

3d Printed Customized Total Scapular Replacement With Constrained Shoulder Joint After Wide Resection For Scapula Chondrosarcoma Treatment: A Case Report

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Case report

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Abstract

Introduction

Total scapular replacement surgery with constrained reverse shoulder joint a complex surgery, especially in the selection of replacement materials. The advent of three-dimensional (3D) printing technique contribute significantly to the success of surgery

Case presentation

A 62-year-old female patient discovered a scapular tumor at the end of 2017. She underwent curettage and bone graft surgery 3 times. The histopathological diagnosis was enchondroma. However, tumor recurrence and malignant transformation unfortunately presented later. She visited our center and total scapular replacement with constrained reverse shoulder joint was then performed. After 6 months, the patient's functional outcome was evaluated by using Musculoskeletal Tumour Society Score (MSTS) with a score of 26/30. There was no sign of local tumor recurrence. The functional of elbow and hand was preserved almost completely. The range of active movement of the shoulder was 60° for forward flexion and abduction, and 45° for backward flexion.

Discussion

Despite the follow-up time was still short, the use of the constrained shoulder joint with the applications of patient-specific 3D printing showed very positive outcomes. Patient was able to start rehabilitative early without any problems. More follow-up time would be needed to evaluate the long-term effectiveness of this method.

Conclusion

Total scapular replacement with constrained reverse shoulder joint using 3D printing technology is a suitable option for the patients with scapular tumor invading the muscles of the rotator cuff.

Introduction

Chondrosarcoma is the second most common primary bone cancer, after osteosarcoma [1]. This cancer usually occurs in middle-aged adults.

Chemotherapy and radiation therapy have limited efficacy in the treatment for chondrosarcoma. These therapies are often applied to the mesenchymal, clear cell and dedifferentiated chondrosarcomas to improve survival rates and prevent local recurrence [2].

Surgery is the key treatment in chondrosarcoma. If chondrosarcoma is classify as low malignant, small in size without bone destruction and invading surrounding soft tissue, the preferred treatment is thorough curettage with local adjuvant treatment such as 90-degree alcohol, liquid nitrogen...The patient was then

required periodic follow-up. In chondrosarcoma 2 and 3-grade with high malignancy, en bloc with wide resection is the principle of surgery in order to make sure all malignant tissues have been removed completely [3].

Total scapular replacement with constrained reverse shoulder prosthesis in a patient with scapula cancer was recently reported by James C Wittig in 2002 [17]. In case the scapular tumor is extensive, radical resection was required to complete removal of the whole scapula and adjacent soft tissues such as the rotator cuff that results in functional deficits in both the shoulder and elbow joints and the wrists [4]. Due to the removal of the total scapula and surrounding soft tissue, this treatment brings effective outcomes in eliminating cancer. The replacement with a scapular prosthesis designed with constrained reverse shoulder joint help to maximize shoulder function in case of irreparable rotator cuff tears. It may lead to a high success rate and is the preferred surgical treatment [5].

The scapula is a special bone. Before the 1970s, the treatment of replacing scapula in scapula tumors were limited, resulting in most scapular resections requiring forequarter amputation. After the Tinkoff-Lindberg surgical method was introduced, amputation treatment decreased, but the function of the shoulder joint was very limited [6]. With a deeper understanding of the biomechanics of the scapula, scapular reconstruction surgery has evolved. The surgery initially used allogeneic bone grafts, PolyEtherEtherKetone (PEEK) and prosthesis. The application of 3D printing technique support to create an accurate reconstruction of the scapula for specific patient. Besides, the combination with the constrained shoulder joint also helps to maximize the function of the shoulder joint in case of irreparable injury to the rotator cuff tendon and to facilitate the early recovery of patients' function.

In this article, we would like to present a case of total scapular replacement with constrained shoulder joint for a patient with bone cancer using 3D printing technology. This case report was made according to the SCARE 2020 guideline [7].

Case Presentation

A 62-year-old female patient presented with dull pain, increasing nocturnal pain in the right shoulder since the end of 2017. The patient did not get better with pain medication. She went to the hospital and detected a scapular tumor. The medical history showed that she underwent curettage and bone graft surgery 3 times in the past. The pathology was diagnosed as an endochondral tumor. The last surgery was conducted in March 2020. However, the patient continued to feel pain at the old surgical incision site. The appearance of a tumor in the old lesion area gradually enlarged and progressed rapidly since February 2021. On examination, we suspected a recurrent scapular tumor and possibly malignant transformation. On computed tomography (CT), the scapula was almost completely destroyed. Therefore, the decision to conduct the total scapular replacement surgery was made.

