

## Early outcome of palliative limb salvage surgery in patient with right proximal humerus osteosarcoma Enneking III at Dr. Soetomo Academic General Hospital: A case report

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### Abstract

**Introduction:** Osteosarcoma is a type of bone cancer that commonly affects the long bones, including the proximal humerus. This case report aims to investigate the clinical outcomes of patients with osteosarcoma of the proximal humerus enneking III. Follow-up assessments were conducted to evaluate post-operative outcomes.

**Case Presentation:** This was a case report of a 33-year-old man who came to an outpatient clinic referred from another island with a chief complaint huge lump in his right shoulder 6 years ago. The patient got several examinations and was diagnosed with osteosarcoma of proximal humerus enneking III accompanied by lung metastases. From the Clinical Pathological Conference (CPC) patient decided to do neoadjuvant chemotherapy followed by restaging and amputation, but the patient refused amputation. Since it was found that the neurovasculature was not encased, limb salvage palliative surgery was performed, in the form of wide resection surgery, reconstruction with bone cement and spacers, and also shoulder reconstruction with prolene mesh, then followed by adjuvant chemotherapy. Follow-up 15 months post-op showed there were no recurrences, stable lung metastases, and improvement in complications, Musculoskeletal Tumor Society (MSTS) score 26 out of 30 and Disabilities of the Arm, Shoulder and Hand (DASH) scores 37.9 out of 100.

**Conclusion:** In this case, the decision to undergo palliative limb salvage surgery is not an ordinary decision. Even though the outcome is not good, the patient is satisfied with the results, and with routine physiotherapy, we can assess the improvement and progressivity of the results of the operation.

**Keywords:** Osteosarcoma; Humerus; Enneking III; Limb salvage palliative surgery; DASH; MSTS

### 1. Introduction

Osteosarcoma is the most common type of malignant bone tumor in children and young adults, arising from mesenchymal stem cells. Osteosarcoma is a type of bone cancer that commonly affects the long bones of the body, including the proximal humerus. With an annual incidence rate of 3.1 cases per million in the US, osteosarcoma accounts for less than 1% of all newly diagnosed cancers in adults and 3–5% of those in children [1]. Osteosarcoma incidence rate at the humerus is about 10%, with 90% of tumors in the proximal humerus. A study reported 15 proximal humeral osteosarcoma tumor recurrences in 100 cases. Another study found a local recurrence rate of 13% in patients with proximal humeral osteosarcoma. In high-income countries, synchronous metastases are detected in 10% to 30% of patients with osteosarcoma [1,2]. However, in low and middle-income countries with delayed diagnosis, up to 40% of patients have metastatic disease at presentation.

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Limb salvage in osteosarcoma refers to the surgical procedure aimed at preserving the affected limb by restoring bone and joint function after the extensive resection of malignant bone tumors in the limbs. Osteosarcoma is the most common primary malignant bone tumor, mainly affecting children and adolescents, with the limbs being the primary site of occurrence (2,3). Limb salvage treatment is considered an effective standard treatment for osteosarcoma of the limb, with a success rate of 60%–80% [4]. This approach involves replacing the diseased bone and reconstructing a functional limb using techniques such as metal implants, bone grafts, or a combination of both. The goal is to remove the tumor with minimal complications while maintaining acceptable function, durability, and appearance of the limb [5]. Advances in limb salvage treatment, such as cryoablation, have been shown to be safe and effective in patients with osteosarcoma [3,6]. This approach is crucial in improving the success rate of limb salvage treatment and promoting the development of limb salvage surgery technology [4].

