

# Inverse shoulder tumor megaprosthesis after large bone resection in massive metastasis of the proximal humerus

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**Abstract.** *Background and aim:* Bone metastases are a significant prognostic factor in the history of cancer and often involve pain and a great impairment of the quality of life. The complete resection of tumor tissue is increasingly performed in patients with solitary bone metastases, to improve the survival of patients and their functional outcomes *Methods:* We report the case of a 65-year-old man with a painful, massive, highly perfused osteolytic lesion localized at the proximal third of the humerus, associated with extensive lesions of the rotator cuff tendons was diagnosed with keratoblastic squamous cell lung cancer metastasis. *Results:* The patient underwent shoulder and proximal humerus reconstruction with inverse tumor megaprosthesis after embolization of the lesion. At 3 and 6 months FU, a nearly complete resolution of painful symptoms, a significant improvement in functional skills, and better execution of most of the activities of daily life have been reported. *Conclusions:* Accordingly with the literature the inverse shoulder megaprosthesis seems able to restore a satisfactory function and the silver-coated modular tumor system appears as a safe and viable treatment option in metastases tumor of proximal humerus. ([www.actabiomedica.it](http://www.actabiomedica.it))

**Key words:** Bone metastasis, limb reconstruction, shoulder megaprosthesis, inverse proximal humerus replacement, silver-coated

## Introduction

Bone metastases (BM) are a significant prognostic factor in the history of cancer and often involve pain and a great impairment of the quality of life (QoL).

The complete resection with curative intent of tumor tissue involving bearing bones and enclosed soft tissues is increasingly performed in patients with solitary BM, to improve the survival of patients and their functional outcomes (1, 2).

The proximal humerus is the third most common site for primary bone tumors and secondary malignancies like BM of carcinoma (3).

Nowadays, limb reconstruction using modular tumor megaendoprostheses is a standard procedure in patients with malignant tumors (4). When the tumor

involves the proximal humerus and soft tissues of the rotator cuff, with functional impairment of the upper limb, the reconstruction reverse shoulder is indicated. In this scenario, the medialized and semi-constrained artificial joint restores stability and motility, achieving better functional results in comparison to anatomical shaped prostheses (5)(6).

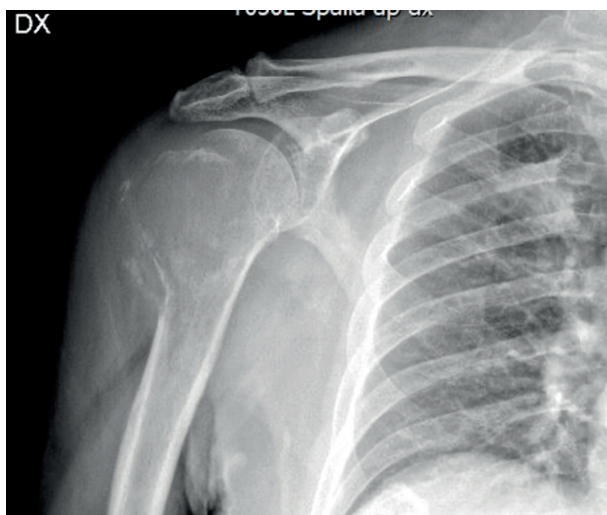
The Modular Universal Tumour and Replacement System (MUTARS, Implantcast Ltd., Buxtehude, Germany) inverse proximal humerus replacement (IPHR) is indicated in young patients in whom the axillary nerve can be preserved, and little muscle resection is necessary. In young patients with high demands on shoulder function a low degree of wear on the polyethylene is desirable. A special type of polyethylene with shock-absorbing properties has been developed to

minimize polyethylene wear in the MUTARS IPHR (5). The aim of our case-report is to share some surgical considerations and the clinical outcomes in IPHR due to a massive metastasis of the proximal humerus.

