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Naskah tinjauan pustaka dan artikel asli hendaknya disertai abstrak berbahasa Indonesia dan Inggris, ditulis pada halaman pertama di bawah nama dan institusi. Panjang abstrak 100-150 kata untuk naskah panjang atau 50-100 kata untuk naskah pendek.

Tabel dan Gambar

Tabel harus singkat dan jelas. Judul table hendaknya ditulis di atasnya dan catatan di bawahnya. Jelaskan semua singkatan yang dipergunakan. Gambar hendaknya jelas dan lebih disukai bila telah siap untuk dicetak. Judul gambar ditulis di bawahnya.

Asal rujukan table atau gambar dituliskan di bawahnya. Tabel dan gambar hendaknya dibuat dengan program Power Point, Free Hand, atau Photoshop, (menggunakan format jpeg).

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Daftar Isi

A Modified Buttockectomy as a Limb Salvage Procedure in Ischium Osteosarcoma: A Case Report

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ABSTRACT

Osteosarcoma, which rarely originates from the axial bones such as pelvis, presents a major challenge in limb preservation. We report a 9-year-old girl with osteosarcoma on her buttock. She underwent wide excision "modified buttockectomy" to resect the tumour with sciatic nerve involvement. One year after surgery, she was able to walk and no recurrence was found.

Keywords: *buttockectomy, osteosarcoma*

ABSTRAK

Osteosarkoma, tulang aksial seperti pelvis jarang sekali ditemukan, dan menjadi tantangan besar dalam upaya penyelamatan ekstremitas. Kami laporkan satu kasus osteosarkoma pada tulang panggul-daearah bokong pada seorang anak perempuan berusia 9 tahun. Pasien menjalani eksisi luas "buttockectomy" yang dimodifikasi untuk mengangkat tumor beserta nervus ischiadikus yang terlibat. Pada pengamatan satu tahun pasca pembedahan, pasien dapat berjalan dan tidak ditemukan tanda-tanda rekurensi.

Kata Kunci: buttockectomy, osteosarkoma

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INTRODUCTION

Pelvis is a quite common site for bone tumour in which the pelvic bone tumours constitutes around 10-15% from all bone tumours. Among them, chondrosarcoma, osteosarcoma and Ewing sarcoma are the most common bone tumour found.[1]

Osteosarcoma is most commonly found in metaphysis of long bones. It rarely originates from the pelvis or spine. Pelvis only represents 5% of osteosarcoma of all sites. Despite that, the osteosarcoma and other types of bone tumours of the pelvis presents a major challenge in orthopaedic oncology.[2] These tumours usually present late and their sizes are relatively large with rapid extension to gluteal muscles. In some cases that involves the centrally located pelvic tumour or sacrum, complete resection is not possible. Local control is also hard to achieve and have a poor outcome.[3]

Buttockectomy is originally the limb salvage procedure indicated for high and low-grade-soft tissue sarcomas of the gluteus maximus which most of them do not extend to the underlying retrogluteal space or involving the femur or sacrum.[4] We present a case of nine-year-old girl presenting with osteosarcoma of the right ischium

treated by using limb salvage procedure using modified buttockectomy approach instead of hemipelvectomy or standard utilitarian approach.