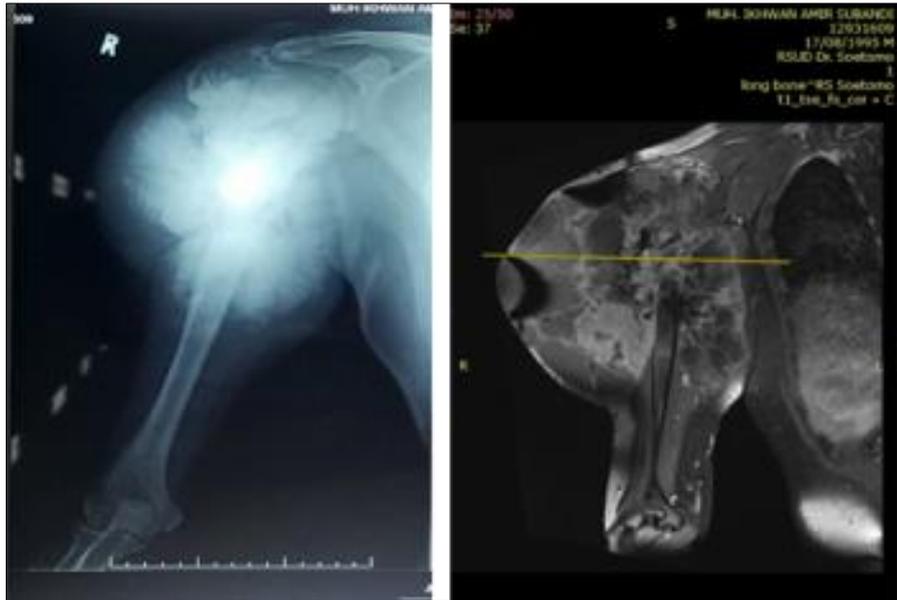
## 2. Case Report

In our case, the patient is a 33-year-old male working as a teacher with a chief complaint of a huge lump on the right shoulder since 6 years ago. Initially, the patient fell from a motorcycle in 2018 (6 years ago) and hit his right shoulder on the road. A few weeks later a lump appeared on the right shoulder the size of a marble. Initially, the lump was small at first and grew larger in the last 4 months (Figure 1). The patient came from outside of Java Island, seeking treatment at Mataram City Regional Hospital, due to limited equipment, the patient was referred to Dr. Soetomo General Academic Hospital.



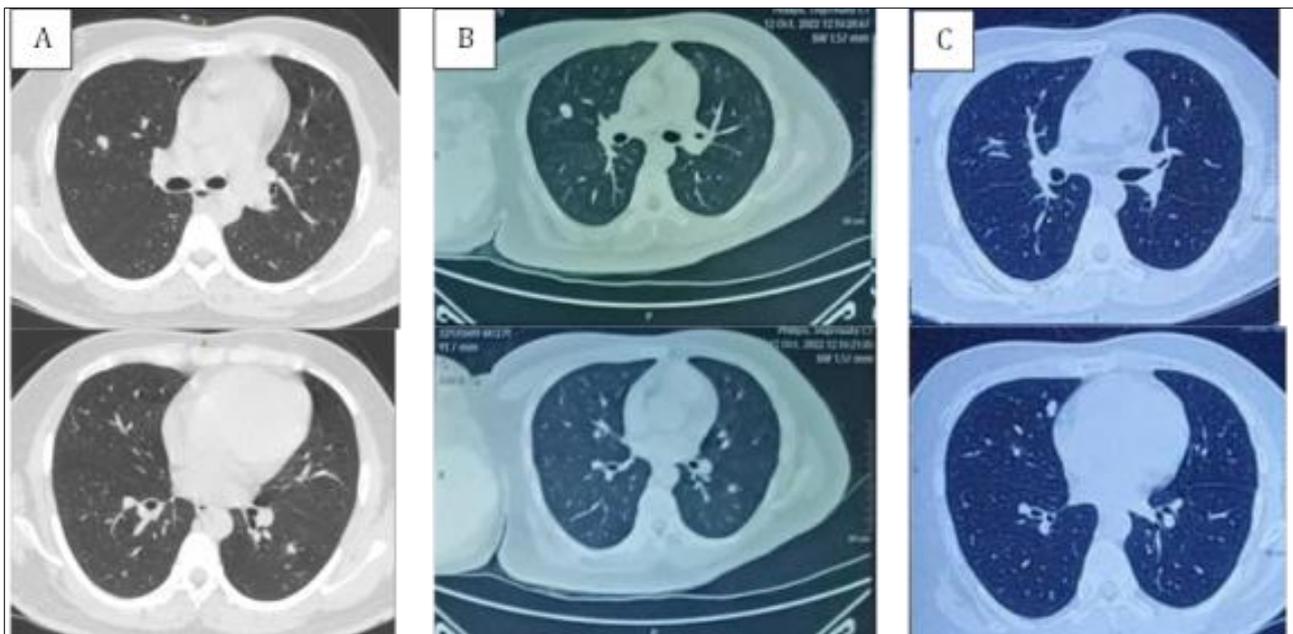
**Figure 1** Clinical picture before surgery

