicin and Cisplatin before the surgery. Imaging reevaluation prior to surgery confirmed a decrease in mass size with clearer border (Fig. 3).

We planned to perform a salvage procedure by wide excision of the mass by pelvic resection type I-II followed by reconstruction with plates and cement for the iliac wing, and arthroplasty for the hip joint. All stages of the operation are explained schematically through the illustration (Fig. 4). The patient was placed in a semi-recumbent position, a utilitarian approach was utilised (Fig. 5A). Upon incision, we found an enlarged lymph node and it was resected. The approach was continued down to internal and external obliquus muscles. The lateral circumflex, obturator, femoral nerve and the femoral vessels were identified and preserved (Fig. 5B). The Sartorius and direct head of the rectus femoris was identified (Fig. 5C) and detached from the tumour (Fig. 5D). The exposure was continued down the base of the tumour. We identified the superior margin as the iliac wing, the medial border at the superior pubic ramus and sacroiliac joint, and the inferior border at the hip joint. We excised the tumour along with infiltrated structures, leaving a dead space at weight bearing area of the hip joint (Fig. 5E), which made reconstruction difficult. We proceed according to plan by marking and resecting the femoral head (Fig. 5F) and reamed the femoral canal. We then placed the resected femoral head as a massive autograft at the acetabulum; however, some modifications were done to accommodate the femoral head into the medial crest of the sacrum, with the remaining defect filled with bone cement (Fig. 5G). We secured the femoral head to the sacrum and pubic rami with two reconstruction plates. We inserted

