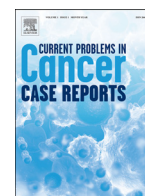


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## Forequarter amputation for local recurrence of sarcoma after previous amputation through the shoulder in a female patient – A case report

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### ABSTRACT

Forequarter amputation is a very demolitive surgical procedure that affects the quality of life and it is performed when the tumour involves the proximal end of humerus and the shoulder. We describe here the case of a female patient with a recurrent dedifferentiated liposarcoma of the upper limb already treated with tumour asportation, isolated limb perfusion and upper limb amputation through the shoulder, who returned to our attention for a recurrence of the sarcoma in the stump. We then performed a forequarter amputation in a previous amputated limb. Since the patient underwent numerous surgical procedures, our last surgical approach was a quite challenging one. To our knowledge, it is the first described case of an interscapular-thoracic disarticulation after a previous amputation to the shoulder.

### Introduction

Sarcomas are rare malignancies of the soft or bone tissues. Soft-tissue sarcomas can arise both in the limbs/trunk and less frequently, in the retroperitoneum. Sarcomas of the limb can be treated with combination of chemotherapy, radiotherapy, and surgery, that remains a key aspect of the treatment of these diseases. Surgical approaches include also loco-regional treatment to achieve local control in terms of limb-sparing surgery, such as isolated limb infusion or isolated limb perfusion. Rarely, the extension of the tumour requires limb amputation. Forequarter amputation is a very mutilating surgical procedure that negatively affects the quality of life, and it is performed for malignancies involving the proximal end of humerus and the shoulder or for malignancies invading proximal nerves and vessels.

### Case report

We report the case of a 60-yo Caucasian female patient who came to our attention about 13 years ago with two nodular lesions (1 cm and 1.2 cm) of the third superior part of the right upper limb that were removed, with a histological diagnosis of a dedifferentiated liposarcoma. The margins status was negative, so she underwent a surgical radicalization of the lesions with a wound closure with skin graft conducted

by the Plastic Surgery Department. She then was treated by adjuvant radiotherapy (50 Gy in 25 fractions). Subsequent follow up was conducted with a magnetic resonance every 6 months and it was always negative but 3 years later, when it was found a local recurrence enveloping the neurovascular bundle of the arm, normally requiring an amputation. Instead, in our centre we performed an hyperthermic isolated limb perfusion with infusion of TNF- $\alpha$  and Melphalan, to try obtaining a limb-sparing surgery. We achieved a partial response, so the patient underwent to necrosectomy and subsequently a three-cycles Epirubicine-Ifosfamid adjuvant chemotherapy, that she concluded with no complications. 10 months later, she developed another recurrence in the site of surgery, so we proposed and performed an amputation through the shoulder. The subsequent follow up was negative, but about 4 years after the disarticulation she developed another recurrence in the site of surgical amputation. The recurrence was located inside the scapular glena with erosion of both the scapula itself and the clavicle anteriorly. We proposed and performed another surgical intervention of forequarter amputation.

After three years of follow-up, the patient is still disease-free.

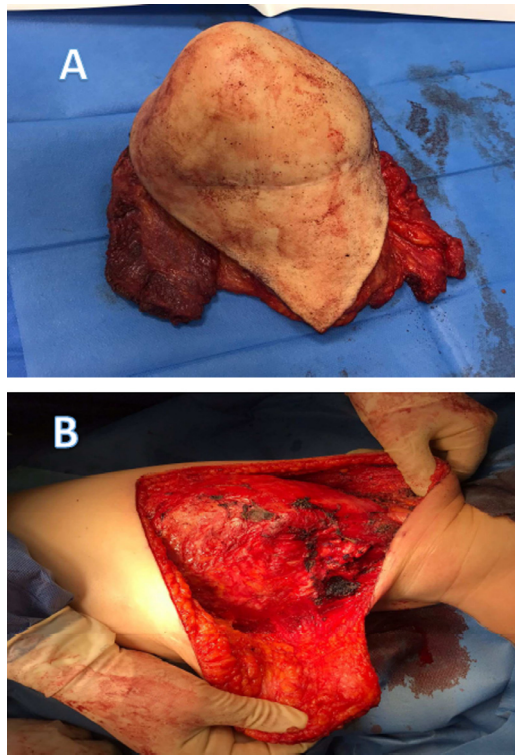
### Surgical technique

Since we had to deal with the stump of the upper limb, we had to perform a skin incision all along the stump itself in a racquet fashion, beginning at the sternoclavicular joint up to the external third of the

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**Fig. A-B.** (A) shoulder and sarcoma removed en bloc. (B) chest wall after forequarter amputation.

clavicle, prolonged anteriorly to about 10 cm below the pectoralis major muscle tendon, below the inferior angle of the scapula, and posteriorly on the external third of the scapula until it reconnected to the clavicle.

Posterior skin flap was retracted medially toward spine, so trapezius muscle was completely exposed. Section of the trapezius was performed through muscular portion in cephalic one-third and through aponeurosis in lower two-thirds. By reflecting trapezius muscle, elevator scapulae, minor and major rhomboid, and medial border of the scapula were exposed. Dorsal scapular nerve and vessels, and accessory nerve were also exposed. All these structures were sectioned, to separate the chest wall and the scapula, which was retracted laterally. At this point, the last remaining direct attachment of the scapula to the chest wall was the digitation of the serratus anterior, which were then sectioned. Carefully dropping the scapula laterally as far as possible, the subclavian vessels and the trunks of the brachial plexus were seen emerging between the scalenus medius and anterior muscles, and then ligated and sectioned. Retracting the anterior flap, the pectoralis major muscle was exposed, then detached from chest wall and retracted laterally. Pectoralis minor was previously sacrificed during isolated limb perfusion. Disarticulation of the clavicle from the sternum was then performed. Shoulder was then removed *en bloc* (Fig. A-B). Two drain was inserted beneath the flaps.