Based on the patient's most recent CT scan, we measured the parameters for preoperative planning and obtained a customized scapular prosthesis with a reverse shoulder joint using 3D printing technique. The

prosthesis had the same size as the actual patient's scapula. Many holes with a diameter of 5mm were drilled at the edge of the prosthesis for soft tissue reconstruction.

The patient was in the lateral position. She was placed under general anesthesia for the surgery. A Z-shape incision along the spine and the medial border of the scapula.

Each muscle with its attached tendon around the scapula was exposed, cut and marked. Spinal accessory nerve and suprascapular nerve were carefully preserved. Next step, the attachment point of the rotator cuff muscles to the scapula was identified and tagged after wide resection of scapula chondrosarcoma. The whole scapula was then released and removed. After that, tumor margins were obtained by intraoperative biopsy during the surgery to make sure all of the cancer was eliminated.

The humerus head was removed. The canal was edited to match the size of the humeral stem in the scapular prosthesis. The correlation between the position of the scapula and the constrained reverse shoulder prosthesis were checked and evaluated after inserting the humeral stem into the humeral canal. The appropriate positions should be marked. The humerus stem was then fixed to the humeral canal with bio-cement. When the cement solidified, reconstruction of the scapula prosthesis to the anatomical position was proceed.

Attention was taken with our greatest effort to preserve the soft tissues. Based on preoperative data, many holes were drilled on the scapula prosthesis same as the point of attachment of muscle groups to the scapula. After stabilizing the scapula prosthesis, we sutured the surrounding muscle tissue to the prosthesis through the holes at the edge of the prosthesis with undigested surgical sutures. The restoration of the position of the attachment points ensures the anatomical position of the muscles. In addition, it helps to ensure balance for normal movement and function between opposing muscle groups.. As a result, the scapula prosthesis will be balanced in its original position, with no dislocation as well as being able to move flexibly, helping to perform the function of the shoulder joint after surgery smoothly.

A postoperative CT image obtained revealed the status of the scapula and reverse shoulder prosthesis in a good position. The structure of the scapula and the reverse shoulder prosthesis is well shaped as expected before surgery.

On the first day after surgery, the patient was able to sit up, passively move her shoulder and check the movement well. There were no signs of numbness, paralysis of the deltoid, supraspinal and subspinal muscles. The patient was instructed to rehabilitate immediately. She was discharged after 7 days and performed regular outpatient rehabilitation. We re-examined patients every 3 months. The patient's postoperative pathology was low-grade chondrosarcoma.

After 6 months, the patient's MSTS score reached 26/30 and there was no sign of local tumor recurrence. The functional elbow and wrist was preserved almost completely. The range of active movement of the shoulder was 60° for forward flexion and abduction and 45° for backward flexion. The range of passive

motion is normal. The patient did not have shoulder pain. She could use her operated hand for daily activities.

Discussion

Chondrosarcoma usually occurs in adults over the age of 50. It is more common in men than women [8]. It commonly presents on the cartilage cells of the thigh bone (femur), shoulder, or pelvis. In contrast, it rarely starts in the knee, ribs, skull. The most common clinical symptoms of chondrosarcoma may include the following: Dull pain that increases gradually over time, pain is usually worse at night, and a large lump (mass) presents on a bone.. Chondrosarcomas are divided into three grades [9]:

Chondrosarcoma grade 1: Low malignancy, lesions/atypical cartilaginous tumors rarely metastasize, rarely recur, and have a 10-year survival rate of >80%.

Chondrosarcoma grade 2, 3: A malignant tumor is associated with a poor prognosis, high local recurrence rates, a lung metastasis rate of >50%, and a 10-year survival rate of <30%.