## Patient and methods

A 65-year-old right-handed man attending to our ward for a few months history of right shoulder pain and dysfunction, which began abruptly when lifting a heavy load. At the initial evaluation, a muscular hypotrophy, and a limitation in all the planes of ROM were observed. Moreover, a severe pain on palpation was evocable in the right shoulder. The numeric pain rating scale (NPRS) score was 8, the visual analogue scale (VAS) was 8, and the Constant-Murley Score was 35 (7).

Antero-posterior radiograph of the right shoulder (Fig. 1) showed a massive osteolytic lesion at the proximal third of the humerus, with bone rarefaction at the acromio-clavicular and scapula-humeral joints. To better investigate the lesion, a 3D CT scan was performed (Fig. 2) (8). To better understand the nature of that tissue, a total body CT with contrast, a positron emission tomography (PET), and a bone biopsy were performed. A keratoblastic squamous cell lung cancer metastasis with extensive areas of necrosis was diagnosed. To complete the pre-operative assessment an



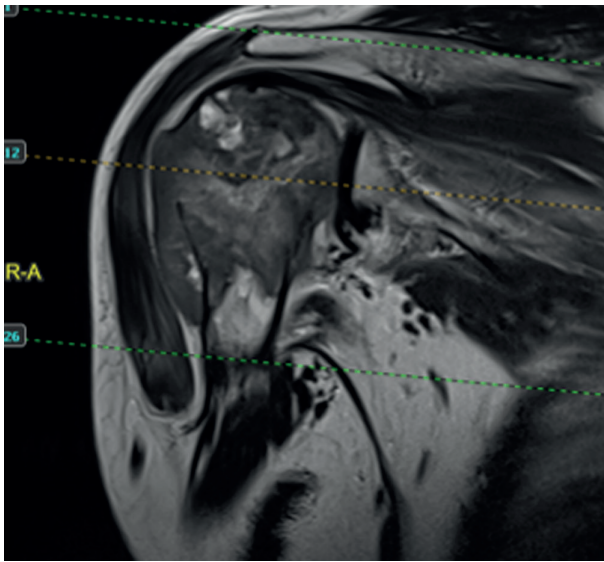
**Figure 1.** massive osteolytic lesion at the proximal third of the humerus



**Figure 2.** CT scan confirmed a massive lytic lesion (75, 90, 100mm antero-posterior, lateral and cranio-caudal diameters, respectively) which led to an obvious disruption of the bone architecture, with the proximal humerus completely replaced by pathological tissue.

MRI (Fig. 3). with intravenous contrast agent was performed to assess functional tumor information, such as vascular permeability, vessel density and perfusion. The highly vascularized tissue in the metaphyseal area was embolized with selective occlusion of the posterior circumflex and subscapular arteries.

The extent of the lesion was measured preoperatively on radiographs and MR images. The measurements were repeated using the most proximal part of the humerus and the surgical neck as a reference point. The double assessment was performed because of the risk of displacement of the head during exposure (which would have made the measurement from the humeral head unreliable) and because of the more approximate anatomical rather than radiographic identification of the surgical neck. A 2 cm tumor-free margins osteotomy was planned. Due to the planned extensive resection which should have involved the muscular insertions of the rotator cuff, it would not have been possible to reconstruct the joint



**Figure 3.** MRI showed the extent of the damage to surrounding soft tissues, with edema and extensive lesions to the rotator cuff tendons.

capsule and with it the passive and active stabilizers of the joint.

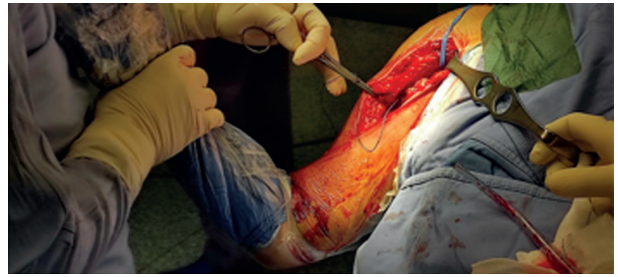
An inverse shoulder prosthesis was chosen, because its nonanatomic design frees the implant from the dependence on dynamic and capsular stabilizers for its function (9). We opted to implant a modular prosthesis because modularity allows to modify the planned resection in case of discrepancy between preoperative planning and intraoperative findings, and can also help to manage possible intraoperative complications (such as a diaphysis fracture). A MUTARS IPHR was implanted through a deltopectoral approach.