The patient went to Dr. Soetomo General Academic Hospital outpatient polyclinic and got a laboratory examination, a right humerus x-ray (Figure 2A), a humerus MRI with contrast (Figure 2B), a CT scan of the thorax, and FNAB. From MRI examination results were obtained visible bone destruction with bone marrow replacement in the epi-meta diaphysis 1/3 proximal to medial of the right humerus accompanied by soft tissue bulging with solid components, with cystic and necrotic components in it, size +/- 16,3 x 16,2 x 21, 6 cm which appeared isointense on T1WI/T2WI restricted diffusion area on DWI which on contrast administration showed heterogeneous contrast enhancement, sunburst type peritoneal reaction, mass infiltrating deltoid, biceps brachii, brachialis, brachioradialis, and right triceps brachii muscles, visible mass extending to the inferior side of glenohumeral joint, no visible involvement of the brachial artery. The distance of bone marrow replacement from the humeral head to the proximal mass is +/- 20.2 cm.

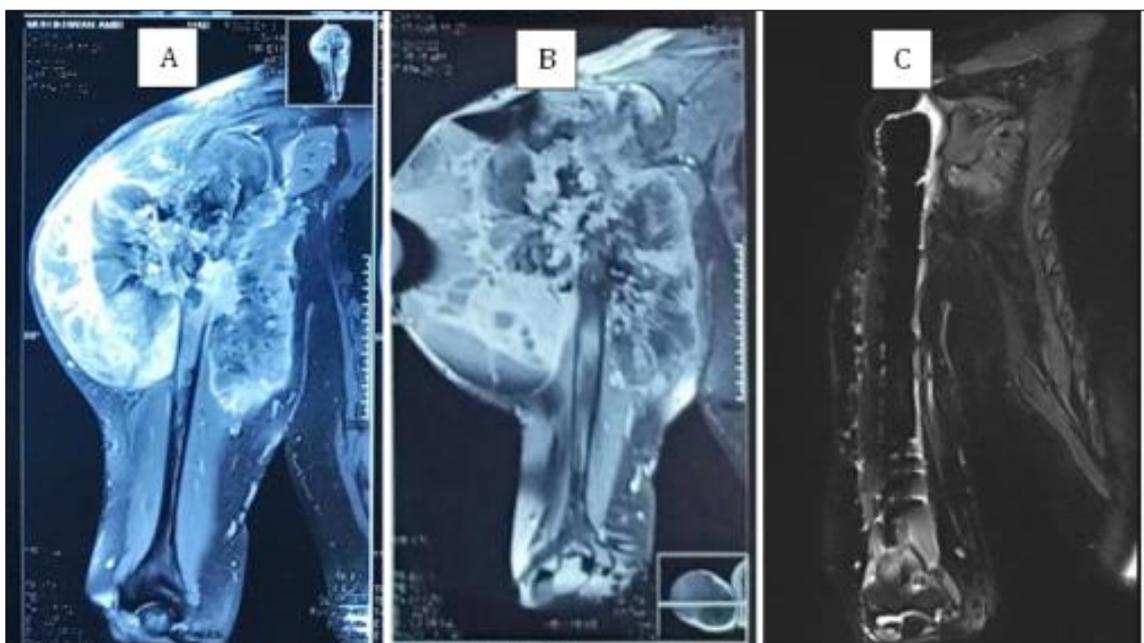


**Figure 2** (A) Pre-operative x-ray; (B) Pre-operative MRI

From the examination results obtained lab results in the form of increased ALP and LDH (Table 1), CT scans thorax there are multiple nodules in the lungs impression of metastases (Figure 3), and FNAB with the result of Osteosarcoma. DASH score measurement pre-operative results obtained a value of 8.3/100 (minimum disability) Then carried out the first Clinical Pathological Conference (CPC) was held on July 18th, 2022 with an assessment of right proximal humerus osteosarcoma enneking III and it was decided to plan for extra-articular disarticulation with partial removal of the glenoid taken.