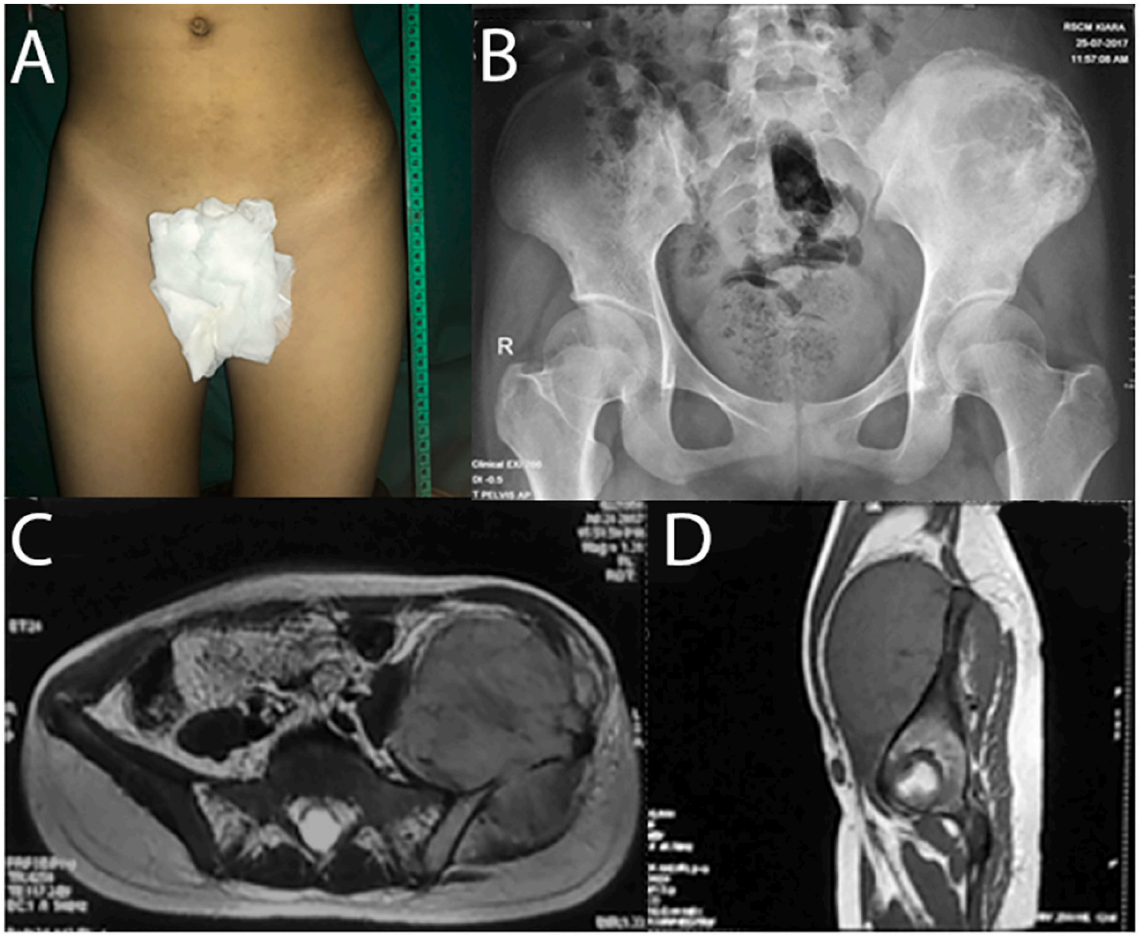

Fig. 1. (A) Clinical picture at first presentation. (B) pelvic AP X-ray showing lytic mass at iliac crest. (C-D) Axial T2-WI MRI showing tumour mass engulfment of iliac wing and extending to anterior border of acetabulum.