## Discussion

In sarcomas of the limb, the surgical approach is aimed to achieve local control of the disease with the lower mutilating outcome as possible. In the past, radical amputation was often proposed as a potentially curative treatment (Clark and Thomas, 2003a; Clark and Thomas, 2003b; Maduri and Akhondi, 2021; Amir et al., 1993; Marinescu et al., 2010; Qadir et al., 2014), but nowadays the progress of modern medicine led to multimodality treatments with the introduction of radiation therapy and chemotherapy and the modification of the surgical techniques, aimed to reduce the rates of amputation. So, a limb-sparing technique can be pro-

posed in well-confined and early lesions, while a first-step amputation is proposed when the lesion is in an advanced stage (typically in case of tumoral invasion of proximal nerves and vessels) or the symptoms caused by the mass are extremely invalidating (typically loss of functioning and scarcely controlled pain). Since the amputation itself does not have influence on the overall survival, in tertiary care centres patients are preferably treated with isolated limb perfusion (Rastrelli et al., 2016). Forequarter amputation is the most mutilating amputation regarding the upper limb, and it is proposed rarely, for it has a negative impact in quality of life, since prostheses are often poor in terms of aesthetics and function, and the patient itself often refuses this kind of aggressive surgical approach. In our case, the patient had no other systemic therapeutic options since she already underwent both chemotherapy and radiotherapy and the surgical intervention could not be avoided, since on one hand the lack of the arm made it impossible to perform an isolated limb perfusion and on the other hand the tumour was already spreading locally with infiltration of the scapula and the clavicle.

It is also true that these kinds of surgical procedures are characterized by an increased risk of complications, (typically wound infections or flap necrosis), so it must be carefully evaluated the risk-benefit ratio. Another important aspect of amputation is pain control. Before surgery, patients have often a poor pain control due to the mass, but after the surgery pain control could also be difficult for the raise of phantom limb pain. Our patient already partially suffered from this condition since she had undergone a previous limb amputation. In our Institute, we usually start treatment with pregabalin the night before surgery and we administer naropin locally into the nerves before cutting them.

Since this kind of procedures is often proposed in locally advanced or metastatic diseases for symptom relief and the few studies conducted are characterized by small groups of patient and extreme variability in the diagnosis, it is also difficult to estimate disease-free and overall-free survival. Parsons et al. (2012) described their experience regarding 40 major amputation (30 of which were forequarter amputation), concluding that in their group of patients the recurrence rates were extremely high (79% of recurrences either locally or a distant sites), but that in some cases the amputation led to prolonged survival, in absence of other available treatment. Puhaindran et al. (2012) described the survival results of 43 patients undergoing 45 major amputations (2 of them received a second more proximal amputation, described as “above the elbow”), with a 6-month cumulative incidence of recurrence of 22% and a 2-year OS less than 20%. Rickelt et al. (2009) described the results of 40 forequarter amputations for different malignancies performed in more than thirty years of activity in three Dutch centres, where OS in the sarcoma group at 1–2–5 years was 89%, 74% and 41%, respectively, while DFS at 5-years was 26%. Other studies have been evaluated DFS and OS in their set of patients, with varying results in terms of DFS and OS according to histology, intent to amputation, initial stage of diseases, thus making quite difficult to confront the different results.

Regarding the surgical procedures, in the past decades the forequarter amputation’s techniques have been notably modified since the first amputation (Tukiainen et al., 2020; Lesenský et al., 2017; Scaglioni et al., 2016), which it is believed to have place back in 1808 in a traumatic setting. Nowadays, the various well-established techniques are the one proposed by Berger (linear incision at the middle third of the clavicle to secure firstly vessel’s control), the one proposed by Kocher (anterior approach) and the one proposed by Littlewood (posterior approach). We had to deal with a quite modified anatomy of the shoulder due to the previous multiple surgical intervention. Since the patient had underwent isolated limb perfusion, we had to deal also with an augmented fibrosis of the vessels, so we wanted to have the vessels secured in safety. We decided to perform a combined approach resembling the one Ferrario et al. (2004) described in 2004. We started posteriorly, but the neurovascular bundle’s ligation was performed as the last step after completing the dissection anteriorly, in a position where we had more control of them. Other authors described the use of this modified approach as well (Amir et al., 1993; Dimas et al., 2007; Mansour and

Powell, 1978), justifying the convenience to ligate the bundle in direct vision.

To our knowledge, this is the first described case of interscapulothoracic amputation after previous amputation through the shoulder.

#### Patient consent statement

Written informed consent was obtained from the patient regarding the publication of anonymized material.

#### Declaration of Competing Interest

The authors declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

#### CRediT authorship contribution statement

**Claudia Di Prata:** Writing – original draft. **Nicolò Zilio:** Writing – original draft. **Paolo Del Fiore:** Writing – review & editing. **Jacques Gowon Souffo Sonkoue De Tamoki:** Writing – review & editing. **Simone Mocellin:** Writing – review & editing. **Marco Rastrelli:** Writing – review & editing.

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