The prognosis of a patient with low-grade malignancy is very good, the 5-year survival rate is over 90%. There are 1-9% of Enchondroma that transform into malignancy. [1]. Treatment of endochondral tumors can be wide resection combined with bone grafting.. However, it is necessary to carefully evaluate the size of the tumour and determine the extension of invasion, and remove all of a cancerous tumour. Our patient was a typical case of secondary bone cancer which appeared in middle-aged people. Her tumor was the malignant transformation from the benign endochondral tumour. Fortunately, she went to our centre to conduct a medical examination and was treated promptly when the tumour just transformed into malignant. She underwent the radical resection of the tumor with a safe margin which means that no malignant cells tumor cells are seen at the resection surface with a microscope. There was no sign of metastasis.

Although the patient underwent 3 times of tumour resections in the past, the tumour still relapsed and converted to chondrosarcoma. The reason was that because the patient was treated at a non-specialist facility, the resection surgeries were not effective in completely removing the tumour cells. In addition, it was challenging to differentiate between enchondroma and low-grade chondrosarcomas. Therefore, periodic examination plays an important role in the early detection of disease progression for patients with tumor bone.

After recognition of malignant transformation to chondrosarcoma, it was necessary to have a plan of wide surgical resection. Because our patient presented large lesions that caused significant bone destruction, the total scapular replacement was compulsory. Total scapular resection is the treatment for tumors of the scapula and has succeeded in preserving elbow and hand function. Limited range of motion and shoulder instability are the major complications of this method. Despite several reconstruction methods have been tested such as bone suspension, prosthetic replacement, bone grafting or soft tissue reconstruction, functional outcomes were poor.

Before the 1970s, surgical tumor resections at the shoulder were almost always treated with amputation. **However,** when the Tickhoff-Linberg procedure was introduced, the indication for amputation was gradually reduced [10]

Scapular prostheses were presented with an acceptable range of motion and function in recent reports [11] [12]. However, long term positive outcomes were still not confirmed. Furthermore, a scapular prosthesis was not popularly available, and its design is not well manufactured yet.

The development of artificial materials offers many options for the reconstruction of the shoulder joint. The reconstruction materials were originally made simple from allograft, PEEK or industrial plastic. [11] [12].

In the past, most scapular prostheses do not match the patient's anatomy and may affect shoulder function. Nowadays, 3D printing technology represents a big opportunity to allow for customized and printed patient-specific scapular prostheses. These prostheses may increase the patient's chances of functional improvement. In recent years, using 3D printing techniques for reconstruction of the scapula with similar patient's anatomy has been applied more and more frequently and has very positive initial results. A study by Beltran et al. [13] showed that the MSTS score for a personalized scapula prosthesis replacement after tumour resection was 87%. Liu et al. [14] described the treatment of scapula tumours that applied the 3D-printed PEEK scapular prosthesis and achieved satisfactory shoulder joint function. 3D printing allows for accurate reconstruction, improving mobility, and facilitating the early recovery of patients' functions.

The shoulder prosthesis was designed to match the patient's anatomy and combined with the constraint structure to help stabilize the shoulder joint. Although the use of 3D printing technology for scapular reconstruction had not yet been reported clinically and needs more time to evaluate, it would be a new trend that will be popularized in the future [15]. The material was a titanium alloy, which was widely used in the artificial joints and other implant materials. Titanium alloy had very good resistance to bending and compression forces. Furthermore, an advantage of this material is excellent compatibility [13].

In addition, the scapula closely connected to the shoulder joint and surround complex system of muscles. Reconstruction in an effort to optimally restore the shoulder function requires understanding the biomechanics and anatomy of each muscle group. The artificial shoulder joints are designed with suture holes corresponding to the original attachment points of muscles to restore the anatomy and function for each muscle group. In our intraoperative, after identifying and cutting the muscles, they were immediately sutured to mark their attached point for reattaching to the the prosthesis later. Because the tumor invades some muscle groups, there may be a lack of length for tendon reconstruction, which can cause pain and imbalance in the soft tissues around the scapula. To overcome this challenge, it is extremely important to move the attachment point of muscles as little as possible, and at the same time the muscle groups were carefully released from the surrounding soft tissues. However, functional

impairment caused by tendon insufficiency is inevitable. Accompanied by a lot of damage to the rotator cuff tendons. This affects the arc of motion of the shoulder joint. When raising the arm, the rotator cuff no longer responds well to keep the rotator cuff at the center of the socket, instead, the deltoid muscle tends to pull the rotator upward, causing the arm to not be raised further.