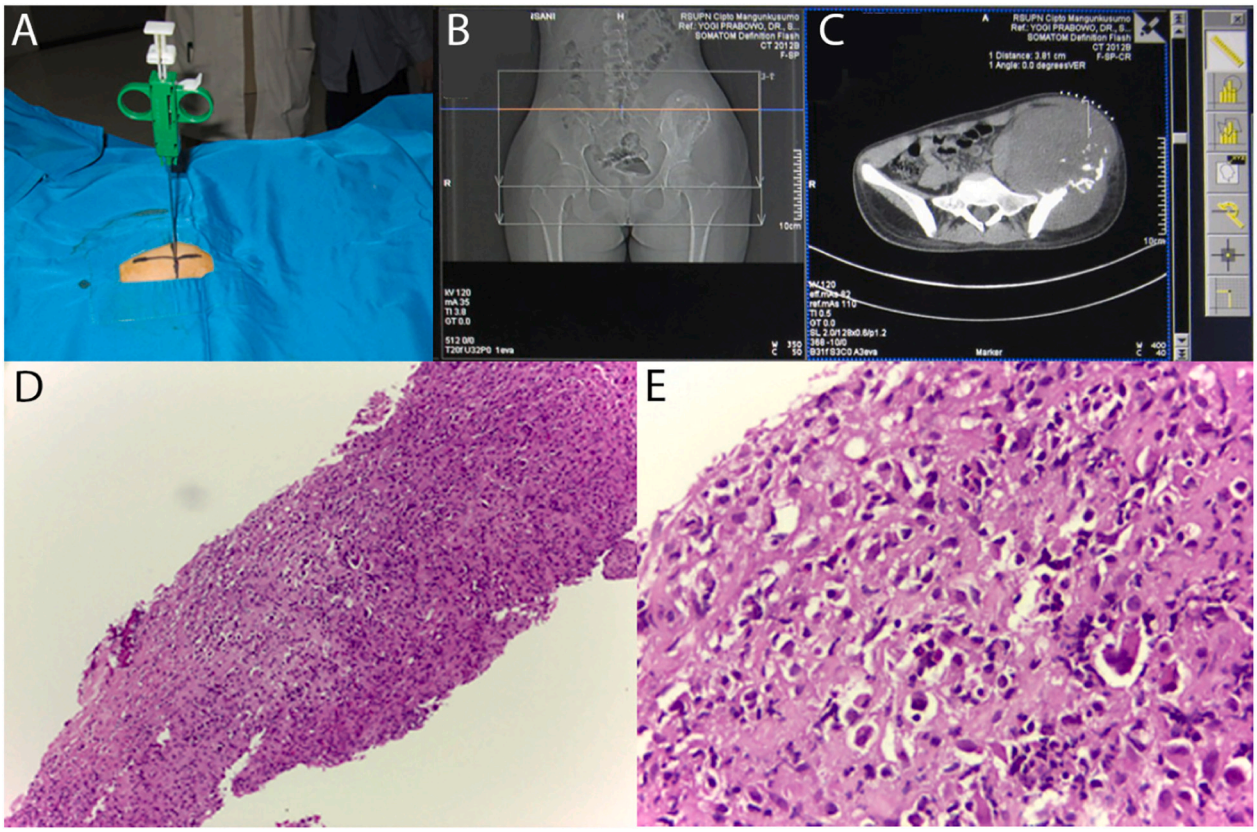

Fig. 2. (A) Location of the core biopsy (B-C) Confirmation of biopsy location by CT-scan. (D-E) histopathological results of the mass with HE 10× (D) and 100× magnification (E) showing round and oval cell, pleomorphic, hyperchromatic, and matrix osteoid like appearance.

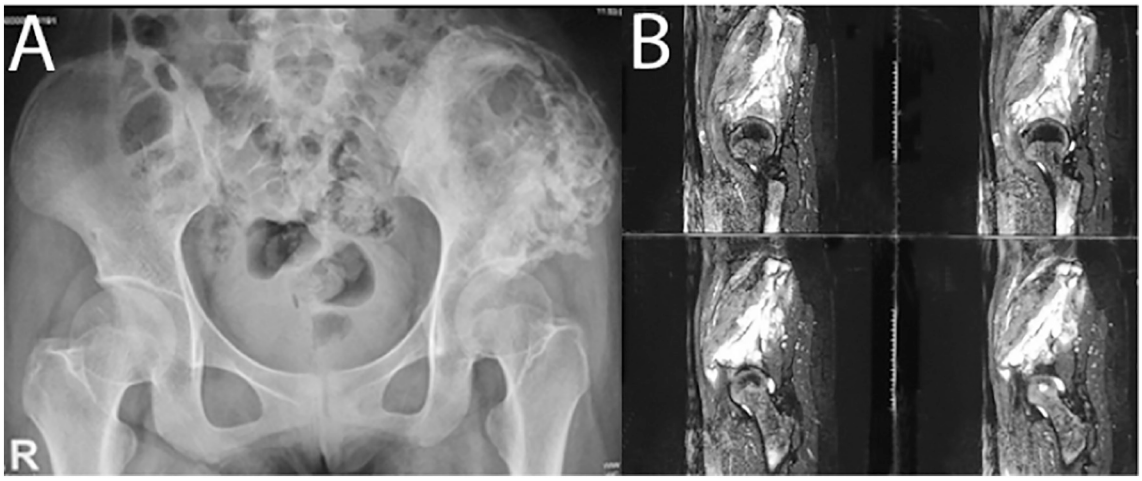

Fig. 3. (A). Post-chemotherapy pelvic X-ray showing more sclerosis on the margins of the tumour and (B) post-chemotherapy MRI.

the acetabular cup secured with two screws to the new acetabulum. The femoral stem was inserted, and after the whole construct was in place, we add polyethene mesh to act as the new capsule.

We tested the stability of the construct, and it was stable in all extent of hip joint movements. We then reattached the Sartorius and rectus, and we closed the wound in layers. The intraoperative blood loss was 1500 ml with 0.5 cm of leg length discrepancy. Postoperatively, the patient was admitted to the intensive care unit for stabilisation as the duration of surgery were 10 h. After a day, the patient was sent back to the hospital ward, and an x-ray was ordered (Fig. 6). On the third postoperative day, the pain had decreased and the patient started to exercise the left extremity in a sitting position. The day after, the patient could stand with minimal pain, and she was walking, albeit limping. After 2 weeks, on routine follow-up in the outpatient clinic, the patient seemed happy, and walking was improved (Fig. 7).

Ten days after the surgery, the histopathological results came back, shockingly, the result was miles away from the previous conclusion, that the patient was diagnosed with malignant Hodgkin's lymphoma (Fig. 8), and we had to change the chemotherapy regiment accordingly. At six months of follow-up, CT scan of the pelvis demonstrated no tumour. No complications occurred during 24 months of follow-up (Fig. 9).