**Figure 3** CT scan thorax (A) Pre-op, (B) Post Neoadjuvant chemotherapy, (C) Post-op



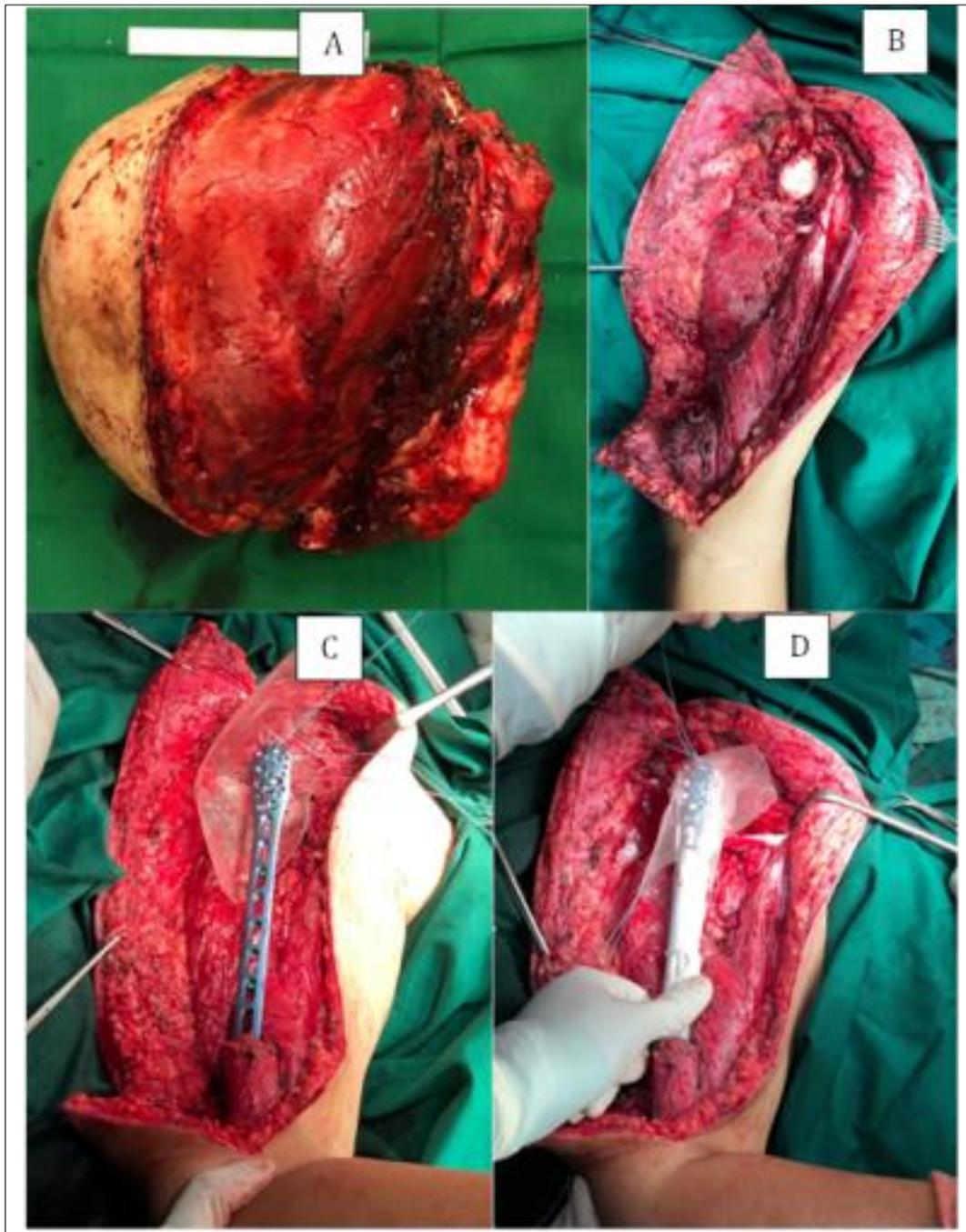
**Figure 4** Right Humerus MRI (A) Pre-neoadjuvant chemotherapy (B) Post neoadjuvant chemotherapy (C) 4 months post-operative

**Table 1** Laboratory result

Laboratory Result	Normal Range	Pre-operative	Postoperative
ALP	46-116 U/L	444	64
LDH	100-190 U/L	260	220

After the first CPC the patient was given neo-adjuvant chemotherapy 3 times with a regimen of doxorubicin + cisplatin in August 2022, after completion of neoadjuvant chemotherapy, a re-evaluation was carried out in the form of an MRI examination of the shoulder and CT scan of the thorax in October 2022. Right humerus MRI results when compared with MRI before neoadjuvant chemotherapy, morphological changes were found, primary malignant bone tumor most likely osteosarcoma in the epi-meta diaphysis 1/3 proximal to the middle of the right humerus measuring +/- 18 x 19.3 x 20.1 cm (impression of increase) with the proportion of necrotic tissue +/- 80% (same impression) (Figure 4). CT scan of the thorax showed the impression of increasing multiple nodules when compared to the previous CT scan.