All procedures were performed following written informed patient consent and in accordance with the ethical standards of the institutional and/or national research committee and the 1964 Declaration of Helsinki and its subsequent amendments or comparable ethical standards.

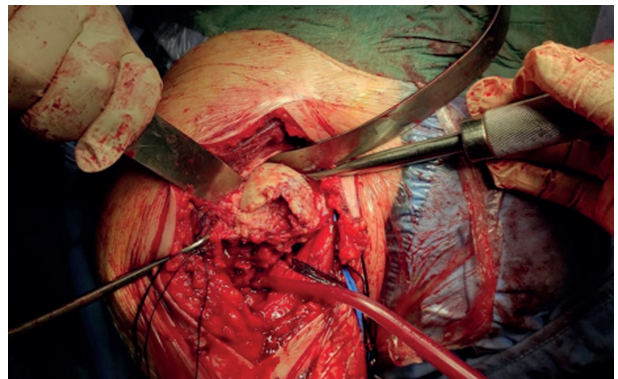
### *Surgical procedure*

The patient was placed in the beach-chair position under general anesthesia. A distally extended deltopectoral approach was performed (Fig. 4).

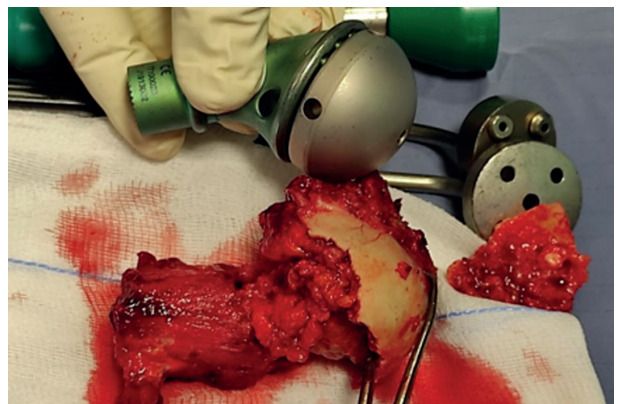
The proximal humeral epiphysis appeared fragmented and filled by tumoral tissue; the rotator cuff



**Figure 4.** Extended deltopectoral approach; once isolated the circumflex nerve, the pectoralis major, the latissimus dorsi and the distal part of the deltoid muscle, were released from their insertion.



**Figure 5.** The tumor with proximal humerus was resected



**Figure 6.** To obtain a safe surgical zone, the resection length was 12 cm, 2 cm distal to the tumor margin based on magnetic resonance and intraoperative fluoroscopic images.

was degenerated but not involved in the tumor process. The preoperative assessment of lesion extent was confirmed with direct visual measurement and intraoperative arthroscopic check. The large destructive

lesion of the humeral head required the intraoperative measurement with reference to the surgical neck. A radical excision of the pathological bone and soft tissues with free margins was performed (Figs. 5, 6). The histological study confirmed large tumor free margins (> 2 cm) from the distal osteotomy.

Due to previous embolization intraoperative bleeding was minimal and the patient did not need blood transfusions in the postoperative period.

The resection of proximal humerus facilitated the exposure of the glenoid and the positioning of the glenoid component.