At the same time, we evaluated preoperatively the patient's tumor and recognized that it invaded extensively the joint capsule and the surrounding soft tissue. The large lesion may lead to soft tissue deficiency after wide tumor resection and destabilize the artificial shoulder joint. Therefore, total scapular replacement with constrained reverse shoulder prosthesis was our surgery plan and implemented for this patient.

Both constrained reverse shoulder prosthesis and non-constrained reverse shoulder prosthesis are designed with medialisation of the centre of rotation of the glenohumeral joint to enhance range of shoulder motion. However, constrained reverse shoulder prosthesis has the glenosphere retained in the humeral head without affecting shoulder rotation.

In cases of non-constrained reverse shoulder replacement, it is necessary to reconstruct the joint capsule firmly to prevent glenohumeral dissociation and stabilize the entire construct for shoulder active function. However, shoulder instability often occurs in cases of large soft tissue lesion in extensive tumor resection [16].

The constrained total scapular prosthesis had a locking mechanism between the scapular part and humeral part. The constrained prosthesis stabilized the shoulder by restoring the active function of the rotator cuff, compensating for the lack of the joint capsule as well as the point of contact support to convert the deltoid straight upward pull into abductor motion. [17]

Compared with the non-constrained structure, the constrained reverse shoulder prosthesis has several advantages: a) create a similar structure as a joint capsule, b) stabilize the humeral head and the scapula and prevent upward movement of the humerus, and c) simulate the normal joint force vector of the muscles at the artificial shoulder joint.

Up to now, there have not been many reports of scapula replacement with a constrained total scapula prosthesis. Most authors agreed that this treatment was a safe and reliable method for scapula reconstruction after resection of high-grade sarcomas. This treatment also provided acceptable functional outcomes and a low complication rate [18] [19].

The first report on constrained total scapula replacement in patients with scapular tumours was done by James C Wittig et al. in 2002. They described three patients who identified as a high-grade sarcoma. At latest follow-up, the MSTS score was from 80-90%, the shoulder joints were stable, and there was no shoulder pain [18].

William J. Cundy et al. retrospectively compared outcomes of 41 patients of reverse shoulder replacement, of which 21 were unconstrained prosthesis and 19 were constrained shoulder replacements. At a mean

follow-up of 4.2 years, there were 5 patients in the constrained group (26%) who had to undergo surgery again due to dislocation, none of which were in the unconstrained group. The authors believed that the impact of repeated movement lead to the polyethylene being deformed causing dislocation.

However, the constrained implants used in the author's report were semi-constrained prosthesis, which had a high dislocation rate. Our patient was reconstructed by a fully-constrained implant which provided greater stability. Most of the authors reporting on the constrained reverse shoulder prosthesis used the fully-constrained type and had good results.

Mavrogenis et al. (2009) reported a case study treated with total constrained shoulder prosthesis and reverse-linked proximal humerus arthroplasty in a patient with Ewing's sarcoma of the scapula. Twelve-month follow-up after surgery, the shoulder joint was stable and painless. Forward flexion and abduction ranged about 30 degree [10].

Ten patients who underwent the total scapular replacement with constrained reverse shoulder prosthesis for bone malignant tumor were retrospectively reviewed by Tang et al. (2011). The implant was manufactured at ChunLi Co (Beijing, China). At latest follow-up, there was one case of dislocation, one case of infection and the average MSTS score was 76.7% [19].

Wang B et al. (2018) reported a series of 8 cases treated with total scapular replacement with a constrained reverse shoulder prosthesis after malignant tumor resection. At the final mean follow-up of 61.8 months, the mean MSTS score was 23.5. There were no cases of dislocation or loose joints [20].

Abdulla I et al. (2018) implemented a test on six cadaveric shoulders. He used an instrument of measuring forces across the joint with varying constraints to record the joint kinematics, loads and muscle forces. Finally, he concluded when the constraint joint was implanted to improve the joint stability, joint load and deltoid forces during active abduction were not affected significantly. Simultaneously, the range of motion was also not significantly different from that of a conventional reverse shoulder [21].

William J.Cundy et al. conducted a systematic review on functional shoulder assessment of inverse shoulder joints. He showed that there was no functional difference between the two types of joints. MSTS scores of the unconstrained inversion shoulder joint and constrained inversion shoulder joint fluctuated 63 - 90% and 60 -77.7%, respectively [22].