The second CPC was conducted on October 30th, 2022 and it was decided to inform the patient to do a palliative therapy plan and consideration of forequarter amputation. The patient refused to do amputation because of the patient's job as a teacher. However, the patient refused amputation and the MRI results showed that the vasculature was still intact even though some of the nerves had encasement, so it was still possible to carry out limb salvage palliative surgery. The patient decided to do palliative surgery on November 21st, 2022 with the procedure of wide resection + open reduction internal fixation (ORIF) plating + bone cement spacer + shoulder reconstruction with prolene mesh (Figure 5). During the operation, it was found that a lot of soft tissue was affected by the tumor mass, including the deltoid, biceps brachii, brachialis, brachioradialis, and right triceps brachii muscles, so no massive reconstruction was carried out, only reconstruction with bone cement and spacers, and also shoulder reconstruction with prolene mesh.



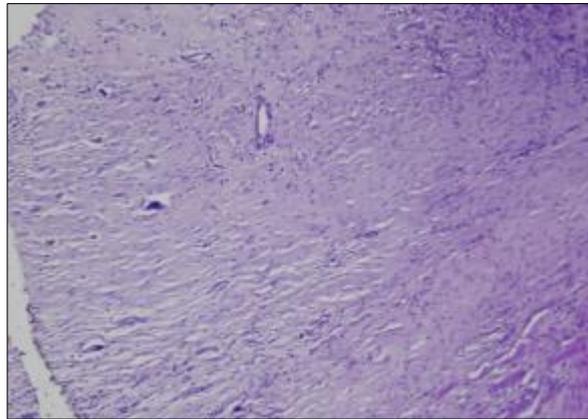
**Figure 5** Clinical picture during surgery (A) the tumor mass, (B) after wide resection, (C) ORIF Plating and shoulder reconstruction with prolene mesh, (D) implantation of bone cement spacer

The results of the x-ray after surgery (Figure 6) including histopathological examination results from wide excision were discussed again at the 3rd CPC on December 19th, 2022. The histopathological examination results showed that the cell types are osteosarcoma, osteoblastic and giant cell-rich type (Figure 7) with tumor size 20 x 19 x 14,5 cm, no lymphangioinvasion, no perineural invasion, Huvos grade 2, distance of tumor to anterior resection of 1 mm, distance of tumor to lateral resection of 2,5 mm, and edge of distal end resection, medial and posterior free of tumor cells, so it was decided to continue with adjuvant chemotherapy with doxorubicin + cisplatin regimen from January until March 2023. 5 months after surgery, the patient returned to the clinic (Figure 8) and evaluations were carried out starting with a laboratory, right humerus MRI, and thorax CT scan. When the patient-controlled, he had no complaints of pain, but the patient was still unable to extend the thumb and wrist (Figure 9). Laboratory results showed a decrease in ALP and LDH (Table 1), from MRI of the shoulder showed no impression of residual mass in the surgical bed (Figure 4), and a CT scan of the thorax showed thorax still obtained multiple nodules in the right and left lungs that suggested metastase process

(Figure 3). The evaluation of 5 months after surgery was discussed again at the 4th CPC on April 17th, 2023 resulting in a decision to evaluate clinical, x-ray, and laboratory in the next month.