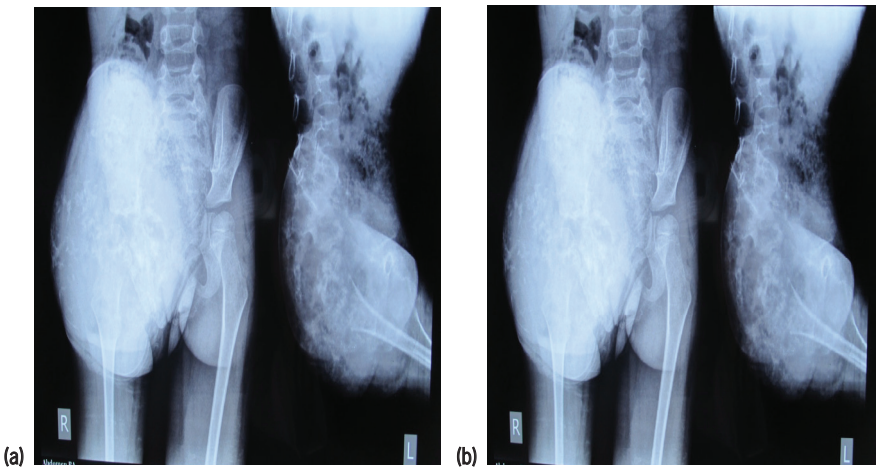
CASE

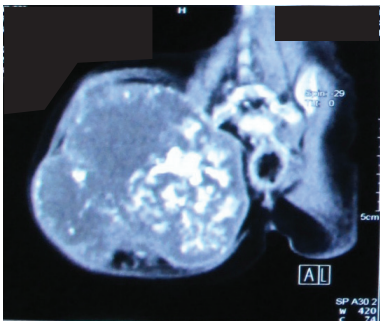
A 6 year old girl was referred to our institution with an enlarging mass on her buttock since 2 years before admission. The mass grew slowly without any pain up until 4 months ago, by which the pain started. Before the pain ensues, she was treated with an alternative medicine (herbs), but there were no improvements and the mass still enlarged. After the pain occurred, she was brought to local general hospital and then was referred to our institution. Besides the pain and enlarging mass on the buttock, her parents also complained about the patient's inability to stand since about 3 weeks ago due to the pain itself.

On physical examination, there was a hard mass on her right pelvis with a diameter approximately 11 cm (Figure 1). There was no tenderness on the lump or any disturbances on neurovascular function distal to the lump. The movement of the hip joint is limited, but still in the acceptable zone. No limitation was found in the knee and ankle range of motion. The other problem that we found is that the patient has a very low body mass index (malnutrition).

Figure 1: Clinical presentation the mass on the right buttock of the nine year old female patient (AP and lateral)

From the previous hospital, the patient has undergone a series of radiological examination including pelvic plain radiograph and computed tomography (CT) scan (Figure 2). From the pelvic plain radiograph, we suspected a malignant chondroid tumour on her right ischium with a possible extension to the femoral head.





(c)

Figure 2: Pelvic X-ray showing a malignant chondroid lesion of the right ischium with the possible involvement of the right femoral head (AP and lateral) (a and b). Axial CT scan showed suggestic a sarcoma on the pelvis (c)

Meanwhile on the abdominal computed tomography scanning, a soft tissue tumour was suspected on her right ischium along with the destruction of the right proximal femur and pelvis. No metastasis was found on the liver or on the lymph nodes. The magnetic resonance imaging (MRI) showed a clearer image on the pelvis. A malignant chondroid tumour was found on right ischium with soft tissue mass which extended to the gluteus maximus, medius, minimus muscle and piriformis muscle. The mass was also extending to the interior pelvis and involving the ischiadicus nerve [Figure 3].

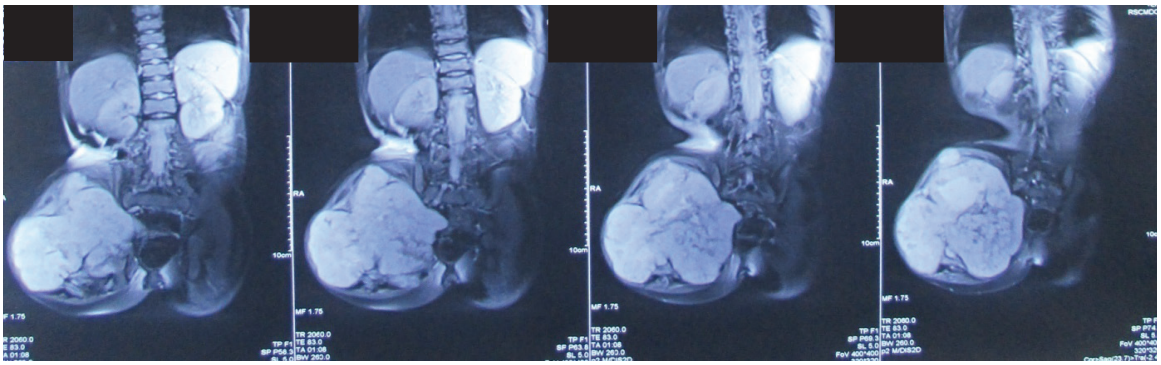


Figure 3: Coronal MRI showing the malignant chondroid lesion of the right ischium along with soft tissue tumour that involved gluteus maximus muscle, medius and minimus along with right piriformis. There was also an extension to intrapelvic organ and sciatic nerve.

