

# Unplanned resection of a soft tissue sarcoma: clinical characteristics and impact on oncological and functional outcomes

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Received: 30 April 2014 / Accepted: 15 December 2014 / Published online: 23 January 2015  
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## Abstract

**Background** Over the past three decades, several studies have reported worse outcomes with unplanned resection for malignant soft tissue tumors. However, the impact of these studies on preventing unnecessary unplanned resection remains unknown.

**Methods** In a retrospective survey on the treatment of soft tissue sarcomas, we compared cases of unplanned resection with cases of planned resection in terms of the properties of unplanned resection and the oncological and functional outcomes. For the unplanned resection cases, an additional wide resection was performed.

**Results** Of 92 cases, unplanned resection was performed in 24 (26 %). Small or subcutaneous tumors were significantly more frequently subjected to unplanned resection. In 17 of 24 unplanned resection cases, residual tumors (70.8 %) were noted. Plastic surgery was more frequently needed for unplanned resection cases. There was no significant difference between the unplanned resection and control cases with regard to oncological outcome. However, as to local recurrence and overall survival, the events occurred only in the cases with residual tumors in the additional wide resection specimen in the unplanned resection group. There was no significant difference in functional evaluation, except for emotional acceptance, which had a better score in the unplanned resection group.

**Conclusions** Despite repeated cautions regarding unplanned resection in terms of its inadequate procedure which contradicts the principles of soft tissue sarcoma

treatment, unplanned resections are still frequently performed. Perhaps the small size and subcutaneous location of the sarcomas in the unplanned resection group did not evoke the probability of malignancy for the surgeons who initially managed them. Even though an additional wide resection was performed, a residual tumor would lead to a worse outcome. An effective awareness program to avoid unnecessary unplanned resections for soft tissue sarcoma should be considered.

## Introduction

The principle of treating malignant soft tissue includes secure evaluation of the lesion using magnetic resonance imaging (MRI) scans, accurate pathological diagnosis with an appropriate biopsy procedure, and the complete resection of the primary lesion with an adequate margin. However, sometimes orthopedic oncologists are required to treat cases in which physicians in other departments do not observe an indication for the standard procedure during the initial stage of treatment. The unplanned resection of a malignant soft tissue tumor is defined as excision without removing a margin of normal tissue covering the tumor, and without histologically diagnosing biopsy specimens [1, 2]. Such diagnostic failure is believed to be, at least in part, due to the rarity of a soft tissue sarcoma [3]. The authors' institution is a regional referral hospital with specialists who provide orthopedic oncology services. Although numerous studies have reported on the unplanned resection of sarcomas since the late 20th century [1, 2, 4–8], we still frequently manage unplanned resection cases. Many questions are raised regarding unplanned resections. First, has the incidence of unplanned resections decreased during the past two decades? In other words, did the

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recent enlightening studies have an impact on preventing unplanned resections? Second, what are the properties of the cases that undergo unplanned resection today? Finally, does an unplanned resection affect the oncological and functional outcomes of treatment for soft tissue sarcomas? Thus, we aimed to re-evaluate unplanned resection cases treated at our institution in order to assess the properties of cases with unplanned resection, as well as their oncological and functional outcomes.

## Materials and methods

We designed a retrospective uncontrolled study based on data obtained from medical records. The collection of clinical data and the publication of data are in accordance with the local guidelines for research ethics, and the institutional review board approved the study design. The inclusion criteria were the following: (1) soft tissue sarcoma treated between 2006 and 2012; (2) cases initially resected at our institution with standard oncological resection based on the surgical margin theory [9] or cases with unplanned resection at another institution followed by additional wide resection at our hospital; (3) follow-up for at least 12 months from the surgery at our institution or until death; and (4) the availability of sufficient clinical data for analysis. According to previous reports, unplanned resection of soft tissue sarcoma is defined as excision without regard for the necessity to remove a margin of normal tissue covering the tumor, and without histological diagnosis by biopsy [1, 2]. Cases diagnosed with well-differentiated liposarcoma were excluded because of the good overall survival in these patients.

We enrolled 92 patients (46 men, 46 women) with a mean age at diagnosis of 59.1 years. Sixty cases had malignant tissue in the lower extremity, 15 in the trunk, and 17 in the upper extremity. Diagnoses were undifferentiated pleomorphic sarcomas in 27 cases, liposarcomas in 17 cases, leiomyosarcomas in 8 cases, myxofibrosarcomas in 7 cases, malignant peripheral nerve sheath tumors in 6 cases, synovial sarcomas in 4 cases, dermatofibrosarcomas protuberance in 4 cases, and other types in 19 cases.