**Figure 6** (A) Pre-op x-ray and (B) Post-op x-ray right humerus ap and lateral



**Figure 7** Histopathological examination post wide resection



**Figure 8** Clinical picture after 5 months of surgery



**Figure 9** Clinical picture of range of motion right upper extremity

After 8 months post-surgery, a CPC discussion was conducted again to evaluate the patient's condition, and it was decided that there was no residual mass. The patient was educated to control back if he suspected a lump. Because the patient has a very high lesion of the radial nerve, the procedure of EMG NCV examination was carried out on October 16th, 2023 and the results obtained for the plexus lesion right denervated brachial plexus on the posterior cord (at the level between the branches of the thoracodorsal nerve to the branching of the radial and axillary nerves) and incomplete lesion of C5-T1 postganglion right irritative brachial plexus lesion. Apart from the results of the MMT evaluation, there are several muscles that have a strength of 0, including the supraspinatus, infraspinatus, subscapularis, teres major, teres minor, deltoid, extensor carpi radialis longus, supinator, extensor carpi ulnaris, and extensor digitorum muscles. While the trapezius and palmaris longus muscles have a motor strength of 5, the pronator teres and flexor carpi radialis have a motor strength of 4, from the results of the MMT evaluation it was concluded that there were no contraindications for tendon transfer. The patient will then be planned for Trapezioplasty surgery for shoulder abduction function and Jones Tendon Transfer for wrist extension.

After follow-up for 15 months post-op, there were no recurrences, stable lung metastases, and improvement in complications. Evaluation of the current condition after 15 months of surgery the MSTS score in this patient is 26 out of 30 (Table 2), and the DASH score in this patient is 37.90 out of 100 (Table 3), showing improvement in daily activities. The patient is currently going off as a teacher and working as a self-employed person. The patient is still in the follow-up phase for 5 years and will undergo further reconstruction in the form of tendon transfer.

**Table 2** MSTS Score

Score	Pain	Function	Emotional	Hand Positioning	Manula Dexterity	Lifting Ability
5	No pain	No restriction	Enthusied	Unlimited	Unlimited	Normal load
4	Intermediate	Intermediate	Intermediate	Intermediate	Intermediate	Intermediate
3	Modest/ Non-disabling	Recreational restriction	Satisfied	Not above the shoulder of no/ Prosupination	Loss of fine movement	Limited
2	Intermediate	Intermediate	Intermediate	Intermediate	Intermediate	Intermediate
1	Moderate/ Disabling	Partial restriction	Accepts	Not above waist	Cannot pinch	Helping only
0	Severe disabling	Total restriction	Dislikes	None	Cannot grasp	Cannot help

**Table 3** DASH score

1	Open a tight or new jar	No difficulty	Mild difficulty	Moderate difficulty	Severe difficulty	Unable
2	Write	No difficulty	Mild difficulty	Moderate difficulty	Severe difficulty	Unable
3	Turn a key	No difficulty	Mild difficulty	Moderate difficulty	Severe difficulty	Unable
4	Prepare a meal	No difficulty	Mild difficulty	Moderate difficulty	Severe difficulty	Unable
5	Push open a heavy door	No difficulty	Mild difficulty	Moderate difficulty	Severe difficulty	Unable
6	Place an object on a shelf above your head	No difficulty	Mild difficulty	Moderate difficulty	Severe difficulty	Unable
7	Do heavy household chores (eg. wash walls, wash floors)	No difficulty	Mild difficulty	Moderate difficulty	Severe difficulty	Unable
8	Garden or do yard work	No difficulty	Mild difficulty	Moderate difficulty	Severe difficulty	Unable
9	Make a bed	No difficulty	Mild difficulty	Moderate difficulty	Severe difficulty	Unable
10	Carry a shopping bag or briefcase	No difficulty	Mild difficulty	Moderate difficulty	Severe difficulty	Unable
11	Carry a heavy object (over 10 lbs)	No difficulty	Mild difficulty	Moderate difficulty	Severe difficulty	Unable
12	Change a lightbulb overhead	No difficulty	Mild difficulty	Moderate difficulty	Severe difficulty	Unable
13	Wash or blow dry your hair	No difficulty	Mild difficulty	Moderate difficulty	Severe difficulty	Unable
14	Wash your back	No difficulty	Mild difficulty	Moderate difficulty	Severe difficulty	Unable
15	Put on a pullover sweater	No difficulty	Mild difficulty	Moderate difficulty	Severe difficulty	Unable
16	Use a knife to cut food	No difficulty	Mild difficulty	Moderate difficulty	Severe difficulty	Unable
17	Recreational activities which require little effort (eg. cardplaying, knitting, etc.)	No difficulty	Mild difficulty	Moderate difficulty	Severe difficulty	Unable
18	Recreational activities in which you take some force or impact through your arm, shoulder, or hand (eg. golf, hammering, tennis, etc.)	No difficulty	Mild difficulty	Moderate difficulty	Severe difficulty	Unable
19	Recreational activities in which you move your arm freely (eg. playing frisbee, badminton, etc.)	No difficulty	Mild difficulty	Moderate difficulty	Severe difficulty	Unable
20	Manage transportation needs (getting from one place to another)	No difficulty	Mild difficulty	Moderate difficulty	Severe difficulty	Unable
21	Sexual activities	No difficulty	Mild difficulty	Moderate difficulty	Severe difficulty	Unable
22	During the past week, to what extent has your arm, shoulder, or hand problem interfered with your	Not at all	Slightly	Moderately	Very limited	Unable