Core needle biopsy showed a tendency to chondrosarcoma with differential diagnosis low grade osteosarcoma. Thus, we decided to do a wide excision – buttockectomy followed by adjuvant chemotherapy. A curvilinear incision was performed starting from the posterior iliac crest curving distally following the gluteus maximus muscle until reach the greater trochanter about 3 cm distal. Then the incision was curved posteriorly back to the inner aspect of the thigh to the gluteal fold forming a large posterior skin flap. Identification of the sciatic nerve was taken place. Since the sciatic nerve was unsalvageable we decided to resect it along with the tumour mass [Figure 4].

An osteotomy was performed on the ischium proximally and ilium distally. The fragment on ischium and ilium then was stabilized using ethibone 2.0. The tumour mass then was brought to pathological department and examined. On microscopic view, the tumour mass was consistent with osteosarcoma due to the presence of malignant osteoid and pleomorphic cells [Figure 5] .

After surgery, the patient underwent rehabilitation and several chemotherapy session. One year after operation, there was no recurrence and she was able to walk eventhough with a high step page gait [Figure 6].

Figure 4: A posterior skin flap was made to expose the gluteus maximus covering the bony tumour mass. Identification of the sciatic nerve which in exits from the tumour mass and therefore is unsalvageable

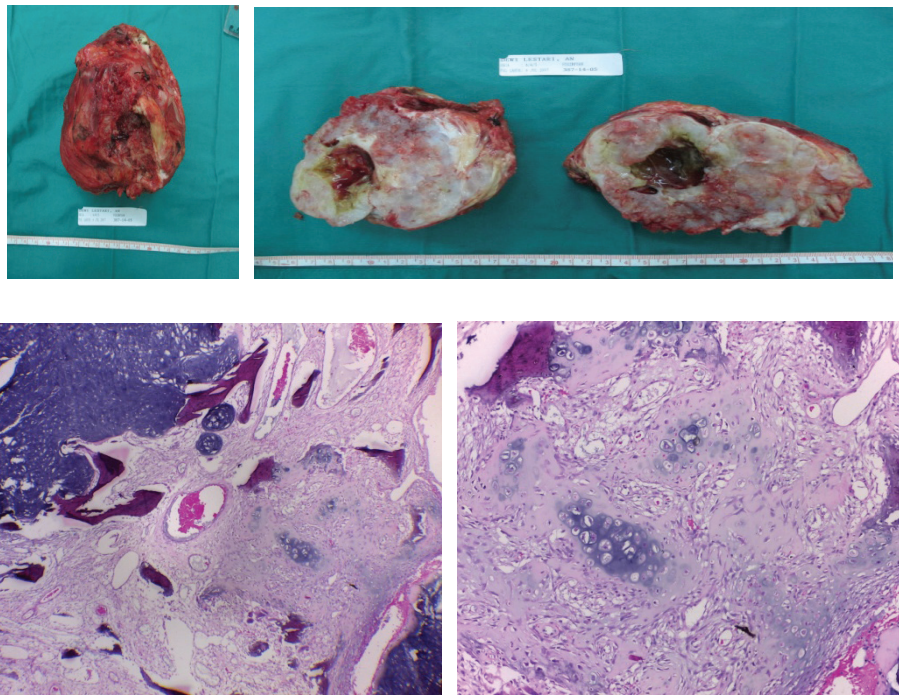


Figure 5: Gross pathology of the tumour showed the lesion is slight chondroblastic which is suggestive to chondrosarcoma. but the existence of malignant osteoid showed that the lesion was osteosarcoma. The histopathology features showed dominant neoplastic cartilaginous component with osteoid production. (H&E 100x , 400x)

Figure 6: Clinical picture of the patient one year after operation

DISCUSSION

The treatment of osteosarcoma of the pelvis varied from limb ablation (hemipelvectomy) or limb salvage. As mentioned before, limb ablation was the preferred choice until around early 80s. Despite that, it's still regularly performed in cases where it is not possible to obtain both adequate margin of tumour resection and a functional limb.[5] The advancement of the diagnostic procedure had enabled the surgeons to preoperatively evaluate the extent of the tumour thus made it possible to perform the limb salvage surgery.

Limb sparing procedure combined with adjuvant or neoadjuvant chemotherapy is the gold standard of malignant pelvic bone tumour treatment. Meanwhile some tumours are resistant to chemotherapy, such as chondrosarcoma. It is largely resistant to either conventional chemotherapy or radiation. [6] In our case, the core biopsy result was suspicious chondrosarcoma or low grade osteosarcoma. Thus, wide en-bloc excision is the treatment of choice.