Our patient had a post-operative follow-up for 6 months. The MSTS score reached 26/30, and the functional elbow and hand were completely preserved. The range of active movement in the abduction and forward reaches 60 degrees, back 45 degrees. The range of passive motion was normal. The patient did not have shoulder pain and could use her operated hands for daily activity. The function of the shoulder joint was restored well because of our effort to optimally preserve the tendon and muscle system around the scapula during surgery. The design of constrained shoulder combined with 3D printing technology in accordance with the patient's parameters helped to stabilize the shoulder joint, thereby preserving almost completely the function of the elbow and hand.

Conclusion

Total scapular replacement with constrained reverse shoulder prosthesis using 3D printing technology for Chondrosarcoma patients showed good postoperative function and a low complication rate. This treatment is a suitable option for the patient with chondrosarcoma and the lesion appeared to have invaded the rotator cuff muscles.

Declarations

Ethics approval and consent to participate

The procedures used in this study adhere to the tenets of the Declarations of Helsinki.

Case reports are approved by the Joint- Consultation for Approval of Surgery and are granted an exemption from requiring ethical approval at Vinmec Healthcare System.

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal when needed.

Consent for publication

Consent for publication had been obtained from that person already

Availability of data and materials

Not applicable

Competing interests

The authors declare that they have no competing interests

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Authors' contributions

DTT: the main doctor conceived the original idea and operated the patient

STQN and DMQ: summed up, operated the patient, revised manuscript

TDT and TVC: wrote the manuscript

All the authors read and approved the final manuscript.

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Not applicable.