	normal social activities with family, friends, neighbours, or groups?					
23	During the past week, were you limited in your work or other regular daily activities as a result of your arm, shoulder, or hand problem?	Not limited at all	Slightly limited	Moderately limited	Very limited	Unable
Please rate the severity of the following symptoms in the last week						
24	Arm, shoulder, or hand pain	None	Mild	Moderate	Severe	Extreme
25	Arm, shoulder, or hand pain when you performed any specific activity	None	Mild	Moderate	Severe	Extreme
26	Tingling (pins and needles) in your arm, shoulder, or hand	None	Mild	Moderate	Severe	Extreme
27	Weakness in your arm, shoulder, or hand	None	Mild	Moderate	Severe	Extreme
28	Stiffness in your arm, shoulder, or hand	None	Mild	Moderate	Severe	Extreme
29	During the past week, how much difficulty have you had sleeping because of the pain in your arm, shoulder, or hand?	No difficulty	Mild difficulty	Moderate difficulty	Severe difficulty	Unable
30	I feel less capable, less confident, or less useful because of my arm, shoulder, or hand problem	Strongly disagree	Disagree	Neither agree or disagree	Agree	Strongly agree

### 3. Discussion

Osteosarcoma is a deadly cancer. If the patient doesn't get any proper and prompt treatment, lung metastases can be a deadly complication. The daily lifestyle of patients with osteosarcoma can be significantly disturbed because of various limitations. Currently, the primary approach for treating osteosarcoma involves administering neoadjuvant chemotherapy, followed by extensive surgical removal of the tumor, and then providing additional adjuvant chemotherapy. Research has demonstrated that the use of neoadjuvant chemotherapy has resulted in a 5-year survival rate of 50-60% for osteosarcoma, as opposed to a rate of 20% when surgery is the sole treatment. In the same condition, the patient who received adjuvant treatment had significant improvement in 2 years of Disease Free-Survival (DFS) (55% vs 20% only operation) and Overall Survival (OS) (80% vs 48 % only operation) (7,8).

In this patient with a late case of osteosarcoma accompanied by metastases to the lung, the patient has entered enneking III. Generally, patients with enneking III osteosarcoma have appropriate actions in the form of palliative limb ablation (amputation). However, in this patient, the patient refused to undergo amputation, so it was decided to undergo palliative limb salvage surgery. Even though the results of the operation were not very good, the patient was quite satisfied with the results of the operation and the comparison of the pre-operative and post-operative DASH score evaluation results was not too far apart (minimum to moderate disability). With the current ROM condition, the patient is still being followed up for the next 5 years to plan for the next operation in the form of trapezioplasty and Jones transfer.

### 4. Conclusion

Osteosarcoma is one of the leading causes of disability due to amputation and decreased function in daily life. In patients with osteosarcoma enneking III, the decision to undergo palliative limb salvage surgery is not an ordinary decision. By carrying out palliative limb salvage surgery, even though the outcome is not good, the patient is satisfied with the results of the operation, and with complete training and good follow-up, we can assess the improvement and progressivity of the results of the operation as indicated by the MSTS score of 26 out of 30 (indicating ability for daily activities) and an average DASH score of 37.9 out of 100 (moderate disability). This palliative limb salvage procedure for osteosarcoma showed good results in improving light daily activities.

## Compliance with ethical standards

### *Disclosure of conflict of interest*

The authors declare that they have no conflict of interest.

### *Statement of informed consent*

Informed consent was obtained from all individual participants included in the study.

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