For the unplanned resected cases, we routinely applied an additional wide resection. The surgical margin for additional wide resection was determined based on MRI imaging before the initial unplanned resection (if available), on the post-operative MRI images performed at our institution showing the operation scar of the initial surgery and any inflammatory change induced by the unplanned resection, and on initial operation scar by unplanned resection on physical examination, in order to resect the operation scar, the residual tumor, trace of inflammation, which might show the trace of residual tumor, trace of operation scar, or

inflammation and hemorrhage by initial unplanned resection. Metastasis was routinely surveyed by chest and abdomen computed tomography (CT) scans at presentation followed by 3 and 6 months of follow-up until 2 and 5 years after the initial surgery, respectively. The tumor grade was determined based on the French Federation of Cancer Center (FNCLCC) Grades. Cases with grades 2 and 3 were classified as high grade [10]. Among the high-grade cases, patients who were between 20–70 years and had a tumor >5 cm in diameter were subjected to adriamycin- and ifosfamide-based post-operative systemic chemotherapy based on the Japan Clinical Oncology Group's 0304 regimen [11].

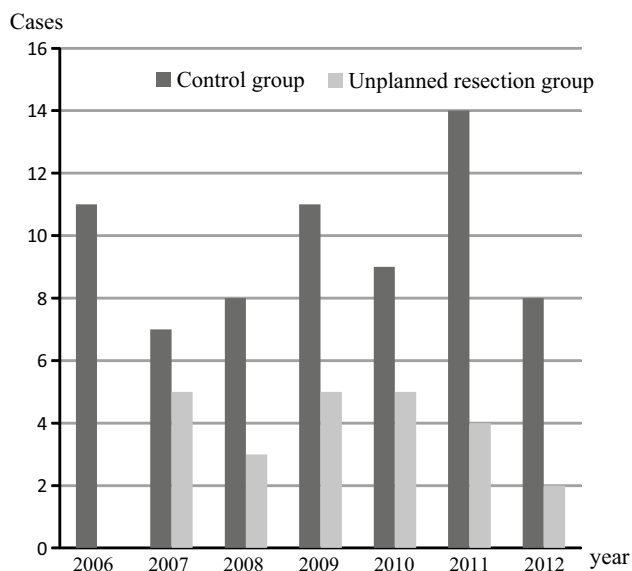
We assessed the annual incidence of unplanned resected cases and the performance of pre-operative MRI in unplanned resection cases, and we surveyed the departments in which unplanned resections were performed. The surgical margin and the incidence of residual tumor in the specimens from the additional wide resections were investigated. The patients' characteristics such as age at diagnosis and sex, information on the tumor such as site, location (subcutaneous vs deep), tumor volume, metastasis at presentation, pathological grade, and properties of the treatment modalities [e.g., application of systemic chemotherapy, needs for reconstruction surgery, intraoperative blood loss, duration of operation, volume of blood transfusion, incidence of surgical site infection (SSI), surgical margin, and incidence of amputation] were recorded and compared between the initial wide resection group and unplanned resection group. Tumor volume was calculated based on three-dimensional MRI scan data. The definition of SSI was based on the Center for Disease Control and Prevention guidelines [12].

We assessed the impact of unplanned resection on the oncological and functional outcomes. Additionally, the impact of the residual tumor on oncological outcomes in the unplanned resection group was analyzed. For oncological outcome, the endpoints in this study were local recurrence-free survival, metastasis-free survival, and overall survival. Based on the method by Enneking et al. [13, 14], functional analysis was performed.

We used the Mann–Whitney *U* test, Fisher's exact test, Kaplan–Meier analysis, and the log-rank test for data analysis. Values of *P* less than 0.05 were considered to denote significance.