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Figures



Figure 1

Preoperation CT images

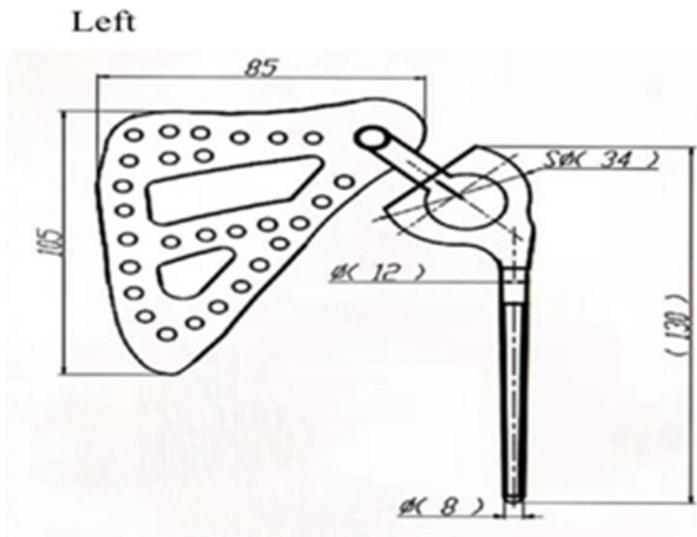


Figure 2

Scapula and constrained reverse shoulder prosthesis

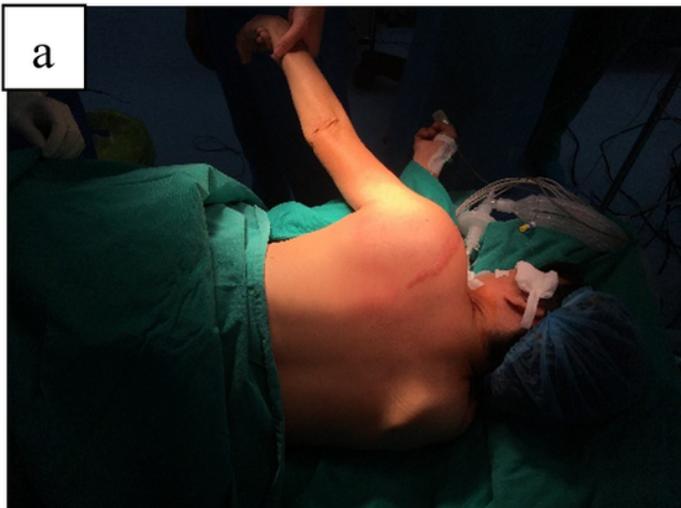


Figure 3

Patient position and surgical site

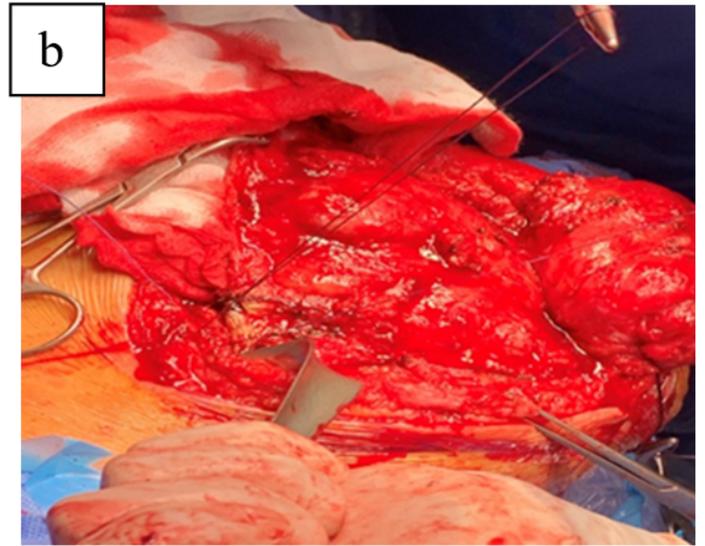
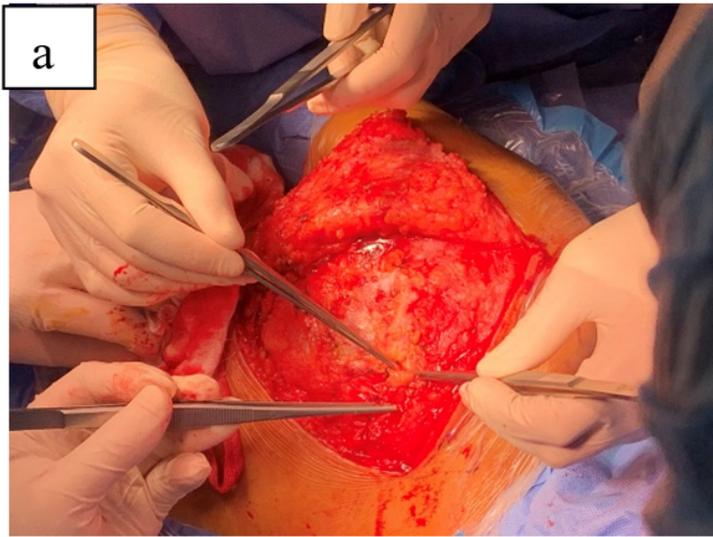


Figure 4

Muscles were exposed and marked.