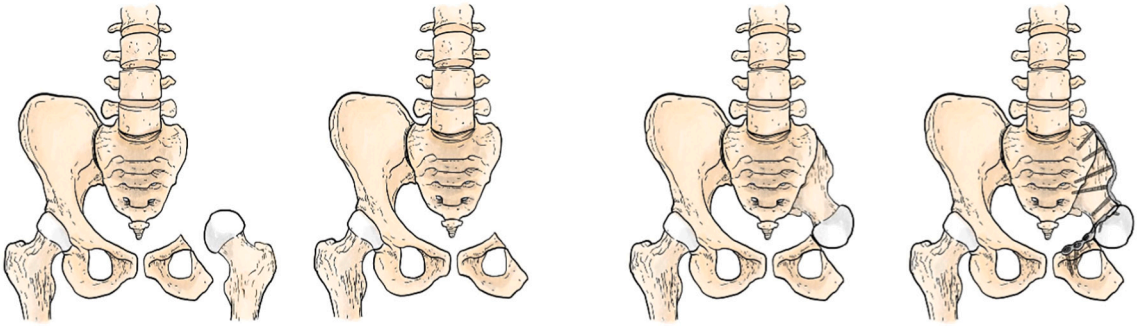

Fig. 4. Stages of operation illustration scheme.