## Results

Among the 92 cases, unplanned resection was performed in 24 cases (26 %). The annual incidence of unplanned resection was 0–42 %, and there was no trend in the incidence of unplanned resection during the past 7 years (Fig. 1). Of



**Fig. 1** Annual incidence of unplanned resection cases from 2006 to 2012

the 24 cases, pre-operative MRI was performed in only 15 (62.5 %). The unplanned resections were performed by general orthopedic surgeons in 10 cases, by plastic surgeons in 8, by general surgeons in 4, by a dermatologist in 1, and by a gynecologist in 1. Pathological examination of specimens for which additional wide resection was performed revealed that 17 of 24 unplanned resection cases had a residual tumor (70.8 %). Of 24 cases, 20 were evaluated as wide while the rest were considered to have a marginal margin post-operatively. The application of radiotherapy was considered in cases with a marginal margin; however, these patients refused the treatment. At the final follow-up, no local recurrence was detected in these cases. In the present study, no radiotherapy was performed over the follow-up period.

The differences in the patients' characteristics, tumor properties, and properties of the treatment modality between the unplanned resection group and the control group are shown in Table 1. Among the factors analyzed, the significant clinical characteristics in the unplanned resection group were subcutaneous location ( $P < 0.001$ ) and small tumor volume ( $P < 0.001$ ). In 19 subcutaneous cases with unplanned resection, the diagnoses included undifferentiated pleomorphic sarcomas in 5 cases, leiomyosarcomas in 4, myxofibrosarcomas in 4, and other types in 6. Compared with the control group, soft tissue or vessel reconstruction was more frequently performed in the unplanned resection group ( $P = 0.0006$ ). Intraoperative blood loss was significantly lower in the unplanned resection group than the control group ( $P = 0.007$ ). Thus, cases that were subjected to unplanned resection had a

considerably smaller tumor in the subcutaneous location, which would need more frequent and additional reconstruction surgery.

Oncological outcomes between the unplanned resection and control groups were assessed. As shown in Table 2, there was no significant difference between the two groups in local recurrence-free survival and overall survival rates, although some had a trend of better prognosis in the unplanned resection group in terms of metastasis-free survival. The residual tumor in the additional wide resection specimen did not show a significant effect on oncological outcomes in the unplanned resection cases (Table 3). However, as for local recurrence and survival rates, these events occurred only in the cases with residual tumors in the additional wide resection.

Functional evaluation between the unplanned resection and control groups was performed (Table 4). In most evaluation items, there was no significant difference in the evaluation points between the two groups. Unexpectedly, for emotional acceptance, patients with unplanned resection had a better evaluation point than the control patients ( $P = 0.049$ ). In addition, better outcomes were noted for pain in the unplanned resection group than the control group ( $P = 0.06$ ).

## Discussion

In the present study, we analyzed the clinical characteristics, oncological outcomes and functional outcomes of cases that had undergone unplanned resections. First, we determined the actual incidence of unplanned resection. There have been many reports on unplanned resection in the last three decades, suggesting that the problem in this field is universal. In 1985, Giuliano raised caution regarding the presence of gross residual tumor tissue in unplanned resection cases [1]; several reports in the 1990s also indicated the risk for residual tumors with such an inadequate procedure [2, 4]. If the incidence and the demerits of unplanned resection had been well recognized, the incidence of unplanned resection would have decreased. However, to date, several reports have found that the incidence of unplanned resection ranges from 33 to 53 % during a study period ranging from 1985 to 2008 [5–8]. In our data, we also did not find a decreasing trend in the incidence of unplanned resection during the period of 2006–2012. These data suggest that unplanned resections of soft tissue sarcomas are still frequently performed. Therefore, more widespread recognition of such an inadequate procedure is required.

Second, we analyzed the clinical characteristics of cases with unplanned resection. We found that small size and subcutaneous location were significantly present in the

**Table 1** Demographic data of the study subjects

	Control group	Unplanned resection group	<i>P</i>
Age	58.1 ± 19.4*	61.8 ± 17.1*	0.43
Sex			
Male	35	11	0.81
Female	33	13	
Site			
Upper extremity	11	6	0.62
Lower extremity	46	14	
Trunk	11	4	
Location			
Deep	50	5	<0.001
Subcutaneous	18	19	
Volume (cm <sup>3</sup> )	384 ± 603*	54 ± 64*	<0.001
Metastasis at presentation			
No	61	23	0.68
Yes	7	1	
Grade			
Low	13	7	0.39
High	55	17	
Systemic chemotherapy			
No	48	18	0.8
Yes	20	6	
Plastic/vessel reconstruction			
No	46	6	0.0006
Yes	22	18	
Intraoperative blood loss (mL)	359 ± 547*	89 ± 71*	0.007
Duration of operation (min)	279 ± 166*	256 ± 117*	0.88
Blood transfusion (mL)	264 ± 570*	217 ± 760*	0.16
Site of surgical infection			
No	63	23	0.99
Yes	5	1	
Surgical margin			
Wide	59	20	0.74
Marginal/intralesional	9	4	
Limb salvage			
Yes	63	21	0.43