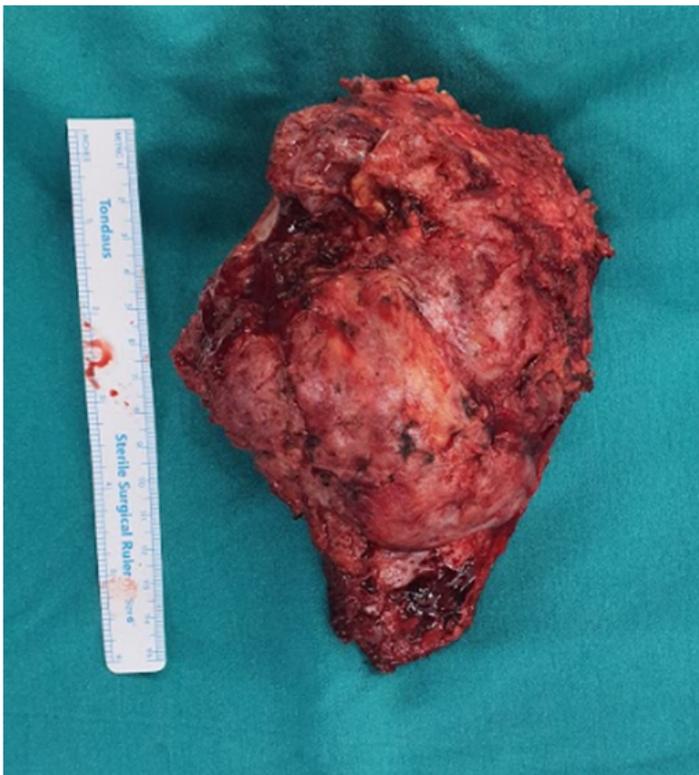


Figure 5

Chondrosarcoma scapula after removing

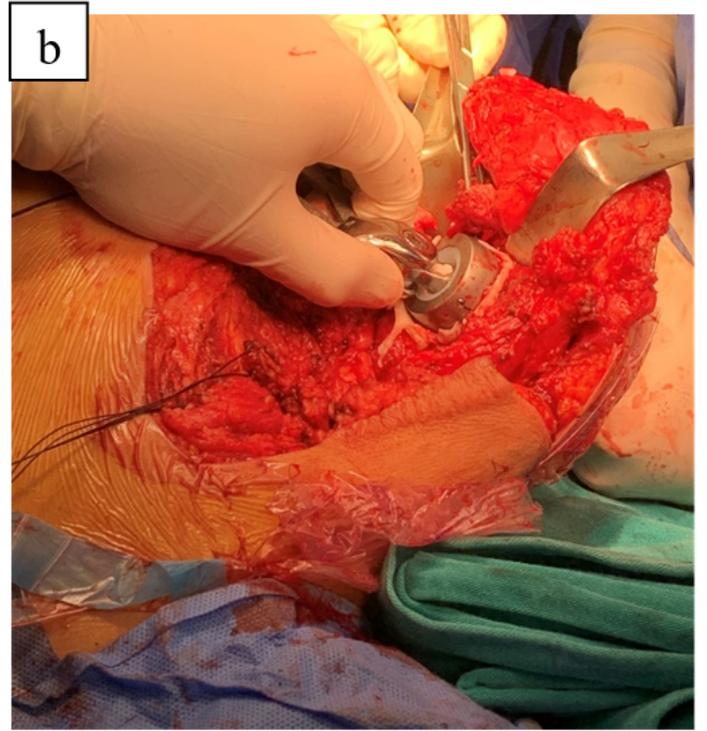
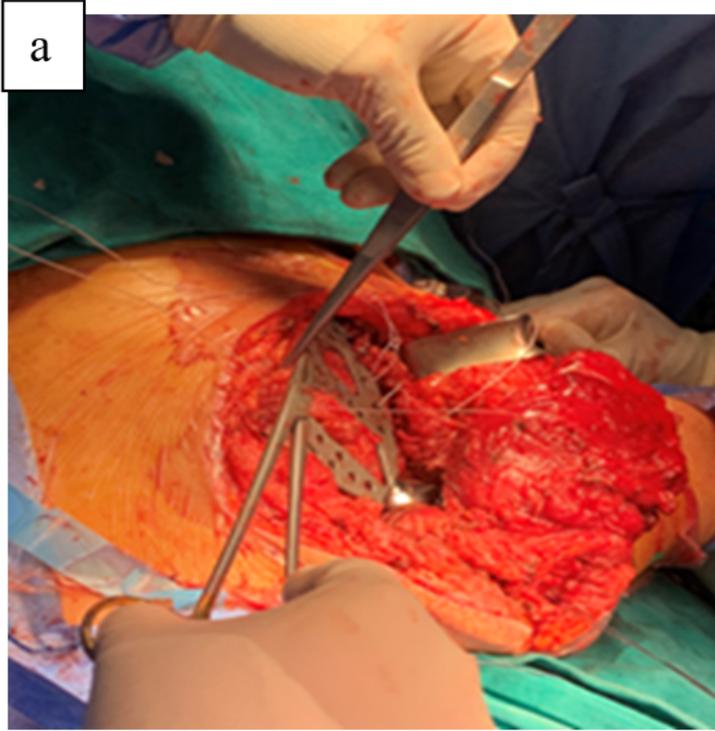


Figure 6

Stitching to restore anatomical attachment points of soft tissues.