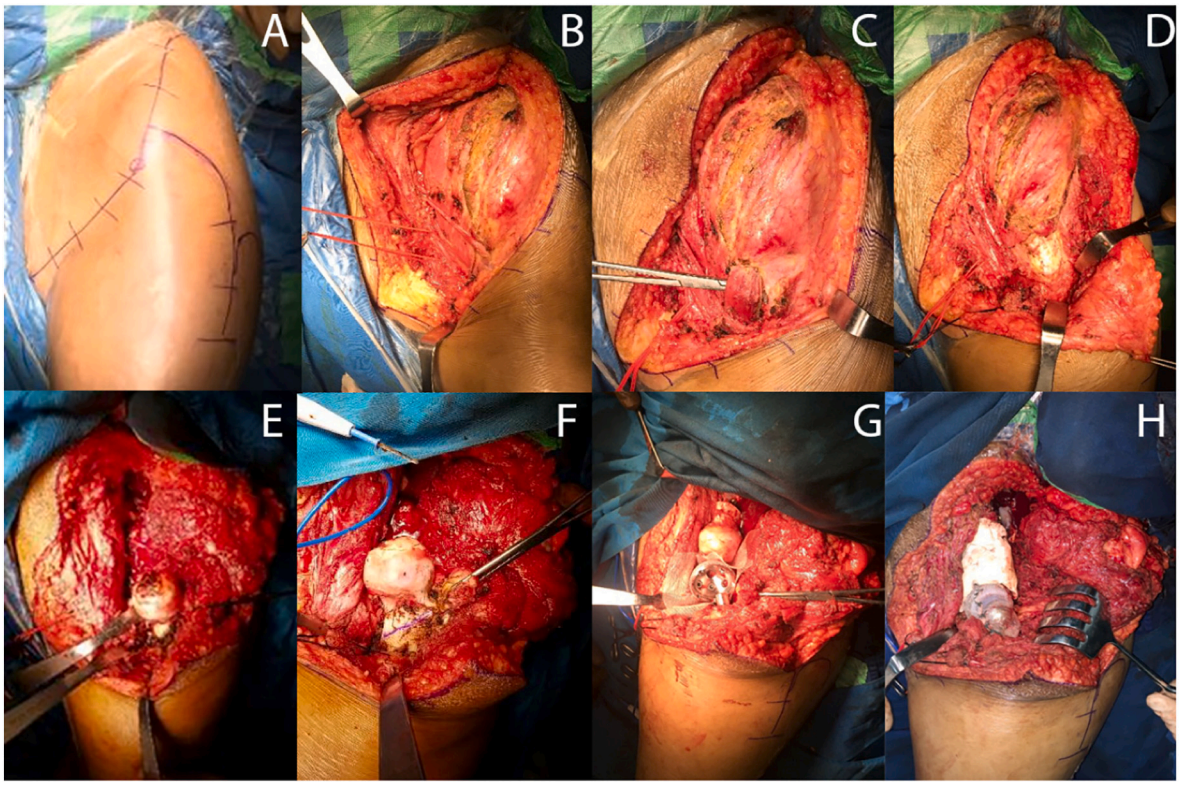

Fig. 5. From top left, (A) Utilitarian design and approach (B) identification and preservation of femoral nerve, artery and vein, (C) identification of Sartorius and rectus femoris muscles (d) after detachment of rectus and Sartorius, exposure down the base of tumour (E) after tumour removal, (F) femoral head marked and resected (G) femoral head in place as a autograft, the femoral stem and acetabular cup in situ (H) final construct, the femoral head was supplemented by cement, with polyethylene mesh acting as capsule.

3. Discussion

Numerous methods have been reported for the reconstruction of lower limb function after pelvic resections. Endoprosthetic reconstructions such as partial femur and total hip replacement with a constrained or non-constrained joint, or saddle-type implant and artificial pelvis have been attempted, and some showed good function. However, most of these reconstructions have not been successful over the long term, and major postoperative complications are common, such as dislocation of the joint, breakage of the implant and serious deep infection. Allografts and autoclaved or radiated autografts of the pelvis, which have been used for hip reconstruction require long term fixation before full weight-bearing is possible [20]. Our procedure maintains rigid fixation between the sacral crest and the prosthesis through the proximal femur autograft to performed early partial weight-bearing postoperatively with no pain and short time surgery that lasts about 10 h with minimal blood loss around 1500 cc.

Reconstruction after resection of neoplasms involving the acetabulum remains one of the most challenging procedures. Pelvic reconstruction is indicated in young patients, cases which involves resection of weight-bearing or -moving elements (a prime example of which would be the hip joint), primary sarcomas, and solitary pelvic bone metastasis with cancers that are considered to be “favorable” and carries with them a long life expectancy, such as thyroid, renal, and breast cancers [13]. Many techniques have been used to preserve the hip function; the simplest one is by leaving the limb hanging and allow a pseudoarthrosis to form. Some have attempted to do arthrodesis of the hip joint; however the success rate was rather low, even the successful one results in a substantial limb length discrepancy and also difficulty with sitting and walking [21].

Major pelvic resections have been classified by the Musculoskeletal Tumor Society into 3 resection types: type I (iliac), type II

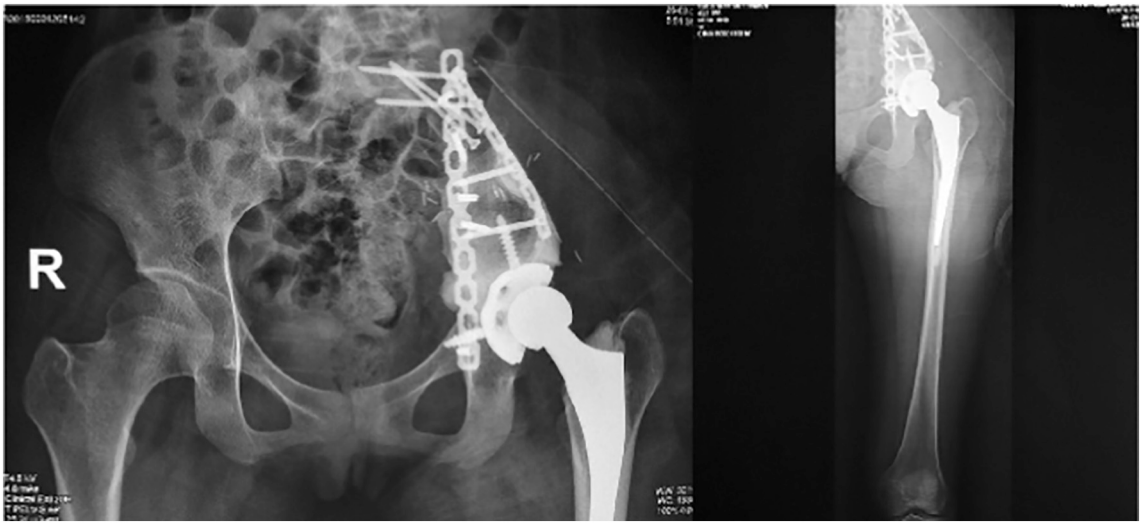

Fig. 6. Post-operative X-ray.