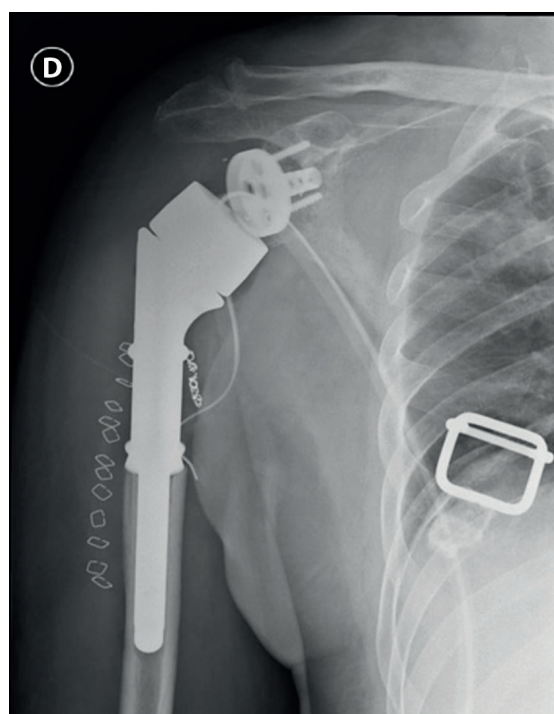
The length of the resected portion of the humerus was used to determine the length of the prosthesis, the augments of the prosthesis (Fig. 7) were used to achieve proper length and tension. Capsular reconstruction was not necessary; due to the wide resection pectoralis major and latissimus dorsi muscles were reinserted just distal to their anatomical insertion, with transosseous sutures on the most proximal part of the residual humeral shaft. The complete articular excursion and stability have been verified. One drain tube was placed.

### *Rehabilitation program*

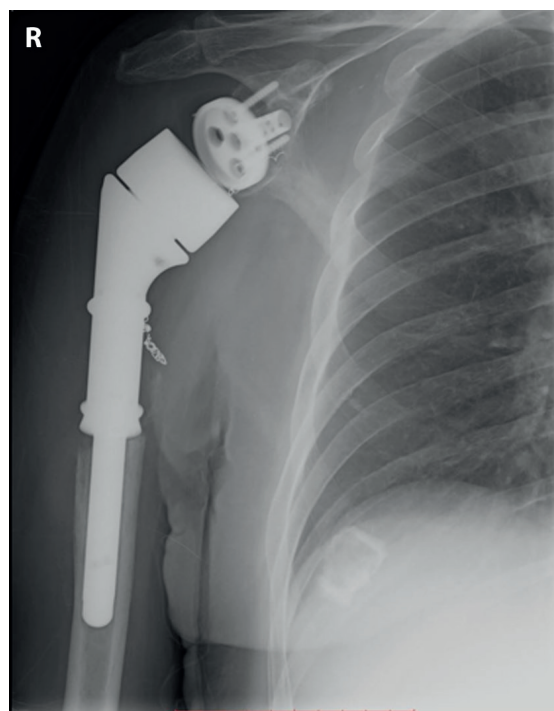
An arm sling with wedge in 15° abduction was positioned after surgery and maintained for the next three weeks. 24 hours after surgery, the drain tube was removed, and the isometric and low grades active assisted mobilization of the shoulder were allowed. The hospitalization lasted 72 h and a home-rehabilitation was prescribed. The rehabilitation program concerned: a) maintenance of the ROM of the elbow, wrist and hand; b) increase of the degrees of internal and external rotation and of the entire ROM on all the planes of the shoulder; c) proprioception exercises.

### **Results**

The last follow-up considered in the study was at 6 months. At 3 and 6 months clinical and radiographic FU (Fig. 8) significant improvement in painful symptoms and functional skills, and better execution of most of the activities of daily life have been reported.



**Figure 7.** A cemented humeral stems was used (TiAlV stem, TiAlV silver coated augments and head, TiAlV inverse humeral cup with titanium-nitride ceramic coating).



**Figure 8.** The x rays showed the good and stable positioning of the implant at 6 months FU

At 3 months the clinical scores were VAS 3, NPRS 3/10, ROM was increased on all planes during active and passive mobilization (active abduction 115°, active flexion 120°). The absolute value of the Constant score at 6 months was 58 points (a “fair” result), in detail the patient reported: the maximum score in the pain sub-scale (15 points), 10 points (of max 20) for activities of daily living, 22 (max 40) in range of motion sub-scale, and 11 points in power subscale (max 25); the difference between the normal and abnormal sides was of 18 points which represents a good result (10). Moreover, the patient reported a subjective benefit well above his expectations.