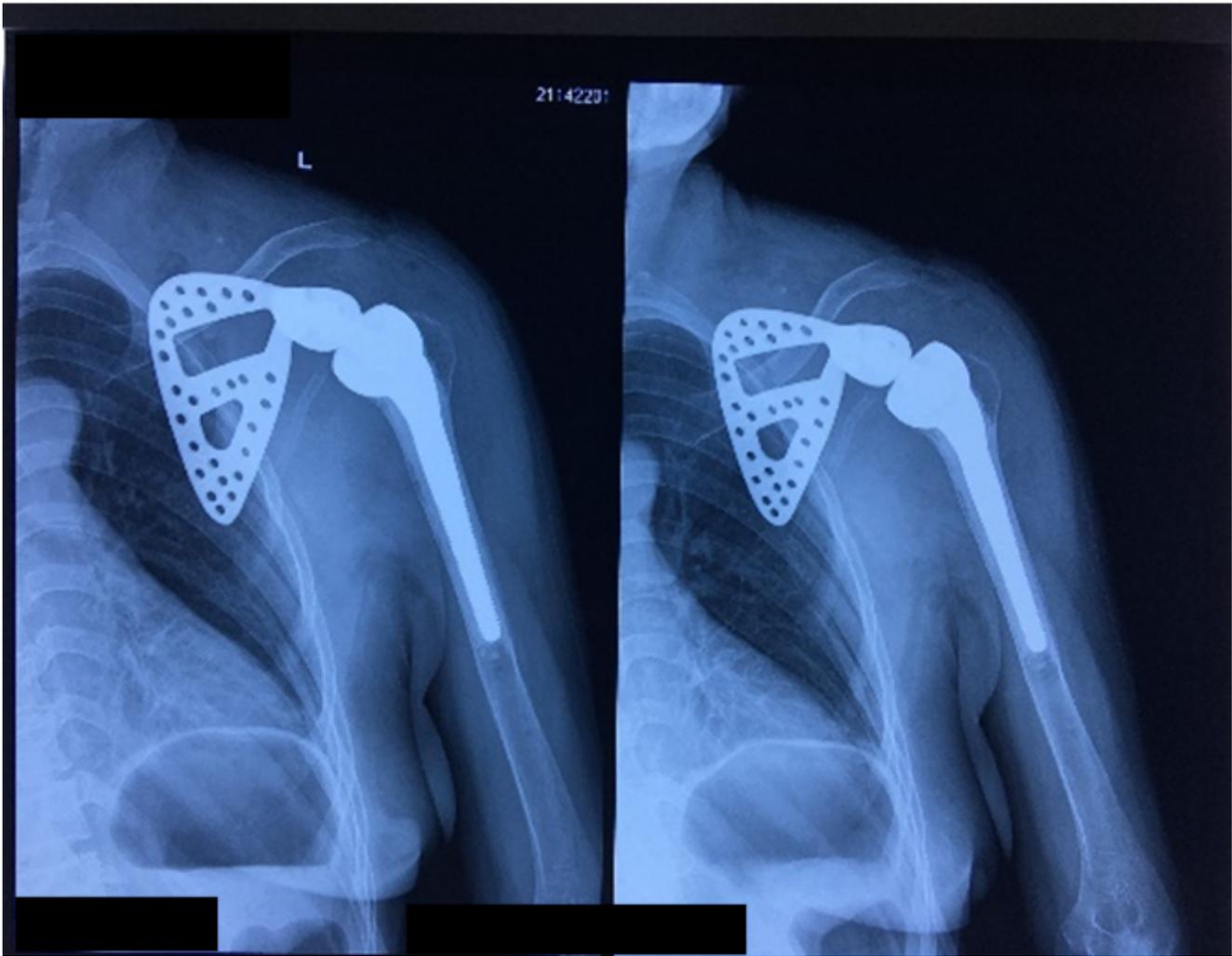


Figure 7

Postoperative X-Ray film

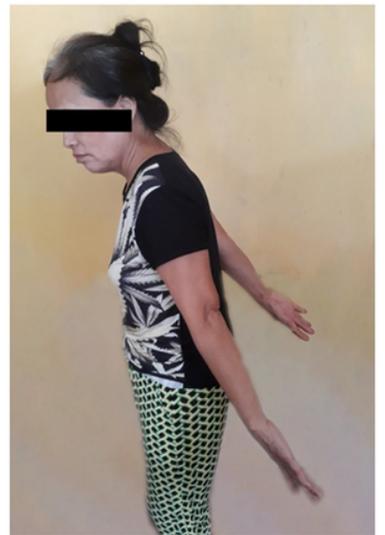


Figure 8

Clinical result of the patient's shoulder function after 6 months

Supplementary Files

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