Fig. 7. Left: 3 days postoperative, the patient could stand with bilateral crutches, Right: The patient could walk with bilateral crutches and stand up without any complaint.

(periacetabular), and type III (obturator). Resections that involving the sacrum are type IV resections, or combinations thereof, based on the system established by Enneking and Dunham [18,22,23]. Pelvic resections that include the femoral head have been designated as type H and are classified into 3 types: type H1 (femoral head), type H2 (pertrochanteric area), and type H3 (subtrochanteric area) as in this patient we performed type H1–2 resections of proximal femur because the plan was to do a reconstruction using total hip replacement, and also the proximal femur used for an autograft parts of the reconstruction [24].

Type II procedure poses the greatest reconstructive challenge because of the resection a functional hip joint [21]. Limb salvage procedure is generally performed to obtain wide margins without compromising survival and function [24]. We performed a combined resection of type I and II at left pelvic due to extended of the tumour form previous CT-guided biopsy which have been discussed and was established as osteosarcoma. From the mechanical aspect, it is a nightmare because of the weight-bearing area that passes the load of the body came through the periacetabular area; it is clear that after resection there would be a great challenge to reconstruct and overcome the dead space.

Arthroplasty reconstruction after pelvic resection are recommended when adequate sacral crest and pubic rami remain for fixation of a pelvic allograft composite with total hip replacement; however in this patient, we used an autograft from her femoral head after being osteotomised. The periacetabular soft tissues and gluteal fascia should be reattached to maximise postoperative hip stability.

The standard pelvic bone resection employs the utilitarian pelvic resection approach, which involves extending the ilioinguinal approach, extending from the pubic tubercle and running through the anterior superior iliac spine, as well as aling the iliac crest up to the superior iliac spine [24] for the periacetabular resections require a lateral extension of the incision to the thigh. As in this patient, we performed layer by layer anatomically incision including abdominal musculature carefully dissected off of the iliac crest. The femoral arteriovenous nerve bundle was identified and protected. Rectus femoris muscle and Sartorius muscle were detached to have a good exposed of the tumour to be resected, also the sacroiliac joint as the medial border. In our series, augmentation with artificial ligaments or synthetic mesh was used to improve primary stability [25].

Restoration of hip joint mobility results in more satisfying functional outcomes than the pseudoarthrosis technique or amputation does, although it is associated with higher complication rates. We attempted to restore hip joint mobility in most of the cases with acetabular involvement.

In this patient, we performed reconstruction using her own proximal femur as an autograft which preoperatively and intraoperatively was a free margin of the tumour. The autograft is function as a gap closer in which the left ilium until the periacetabular were removed leaving disconnection of load sharing mechanism from upper to the lower extremity, and also leaving the left limb hanging. The osteotomized proximal femur was placed 180^{o} to the sacral crest from posterior to the anterolateral and fixated with double reconstruction plate laterally and medially. The lateral one was fixated through the autograft and the proximal sacral, and the medial one was fixated between the remaining pubic rami to the sacrum and also fixated the autograft. At the end the bone cement was placed to the surrounding autograft after the acetabular shell was already placed and the projection already being calculated, also fixated with 2 screws to the autograft and 1 screw to the remaining pubic rami made it a very rigid fixation system surrounding