Partial recovery of deltoid, pectoral and bicipital muscular trophism was observed. Moreover, an improvement of the mood and psychological picture was reported.

## Discussion

Proximal humerus metastases are a challenge to both the surgeon and the patient alike. The late detection of these tumors coupled with few operable cases makes it a controversial topic with differing views across schools of thought. Patient survival depends on the grade and stage of the primary tumor and the tumor response to chemotherapy. A careful evaluation of the cost-benefit ratio is mandatory, especially in the delicate field of palliative care. Indeed before proceeding with aggressive surgical treatments, we carry out a collegial consultation with the oncologist and discuss treatment options with the patient and his relatives, who in our society still play a fundamental role in ensuring the best possible quality of life for these patients. Moreover, palliative care is of paramount ethical importance and is a legal obligation in Italy (Italian law 38/2010) (11).

The patient characteristics, the implant design, tumour-related complications and functional outcomes, influence the choice between different forms of reconstruction (12). In the reported case before surgery, the response to primary tumor therapy was encouraging, and shoulder pain, defined by the patient as an “agony”, was unresponsive to analgesic therapy.

The surgeons decided, taking into account the perspectives made by the oncologist, and the patient’s expectations, to reconsider the prognosis and consequently the indications, having very clear the ethical value of allowing a cancer patient to live with dignity the time left to live.

Basic requirements for preoperative planning are the CT scan and the MRI. We decided to use the combination of these data for the pre-operative planning. It allowed to planning the resection margins with respect to joint axes and spatial implant fixation (13,14). The postoperative radiographic result shown in figure n 7 does not deviate from the preoperative planning, but the modularity of the chosen implant ensured, if necessary, possible changes in the intraoperative route.

The prosthetic surgery aims to offer to the patient a good QoL with a definitive treatment, as the advances in the management of cancer prolong the survival of these patients (15,16).

In our case there were several reasons to choose this kind of prosthesis over other options: because of the large size and high grade of the tumor, *en bloc* excision had to be performed near the shoulder joint. Therefore, replacement of an articular surface would be insufficient to reconstruct the entire anatomical region. IPHR has several characteristics that make the implant intrinsically stable: a) a semiconstrained design due to deeper and more containing concavity (which in the inverse prosthesis is located on the humeral side and not on the glenoid side); b) a high congruence of the articular surfaces (which have the same radius of curvature); a fixed, medialized joint’s center of rotation that converts shear forces into compressive forces. This high degree of intrinsic stability frees the reverse total shoulder prosthesis from dependence on rotator cuff active stabilization and capsular reconstruction (9). On the one hand the wide resection increases the risk of bleeding and vascular-nerve injuries and complicates muscle-tendon reconstruction. On the other hand, a more distal humerus osteotomy increases glenoid exposure and simplifies the implantation of the prosthetic components as long as sufficient residual bone is maintained. Intraoperative bleeding after embolization of the hypervascular bone metastasis was minimal in accordance with the results of the study by Pazonis et al. (17)

Despite many studies describe the use of MUTARS tumor endoprotheses in the lower limb (4, 18, 19) not so many studies describe this prosthesis design for the proximal humerus (4, 20).

One hundred patients with endoprosthetic replacement of the proximal humerus were followed up by Kumar et al. (21) The study reported mean shoulder flexion of 55° and abduction of 45° in combination with a mean Musculoskeletal Tumor Society Score (MSTS) of 79%. The overall survival at 10 years was 42%, and the survival of the implant without further surgery for mechanical failure was 86.5% at 20 years. At 6 month FU, the patient of the case report, performed a flexion of 120° and abduction of 115°, showing a satisfying ROM. The absolute Constant score was fair considering the operated side but resulted good when comparing the two limbs. Since the subjective evaluation of shoulder function, especially in the elderly patients, often deviate from the objective score result, a relativization of the absolute score is possible by a comparison with age- and gender-specific norms or the contralateral side (22). Despite the fair score on the constant scale, the patient was immediately extremely satisfied with the result obtained considering the starting point. Preoperatively, the patient reported unbearable pain and complete functional impotence with a pseudo-paralytic limb due to pain, and an immediate (already assessed 48 hours after surgery) recovery of active mobility with a pain-free ROM of about 20 degrees in flexion- extension and 15 degrees of active abduction. Already at three months of follow-up the patient was fully satisfied with the result, reporting minimal pain, and a gradually recovered ability to carry out daily life activities and some hobby activities.