Meanwhile, there's still some controversies whether the involvement of the sciatic nerve and/or femoral vessels are also the contra-indications for limb salvage surgery. In general, the involvement of one or both of these structures is the contraindication for limb sparing procedure since it will further impair the function of the limb.[5] A functionally impaired limb however is much preferred rather than an amputation. These patient whose sciatic or femoral nerve has to be sacrificed, will do well with proper rehabilitation and orthosis.[1] A series of study even reported that a limb salvage procedure has significantly better prognosis compared with patient treated with hemipelvectomy, but it might be related to the extension of the disease that requires a more aggressive treatment.[7] In this case, the MRI have shown that the sciatic nerve was involved in the tumour and have to be sacrificed along with the tumour. The margin of the tumour itself involves most soft tissues around the gluteus in which the clear margin of tumour was obtainable. Complete resection is always more preferred despite which technique was used to obtain it and salvage surgery is our preference in this case after evaluating the extent of the tumour preoperatively.

Instead of using conventional pelvic resection using the utilitarian approach, we preferred buttockectomy technique due to the extensive involvement of the gluteus maximus muscle. The technique which was proposed by Sugarbaker consist of these following steps: incision of the skin around the tumour, making a skin flap to expose the entire gluteus maximus muscle; identifying the inferior rolled edge of the muscle and traced laterally to its insertion on the iliotibial tract. Then we proceeds counter clockwise starting at inferior aspect of gluteus maximus. [8]

Buttockectomy was initially performed in soft tissue sarcoma cases which occurred in gluteus maximus and was contraindicated if there was an

extension of the sarcoma to the pelvic bone or sacral nerve.[4] Our approach by using buttockectomy for osteosarcoma of the pelvis was due to the vast soft tissue extension to major muscle in the gluteus region, especially the gluteus maximus. By using the buttockectomy approach, the entire gluteus maximus muscle can be obtained better compared by the utilitarian approach. Our buttockectomy approach is also modified in order to make a pelvic resection type 1 after the gluteus maximus incision was performed.

Prognosis of this patient remains poor despite the adequate tumour resection and chemotherapy given. Therefore, wide surgical margin is usually hard to obtain. Ham et al[7], analyzing the factors which influence the prognosis of the pelvis osteosarcoma, mention that positive prognostic factors are complaint less than three months before initial presentation, tumour size less than 8 cm, surgical resection and osteoblastic subtype. Saab et al [9] mentioned that metastatis at initial presentation contributes to the poor prognosis in pelvic osteosarcoma patients.[9] In our case, the prognosis remains indeterminate despite the clear surgical margin and no metastasis was found during preoperative examination. The nature of the tumour was chondroblastic thus it might have some resistance to either chemotherapy for systemic control. The size of tumour was also bigger than 8 cm in diameter and late presentation also contributes to the prognosis of our patient.

Wide surgical margin was consistently a significant prognostic factor in osteosarcoma series.[5,7,10] Ferari et al emphasized the importance of local control in pelvic osteosarcoma due to poor pathological response to chemotherapy and high incidence of chondroblastic variant.[11] Complete surgical resection is extremely important in pelvic osteosarcoma, but in the practice, it is very hard to obtain because of the complex anatomy of the pelvis which is associated with the association of the large vessel, nerves and visceral organ with minimal compartmentalization. Even some studies or author regard it is impossible to achieve adequate margin in difficult pelvic area.[12] Furthermore it is not the only deciding factor corresponding to the fact that wider surgical margin in hemipelvectomy does not yield a better survival in several studies.[5,7,10] Hemipelvectomy itself is reserved for a more advanced tumour or for the potentially curable cases since the devastating functional result and psychological effect to the patients.

Unlike the osteosarcoma of the extremity, the advance of chemotherapy has a limited value in treating pelvic osteosarcoma. Poor response of pelvic osteosarcoma is due to the chondroblastic nature of the tumour.[13] Recurrence rate was significantly lower in those patients who were treated by wide excision compared with intralesional excision.[14] Five year survival rate of Enneking stage IIB osteosarcoma patients despite surgical and chemotherapy was approximately around 21-40%.[7,10] Our case have been followed up for about one year, there was no recurrences found clinically eventhough the patient only underwent one series of chemotherapy due to the lack of compliance. As for the functional level, the patient already can performed her daily activities with minimal complaint. In conclusion modified buttockectomy is a considerable approach in treating ischium osteosarcoma with massive soft tissue extension to the buttock.

CONFLICT OF INTERESTS

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

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