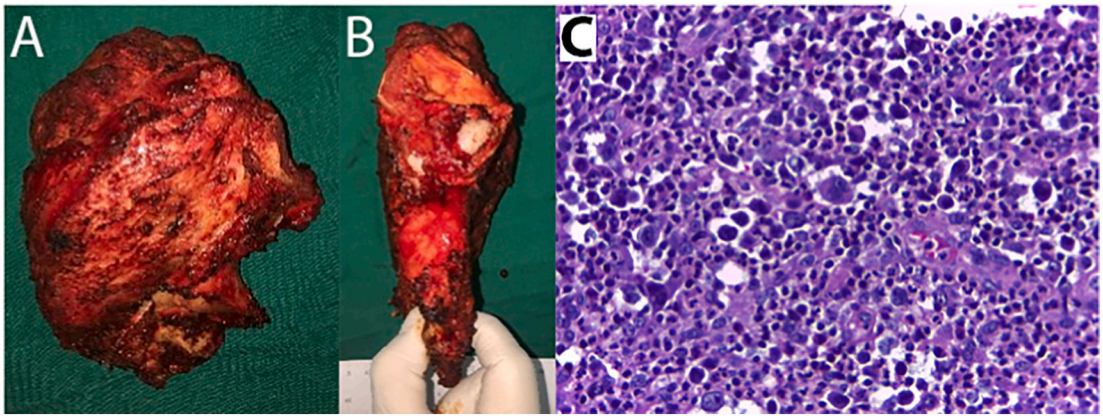

Fig. 8. Gross pathology of the tumour from anterior (A) and medial (B). (C) Microscopic appearance of the tumour on 100× magnification with HE staining showing atypical lymphoid cells and connective tissue, along with binucleated cell (Reed-Sternberg cell) which is pathognomonic of lymphoma Hodgkin.

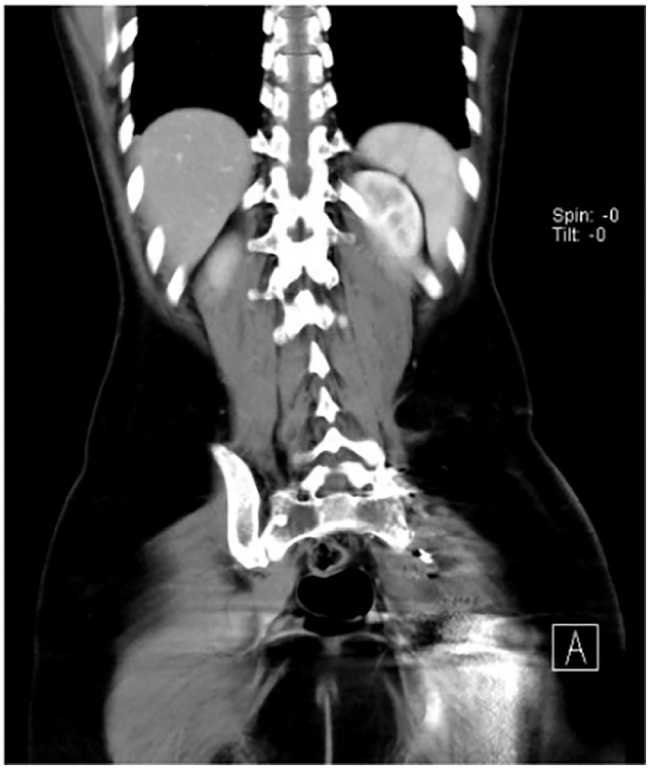

Fig. 9. CT scan of the pelvis at 6 months of follow-up demonstrated no tumour.

the arthroplasty. The autograft actually function as a holder for the acetabular shell. With the prosthesis in place, we tried and evaluated that the acetabular prosthesis should be symmetric with the contralateral side in height, lateral distance, and also the orientation [24].

The pelvis functions as the axis that transmits the weight of the upper body to the legs and the pelvis contains the hip joint. If bone stock loss occurs due to wide resection of sarcomas, then the focus of the treatment should be set on restoring femorosacral continuity for weight-bearing and obtaining mobility of the hip joint. In this study, satisfactory functional results could be achieved in those cases where pelvic ring continuity was restored through reconstruction [18].