\* Mean ± standard deviation

**Table 2** Oncological outcomes of the patients in the unplanned resection and control groups

	5-year survival	<i>P</i>
Local recurrence		0.95
Control	76.3 %	
Unplanned	79.8 %	
Metastasis		0.051
Control	63.0 %	
Unplanned	87.5 %	
Overall survival		0.31
Control	82.0 %	
Unplanned	87.8 %	

**Table 3** Oncological outcome of patients in the unplanned resection group who had and those who did not have residual tumors

	5-year survival	<i>P</i>
Local recurrence		0.19
Residual tumor (–)	100 %	
Residual tumor (+)	72.1 %	
Metastasis		0.85
Residual tumor (–)	85.7 %	
Residual tumor (+)	88.2 %	
Overall survival		0.44
Residual tumor (–)	100 %	
Residual tumor (+)	84.7 %	

**Table 4** Functional evaluation (mean  $\pm$  standard deviation) using the International Society of Limb Salvage scoring system in each group

Common	Pain	Function	Emotional acceptance
Control	4.6 $\pm$ 0.66	4.4 $\pm$ 1.17	4.5 $\pm$ 0.85
Unplanned	4.9 $\pm$ 0.25	4.8 $\pm$ 0.39	4.8 $\pm$ 0.49
<i>P</i>	0.06	0.27	0.049
Upper extremity	Hand position	Dexterity	Lifting ability
Control	4.9 $\pm$ 0.31	4.5 $\pm$ 0.84	4.7 $\pm$ 0.44
Unplanned	5.0 $\pm$ 0	5.0 $\pm$ 0	5.0 $\pm$ 0
<i>P</i>	0.48	0.21	0.22
Lower extremity	Support	Walking	Gait
Control	4.4 $\pm$ 1.3	4.3 $\pm$ 1.27	4.4 $\pm$ 1.24
Unplanned	4.8 $\pm$ 0.39	4.7 $\pm$ 0.49	4.7 $\pm$ 0.49
<i>P</i>	0.6	0.7	0.99
Total score			
Control	26.8 $\pm$ 5.6		
Unplanned	29.0 $\pm$ 1.8		
<i>P</i>	0.12		

unplanned resection cases compared to the control cases. In the present study, we found elevated levels of intraoperative blood loss in the control cases that were probably because of the large size and deep location of the tumors. In general, large size, acute progression, and deep location are a sign for malignancy in soft tissues, and such cases should be consulted by a specialist. Cases without these properties should be managed in a non-specialist hospital, where they would be treated by unplanned resections. In fact, our data and previous reports indicate that a significant small size and elevated incidence of subcutaneous location are tumor conditions for which unplanned resection is used [6, 8]. To reduce the incidence of unplanned resection cases, more awareness of the possibility of malignancy in soft tissue masses with a small size and subcutaneous location is needed. Our study revealed that MRI before unplanned resection was performed only in 62 % of the cases. Likewise, a previous study reported that MRI was performed only in 6 out of 38 cases [3]. In the absence of an MRI, it would definitely be difficult to define the safety margin for additional wide resection. If surgeons understood post-unplanned resection management theory (i.e., the surgical margin theory for preventing local recurrence), such a procedure would never be performed. Next, our data along with previous data suggest that unplanned resections were performed most frequently in the general orthopedic department, followed by the plastic and general departments. Venkatesan et al. [15] reported that general, plastic, orthopedic, and vascular surgeons and general practitioners most commonly performed unplanned resections. In Siegel et al.'s [16] study, patient referral after unplanned resection

was most commonly done by general surgeons without specialty training in oncology. These data suggest that unplanned resections are mainly performed by orthopedic surgeons who are not bone and soft tissue tumor specialists, followed by general and plastic surgeons.

For the management of cases with unplanned resection, additional wide resection, which aims to remove the scar tissue and suspicious soft tissue contaminated during the previous inadequate procedure and probable residual tumor tissue, has been recommended in many reports. Additional wide resection results in a high incidence (range from 31 to 74 %) of residual tumor in the specimens [1, 4, 7, 15, 17–19]. In the present study, the rate of residual tumor in the specimens was 71.0 %, supporting the significance of additional wide resection for cases in which unplanned resection is performed. In previous retrospective data on the oncological outcome of unplanned resected high-grade sarcoma treated with or without additional wide resection, additional wide resection significantly resulted in better oncological outcomes, including local recurrence-free survival, metastasis-free survival, and overall survival rates [20].