Different rates of complications and revision have been reported in literature (21,23,24).

Moreover, better clinical outcomes and less number of complications with a longer follow-up were found in young patients using IPHR (25). To mention, wear on the IPHR is probably associated with loosening of the stem (26) but to date there have been no large studies examining retrieved glenoid-components from reversed shoulder prostheses. An *in vitro* tribological comparison between MUTARS IPHR and conventional anatomical shoulder arthroplasty (5) showed a polyethylene wear comparable between

the two models. The similar rate of polyethylene wear is considered, in that study, as an indicator of the good functioning of MUTARS system. Despite a lower wear rate than the wear reported by Terrier et al. (27) the glenoid component of the MUTARS IPHR has wear properties comparable to others reversed shoulder prostheses. The wear of the prosthesis is, in the first instance, correlated to the level of use: a comparable wear indirectly demonstrates a comparable functional result between anatomic prosthesis, inverse prosthesis and oncological reverse megaprosthesis. This data suggests the possibility of a good functional recovery after a shoulder replacement with MUTARS IPHR even in young patients with high demands.

Infections represent the most severe complications of tumour arthroplastic treatments (28). Indeed the long surgery time, the large incisions, and the immunosuppression due to chemotherapy and radiotherapy as well as the increasing resistance of the bacteria against antibiotic drugs are all risk factor for early or late infectious complications. The anti-infective effect of silver ions has been known for centuries (29, 30).

The silver coating compared with standard titan megaprosthesis, shows a protective role, especially in the first 6 months after surgery (29).

In reverse shoulder arthroplasty, intact deltoid function leads to better functional outcomes (31). On the other hand a good clinical shoulder function, requires not only active elevation but also control of active external rotation. (32) For this reason the reconstruction of the capsule and refixation of the muscles can contribute to a better functional result especially in the anatomical prosthesis where their action is essential for proper function and stability. On the one hand, the integrity of the rotator cuff is not essential to guarantee the stability and an acceptable function of an inverse prosthesis, on the other hand, in the absence of a rotator cuff, the action of the deltoid determines an increase in the shear forces between the glenosphere and the bone. For this reason it is important to reconstruct (where it had been detached) the insertions of the latissimus dorsi, which can partially counteract the shear forces exerted by

the deltoid, reducing the risk of early mobilization of the glenosphere (9).

The trevira or dracon tube allows for better anchoring of the muscle component in order to achieve an anatomical reinsertion of remaining soft tissue following resection (33, 34). On one hand there is no statistically significant increased risk of infection by using trevira tube even among immunosuppressed patients, on the other hand, it can be a risk factor for cranial migration or subluxation of the prosthesis. (35). In agreement with the above in this case report we did not observe any early complications and the patient achieves a satisfying clinical outcome at short FU.

## Conclusions

Accordingly with the literature our clinical results suggest that the silver-coated MUTARS IPHR represents a safe and viable treatment option in metastases or primary malign tumor of proximal humerus with acceptable functional outcome. A mid to long-term follow-up will be essential for a more comprehensive evaluation. A careful cost-benefit analysis should be always well considered, but especially in the delicate field of palliative care we still give a lot of weight to the anonymous quote: “You can’t give your life more time, so give the time you have left more life”.

**Conflict of Interest:** Each author declares that he or she has no commercial associations (e.g. consultancies, stock ownership, equity interest, patent/licensing arrangement etc.) that might pose a conflict of interest in connection with the submitted article

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