The complication rate after removal of a pelvic tumour and reconstruction with pelvic allografts or prostheses is high, ranging from 30 to 90 % [25]. Frequent complications after such procedure include intraoperative haemorrhage; sciatic and femoral nerve injuries on the opposite side; ureter, bladder, and bowel injuries; wound-healing problems; infection and dislocation of prostheses; infection and fracture of allografts; lower quadrant hernia; bowel ischemia; and late venous thrombosis [23,24]. However, in this patient there is no complication found, also postoperatively she could standup and begin to walk with crutches as a partial weight-bearing just after 2–3 days postoperative.

After the result from pathology anatomy department came of the resected tumour, shockingly was different from the previously concluded biopsy. From the histopathological review, the cell appearance showed atypic cell with massive distribution of lymphoid cell and connective tissue; also several Reed-Sternberg cell were found. From the CPC forum it was concluded that the patient is having primary lymphoma malignum and need to be confirmed using immunohistochemical examination, which the result is confirmed to be the classic Hodgkin lymphoma nodular sclerosis from the positive results of CD30, MUM1, and KI-67 [26–28]. Although both Hodgkin's and non-Hodgkin's lymphomas are not rare entities, involvement of the bones is unusual and is most often associated with diffuse disease. The tumours may be easily diagnosed histologically, even after a needle biopsy. But because of a resemblance to other types of tumors such as Ewing's tumour, myeloma, osteosarcoma, chondrosarcoma, or metastatic carcinoma, it may be difficult to diagnose even after an open biopsy is performed [29,30].

Hodgkin lymphoma (HL) is a rare B-cell malignant neoplasm that affecting approximately 9000 new patients annually. It represents 11 % of all lymphomas seen in the United States (US), bone involvement has been found in 10 % to 20 % of cases with <2 % of cases showing skeletal lesions as the initial presentation. Classical Hodgkin lymphoma is defined subgroups as: nodular sclerosis, mixed cellularity, lymphocyte depletion, and lymphocyte-rich Hodgkin lymphoma, is characterised by the presence of Hodgkin and Reed-Sternberg (HRS) cells [31]. Classic symptoms includes fevers, night sweats, or unintentional weight loss are present [32]. Being one of the least common in primary bone tumour malignancies had been a diagnostic challenge due to its variable radiographic presentations, the most common radiographic features include permeative, lytic pattern of bone destruction, metadiaphyseal location, periosteal reaction, and soft-tissue mass [33]. Staging for this disease are so essential for the optimal therapy. Discriminating solitary bone lesion in HL patients from “primary” to “secondary” is challenging as the diagnosis of primary bone HL should be made based on strict histological and clinical manifestation [34–38].

Fourteen months postoperatively, the patient was diagnosed with classical Hodgkin lymphoma of the pelvis with a subtype of nodular sclerosis. The lymph nodes showed no cancer involvement but were confirmed to have reactive hyperplasia. At that time, the patient recovered well, and she was discharged after 5 days with no postoperative complications. However, the acetabular inclination and anteversion was less. The patient could walk with 2 crutches and

sometimes stand up without the external support, she started capable most of activities of daily living needed for self-care. However, 22 months after surgery, the patient died due to respiratory failure.

4. Conclusion

Limb-salvage pelvic resections are starting to gain some ground, propelled by the various advancements in imaging and surgical techniques and instrumentations. The choice of implant due to the limited source as in a developing country remains challenging because of the high cost as megaprosthesis per se. However, in cases of pelvic sarcomas, the utilization of this technique remains limited, as the complex anatomy and the bulk of tumour growth often limits the choice of what procedure can be conducted. Reconstruction techniques have also advanced, albeit difficult and laden with complications, especially when the lesion involves the acetabulum.

Funding

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Ethical approval

This case report did not intervene with patients' treatment plans and hence did not require ethical approval.

Consent

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.

Author contribution

Yogi Prabowo: supervising, data collection, analysis

Sammy Saleh Alhuraiby: data analysis, interpretation, writing the paper

Guntur Utama Putera: providing illustrations

Anissa Feby Canintika: writing the paper, analysis, interpretation

Registration of research studies

Not applicable.

Guarantor

Yogi Prabowo.

Declaration of competing interest

None declared.

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