Our data and those by other authors showed the considerably high rate of soft tissue reconstruction as the management modality in plastic surgery departments [6, 21]. The use of this reconstruction may be because of the widespread contamination reported in the previous surgery, as well as a significantly high rate of unplanned resection performed subcutaneously. In other words, even if the unplanned resection was not performed in such a case, reconstructive surgery was still needed although the area requiring reconstruction was smaller than in unplanned resection cases.

Finally, we analyzed the impact of unplanned resection on oncological and functional outcomes. Unplanned resection cases show better prognosis when tumors have a small size and subcutaneous location in soft tissue sarcoma staging; such cases also show considerably good oncological outcomes. An early study by Lewis et al. [22] suggested that patients with soft tissue sarcomas who undergo additional wide resection after unplanned resection show improved survival rates compared with those who undergo one operation, regardless of patients categorized by the American Joint Committee on Cancer stages. Many authors suggest that unplanned resection is not a significant risk for a worse oncological outcome in terms of local recurrence, metastasis, and overall survival rates [6, 8]. Conversely, several reports suggest worse oncological outcome in terms of local recurrence and metastasis [7, 21]. As for oncological outcome, our data suggest that additional wide resection is not a worse prognostic factor for oncological outcome. We believe that such controversial results are expected because of the probable bias in various aspects, including variations in radiological evaluation after unplanned resection, the definitions of margins for additional wide resection, the rates of residual tumor occurrence in specimens in which additional wide resection is performed, the application of radiotherapy and chemotherapy, the presence of invasive tumors, and the inclusion criteria of the cases (the study by Lewis included only extremity cases but not trunk cases [22]). However, many studies commonly regard residual tumors, in which additional wide resection is performed, as a significant risk factor for worse oncological outcomes [4, 8, 17, 21]. Unexpectedly, few studies have reported on the functional results of unplanned resection cases, and this may be responsible for the limited difference in the results between planned and unplanned resection cases [6], which supports our present data.

Upon reviewing the present data and the results of previous reports, we emphasize that even though unplanned resection itself may not set a striking risk factor for worse oncological outcomes, and that additional wide resection was considered a useful procedure in controlling unplanned resection cases, unplanned resection should be avoided for several reasons: the definite risk for microscopic residual tumors in unplanned resection cases resulting in worse prognosis and worse oncological outcomes; difficulty in defining the margin in additional wide resections [4]; and the inconvenience for patients having to undergo more operations, with subsequent further expenses [23]. We again emphasize the need for widespread awareness for the proper management of soft tissue palpable masses.

To reduce the incidence of unplanned resections, analysis of the actual cause of such inadequate procedures is strongly needed. Perhaps the small size and subcutaneous location of the sarcomas in the unplanned resection

group did not indicate the probability of malignancy for surgeons who performed the initial management. The lack of recognition of malignancy in soft tissue tumors, inadequate knowledge of the initial management of soft tissue sarcoma, and misdiagnoses in studies by radiologists can be considered as underlying factors. However, the actual background of such procedures in each case could not be analyzed in the current study. Therefore, a prospective study on the status of malignancy recognition and the basic knowledge of soft tissue malignancy in a non-referral hospital and in departments including general surgery, plastic surgery and dermatology is needed in the future. In addition, the considerably small sample size, lack of analysis on the impact of variation in the duration from the initial unplanned resection to the additional wide resection on oncological outcomes, and lack of analysis on the quality of life for each group were limitations of the present study.

We analyzed the incidence, clinical characteristics, and oncological and functional results of unplanned resection on malignant soft tissue tumors. The incidence of unplanned resection has not decreased over the past several years. Small tumors located subcutaneously are significantly and frequently subjected to unplanned resection. Unplanned resection followed by additional wide resection results in a non-significant impact on the oncological and functional outcomes.

**Acknowledgments** This study was supported in part by the Health and Labor Sciences Research Expenses for Commission, and the Applied Research for Innovative Treatment of Cancer from the Ministry of Health, Labor and Welfare (H26-084).

**Conflict of interest** The authors have no conflict of interest to declare and no funding sources that might generate a conflict of interest.

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